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Exploration of differences in types of sleep disturbance and severity of sleep problems between individuals with Cri du Chat syndrome, Down's syndrome, and Jacobsen syndrome: A case control study

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ABSTRACT

The prevalence of sleep problems in individuals with intellectual disability (ID) seems to vary between genetic syndromes associated with ID. Different types of sleep disturbances may indicate underlying causes of sleep problems and these types of sleep disturbances may vary between different genetic syndromes. We examined and compared five types of sleep disturbance as well as severity of sleep problems in individuals with Cri du Chat syndrome (CDC), Down's syndrome (DS), Jacobsen syndrome (JS), and individuals with non-specific ID (NS). We used Simonds and Parraga's Sleep Questionnaire (1982) to assess prevalence of types of sleep disturbance and to explore differences in types of sleep disturbance and severity of sleep problems between the four diagnostic groups. In each group, mean scores for Snoring were significantly higher than those for Sleep apnea and Snoring was the most prevalent type of sleep disturbance in CDC, DS, and JS. The mean score on Complaints related to sleep was remarkably high in the JS group. There were no differences in severity of sleep problems between groups. These findings suggest that snoring is an important underlying cause of sleep problems in individuals with CDS, DS, and JS.

1. Introduction

The prevalence of sleep problems in individuals with intellectual disability (ID) seems to vary between genetic syndromes associated with ID (Didden & Sigafoos, 2001; Stores & Wiggs, 2001). For example, of children and adults with Angelman syndrome 37% have severe night-waking problems and 2% have severe settling problems (Didden, Korzilius, Smits, & Curfs, 2004). Of children with Down syndrome 32% have severe night-waking problems and 20% have severe settling problems (Stores, Stores, & Buckley, 1996). These prevalence rates differ in some ways from prevalence rates found in children with mixed or heterogeneous ID (i.e. children having different etiologies or no clear etiology for their ID). Of children with mixed

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or heterogeneous ID 10–26% have severe night-waking problems, 4–26% have severe settling problems (Didden, Korzilius, Van Aperlo, Van Overloop, & De Vries, 2002; Wiggs & Stores, 1996).

During the last decade, a growing number of studies have targeted sleep problems in individuals with genetic syndromes. In order to assess whether certain sleep problems are characteristic features of a genetic syndrome, using one or more control groups of individuals with mixed, heterogeneous or non-specific ID (NS) is strongly recommended (Hodapp & Dykens, 2001). Few studies assessing sleep problems in genetic syndromes have used appropriate control groups, i.e. in Cornelia de Lange syndrome (Hall, Arron, Sloneem, & Oliver, 2008), Cri du Chat syndrome (Maas et al., 2009), Down's syndrome (Cotton & Richdale, 2006; Stores et al., 1996), Tuberous Sclerosis (Hunt & Stores, 1994), and Prader-Willi syndrome (Cotton & Richdale, 2006).

Furthermore, different types of sleep disturbances may indicate different underlying causes of sleep problems (Wiggs & Stores, 2004). These types of sleep disturbances may vary between genetic syndromes (Cotton & Richdale, 2006). For example, obstructive sleep apnea syndrome is commonly observed in children with Down's syndrome and may indicate a specific underlying cause for settling and night-waking problems frequently observed in Down's syndrome (Shott et al., 2006).

Prevalence of sleep problems among samples of individuals with ID is usually assessed by questionnaires. The Sleep Questionnaire by Simonds and Parraga (SQ-SP; 1982) – adapted version for use in individuals with ID (Wiggs & Stores, 1996) – is the sleep questionnaire used most often in prevalence studies in individuals with ID (Maas et al., 2011). Several types of sleep problems (i.e. settling problems, frequent night waking and early waking), as well as their level of severity can be assessed with the SQ-SP. Severity level can be reported for each type of sleep problem separately, but can also be expressed in a total severity score, which is called the Composite Sleep Index (CSI) (Wiggs & Stores, 1998). Furthermore, the SQ-SP enables one to gather data on different types of sleep disturbance. Psychometric properties of part four of the SQ-SP (see Materials) for use in individuals with ID have recently been examined in a sample of 345 individuals with mixed or heterogeneous ID (Maas et al., 2011). Internal consistency (Cronbach's $\alpha = .80$) and test-retest reliability (Spearman's rank correlation = .83) appeared to be good and convergent validity was adequate (Pearson correlation = .79). Factor analysis revealed five sleep factors that were related to five types of sleep disturbance: (a) Snoring, (b) Daytime sleepiness, (c) Complaints related to sleep, (d) Sleep apnea and (e) Anxiety related to sleep. Differences in CSI and factor scores between a group of individuals with ID that were referred to a sleep clinic for individuals with ID (sleep clinic group) and a group of individuals with mixed or heterogeneous ID (control group) were explored. CSI and factor scores on Daytime sleepiness and Complaints related to sleep differentiated between the sleep clinic group and the control group, further demonstrating validity.

Data on types of sleep problems, their level of severity and types of sleep disturbances gathered with the SQ-SP in three genetic syndromes (i.e. Jacobsen syndrome (JS), Cri du Chat syndrome (CDC), and Down's syndrome (DS)) have been published in two studies (Maas et al., 2008, 2009). Severe sleep problems were found in 9%, 20%, and 7% of the syndrome groups, respectively. No data on the CSI were reported. In both studies five types of sleep disturbances were distinguished, which reflect different types of sleep disturbances encountered in clinical practice (Johnson, Wiggs, Stores, & Huson, 2005; Stores, Wiggs, & Campling, 1998). However, the five types of sleep disturbances encountered in clinical practice appeared not to match the five sleep factors in individuals with ID (Maas et al., 2011), resulting in a lack of information on types of sleep disturbance in CDC, DS and JS. Thus the underlying causes for sleep problems remain unclear. Furthermore, data on sleep problems and type of sleep disturbances for CDC and DS have been compared with a NS control group, but so far this had not been done for JS. Therefore, the principal aim of this study was to assess the prevalence of five types of sleep disturbances in individuals with CDC, DS, JS, and NS using the SQ-SP. We also compared the level of severity of sleep problems expressed in the CSI, and different types of sleep disturbance in order to explore differences in severity of sleep problems and the underlying causes of sleep problems between the four diagnostic groups.

2. Methods

2.1. Participants and procedure

The sample included individuals who had participated in two previously published studies (Maas et al., 2008, 2009) and consisted of 100 individuals with ID, of whom 75 suffered from a genetic syndrome and 25 had a non-specific ID (NS). Of the sample of individuals with a genetic syndrome, 25 had CDC, 25 had DS, and 25 had JS. The parents or professional caregivers of individuals with CDC and individuals with JS were recruited via parent organizations (the American 11q Research and Resource Group and the Dutch Cri du Chat Parent Association). The caregivers of the individuals with DS and the individuals with NS were recruited via their child's day care center, special school or adult activity center. All caregivers received a package by mail. Each package consisted of a covering letter, the SQ-SP, a consent form and a stamped self-addressed envelope. Data from individuals for whom more than nine responses were missing on part four of the SQ-SP or for whom more than four consecutive responses were missing on part four of the SQ-SP were excluded from analyses. The individuals with NS were matched to those with CDC, DS and JS on age, level of cognitive functioning, presence of epilepsy and use of medication related to sleep problems. Demographic characteristics are presented in Tables 1 and 2.

Results of one-way ANOVA revealed that there were no significant differences between groups in age in months ($F(3, 96) = 1.25$, $p = .30$). Furthermore, results of two-tailed Fisher's exact tests showed that there were no significant differences between groups in level of cognitive functioning ($p = .88$), epilepsy ($p = .83$) and use of medication related to sleep problems ($p = .39$).

Table 1
Age (years; months) by diagnostic group.

Group	*n*	Mean	SD	Minimum	Maximum
CDC	25	13.5	7.7	3.8	28.5
DS	25	16.4	9.4	4.2	41.5
JS	25	12.9	5.5	3.9	25.10
NS	25	12.9	7.5	4.0	31.6

CDC, Cri du Chat syndrome; DS, Down's syndrome; JS, Jacobsen syndrome; NS, non-specific intellectual disability.

Table 2
Participant characteristics by diagnostic group.

Characteristic	CDC (*n* = 25) *n* (%)	DS (*n* = 25) *n* (%)	JS (*n* = 25) *n* (%)	NS (*n* = 25) *n* (%)	χ^2 value (3)[a]	*p*[b]
Number of males	8 (32)	19 (76)	9 (36)	13 (52)	11.97	.008
Living situation						.19
Family	22 (92)	25 (100)	25 (100)	24 (96)		
Residential facility	2 (8)	0	0	1 (4)		
Level of cognitive functioning						.88
IQ > 70	1 (4)	1(4)	2 (8)	1 (4)		
Mild/moderate ID	6 (24)	10 (40)	9 (36)	11 (44)		
Severe/profound ID	7 (28)	5 (20)	3 (12)	5 (20)		
Unknown[c]	11 (44)	9 (36)	11 (44)	8 (32)		
Epilepsy	1 (4)	1 (4)	1 (4)	1 (4)		.83
Medication use related to sleep	3 (12)	0	1 (4)	1 (4)		.39

CDC, Cri du Chat syndrome; DS, Down's syndrome; JS, Jacobsen syndrome; NS, non-specific intellectual disability; ID, intellectual disability.
[a] Kruskal–Wallis tests.
[b] In case no value for χ^2 is depicted because of low cell frequencies, the *p* value is a result of Fisher's exact test.
[c] No outcomes of tests or scales were available.

The male to female ratio varied across groups. Results of a chi-square test revealed that there were significant differences between groups in gender ($\chi^2(3) = 11.97$, $p = .008$). Pairwise group comparisons using a Bonferroni correction showed that there were significantly more males in the DS group than in the CDC group ($\chi^2(1) = 9.74$, $p = .004$). Due to low cell frequencies it appeared not possible to control for gender differences in further statistical analyses.

2.2. Materials

The SQ-SP was developed by Simonds and Parraga (1982) and was modified by Wiggs and Stores (1996, 2004) and consists of five parts. Part one-addresses demographic information (e.g., name and dosage of current medication and presence of epilepsy). The second part covers current (i.e., last three months) behaviors related to settling to sleep, night waking and early waking. In part three, parents are asked to fill in at what times their child usually goes to bed, wakes up in the morning, among other topics related to the sleep pattern. The fourth part assesses the frequency of occurrence of 45 behaviors related to sleep (e.g., 'Moves around a lot in bed during sleep', 'Snores loudly during sleep', 'Afraid of the dark') on a 7-point Likert-type scale, from 'Never' (1) to 'Daily' (7). The fifth and last part contains items about parents' impression of their child's current or past sleep problems, as well as treatment of the child's sleep problem.

Five types of sleep disturbance or sleep factors (Snoring, Daytime sleepiness, Complaints related to sleep, Sleep apnea and Anxiety related to sleep) were derived from part four of the SQ-SP (see Maas et al., 2011). The factor Complaints related to sleep refers to movements, excessive sweating and episodes of confused behavior during sleep. This factor consists of six items with a possible range from 6 to 42. Snoring consists of five items with a possible range from 5 to 35. Daytime sleepiness consists of four items with a possible range from 4 to 28. Both Sleep apnea and Anxiety related to sleep consist of three items with a possible range from 3 to 21.

To assess the level of severity of sleep problems a CSI was calculated (Wiggs & Stores, 1998). The *frequency* of problems with settling, night waking, early waking and co-sleeping and the *duration* of settling and night waking were derived from the SQ-SP, resulting in the CSI index ranging from 0 to 12. A score of $\geq$4 is indicative of a severe sleep problem (Wiggs & Stores, 1998) and by this the prevalence of severe sleep problems was assessed.

2.3. Statistical analyses

To test for differences within and between the four diagnostic groups nonparametric tests were used since data on the sleep factors and CSIs were not normally distributed and sample sizes were relatively small. To explore factor scores within a

diagnostic group a percentage of maximum score was calculated for each factor and Wilcoxon tests were performed. Because multiple comparisons were involved a Bonferroni correction was applied for the number of comparisons, resulting in $\alpha = .005$. To test differences in CSI and factor scores between groups Kruskal–Wallis tests were performed and a Bonferroni correction was used for the number of comparisons ($\alpha = .005$). Subsequent post hoc Mann–Whitney tests were performed to test for differences in CSI and factor scores between two diagnostic groups and a Bonferroni correction was applied for the number of comparisons ($\alpha = .0083$).

3. Results

3.1. Prevalence of types of sleep disturbance

Mean factor scores are shown in Table 3. To explore factor scores within each diagnostic group mean scores expressed as percentage of maximum score were calculated which are shown in Table 4. Within each group the factor Snoring had the highest mean score while the factor Sleep apnea had the lowest score. Wilcoxon tests within each group using Bonferroni correction revealed that the difference in mean score for Snoring and Sleep apnea was statistically significant within each group ($p < .005$). In the CDC, DS, and NS group, daytime sleepiness showed the second highest score, while in the JS group Complaints related to sleep showed the second highest score. The difference in mean score between the highest and the second highest score was substantial within the CDC, DS, and JS group. p-Values approached significance when snoring and daytime sleepiness were compared within the CDC group ($z = 2.81$, $p = .005$) and the DS group ($z = 2.81$, $p = .005$), but not within the JS group ($z = 1.55$, $p = .12$). Also in the JS group the difference in mean score for Snoring and Daytime sleepiness approached significance ($z = 2.74$, $p = .006$).

3.2. Comparison between individuals with CDC, DS, JS and NS

3.2.1. Severity of sleep problems

Mean CSI and number of individuals with a CSI score ≥ 4, indicating a severe sleep problem, are shown in Table 5. The highest mean CSIs were found in the JS group and the NS group and the lowest mean CSI was found in the DS group. Results of a Kruskal–Wallis test revealed that differences in mean CSI between groups were not statistically significant ($\chi^2(3) = 6.81$, $p = .08$). The highest percentage of individuals with a severe sleep problem was found in the JS group (i.e. 20%, 5/25), followed by the CDC and the NS group (i.e. 12%, 3/25), and the DS group (i.e. 4%, 1/25). Differences in percentage of individuals with a severe sleep problem between groups were not statistically significant ($p = .43$, Fisher's exact test).

Table 3
Mean scores and median scores for sleep factors by diagnostic group.

Factor	n	Mean (SD)	Median	χ^2 value (3)[a]	p
Snoring				7.51	.06
CDC	21	14.95 (7.95)	14		
DS	17	16.47 (9.58)	14		
JS	19	16.05 (9.00)	14		
NS	20	10.52 (7.26)	7		
Daytime sleepiness				3.85	.28
CDC	25	7.72 (4.50)	7		
DS	24	5.83 (3.17)	4		
JS	24	7.21 (3.64)	7		
NS	22	6.95 (3.84)	6		
Complaints related to sleep				11.48	.009
CDC	22	8.77 (3.89)	6		
DS	21	8.00 (2.39)	7		
JS	23	14.17 (8.17)	12		
NS	19	8.53 (3.84)	7		
Sleep apnea				0.44	.93
CDC	23	3.74 (3.12)	3		
DS	24	3.83 (2.84)	3		
JS	24	3.29 (1.08)	3		
NS	25	3.36 (1.60)	3		
Anxiety related to sleep				4.52	.21
CDC	25	3.92 (1.98)	3		
DS	22	3.91 (2.09)	3		
JS	24	4.87 (3.80)	3.5		
NS	23	5.00 (4.46)	3		

CDC, Cri du Chat syndrome; DS, Down's syndrome; JS, Jacobsen syndrome; NS, non-specific intellectual disability.
[a] Kruskal–Wallis tests.

Table 4
Mean scores (expressed as percentage of maximum score) for sleep factors by diagnostic group.

Group	*n*	Mean	SD	Minimum	Maximum
CDC					
Snoring	21	42.72	22.70	14.29	100
Daytime sleepiness	25	27.57	16.06	14.29	67.86
Complaints related to sleep	22	20.89	9.26	14.29	50.00
Sleep apnea	23	17.81	14.87	14.29	85.71
Anxiety related to sleep	25	18.67	9.42	14.29	47.62
DS					
Snoring	17	47.06	27.37	14.29	88.57
Daytime sleepiness	24	20.83	11.33	14.29	60.71
Complaints related to sleep	21	19.05	5.68	14.29	28.57
Sleep apnea	24	18.25	13.52	14.29	66.67
Anxiety related to sleep	22	18.61	9.96	14.29	47.62
JS					
Snoring	19	45.86	25.72	14.29	100
Daytime sleepiness	24	25.74	12.98	14.29	64.29
Complaints related to sleep	23	33.74	19.46	14.29	85.71
Sleep apnea	24	15.67	5.16	14.29	38.10
Anxiety related to sleep	24	23.21	18.11	14.29	100
NS					
Snoring	20	29.29	20.75	14.29	88.57
Daytime sleepiness	22	24.84	13.70	14.29	67.86
Complaints related to sleep	19	20.30	9.13	14.29	40.48
Sleep apnea	25	16.00	7.64	14.29	52.38
Anxiety related to sleep	23	23.81	21.25	14.29	90.48

CDC, Cri du Chat syndrome; DS, Down's syndrome; JS, Jacobsen syndrome; NS, non-specific intellectual disability.

Table 5
Composite Sleep Index (CSI) by diagnostic group.

Group	*n*	Mean	SD	Minimum	Maximum	CSI $\geq$ 4 *n* (%)
CDC	25	1.08	1.75	0	6	3 (12)
DS	25	0.48	1.12	0	5	1 (4)
JS	25	1.68	2.16	0	7	5 (20)
NS	25	1.60	2.31	0	9	3 (12)

CDC, Cri du Chat syndrome; DS, Down's syndrome; JS, Jacobsen syndrome; NS, non-specific intellectual disability. CSI $\geq$ 4 indicates a severe sleep problem.

3.2.2. Types of sleep disturbance

Differences in mean factor scores between diagnostic groups were explored using Kruskal–Wallis tests (see Table 3). Whereas differences between groups on Snoring approached significance ($p = .06$), differences on Complaints related to sleep were statistically significant. Subsequent Mann–Whitney tests revealed that the JS group showed significantly more Complaints related to sleep than the DS group ($z = 2.89$, $p = .004$). p-Values approached significance if mean scores on Complaints related to sleep were compared between the JS group and the CDC group ($z = 2.60$, $p = .009$) and between the JS group and the NS group ($z = 2.49$, $p = .013$).

4. Discussion

We used Simonds and Parraga's (1982) Sleep Questionnaire to assess the prevalence of types of sleep disturbances in individuals with CDC, JS, DS, and NS to explore differences in severity of sleep problems and the underlying causes of sleep problems between the four diagnostic groups. Severe sleep problems were prevalent in 20% of the individuals with JS, 12% of the individuals with CDC and NS, and 4% of the individuals with DS. The differences between groups were not statistically significant and we can conclude that there was no increase in the prevalence rates of severe sleep problems in the three genetic syndromes in comparison with NS. In each group mean score for Snoring was significantly higher than for Sleep apnea. In the CDC, JS, and DS group snoring was the most prevalent type of sleep disturbance, whereas in the NS group no type of sleep disturbance was identified as most prevalent. Thus, Snoring discriminates between NS and the three genetic syndromes, but not between the three genetic syndromes. Parents or professional caregivers of individuals with JS reported significantly more Complaints related to sleep than those of individuals with DS, and tended to report more complaints related to sleep than those of individuals with CDC and individuals with NS.

Closer inspection of the nature of the Complaints related to sleep in the JS group revealed that restless legs, excessive sweating while falling asleep and episodes of confused behaviors were not particularly common behaviors in the JS group (17%, 8%, and 0%, respectively), but restless sleep, quick movements of arm or legs and startles or jerks part of the body while falling asleep were commonly reported (60%, 29%, and 25%, respectively). Prevalence rates of the last two behaviors differed significantly between diagnostic groups, and the differences in prevalence rate of restless sleep approached significance, with the JS group having the highest score. These results broaden our knowledge of sleep in individuals with JS, since the prevalence rates of restless sleep and quick movements of arms and legs and startles or jerks while falling asleep have not been compared to those of individuals with NS or other genetic syndromes before, and these behaviors appear to be characteristic sleep features of JS. However, the nature, timing and duration of these complaints related to sleep have not yet been clarified by objective measures, as mentioned by Maas et al. (2008). Audiovisual recordings in the home setting can serve this purpose, because both timing and duration as well as the nature of the complaints can be observed using that method.

The prevalence rates of severe sleep problems in the CDC, DS, JS, and NS groups were lower than those in a sample of 345 individuals with mixed or heterogeneous ID referred to a sleep clinic (see Maas et al., 2011). Although some individuals from each diagnostic group had a severe sleep problem, we can conclude that the prevalence rates in the diagnostic groups on the whole were not clinically elevated. However, the prevalence rates of Snoring in the CDC, DS, and JS groups were somewhat higher than the prevalence rates found in a sleep clinic group. These results indicate that snoring is a sleep disturbance particularly prevalent in individuals with CDC, DS, and JS. Finally, the prevalence rate of Complaints related to sleep in the JS group was similar to that of a sleep clinic group, suggesting that on average individuals with JS show clinically elevated complaints related to sleep.

Snoring is the most prevalent sleep disturbance in individuals with CDC, DS, and JS. It is a well-known fact that snoring is highly prevalent in individuals with DS, and another study has shown that snoring is quite prevalent in individuals with CDC (Maas et al., 2009), but it had not been reported in individuals with JS previously (Maas et al., 2008). Primary snoring (i.e. loud upper airway breathing sounds in sleep without episodes of apnea) is at the least severe end of the Sleep Related Breathing Disorders continuum (SRBD) (Huynh, Morton, Rompré, Papadakis, & Remise, 2011). Since SRBD is associated with craniofacial anomalies it is not surprising to find that snoring is a prevalent type of sleep disturbance in individuals with a genetic syndrome presenting with anomalous craniofacial features. Typically developing children with primary snoring have a higher risk of exhibiting hyperactive and inattentive behavior, daytime sleepiness, and poor school performance than children who have never snored (Brockmann, Urschitz, Schlaud, & Poets, 2012). Snoring in individuals with CDC, DS, and JS should not be ignored and some individuals might benefit from treatment by a sleep medicine specialist and an ear, nose and throat specialist (ENT).

This study has several limitations. The first limitation relates to the small sample size as a result of which we had to use a simple statistical model and did not attempt to explore associations between individual-related variables (e.g. age), sleep factors scores and severity of sleep problems. Diagnostic groups were matched on the most important variables, i.e. age, level of cognitive functioning, epilepsy and use of medication related to sleep problems to control for some of the individual-related variables. The second limitation concerns the inclusion of both children and adolescents as well as adults in our sample, leading to a sample with a wide age range, which may limit the generalizability of the results. However, the age range is comparable with the sample used to assess the psychometric properties of the SQ-SP (Maas et al., 2011) and therefore the SQ-SP is suitable for this sample. Future studies should use samples consisting of either children and adolescents or adults with ID to control for developmental effect on the prevalence of different types of sleep problems. The last limitation has to do with the fact that the sample of individuals with DS deviates from other studies on sleep in individuals with DS in terms of age and gender. In this study more males were included than in other studies. This indicates that our sample is not representative for individuals with DS. Furthermore, our sample included both children and adults with DS and therefore it is impossible to compare this study with previously published studies (e.g. Stores et al., 1996) including samples of only children with DS.

In conclusion, our study shows that sleep problems in individuals with CDC, DS, and JS are not more common than in individuals with NS. However, the underlying causes of specific sleep disorders in individuals with CDC, DS, and JS seem to be different from those in individuals with NS. Clinicians faced with sleep problems of individuals with CDC, DS, and JS should be alert to specific types of sleep disturbance, specifically snoring. They may want to make eliminating snoring their first aim when treating clients with sleep problems.

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