



# Sleep problems and language development in toddlers with Williams syndrome



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## ABSTRACT

Sleep and related maternal beliefs were assessed in a narrow age range of 18 children with Williams syndrome (WS) and 18 typically developing (TD) children. WS is a rare genetic disorder characterised by a complex physical, cognitive and behavioural phenotype. High prevalence of sleep difficulties in older children and adults with WS have been reported. Parents completed 6 questionnaires: the Brief Infant Sleep Questionnaire, Infant Sleep Vignettes Interpretation Scale, Pittsburgh Sleep Quality Index of Parents, Child Behaviour Checklist, MacArthur Communicative Development Inventory for Infants – Words and Gestures, and the Major (ICD-10) Depression Inventory. Compared to TD children, those with WS had shorter night sleep, more night wakings and wakefulness according to parental report. Regression analyses revealed that a proportion of the variance in language development scores in WS children could be explained by night sleep duration. Compared to control parents, the mothers of the WS group were more likely to describe their child's sleep as problematic and had higher rates of involvement with child sleep, yet they had a lesser tendency to interpret sleep problems as signs of distress and a greater tendency to emphasise limit setting. Approximately half of both groups of mothers experienced poor sleep quality. This was also related to maternal mood, and night wakefulness in the children with WS. This is the first study to quantify sleep difficulties in young children with WS in a narrow age range using maternal report. The possible negative effects on maternal sleep and mood, and the link between night sleep and language development in young children with WS, requires further detailed investigation.

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## 1. Introduction

Sufficient sleep is defined as an amount that is conducive to effective social and neuropsychological functioning and is important to healthy development in children (Dahl, 1999; Hill, Hogan, & Karmiloff-Smith, 2007). There is an extensive body of evidence demonstrating that insufficient sleep interferes with higher cognitive functioning (e.g., Randazzo, Muehlbach, Schweitzer, & Walsh, 1998; Walker & Stickgold, 2006), and contributes to child behavioural problems (e.g., Sadeh, Gruber, & Raviv, 2002). There have been fewer studies examining the effects of sleep quality in typically developing (TD) infants and

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toddlers but mounting evidence suggests that insufficient sleep, and frequent night wakings in the early years are associated with a variety of short- and long-term negative outcomes such as lower performance in language and spatial tasks, higher levels of impulsivity and hyperactivity (Dearing, McCartney, Marshall, & Warner, 2001; Touchette et al., 2007), larger weight for length (Tikotzky et al., 2010), lower scores on measures of mental and motor development (Dearing et al., 2001; Freudigman & Thoman, 1993; Scher, 2005).

Estimates of sleep difficulties amongst TD infants and toddlers in Australia, UK, US, Canada, and a number of Asian countries suggest that 20–50% of parents report some degree of difficulty with their child's sleep, posing a large cost to health services (e.g., Armstrong, Quinn, & Dadds, 1994; Hiscock & Wake, 2001; Johnson, 1991; Martin, Hiscock, Hardy, Davey, & Wake, 2007; Mindell, Sadeh, Wiegand, How, & Goh, 2010), and reports of associated negative mental and physical health for parents (e.g., Martin et al., 2007). Early sleep problems are often presumed to be temporary however, this is typically not the case, and may persist into childhood (Touchette et al., 2007; Zuckerman, Stevenson, & Bailey, 1987). Touchette et al. (2005) found that a third of infants who were sleeping less than 6 h per night at 5 and 17 months, continued to have this problem at 29 months. Even when insufficient sleep at 2.5 years increased to sufficient levels by 3.5 years, lower scores on a spatial task, and higher scores on a hyperactivity–impulsivity measure were found at age 6 compared to those sleeping more than 10 h a night. This is of particular concern not only for TD children, but also those with developmental disorders as sleep problems may compound their existing cognitive and behavioural difficulties, and they are inherently more vulnerable to sleep difficulties (Bartlett, Rooney, & Spedding, 1985; Lancioni, O'Reilly, & Basili, 1999).

The main aim of the current study was to investigate sleep in toddlers with WS. To date, there appear to be no published studies providing quantifiable reports investigating sleep specifically in toddlers with this rare developmental disorder. WS is caused by a hemizygous microdeletion of some 28 genes on chromosome 7q11.23 (Tassabehji, 2003). The incidence of WS is approximately 1 in 20,000 live births (Morris, Demsey, Leonard, Dilts, & Blackburn, 1988). The main cognitive characteristics of WS include overall IQ scores between 55 and 69 (Mervis et al., 2000; Searcy et al., 2004), a 'hyper-social' personality profile, relatively good face recognition and language skills, and poor visuo-spatial skills (Annaz, Karmiloff-Smith, Johnson, & Thomas, 2009; Donnai & Karmiloff-Smith, 2000).

Studies examining sleep in individuals with WS indicate that sleep is problematic for this population. Annaz and colleagues (2011) reported that 97% of school-aged children with WS experienced bedtime resistance, sleep anxiety, night wakings, and daytime sleepiness. More recently, actigraphy measures revealed that, compared to TD children and children with Down syndrome, children with WS (age range: 6–12 years) had significantly longer sleep latencies as well as parental reports of bed-wetting and body pains (Ashworth, Hill, Karmiloff-Smith, & Dimitriou, 2013). A large proportion of adolescents and adults (age range: 17–35 years) with WS are also reported to experience daytime sleepiness, nocturnal leg discomfort, and fragmented sleep as measured by actigraphy (Goldman, Malow, Newman, Roof, & Dykens, 2009). Polysomnography studies have demonstrated differences in sleep architecture between TD and WS children. Arens et al. (1998) found that children with WS spent double the amount of time awake after sleep onset, more time in stages 3 and 4 (slow wave sleep), and less in stages 1 and 2. Reduced sleep efficiency (sleep onset to offset time as a proportion of time in bed), and REM sleep, as well as increased slow wave sleep, respiratory-related arousal, and restlessness have been seen in studies with children (age range: 2–18 years, Mason et al., 2011), and adolescents and adults (age range: 14–29 years, Bódizs, Gombos, & Kovács, 2012; Gombos, Bódizs, & Kovács, 2011).

Due to the mounting evidence seen in children and adults with WS, Annaz, Hill, Ashworth, Holley, and Karmiloff-Smith (2011) argued that sleep difficulties should be considered one of the defining symptoms of WS, a sentiment also shared by some clinical professionals. The ages of the participants in the studies reviewed above were wide in range, and there appear to be no studies investigating sleep specifically in WS in the first few years of life. Investigating sleep in the early years is essential as it is unclear if sleep problems are a phenotypic characteristic of WS (Bódizs et al., 2012) or whether sleep difficulties develop as a result of other domain specific factors associated with WS such as issues with attention (e.g., Scerif, Cornish, Wilding, Driver, & Karmiloff-Smith, 2004), hypersociability or behavioural problems (Bódizs et al., 2012; Mason et al., 2011).

Sleep involves finely tuned multidimensional processes such as psychological processes, biochemistry, genetics, and responses to external environmental cues (Hill, 2011). Therefore multiple types of regulation may affect the complex sleep-wake system, which consequently may impact on waking life and family functioning. Sleep problems in early childhood may have a negative impact on multiple factors such as behaviour, cognition, language, and health of a child (e.g., Dearing et al., 2001; Scher, 2005; Tikotzky et al., 2010; Touchette et al., 2007). Furthermore, parental involvement may contribute to infant sleep disturbances and infants' abilities to self-soothe (Sadeh, Flint-Ofir, Tirosh, & Tikotzky, 2007). This may particularly be the case in primary caregivers of children with developmental disorders (e.g., Meltzer, 2008) as they may be at increased risk of associated stress, and experience more parental anxiety. There may be bidirectional effects where sleep difficulties in children or parents may impact on each other (e.g., Meltzer & Mindell, 2007; Wayte, McCaughey, Holley, Annaz, & Hill, 2012). Therefore, in the current study, a range factors were explored in six questionnaires assessing child sleep, parental cognition in relation to early child sleep, maternal sleep, and mood measures. The main goals of the study were: (i) to compare sleep in TD toddlers and those with WS from a narrow age range, using parental reports; (ii) to explore the relationships between sleep and behaviour and language outcomes; and (iii) to examine the relationships between child sleep and related maternal beliefs, maternal sleep, and maternal mood.

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