

Title:

Sleep problems in adults with Down syndrome and their family carers

Running title:

Sleep problems and Down syndrome

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Key words:

Sleep problems, adults, Down syndrome, carers

Abstract

Background

Sleep disturbance is common and can have harmful psychological and physical effects.

Whilst sleep problems in children with Down syndrome (DS) have received a reasonable amount of attention, very little has been written about this topic in adults with DS.

Method

The present study consisted of an online survey completed by 100 family carers of adults with DS.

Results

High rates of sleep problems of different types were reported in the adults with DS comparable to those found in children with DS in previous research. Significant associations were found between sleep problems and body mass index, excessive daytime sleepiness and a range of health and psychological problems. Low rates of treatments for sleep problems were reported. The majority of family caregivers felt their own sleep was affected.

Conclusions

Sleep problems in adults with DS are common and varied. Assessment and treatment of such problems are likely to improve quality of life.

1 Introduction

There is convincing evidence that persistent sleep disturbance has harmful psychological and physical effects for any individual (Kryger, Roth & Dement, 2017). Inadequate or poor quality sleep is associated with disturbed emotional state or behaviour such as anxiety, depression and irritability and also impaired cognitive function and performance. Such disturbance is common in the general population but certain groups are at particularly high risk, including people with intellectual disabilities (Gunning & Espie, 2003; Brylewski & Wiggs, 1998; van de Wouw, Evenhuis & Echteld, 2012; Surtees, Oliver Jones, Evans & Richards (2018). The common condition of Down syndrome (DS) affecting around 1 in 1,000 live births worldwide, is a case in point. Sleep disturbance in children with DS has received a reasonable amount of research attention which has found high rates of both behavioural and physical sleep problems, including obstructive sleep apnoea (Joyce & Dimitriou, 2017; Fernandez et al, 2017; Hill et al, 2016; Stores & Stores, 2013, 2014; Stores, Stores & Buckley, 1996). However, relatively little has been written about sleep in adults with this condition, especially those of an older age.

The limited existing literature on sleep in adults with DS has found high rates of sleep disordered breathing, especially obstructive sleep apnoea (OSA) (between 35 and 42%) (Resta et al, 2003; Trois et al, 2009; Hill 2016) attributed to physical factors such as midfacial and mandibular hypoplasia, relatively large tongue, abnormally narrow airway and relative tonsillar and adenoidal encroachment, obesity and generalized hypotonia causing collapse of the airway on inspiration (Hill, 2016; Hill et al, 2015).

Behavioural sleep disturbance in adults with DS has received limited attention. Esbensen (2016) explored both behavioural sleep problems and sleep disordered breathing (as reported by family care givers) and associated comorbidities in this group. Different

physical and behavioural correlates for behavioural sleep problems and sleep apnoea were apparent. Sleep apnoea was associated with more common respiratory concerns and more frequent visits to physicians. By comparison, behavioural sleep disturbances were associated with poorer health, more frequent overnight hospital and emergency department visits, more common cardiac conditions, less common thyroid condition, more mental health conditions and a higher rate of daytime behavior problems.

Associations between sleep disruption (mainly related to sleep disordered breathing) and impaired cognitive functioning including impaired executive function and visuospatial skills, have also been described in adults with DS by Chen, Spano & Edgin (2013) and Andreou et al (2002).

The limited literature on sleep disturbance in adults with DS has four main shortcomings. The first is the small number of individuals studied. The second is the limited information about the nature of the reported sleep disturbances, the emphasis being on sleep problems rather than sleep disorders as described in the International Classification of Sleep Disorders ICSD-3 (American Academy of Sleep Medicine, 2014). As an individual's sleep disorder indicates appropriate choice of treatment, ultimately studies of sleep disturbance in adults with DS should aim to identify sleep disorders. However, this usually calls for detailed enquiry possibly including special investigations. The present study is best seen as an exploratory survey of a neglected topic intending to raise possible issues to be explored in greater clinical detail in future research.

The third shortcoming is that little account is taken of the range of possible coexisting medical and psychological conditions in DS (Bittles & Glasson, 2010; Smith, 2001) many of which are capable of contributing to sleep disturbance (Kryger et al, 2017). Examples include upper airway obstruction, cardiac disease, obesity, thyroid disease,

sensory deficits, epilepsy, gastro-oesophageal disease, musculo-skeletal problems, anxiety, depression and dementia. It is possible such conditions may exacerbate or be exacerbated by sleep problems.

Finally, to the author's knowledge, the sleep of family caregivers has not been the subject of previous research. Sleep disruption in these individuals could have harmful effects on their health and general wellbeing resulting in impairment of their caring abilities.

With these limitations in mind, the aims of the present project were as follows:

1. To document the occurrence and nature of a wide range of sleep problems and behaviours, as reported by family carers, in a population based sample with a wide age range. As already emphasized, in this exploratory study, it was not feasible to diagnose sleep disorders underlying the reported sleep problems as this would have required detailed clinical assessments which was outside the scope of this research.
2. To investigate associations between sleep problems and a wider range of factors than has been looked at previously including age, sex, body mass index (BMI), level of intellectual disability, daytime sleepiness and co-existing health and psychological problems also as judged by family carers.
3. To describe current and past treatments for each individual's sleep problem(s) in the adults with DS.
4. To explore possible sleep problems of family carers and other family members to see if their sleep is affected by their relative with DS.

The author recognises the importance of obtaining information from adults with DS themselves about their sleep and has carried out a separate survey on sleep which adults with DS have completed. This will form the basis of a separate article submission.

2 Methods

2.1 Participant recruitment

Family carers with an adult relative with DS aged 16 years or over were recruited through the UK Down's Syndrome Association via the Association's electronic newsletter, blog, Facebook and Twitter accounts where a short article about the nature of the research was provided together with a link to the online survey. The survey was compiled using the Bristol Online Survey platform. Study information sheets for both the family carers and the adults with DS (should the family carer have wished to discuss the research with their relative with DS) were embedded within the online survey.

2.2 Design

The present study consisted of a cross-sectional online survey completed by family carers of adults with DS.

2.3 Assessments

Each individual's sleep was assessed from the perspective of the family carer using an online version of the Simonds and Parraga Sleep Questionnaire (Simonds and Parraga, 1982). This questionnaire has been used in previous studies of children with DS and other forms of intellectual disability (Stores & Stores, 2014, 2004; Stores, Stores & Buckley, 1996) but has been adapted for use with adults with intellectual disability (Brylewski & Wiggs, 1998; 1999) and has been shown to have satisfactory psychometric properties (Mass et al, 2011). It is completed by primary carers. Although the findings do not translate into ICSD-3 sleep disorders, they provide comprehensive information about clinical symptoms and signs.

Separate sections of the survey enquire about the sleeping arrangements and habits of the adult with DS, their typical sleep duration and the frequency of various sleep disorders and behaviours over the past month on the following five point scale: *never, about once a month, about once a week, several times a week, daily* and an additional possible response of *don't know*. Thus clinically important degrees of sleep problems could be distinguished from less frequent sleep disturbance. Carers were also asked about any current or past treatments for sleep problems that their adult relative with DS was or had received.

The Epworth Sleepiness Scale (ESS) (Johns, 1991), a measure of the propensity to fall asleep in eight situations was used to assess daytime sleepiness by proxy from the perspective of family carers. A similar approach has been used successfully with caregivers of adults with DS previously (Trois et al, 2009). The eight situations are: sitting and reading; watching television; sitting inactive in a public place e.g. theatre, meeting; as a passenger in a car for an hour without a break; lying down to rest in the afternoon; sitting and talking to someone; sitting quietly after lunch (when they have had no alcohol); and in a car, while stopped in traffic. Respondents are asked to indicate the chance of dozing from 0 - 'would never doze', 1 - 'slight chance of dozing', 2 - 'medium chance of dozing' or 3 - 'high chance of dozing and a total score obtained.

Additional information was sought from carers on the age, gender, height, weight, level of intellectual impairment and usual place of living of the adult with DS. Carers were also asked whether their relative had specific health and psychological problems from a pre-defined list of conditions which are known to occur in adults with DS.

Carers were also asked about effects on their own and other family members' sleep using the question "Do you think your sleep is affected by the sleep of your relative with DS

in any way?” with a possible ‘Yes’/‘No’ answer. This was followed up with a supplementary open-ended question “If Yes, how does their sleep affect your own?”. Similarly, carers were asked “Is the sleep of other family members affected by the sleep of your relative with DS” with a possible ‘Yes’/‘No’ answer and a supplementary open-ended question “If Yes, how?”.

2.4 Ethical approval

The study was reviewed and given a favourable opinion by the South West Cornwall and Plymouth NHS Research Ethics Committee (REC Reference No: 16/SWE/0055).

2.5 Statistical Analysis

Data were exported from the Bristol Online Survey platform into SPSS V24 (IBM). Descriptive, Chi-squared (χ^2), Odds Ratio (OR), Cohen’s d and independent t-test statistics were used in the analyses and are described in the relevant Results sections.

3 Results

3.1 Respondents

A total of 101 family carers completed the survey fully. However, one response was based on an individual with DS less than 16 years of age so was excluded leaving a sample size of 100. Seventy-four respondents were mothers of adults with DS, one was a father and 21 were siblings. The remaining four were a sister-in-law, a niece, a ‘Shared Lives’ host and a formal carer.

3.2 General characteristics of adults with DS

General characteristics of the adults with DS are shown in Table 1.

Table 1 about here

The sample of adults with DS reported on consisted of 55 males and 45 females with an age range spanning 45 years (16-61 years). More individuals fell into the younger age categories which is representative of the population. The majority of the adults were reported by carers as having a 'moderate' or 'moderate to severe' level of intellectual disability and the majority lived with their family.

3.3 Health and psychological/behavioural problem in adults with DS

The percentage of adults with DS reported as having various health and psychological problems from a pre-defined list are shown in Table 2.

Table 2 about here

The most commonly reported health problems were hearing loss, thyroid problems, heart problems and obstructive sleep apnoea (OSA). The most commonly reported psychological problems were obsessionality, fear of certain things, anxiety and being withdrawn from other people.

3.4 Sleeping arrangements, habits and total sleep time

Ninety-three percent of adults with DS were reported as having a room of their own. Seventy-six percent had a bedtime routine and 40% had a special thing to help them settle. The most common of these were a soft toy or blanket (16%), relaxing music or sounds (6%) having the TV on (5%) and a night light (4%). The mean reported total nocturnal sleep time was 8 hours (SD 1.7) with a range of 4 to 12 hours.

3.5 Sleep problems and behaviours

As an indication of the relative occurrence of clinically significant sleep disturbances, Table 3 shows the number of adults with DS reported as having sleep problems and behaviours under the headings: *disorders of initiating and maintaining sleep, other problems or behaviours occurring during sleep, features associated with obstructive sleep apnoea and sleep related problems or behaviours occurring during the day* in each of the combined categories: (1) 'Daily' or 'Several times a week'; (2) 'About once a week' or 'About once a month' and lastly (3) 'Never'. Sleep disorders and behaviours are ordered by the most to least frequent occurring 'Daily' or 'Several times a week'.

Table 3 about here

Within '*Disorders of initiating and maintaining sleep*', the most common problems occurring daily or several times a week were problems settling at night (37%), awakens during the night and requires attention (33%) and very reluctant to go to bed (30%). For '*Other problems/behaviours occurring during sleep*', the most common problems were teeth grinding (39%) and sleep talking (26%). The highest overall rates of occurrence were found

for *'Features associated with obstructive sleep apnoea'* including breathes through mouth rather than nose (84%), snores loudly (53%), restless sleeper (48%), sleeps with head tipped right back (43%), sleeps in unusual positions (40%), appears to stop breathing for periods of up to 30 seconds (35%) and appears to have difficulty breathing during sleep (33%). The most commonly occurring *'Sleep related disorders or behaviours occurring during the day'* were lacks energy during the day (45%), excessively sleepy during the day (38%), irritable during the day (33%), difficulty staying awake during the day (30%), actually falls asleep during the day (26%), takes naps during the day (25%), drowsy during the day (25%) and miserable during the day (21%).

3.6 Sleep problem categories

In order to explore associations between different types of sleep problems with age, gender, BMI, level of intellectual disability, daytime sleepiness and health and psychological problems, adults with DS were categorised as to whether or not they were reported to have (a) a behavioural sleep problem defined as 'problems settling at night', 'awakens during the night and requires attention' and/or 'wakes before 5 in the morning' occurring either daily or several times a week or (b) obstructive sleep apnoea as reported by family carers. These sleep problems were included as they are known to affect sleep quality and quantity. There was missing data on OSA from three family carers. The proportion of the remaining 97 adults with DS in each of these categories is shown in Table 4.

Table 4 about here

A total of 48% of adults were reported to have a behavioural sleep problem and 25% were reported to have OSA. Thirty-one percent of adults were reported to have a behavioural sleep problem and no OSA, 7% were reported to have OSA and no behavioural sleep problems, 18% were reported to have both a behavioural sleep problems and OSA and 44% were reported to have neither a behavioural sleep problem or OSA. There was a significant positive association between the behavioural sleep problem category and OSA ($\chi^2=6.40$, $df=1$, $p=0.011$, $OR=3.481$) meaning those with a behavioural sleep problem were more likely to be reported as also having OSA.

3.7 Sleep problems and age and gender

No significant associations were found between age and whether an individual was reported to have a behavioural sleep problems ($t=0.391$, $df=96$, $p=0.697$) or obstructive sleep apnoea ($t=1.349$, $df=93$, $p=0.18$). Similarly, no significant associations were found for gender and behavioural sleep problems ($\chi^2=0.66$, $df=1$, $p=0.797$) and obstructive sleep apnoea ($\chi^2=0.48$, $df=1$, $p=0.826$).

3.8 Sleep problems and BMI

The mean BMIs for adults with DS reported as having behavioural sleep problems and OSA are shown in Table 5.

Table 5 about here

No significant association was found between whether an individual was reported to have a behavioural sleep problem and their BMI ($t=1.542$, $df=85$, $p=0.128$). However,

individuals reported as having OSA had a significantly higher BMI than those who were not (mean BMI 32.6 vs 28.7) ($t=2.443$, $df=90$, $p=0.017$, Cohen's $d=0.59$).

3.9 Sleep problems and level of intellectual disability

Associations between different types of sleep problems and level of intellectual disability were explored by comparing the rates of occurrence in those adults reported as having mild or mild to moderate, moderate and lastly moderate to severe or severe intellectual disability. No significant differences were found between these groups for behavioural sleep problems ($\chi^2=0.599$, $df=2$, $p=0.741$) and obstructive sleep apnoea ($\chi^2=1.291$, $df=2$, $p=0.525$).

3.10 Epworth Sleepiness Scale (ESS)

The results of the *ESS* are shown in Table 6.

Table 6 about here

An *ESS* score was available for 91 individuals. The missing nine scores were due to not all *ESS* items being completed and therefore it not being possible to calculate a total score. *ESS* scores covered a wide range from 0-24. Thirty-eight percent of individuals fell into categories of clinically significant excessive daytime sleepiness using the categories defined by Johns (1991), 14% as mild, 14% as moderate and 10% as severe.

Associations were investigated between the sleep problem categories and *ESS* scores. Mean *ESS* scores for these groups are also shown in Table 6. Adults reported as having a behavioural sleep problem had significantly higher *ESS* scores than those without

($t=2.407$, $df=89$, $p=0.018$, Cohen's $d=0.5$). Adults reported as having obstructive sleep apnoea had significantly higher ESS scores than those without obstructive sleep apnoea (11.8 vs 7.6) ($t=3.039$, $df=87$, $p=0.003$, Cohen's $d=0.75$).

3.11 Associations between sleep problems and health and psychological problems

Associations were investigated between the sleep problem categories and the occurrence of the health and psychological problems detailed in Table 2.

For the behavioural sleep problems, a significant positive association was found for hearing loss ($\chi^2=8.032$, $df=1$, $p<0.005$, $OR=3.306$). Significant positive associations were also found between behavioural sleep problems and anxiety ($\chi^2=12.215$, $df=1$, $p<0.001$, $OR=4.50$), fear of certain things ($\chi^2=12.154$, $df=1$, $p<0.001$, $OR=4.373$), obsessionality ($\chi^2=7.542$, $df=1$, $p=0.006$, $OR=3.201$), depression ($\chi^2=6.250$, $df=1$, $p=0.012$, $OR=3.903$), aggressive behaviour ($\chi^2=6.133$, $df=1$, $p=0.013$, $OR=4.889$), destructive behaviour ($\chi^2=5.204$, $df=1$, $p=0.023$, $OR=4.414$), behaviour is difficult to control ($\chi^2=5.659$, $df=1$, $p=0.017$, $OR=3.703$) and injures self ($\chi^2=4.525$, $df=1$, $p<0.033$).

With regards to obstructive sleep apnoea, significant positive associations were found for epilepsy/convulsions/fits/seizures ($\chi^2=6.291$, $df=1$, $p=0.012$, $OR=7.368$), muscular skeletal problems ($\chi^2=5.952$, $df=1$, $p=0.015$, $OR=4.063$). A significant positive association was also found for depression ($\chi^2=4.955$, $df=1$, $p=0.026$, $OR=3.307$).

3.12 Treatments for sleep problems

Fifteen of the 100 carers indicated their relative was having treatment for sleep problems *at the present time or in the last month*. In response to this question, eight indicated they were taking medication: two were taking melatonin, one of whom reported

this as helpful and the other not; two mirtazapine and single individuals were taking haloperidol, fluoxetine, fluticasone and clonazepan, all of which were reported as helpful except for the fluticasone. Six carers indicated their relative was using Continuous Positive Airway Pressure (CPAP) five of which stated this was helpful and one indicated this was not. One carer said their relative was receiving psychological treatment for nightmares which they had not found helpful.

When asked about any treatment for sleep problems that their relative had received *prior to the last month*, four additional carers indicated CPAP, three of whom said their relative had not tolerated this and were therefore no longer using it and one reported this as helpful. Four carers indicated psychological advice relating to promoting good sleep hygiene and one carer said they had received advice relating to changing their relative's sleeping position.

3.13 Family carers' and other family members sleep

Fifty-eight percent of carers indicated they felt their sleep was affected by the sleep of their relative with DS. When asked how their sleep was affected, the vast majority of responses related to sleep disruption as a result of listening out for their relative, checking on them during the night or not being able to sleep while they are awake. Other reasons included disruption caused by having to settle and/or re-settle their relative during the night, their relative shouting or calling out, listening out or checking on their breathing, co-sleeping, managing nocturnal wanderings, managing routines and rituals, the restless sleep of their relative and the noise caused by CPAP machines. Some illustrative responses to the open ended question '*How does the sleep of you relative with DS affect your sleep?*' are shown in Table 7.

Table 7 about here

Twenty-six per cent of carers indicated they felt the sleep of other family members was affected by their relative with DS. When asked how their sleep was affected, most responses related to sleep disturbance caused by their relative being noisy and shouting during the night and having to check on them. Other individual reasons included their talking out loud, being aware of their relative moving around, the noise of CPAP machines, their relative screaming every hour, snoring, coming in to their bedroom, playing music or singing and co-sleeping. Some illustrative responses to the open ended question 'How is the sleep of other family members affected by the sleep of your relative with DS?' are shown in Table 8.

Table 8 about here

4 Discussion

This study was undertaken because of the limited information about the important problem of disturbed sleep in adults with DS. To re-cap, the main objectives of the study were as follows:

1. To document the occurrence and nature of a wide range of sleep problems and behaviours, as reported by family carers, in a population based sample with a wide age range.
2. To investigate associations between sleep problems and a wide range of factors including age, sex, body mass index (BMI), level of intellectual disability, daytime sleepiness and co-existing health and psychological problems also as judged by family carers.

3. To describe current and past treatments for each individual's sleep problem(s) in the adults with DS.
4. To explore possible sleep problems of family carers and other family members to see if their sleep is affected by their relative with DS.

High rates of sleep problems were reported by the family carers in the adults. These problems included disturbed initiation and maintenance of sleep, other problems or behaviours during sleep, features associated with OSA and sleep related problems occurring during the day. Interestingly, rates of occurrence of the majority of sleep problems and behaviours as detailed in Table 3 were very similar to those found in a sample of 91 children with DS aged 4 to 19 years studied previously (Stores et al, 1996) which were shown to be significantly more common than in the siblings of the children with DS and children in the general population. This suggests that childhood sleep problems in children with DS may well persist into adulthood.

No significant associations were found between any types of sleep problems and age, gender or level of intellectual disability. OSA as reported by carers was more common in adults with a greater BMI which is in keeping with previous research in the area (Trois et al, 2009).

A number of significant associations were found between sleep problems of different types and the various health and psychological problems asked about. Most of the factors associated with behavioural sleep problems were psychological in origin indicating these sleep problems may be part of generalized psychological disturbance presenting at night time. The significant association between behavioral sleep problems and hearing loss might be explained by the reduced sensory input interfering with stimulus cues for initiating and maintaining sleep. This possibility warrants further research. The factors significantly

associated with OSA were physical and psychological in origin. These findings are somewhat in keeping with those of Esbensen (2016) who found different profiles for behavioral sleep problems and OSA.

Aetiological factors underlying sleep disturbance in individuals with DS are likely to be multifactorial consisting of intrinsic pathophysiological factors, physical comorbidities, psychological comorbidities, possible medication effects and carers' practices. Comorbidities in DS including those in adults are discussed by Bax & Gillberg (2010). Identification of multifactorial influences on sleep in the individual case is essential in designing an appropriate treatment programme (Stores, 2016).

An important issue arising from the demonstration of sleep problems, of whatever origin, is the psychological effect i.e. what contribution impaired sleep quality might make to the daytime functioning and wellbeing of adults with DS. In keeping with evidence that OSA is often psychologically harmful because of the disturbance of sleep physiology that it causes (Brown, 2005), the present findings included associations between sleep problems and daytime functioning in adults with DS. There is a need for further exploration of the relationship between sleep disturbance and cognitive functioning as suggested by studies in children with DS (Joyce & Dimitriou, 2017; Nixon et al, 2016) and in preliminary studies in adults with DS (Chen, Spano & Edgin, 2013; Andreou et al, 2002).

High rates of excessive daytime sleepiness were found in the cohort with 38% falling into clinically significant categories. Significant associations were found for both behavioral sleep problem and OSA as reported by family carers and increased daytime sleepiness.

Despite high rates of sleep problems, somewhat low rates of current or past treatments were described by carers. Twenty-four adults were reported as having OSA yet only six individuals were currently undergoing CPAP treatment or had used it in the last

month. Unsuccessful past attempts at CPAP were also described. It is possible other adults were offered CPAP but carers thought they would not tolerate it so never attempted it. Very few psychological treatments were reported.

A majority of carers (58%) felt their sleep was affected by the sleep of their relative with DS. Reasons given for this sleep disturbance varied but usually this was caused by a felt need to be vigilant during the night and also disturbance caused by noisy breathing and/or behaviour of their relative with DS. Carers' responses illustrated the serious impact this had in some cases. Similar effects on other family members' sleep were also described. This is a previously unreported complication and an important one as a significant proportion of adults with DS live with their family all or some of the time and carers' and relatives' sleep being affected may in turn affect their relationships with their relatives with DS by, for example, causing them to become more irritable which may impact on the individual with DS.

There are a number of limitations with this preliminary study which need to be taken into consideration when interpreting the results. There are limitations to questionnaire studies in general, mainly concerning the subjectivity of the data obtained and the possibility of bias on the part of the informant. However, in the present study, there is no particular reason to believe family carers completing the survey would be motivated to provide misleading information. However it is possible that unconscious biases or confounds could affect carers responses. For example, an anxious carer may be more likely to consider the sleep of the person they care for to be problematic and to report higher levels of behavioural difficulties. It is also possible the present findings provide an under-estimation of the rate of sleep problems as family carers may not be aware of problems if they are in a separate rooms.

There is also a possibility of a biased sample having been recruited. However, it was made clear that information was requested on both adults with and without sleep problems and that the aim was to get a representative picture of sleep problems in adults with DS in general. Nevertheless, it is possible that those family carers who considered their relative with DS to have sleep problems were more likely to respond leading to an overestimation of such problems. Future research should aim to achieve a more systematic approach to recruitment.

It should also be noted the accuracy of some information obtained from carers including height, weight and level of intellectual disability should be interpreted with caution.

5 Conclusions and future research

This study was undertaken because it deals with a subject that is neglected despite its importance for the health and wellbeing of adults with DS and also their carers. It is intended that the findings lead to further research aimed at clarifying issues that have arisen from this investigation.

The subject matter is inevitably complex not least because adults with DS are a heterogenous group regarding, for example, intellectual level, behaviour, psychological status and also physical health which can be compromised in many ways. All these and other variables, such as carers' own well being and general circumstances can influence the individuals' sleep patterns.

The concepts of sleep problems and sleep disorders add further complexity, the latter being especially important as a guide to choice of appropriate treatment. Identifying the sleep problems of adults with DS sets the scene for more focused sleep disorder studies.

Future research should also include the use of objective methods such as actigraphy to measure sleep quality and quantity. This approach is relatively low cost, well tolerated and concords well with polysomnography (Van De Water Holmes & Hurley, 2011).

The present investigation has demonstrated high rates of sleep problems of different types in adults with DS comparable to those found in children with DS. The point has been made that this suggests that childhood sleep problems may well persist into adulthood in this group. Despite these high rates, relatively low levels of helpful treatments were reported. Significant associations were found between sleep problems of different types and BMI, excessive daytime sleepiness and with a range of health and psychological problems. Of significant practical importance is that the sleep of family carers and other family members was affected by the sleep problems of their relative with DS.

Future research should explore further associations between sleep problems and daytime functioning. As these relationships become clearer, effective treatments for sleep problems should be valuable in reducing some of the daytime psychological consequences in adults with DS who already face cognitive challenges. The success in other settings of behavioural treatment for sleep disturbance (Gunning & Espie, 2003) suggests that similar treatment approaches (if necessary modified) can be expected to improve their quality of life and help them to reach their full potential. In addition, investigation of support and advice for family carers in managing sleep problems (including their own) also deserves priority.

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Table 1
General characteristics of the adults with DS

N	100
Male	55 (55%)
Female	45 (45%)
Age	
Mean	28 years (SD 11.4)
Range	16 - 61 years
Age group (years)	
16-25	53 (54%)
26-35	26 (27%)
36-45	8 (8%)
46-55	6 (6%)
56-61	5 (5%)
missing	n=2
BMI	
Mean	29.6 (SD 6.8)
Range	18.9 - 49.2
Normal (≤ 24.9)	26 (27%)
Overweight (25-29.9)	26 (27%)
Obese (≥ 30)	43 (45%)
missing	n=5
Level of intellectual disability	
Mild	4 (4%)
Mild to Moderate	16 (16%)
Moderate	34 (35%)
Moderate to severe	33 (34%)
Severe	11 (11%)
missing	n=2
Usual place of living	
With family	86 (86%)
Supported Living	9 (9%)
Shared Lives	1 (1%)
Other	4 (4%)

Table 2
Health and psychological problems reported in the adults with DS (n=100)

Health problems		Missing (n)
Hearing loss	42%	2
Thyroid problems	38%	5
Heart problems	32%	3
OSA	25%	3
Muscular skeletal	15%	5
Epilepsy/convulsions/fits/seizures	7%	4
Diabetes	5%	5
Atanlantoaxial subluxation	4%	8
Dementia	5%	4
Psychological		
Obsessional	57%	2
Fear of certain things	43%	1
Anxiety	39%	1
Withdrawn from other people	21%	2
Depression	19%	2
Behaviour is difficult to control	18%	2
Aggressive behaviour	14%	2
Destructive behaviour	13%	1
Injures himself/herself	4%	2

Table 3

Percentage (of those who responded) of adults with DS reported as having sleep problems or behaviours by family carers

Sleep problem/behaviour	'Daily' or 'Several times a week' (%)	'About once a week' or 'About once a month' (%)	'Never' (%)	'Don't know' or missing (n)
Disorders of initiating and maintaining sleep				
Problems settling at night	37	27	35	1
Awakens during the night and requires attention	33	26	41	5
Very reluctant to go to bed	30	19	50	1
Wakes before 5 in the morning	11	30	58	4
Reluctant to go to bed because of fears	7	12	81	5
Other problems/behaviours occurring during sleep				
Teeth grinding	39	14	47	17
Sleep talks in his/her sleep	26	40	33	12
Rolls from side to side rhythmically during sleep or while going off to sleep	12	10	78	9
Complains of nightmares	7	20	73	5
Bites tongue during sleep	4	1	95	12
Night terrors	2	6	92	3
Sleep walks	1	5	94	4
Head during sleep or going off to sleep	0	2	98	6
Features associated with obstructive sleep apnoea				
Breathes through mouth rather than nose	84	9	7	11
Snores loudly in bed	53	28	19	5
Restless sleeper i.e. moves around a lot in bed	48	24	27	9
Sleep with his/her head tipped right back	43	9	49	20
Sleeps in unusual positions	40	15	45	9
Appears to stop breathing for up to 30 seconds	35	17	49	22
Appears to have difficulty breathing during sleep	33	14	53	9
Sweats a lot during sleep	21	14	65	9
Gags or chokes during sleep	7	13	80	15
Complain of headaches on waking up?	3	15	82	2
Makes quick movements of the arms and legs (e.g. kicking or flailing)	3	10	73	14
Wets the bed during sleep	3	10	96	1
Sleep related problems or behaviours occurring during the day				
Lacks energy during the day	45	31	24	2
Excessively sleepy during the day	38	35	28	2
Irritable during the day	33	31	36	2
Difficulty staying awake during the day	30	28	43	2
Actually falls asleep during the day	26	32	41	1
Take naps during the day	25	24	49	4
Drowsy during the day	25	22	53	11
Miserable during the day	21	34	56	3
Irresistible sleep attacks	16	13	70	9
Appears more active than others during the day	14	23	63	6
Become so weak that he/she falls to the ground or has to lie down before falling, usually in response to laughing or crying	1	3	97	2
Feeling of being paralysed upon awakening, even though he/she is aware of his/her surroundings	0	1	99	9

Table 4
Proportion of adults with DS with different categories of sleep problems as reported by family carers

		OSA		Total
		Yes	No	
Behavioural sleep problem	Yes	17 (18%)	30 (31%)	47 (48%)
	No	7 (7%)	43 (44%)	50 (52%)
Total		24 (25%)	73 (75%)	97 (100%)

Note: There was missing data on OSA from family carers for 3 adults with DS.

Table 5
Mean BMI for adults with DS with and without reported sleep problems

	Yes	No
Behavioural sleep problem	30.6 (SD 7.6)	28.5 (SD 5.7)
OSA*	32.6 (SD 6.4)	28.7 (SD 6.7)

*p<0.05 (2-tailed)

Table 6
Epworth Sleepiness Scale (n=91)

Mean	8.5 (SD 5.8)
Range	0-24
0-5 (Lower normal)	35 (38%)
6-10 (Higher normal)	21 (23%)
11-12 (Mild excessive daytime sleepiness)	13 (14%)
13-15 (Moderate excessive daytime sleepiness)	13 (14%)
16-24 (Severe excessive daytime sleepiness)	9 (10%)
Behavioural sleep problem	
Yes	Mean: 9.98 (SD 6.30)*
No	Mean: 7.11 (SD 5.02)
Obstructive sleep apnoea?	
Yes	Mean: 11.8 (SD 5.9)**
No	Mean: 7.6 (SD 5.5)

**p<0.01 *p<0.05 (2-tailed)

Note: A total ESS score was missing for nine individuals as not all ESS items were responded to.

Table 7

Selected family carers' responses to 'How does the sleep of your relative with DS affect your sleep?'

"23 years of sleeping lightly and having to get up to my son. Often up for an hour settling him and then getting back to bed but lying there listening out until I am sure he was back to sleep. Him sleeping propped up against me sitting at top of the bed when he had chest infections and couldn't sleep lying down. After 23 years I have little or no sleep pattern left myself and get very little sleep and I am very restless at night."

"If he's talking too loudly I have to get up or if he's trying to make sense of something he gets stressed and needs reassurance."

"I hear her when she gets up to fix her bed, if she cannot sleep I go in with her until she settles, on holidays she will not sleep on her own which mean I spend more time in her bed rather than my husbands."

"I listen in case she stops breathing, I also get woken up when she has nightmares or if just goes to the loo. She does get up every night for the loo"

"We cannot go to bed until all routines have been completed Then we need to keep checking on her to see if she has fallen asleep, to help her into bed. Sometimes she appears to be asleep in a crouching position but when approached she wakes up and waves us away says "No, I all right. I not tired"."

Table 8

Selected family carers' responses to 'How is the sleep of other family members affected by the sleep of your relative with DS?'

"Screaming awake every hour"

"They are kept awake by the talking out loud"

"The noise of the cpap machine can affect others nearby."

"Whoever is in the house is disturbed by shouting out."

"Each member of the family take it in turn to put her to sleep each night and stay with her. Again, she sleeps in very funny positions so the person with her spends a lot of time turning her around during the night or putting the duvet back on again."