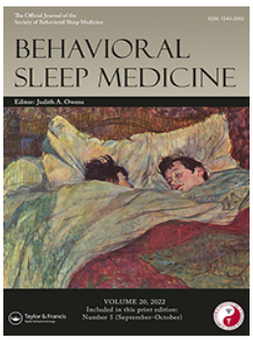

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Direct assessment of overnight parent-child proximity in children with behavioral insomnia: Extending models of operant and classical conditioning

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ABSTRACT

Introduction: Explanatory models of behavioral insomnia typically draw on operant learning theory with behavioral techniques focused on altering parent-child interactions to improve sleep. However, there are no data describing parent-child interactions overnight beyond parent report. In this study we used radio frequency identification technology to quantify parent-child proximity overnight in two groups at elevated risk of behavioral insomnia, Angelman syndrome (AS) and Smith-Magenis syndrome (SMS).

Materials and Methods: Nineteen children aged 4–15 years (8 with AS, 11 with SMS) participated in a week-long at-home assessment of sleep and overnight parent-child proximity. Sleep parameters were recorded using the Philips Actiwatch 2 and proximity data were recorded using custom-built radio frequency identification watches.

Results: Three patterns of proximity data between parent-child dyads overnight were evident: "checking" (six with AS, five with SMS), "co-sleeping" (four with SMS) and those who had "no proximity" overnight (two with AS, two with SMS). In the AS group, 25.45% of actigraphy-defined wakes resulted in a parent-child interaction. In the SMS group, 39.34% of wakes resulted in a parent-child interaction. Children who interacted with their parents when settling to sleep were not significantly more likely to interact at waking.

Discussion: The novel application of radio frequency identification technology is a feasible method for studying overnight parent-child proximity. Profiles of proximity between participants that are not closely aligned with operant models of behavioral insomnia were evident. These results have significant implications for the etiology of poor sleep and the application of behavioral sleep interventions.

Introduction

Behavioral insomnia is common in childhood (Vriend & Corkum, 2011) and associated with difficulties in daytime behavior, mood and physical health (Sadeh, 2007). In individuals with intellectual disability (ID), rates of insomnia are greatly elevated (Agar et al., 2021; Richdale et al., 2000; Van de Wouw et al., 2012) and sleep problems are persistent (Quine, 1991; Wiggs & Stores, 1996). Causal models of insomnia often impute two aspects of behavioral theory – associative and operant learning. According to the associative model, poor sleep results from established sleep-onset associations in the initial settling period not being replicated at waking (Vriend & Corkum, 2011). These are often considered to result from poor sleep hygiene. If children learn to associate initial sleep onset with parental presence, they may be unable to self-soothe at bed time and waking, and demonstrate

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signaling behaviors (such as crying, leaving the bedroom, challenging behavior) that are ultimately rewarded by the reinstatement of initial sleep onset conditions. According to the operant model, these signaling behaviors can be strengthened by environmental reward, particularly parental attention (Didden et al., 2011). As the child's signaling behavior is positively reinforced, caregivers' provision of attention is negatively reinforced by their distress at their child's signaling being relieved and their own sleep reinstated. The behavioral model therefore predicts that children who interact with their parents at settling and sleep onset will be less able to self-soothe and therefore more likely to interact with their parents at waking. This principle underpins many interventions for behavioral insomnia. However, no studies have directly quantified parent-child interactions and child waking overnight.

The paucity of data directly quantifying aspects of the behavioral model of insomnia is likely due to methodological challenges. Asking parents to complete diaries of their child's sleep and nighttime interactions is burdensome and prone to recall error (Dayyat et al., 2011; Sadeh, 1996). Videosomnography can identify interactions in the child's bedroom, in view of the camera, and has been used with typically developing children and those with ID (Agar et al., 2020; Kim et al., 2014; Teti et al., 2010). However, this approach provides a limited picture of overnight interactions by omitting those occurring away from the view of the camera. These interactions are highly relevant to the behavioral model of insomnia. Therefore, in this study we designed a novel assessment approach to quantify parent-child interactions overnight. We asked children and parents to wear radio frequency identification (RFID) sensors in custom-built wristwatch apparel for 3 to 6 nights. These sensors generate a signal strength indicative of the proximity of the child to the parent. Our first aim was to determine the feasibility and tolerability of this approach to assessing parent-child interactions.

To optimize evaluation of this methodology, we focused on children with Angelman syndrome (AS) and Smith-Magenis syndrome (SMS) for whom sleep is very poor with a high likelihood of overnight parent-child interactions. These children are therefore at saturated risk for behavioral insomnia. AS results from disruption to the ubiquitin ligase E3A (UBE3A) gene on the maternal chromosome 15q11-q13, through deletion, mutation, uniparental disomy or imprinting center defect (Dagli et al., 2011). SMS is caused by changes to the RAI1 gene on chromosome 17p11.2, an area implicated in the regulation of several circadian genes, through deletion or mutation (S. R. Williams et al., 2012). This change results in a shifted circadian rhythm with excessive daytime sleepiness and early waking (De Leersnyder et al., 2001; Trickett et al., 2018, 2020).

The behavioral phenotype of both syndromes is well-established, with strong sociability and preference for adult over peer interaction in AS, and preference for a specific caregiver in SMS (Adams et al., 2011; Wilde et al., 2016, 2013). In both groups, social approach behaviors occur when adult attention is unavailable (Heald et al., 2013; Wilde et al., 2016, 2013). Additionally, both groups evidence an elevated prevalence of insomnia, and "general" sleep difficulties which encompass caregiver concerns around sleep and behavior at waking (Agar et al., 2021).

In addition, impulsivity is common (Oliver et al., 2011) resulting in a likely lower threshold for signaling behaviors. Given poor adaptive functioning in these syndromes and elevated rates of self-injury and aggression (C. A. Williams et al., 2010; Arron et al., 2011; Udwin et al., 2001), parents may also be more likely to respond to these signaling behaviors. It is likely that these aspects of the phenotype are related to high levels of parent stress in these groups (Goldman et al., 2012; Heald, 2018). As such, they provide an optimal context to evaluate the feasibility of empirical quantification and description of parent-child interactions, with resultant methodological advances improving clinical outcomes for these high-risk groups.

This study has three aims:

(1) To determine the feasibility of RFID technology to assess parent-child interactions.
(2) To quantify and describe parent-child proximity and sleep overnight in AS and SMS.
(3) To assess the co-occurrence of parent-child proximal episodes during critical windows of actigraphy-defined settling, sleep onset and waking. Given the model of sleep associations, it is hypothesized that children who interact with their parents at settling and sleep onset will be less able to self-soothe and therefore more likely to interact with their parents during night waking.

Materials and methods

Participants

As part of a wider sleep study (approved by the Science, Technology, Engineering and Mathematics Ethical Review Committee at the University of Birmingham), 36 families were invited to participate. Parents provided informed consent for themselves and their child, with ongoing assent sought from the child. Of these, seven children (19.4%, five with AS, two with SMS) were unable to tolerate the RFID watch overnight. Two families delayed starting their sleep week by >4 days, thus missing the window for proximity data collection due to the watch battery life (5.6%). For eight families data could not be retrieved from the child's sensor or from a parent's sensor (22.2%). Thus, 19 children and 35 caregivers were included, eight children (mean age = 8.38 years, SD = 3.15) and 14 caregivers (mean age = 45.64 years, SD = 7.01) in the AS group, and 11 children (mean age = 11.13 years, SD = 2.37) and 21 caregivers (mean age = 43.52 years, SD = 5.87) in the SMS group. In both groups caregivers reported a mode education level of Polytechnic/University degree, National Vocational Qualification (NVQ) 4, or equivalent (range: Fewer than 5 General Certificate of Secondary Educations or O Level's (grades A-C), NVQ 1 or, Business Technology and Education Council First Diploma – Masters/ Doctoral degree, NVQ 5, or equivalent). Table 1 reports the children's sleep parameters and characteristics of each family who agreed to participate.[1]

There were no significant differences in any sleep parameters, age, gender, sleep hygiene or caregiver wellbeing scores between those who had available proximity data and those who did not. However, there was a significant difference identified specifically between those who could and could not tolerate the RFID watches.[2] Those who could not tolerate the RFID watches spent significantly more time in bed (t (19.94) = −3.480, p = .002).

Procedure

Each child and participating caregiver(s) wore the RFID watches overnight for the duration of their sleep study week. Each RFID "watch" consisted of a sensor, powered by a 3 V coin battery, in an enclosed watch face attached to a strap (see Figure 1).

Concurrently, the child wore a Phillips Actiwatch 2 and the parent completed a sleep diary to monitor their child's bed time, get up time and waking behaviors. Actigraphy data were cleaned using a standardized protocol developed by Trickett et al. (2017). This protocol was developed to maximize accuracy of the data and remove artifact which can make actigraphy unreliable (Acebo et al., 1999). For example, sleep intervals would be excluded if the diary indicated that the watch had been removed overnight, or adjusted if the diary suggested the child was sedentary rather than asleep in the early evening (see Appendix 1 for the full protocol). This protocol is intended to standardize and make explicit the visual inspection process that typically occurs as part of cleaning actigraphy using a sleep diary to remove artifact (see, Berger et al., 2008).

As part of the wider study, parents also completed several questionnaires. The Family Inventory of Sleep Habits (FISH, Malow et al., 2009) asks parents to report on their child's bedtime routine and broader sleep hygiene in the past month, using a five point Likert scale. Higher FISH scores are indicative of better sleep hygiene. The Modified Simonds & Parraga Sleep Questionnaire (MSPSQ, Simonds & Parraga, 1982) is a parent-report measure of an individual's poor sleep, adapted for use in individuals with developmental disabilities by Wiggs and Stores (1996). Higher scores on the MSPSQ are indicative of greater sleep disturbance, with a cutoff score of 56 suggested to indicate "poor sleepers" (Johnson et al., 2012). In this study, total scores on both measures were calculated and

[1]It is important to note that children were not recruited to this study on the basis of having two heterosexual parents. Three single parent families were recruited to the study. However, the number of mothers and fathers recruited allows consideration of potential differences in maternal- and paternal- child interaction at night.

[2]Due to the size of the groups, these potential differences were considered regardless of syndrome. However, visual inspection of the data revealed these differences were likely driven by two children in the SMS group who could not tolerate the RFID watches.

Table 1. Demographic and sleep characteristics and actigraphy derived sleep parameters of children in each syndrome group and their participating caregivers.

Participant	Gender	Exact Age (years)	Bed Time (hh:mm) Mean (SD)	Get Up Time (hh:mm) Mean (SD)	Time in Bed (hh:mm) Mean (SD)	Total Sleep Time (hh:mm) Mean (SD)	Sleep Onset Latency (mins) Mean (SD)	Sleep Efficiency (%) Mean (SD)	Wake After Sleep Onset (mins) Mean (SD)	MSPSQ Total Score[a]	Caregivers Participating	Primary Caregiver Anxiety Score[b]	Primary Caregiver Depression Score[b]
ANG3	Male	8.53	19:12 (25)	06:37 (01:21)	11:25 (01:12)	10:44 (01:26)	4.14 (4.53)	93.87 (4.76)	7.36 (6.31)	68	Mother Father*	7	7
ANG11	Female	11.57	21:47 (31)	06:50 (42)	09:02 (54)	08:00 (01:07)	8.42 (10.47)	88.28 (4.40)	7.75 (5.02)	57	Mother* Father	12	8
ANG17	Female	10.02	20:49 (14)	05:50 (02:19)	09:01 (02:09)	07:16 (02:01)	4.57 (6.87)	79.26 (7.15)	82.86 (28.90)	70	Mother* Father	12	8
ANG20	Female	7.32	19:39 (39)	06:14 (01:40)	10:35 (01:12)	05:50 (01:59)	90.83 (95.59)	54.60 (15.63)	168.00 (116.39)	59	Mother* Grandmother	10	10
ANG23	Female	13.07	21:31(-)	06:43(-)	09:12(-)	-(-)	-(-)	-(-)	-(-)	63	Mother* Father	11	7
ANG24	Male	12.78	-(-)	-(-)	-(-)	-(-)	-(-)	-(-)	-(-)	59	Mother Father*	12	7
ANG26	Female	10.79	19:14 (20)	07:17 (34)	12:03 (35)	08:52 (01:07)	82.79 (53.69)	73.48 (8.07)	67.79 (21.34)	47	Mother*	10	11
AS_N_1	Male	5.61	21:01 (42)	04:41 (01:13)	07:40 (01:02)	05:52 (48)	11.57 (14.51)	76.70 (7.77)	75.64 (26.99)	76	Mother* Father	8	11
AS_N_3	Female	8.11	19:20 (13)	04:42 (54)	09:21 (55)	07:17 (31)	31.71 (21.85)	78.37 (6.10)	81.93 (30.06)	81	Mother Father*	11	9
AS_N_4	Female	10.02	20:30 (47)	05:52 (59)	09:21 (01:14)	07:52 (01:00)	27.79 (24.85)	84.26 (3.90)	44.00 (14.91)	94	Mother* Father	12	8
AS_N_7	Female	4.82	19:11 (19)	05:49 (01:23)	10:37 (01:30)	08:55 (59)	21.86 (22.07)	84.49 (4.70)	59.41 (29.19)	73	Mother* Father	11	8
AS_N_8	Male	9.53	21:02 (32)	07:39 (01:54)	10:37 (02:06)	07:25 (02:13)	24.33 (9.33)	70.09 (15.11)	146.50 (104.94)	66	Mother* Father	10	6
AS_N_9	Male	14.18	-(-)	-(-)	-(-)	-(-)	-(-)	-(-)	-(-)	60	Mother* Father	12	9
AS_N_10	Male	6.68	19:11 (33)	07:11 (02:02)	11:59 (01:59)	07:40 (01:22)	13.43 (6.82)	64.18 (5.80)	198.86 (62.81)	47	Mother* Father	14	9
AS_N_11	Female	4.47	19:26 (30)	06:05 (50)	10:38 (43)	07:00 (41)	9.14 (8.35)	65.88 (6.02)	202.43 (38.02)	68	Mother*	13	10
AS_N_12	Female	15.11	-(-)	-(-)	-(-)	-(-)	-(-)	-(-)	-(-)	74	Mother* Step-Father	14	12
AS_N_13	Male	6.64	-(-)	-(-)	-(-)	-(-)	-(-)	-(-)	-(-)	72	Mother* Father	7	13
AS_N_14	Female	7.25	20:02 (47)	06:37 (51)	10:34 (01:01)	07:35 (37)	47.30 (48.21)	71.96 (4.68)	102.10 (35.90)	44	Mother* Father	8	8
Mean (SD)	7:11 (M:F)	9.25 (3.19)	20:08 (56)	06:17 (52)	10:08 (01:15)	07:42 (01:17)	29.07 (28.45)	75.80 (10.74)	95.74 (65.16)	65.44 (12.62)	-	10.78 (2.16)	8.94 (1.89)
SMS2	Male	9.94	20:15 (8)	07:07 (31)	10:52 (29)	07:10 (01:03)	14.38 (3.91)	66.32 (11.65)	193.50 (70.66)	98	Mother* Father	12	10

(*Continued*)

Table 1. (Continued).

Participant	Gender	Exact Age (years)	Bed Time (hh:mm) Mean (SD)	Get Up Time (hh:mm) Mean (SD)	Time in Bed (hh:mm) Mean (SD)	Total Sleep Time (hh:mm) Mean (SD)	Sleep Onset Latency (mins) Mean (SD)	Sleep Efficiency (%) Mean (SD)	Wake After Sleep Onset (mins) Mean (SD)	MSPSQ Total Score[a]	Caregivers Participating	Primary Caregiver Anxiety Score[b]	Primary Caregiver Depression Score[b]
SMS3	Male	10.67	20:13 (26)	04:46 (56)	08:33 (01:00)	07:03 (53)	.07 (0.17)	82.72 (6.79)	65.93 (28.02)	73	Mother* Father	11	10
SMS4	Female	10.48	20:31 (57)	04:55 (01:28)	08:23 (01:04)	06:55 (46)	27.88 (38.84)	82.86 (4.54)	44.69 (11.52)	84	Mother Father*	7	9
SMS11	Female	11.84	20:19 (20)	05:56 (27)	09:36 (16)	07:15 (14)	24.57 (19.83)	75.67 (3.23)	94.07 (15.41)	81	Mother Father*	8	9
SMS12	Female	8.99	19:08 (6)	06:02 (25)	10:54 (29)	08:13 (31)	14.92 (11.78	75.46 (6.29)	127.42 (41.80)	79	Mother* Father	9	11
SMS13	Male	-	-(-)	-(-)	-(-)	-(-)	-(-)	-(-)	-(-)	-	Mother Father*	-	-
SMS14	Female	12.21	17:52 (37)	04:16 (01:08)	10:24 (01:24)	07:08 (56)	9.71 (7.18)	68.84 (3.72)	177.93 (40.84)	79	Mother Father*	6	11
SMS15	Male	14.99	20:44 (25)	06:35 (12)	09:51 (28)	07:26 (19)	15.43 (13.13)	75.73 (4.01)	110.29 (21.48)	90	Mother*	9	10
SMS21	Male	10.87	18:49 (01:35)	05:09 (37)	10:19 (01:58)	07:28 (02:16)	12.25 (13.08)	71.12 (7.49)	134.38 (24.42)	94	Mother Father*	14	10
SMS24	Female	12.07	21:20 (25)	06:21 (53)	09:01 (58)	07:03 (32)	58.17 (24.77)	78.60 (4.44)	43.94 (7.14)	88	Mother* Father	10	8
SMS27	Female	10.38	20:37 (35)	05:46 (32)	09:09 (27)	07:54 (30)	6.38 (7.55)	86.30 (2.98)	46.19 (13.08)	85	Mother* Father	9	14
SMS34	Male	11.02	20:37 (41)	05:51 (57)	09:14 (01:00)	07:38 (01:13)	5.75 (3.68)	82.38 (5.89)	60.25 (10.18)	85	Mother*	10	9
SMS37	Male	11.76	20:03 (18)	04:53 (10)	08:50 (11)	04:56 (01:07)	101.50 (15.25)	55.18 (11.63)	54.20 (19.73)	63	Mother Father*	10	11
SMS41	Female	10.99	19:54 (33)	05:37 (23)	09:43 (46)	07:23 (47)	17.93 (17.90)	76.00 (3.70)	100.00 (18.61)	88	Mother* Father	11	8
SMS_N_1	Male	12.17	19:57 (27)	04:45 (01:29)	08:47 (01:34)	07:36 (01:28)	1.58 (2.37)	86.13 (3.77)	62.83 (13.20)	80	Mother* Father	11	5
SMS_N_2	Female	4.70	19:37 (21)	03:48 (51)	08:10 (53)	06:50 (42)	2.29 (3.61)	83.90 (4.83)	61.57 (20.33)	93	Mother* Father	12	10
SMS_N_3	Male	12.32	21:01 (46)	05:23 (01:33)	08:22 (01:31)	06:45 (36)	2.69 (3.03)	82.55 (11.53)	81.56 (69.82)	70	Mother* Father	7	7
SMS_N_4	Male	8.87	20:45 (21)	04:56 (41)	08:10 (48)	06:19 (01:04)	15.07 (22.42)	77.04 (8.52)	64.86 (23.85)	122	Mother* Father	14	9
Mean (SD)	10:8 (M:F)	10.84 (2.13)	20:06 (52)	05:25 (52)	09:18 (55)	07:07 (43)	19.45 (25.23)	76.87 (8.08)	89.62 (45.71)	84.41 (12.97)	-	10.00 (2.29)	9.47 (1.94)

[a]Total score derived from the Modified Simmons & Parraga Sleep Questionnaire, higher scores indicate greater difficulty. [b]Total score derived from the Hospital Anxiety and Depression Scale, higher scores indicate greater distress. *Denotes primary caregiver. – indicates missing data.

specific items which relate to parent-child overnight interactions were drawn from each measure to consider the validity of the proximity assessment. Parents also completed the Hospital Anxiety and Depression Scale (HADS, Zigmond & Snaith, 1983) to measure caregiver wellbeing in relation to overnight proximity, with higher scores indicative of greater distress. A cut off score of >8 on either the anxiety or depression subscale indicates "caseness" with optimal balance between specificity and sensitivity (Bjelland et al., 2002).

RFID technology

RFID sensors exchange packets of information (tag identifiers, local time, remote time, received signal strength indicator and angle) in one second intervals using ultra-low-power radio with other sensors nearby (Cattuto et al., 2010). Received signal strength indicator (RSSI) is therefore a proxy measure of proximity. Sensors begin recording when the battery is inserted. With the selected recording intervals (1s), battery life lasts four to six days. Sensors were originally designed by the SocioPatterns collaborative to study social dynamics in schools and at conferences (Mastrandrea et al., 2015; Stehlé et al., 2011) and have been used in studies of disease spread, infant interactions and animal social networks (Ozella et al., 2018, 2020; Voirin et al., 2015). These studies have generally used RFID technology with very large samples to conduct network analyses. In this feasibility study a recorded RSSI of ≥-89 was deemed indicative of proximity and hence interaction. Using firmware designed by OpenBeacon (https://github.com/meriac/openbeacon-ng) data were retrieved and analyzed according to the protocol developed for this study by the first author (see Appendix 2) to examine proximal episodes between the child's sensor (Sensor A) and the parents' sensors (Sensor B and Sensor C) overnight.[3]

Data analysis

To describe parent-child proximity overnight, each child's sleep/wake data for each 30 second epoch between 30 minutes prior to bed time and get up time was extracted from their actigraphy.[4] These sleep/wake data were plotted against the RSSI, recorded in one second epochs, between the child's sensor and both parents' sensors. All data were plotted, without imposing definitions of wake or interaction, and visually inspected to classify parent-child triad interactions based on proximity data. Fisher's Exact test and one way analysis of variance (ANOVA) were used to examine the relationships between classification groups, syndrome, sleep hygiene, sleep parameters and caregiver wellbeing.

The percentage of actigraphy-identified settling periods and wakings which overlapped with a parent-child proximal episode was calculated. In order to be considered a proximal episode, RSSI had to be recorded continuously between two sensors at −89 or less for at least a minute. For the purpose of this cutoff, episodes were deemed continuous if these epochs were not greater than 30 seconds apart, given the interval length of actigraphy. Therefore, proximal episodes vary in duration from one minute to much longer periods of proximity. Several proximal episodes may occur in quick succession, where there is a break of more than 30 seconds between recorded RSSI at −89 or less. The settling period was defined as 30 minutes prior to the actigraphy-derived time of sleep onset, to balance the need to capture as much of the bedtime routine and settling to sleep as possible whilst not capturing "playtime" and other interactions which occur long before the child is put to bed. Wakings were defined as distinct periods of movement after sleep onset (see actigraphy protocol;

[3]Sensor B and Sensor C also exchange information with each other, but these were not analyzed for this study.
[4]On eight nights actigraphy data were not usable, so diary data were used to determine the child's bed time and sleep/wake data were not plotted. This affected four participants with AS and one participant with SMS.

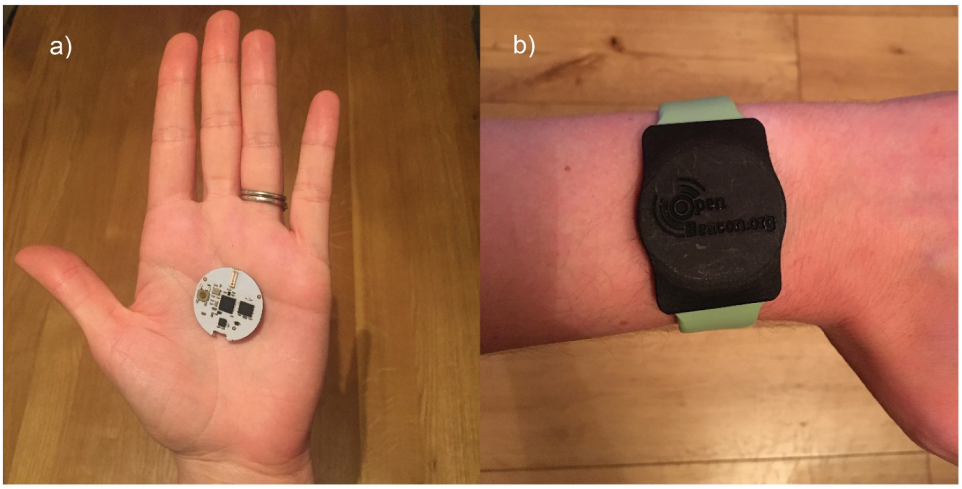

Figure 1. The sensor, with adult hand included for size reference (panel a) and the external strap and casing used to create the "RFID watch" (panel b).

Appendix 1, Trickett et al., 2017). The number of proximal episodes which occurred when the child was asleep according to actigraphy (non-waking) was also calculated, to objectively consider parent monitoring of medical equipment and child safety overnight which has been previously reported in these syndrome groups (Agar et al., 2022, 2020; Foster et al., 2010; Trickett et al., 2017). The number of children for whom a parent was present at the exact time of sleep onset was also calculated. These stringent definitions of settling, waking and proximal episode were used to reduce the risk of type 1 error.

Chi-Square and Mann-Whitney U tests were used to examine differences between syndrome groups in each parent's involvement in, and number and length of, proximal episodes at settling, sleep onset, waking, and non-waking. The relative risk of proximal episode at waking and non-waking given proximal episodes at settling and parent presence at sleep onset was calculated.

Results

Sleep characteristics derived from actigraphy were objectively poor in the nineteen children who took part in the proximity assessment. For the eight children with AS who took part, the mean sleep efficiency was 76%, indicating a substantial amount of time spent in bed not actually asleep. The mean amount of wake after sleep onset was 99 minutes in the AS group. For the 11 children with SMS who took part, the mean sleep efficiency (78%) and wake after sleep onset (81 minutes) were similarly poor, but children also woke very early for the day, with an average get up time of 05:18 am. Some children also experienced early bed times, and reduced total sleep time. Similarly, parent reports of sleep disturbance on the MSPSQ were very high, with six children in the AS group and all 11 in the SMS group scoring above the cutoff for poor sleepers suggested by Johnson et al. (2012).

Describing parent-child proximity overnight

To address the first aim, each child's RFID data, indicating proximity to their parent(s) between actigraphy-derived sleep onset and get up time, were plotted against their sleep/wake actigraphy data. Graphs are presented in order of greatest to least amount of wake after sleep onset, in Figure 2 for the AS group and Figure 3 for the SMS group.

Visual inspection suggests that overall parent-child proximity overnight was limited despite frequent and extended periods of time coded as "wake" in actigraphy. However, this varied between participants with two broad patterns of proximity data identified. The first, represented by short "bursts" of green or blue on the graph, indicates brief interactions which occurred at a physical distance. These bursts occurred in both groups, often in the 30 minutes before actigraphy-defined sleep onset but also through the night. For some children these bursts were relatively brief and isolated while for others they were extended and clustered (see ANG17, SMS14). The second pattern, represented by extended "bars" of green or blue, indicates longer episodes with children and parent(s) in closer proximity, for example, co-sleeping following a presumed waking. These periods were less common and isolated to only one night of the assessment period, but were notable for several children with SMS where proximal episodes lasted up to four hours (see SMS41).

From visual inspection, three categories of overnight parent-child proximity emerged[5]; triads where only *bursts* of proximity occurred (classified as "checking"), triads with at least one *bar* of proximity as well as bursts (classified as "co-sleeping"), and triads where there were no separate bursts or bars after sleep onset ("no proximity", see SMS_N_3 and AS_N_10). The patterns before actigraphy-derived sleep onset were not considered in this classification. Further detail on these classifications is provided in Table 2. Fisher's Exact test revealed there was no association between syndrome group and being classified as showing "checking" (p = .208), "co-sleeping" (p = .085) or "no proximity" (p = .574).

There was a significant difference in overall sleep hygiene scores between classification groups (F (2, 16) =4.747, p = .024). Post-hoc Tukey test revealed the mean sleep hygiene score was significantly higher in the "no proximity" group (M = 51.75, SD = 3.95) than the "co-sleeping" group (M = 42.75, SD = 3.20). However, the sleep hygiene scores for those who showed "checking" did not differ significantly from the "co-sleeping" or "no proximity" groups. Item-level sleep hygiene data are included in Table 2. There were no significant differences between classification groups for caregiver anxiety (F (2, 16) =.836, p = .451) or depression (F (2, 16) =.850, p = .446). The classification groups did not significantly differ in terms of time in bed (F (2, 15) =.234, p = .794), total sleep time (F (2, 15) =1.141, p = .346), sleep onset latency (F (2, 15) =1.843, p = .192), sleep efficiency (F (2, 15) =.689, p = .517), wake after sleep onset (F (2, 15) =.169, p = .846) or parent reported poor sleep according to the MSPSQ (F (2, 16) =.2.194, p = .144). However, there was a trend for co-sleepers to experience poorer sleep in all parameters except wake after sleep onset (see Appendix 3).

Co-occurrence of proximal episodes in crucial sleep/wake periods

To address the second aim, the number and length of proximal episodes ≥1 minute which occurred between a child and their mother or father (or both) was calculated, and the co-occurrence of these with periods of settling, waking and non-waking periods defined by actigraphy[6] was quantified (see Table 3). The number of wakes which resulted in a proximal episode is also included.

Overall, 310 operationally-defined proximal episodes between parents and children were recorded between settling and get up time in 19 families. Chi-Square analyses revealed there was no significant difference between syndrome groups on number of proximal episodes at settling ($\chi 2$ (1) = .757, p = .384) or having a parent present at the time of sleep onset ($\chi 2$ (1) = .135, p = .713). However, there was a significant difference between syndromes for the number of proximal episodes at waking ($\chi 2$ (1) = 12.012, p =.001), with individuals with SMS interacting more with their parents at waking (44.8% of all SMS interactions)

[5]For the purpose of classification the proximity between a parent and child was considered a "bar" if it was represented by a continuous and largely flat line lasting ≥1 hour. In contrast, "bursts" of proximity were characterized by rapid changes in RSSI value, represented by spikes on the graph.

[6]Due to missing actigraphy data, four settling interactions across three participants with AS were defined using sleep diary data. These are presented in Table 3, but are not included in later relative risk analyses.

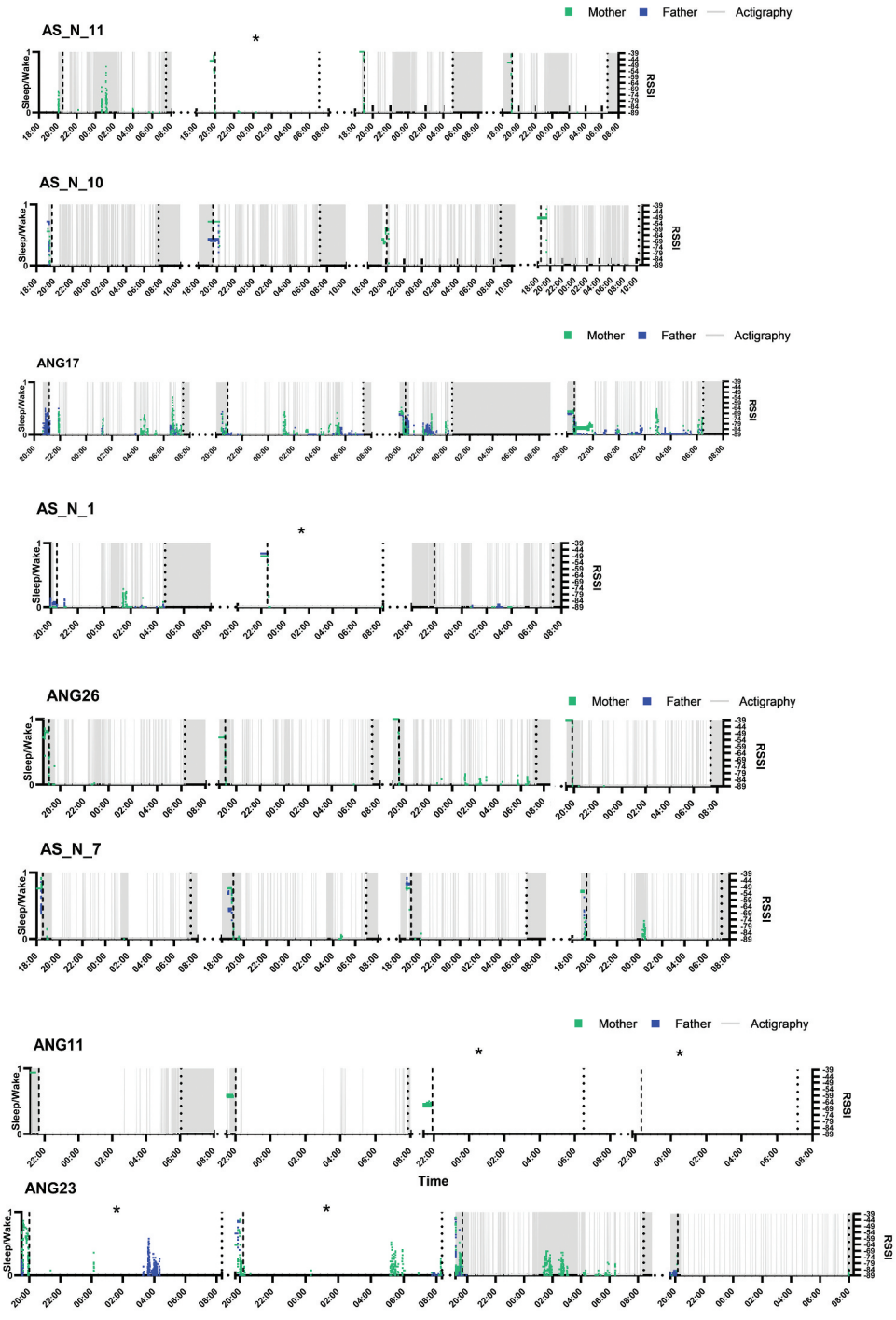

Figure 2. Proximity of children in the AS group and their parent(s) for each night (with time on the x axis) according to RSSI, with proximity to mother presented in green and proximity to father presented in blue. Higher RSSI indicates closer proximity. The gray lines indicate epochs defined as "wake" according to actigraphy. The dashed black line indicates the actigraphy-derived bed time for that night, and the dotted line the actigraphy-derived get up time. * denotes night where actigraphy data were missing, thus bed time and get up time are derived from the sleep diary.

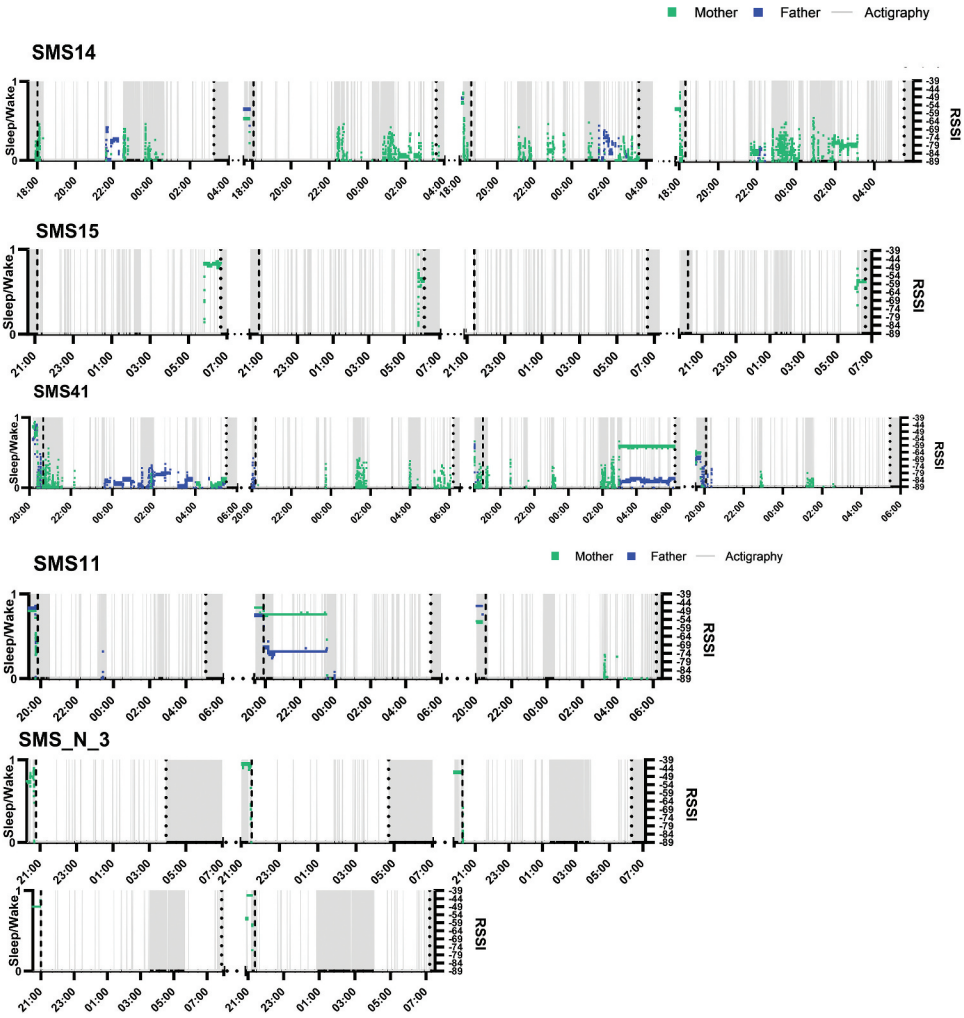

Figure 3. Proximity of children in the SMS group and their parent(s) for each night (with time on the x axis) according to RSSI, with proximity to mother presented in green and proximity to father presented in blue. Higher RSSI indicates closer proximity. The gray lines indicate epochs defined as "wake" according to actigraphy. The dashed black line indicates the actigraphy-derived bedtime for that night, and the dotted line the actigraphy-derived get up time.
*denotes night where actigraphy data were missing, thus bed time and get up time are derived from the sleep diary.

than individuals with AS (25.7% of all AS interactions). There was also a significant difference between syndromes for the number of proximal episodes at non-waking ($\chi 2$ (2) = 16.663, p = <.001), with individuals with AS having more (55.9% of all AS episodes) than the SMS group (32.8% of all SMS episodes).

To address the third aim, to further consider the co-occurrence of parent-child proximal episodes with critical windows of actigraphy-defined settling, sleep onset and waking, a series of relative risk analyses were conducted. Table 4 presents the relative risk of children interacting with a parent at waking, and also at "non-wake", *given* that they had a parent present at settle, sleep onset and waking.

In summary, these analyses demonstrate some differences in the frequency and length of episodes of parent-child proximity between syndrome groups. Individuals with SMS interacted more with their parents at waking, and individuals with AS more at non-waking. However, children who had proximal episodes with their parents at settling, sleep onset and waking were not significantly more likely to

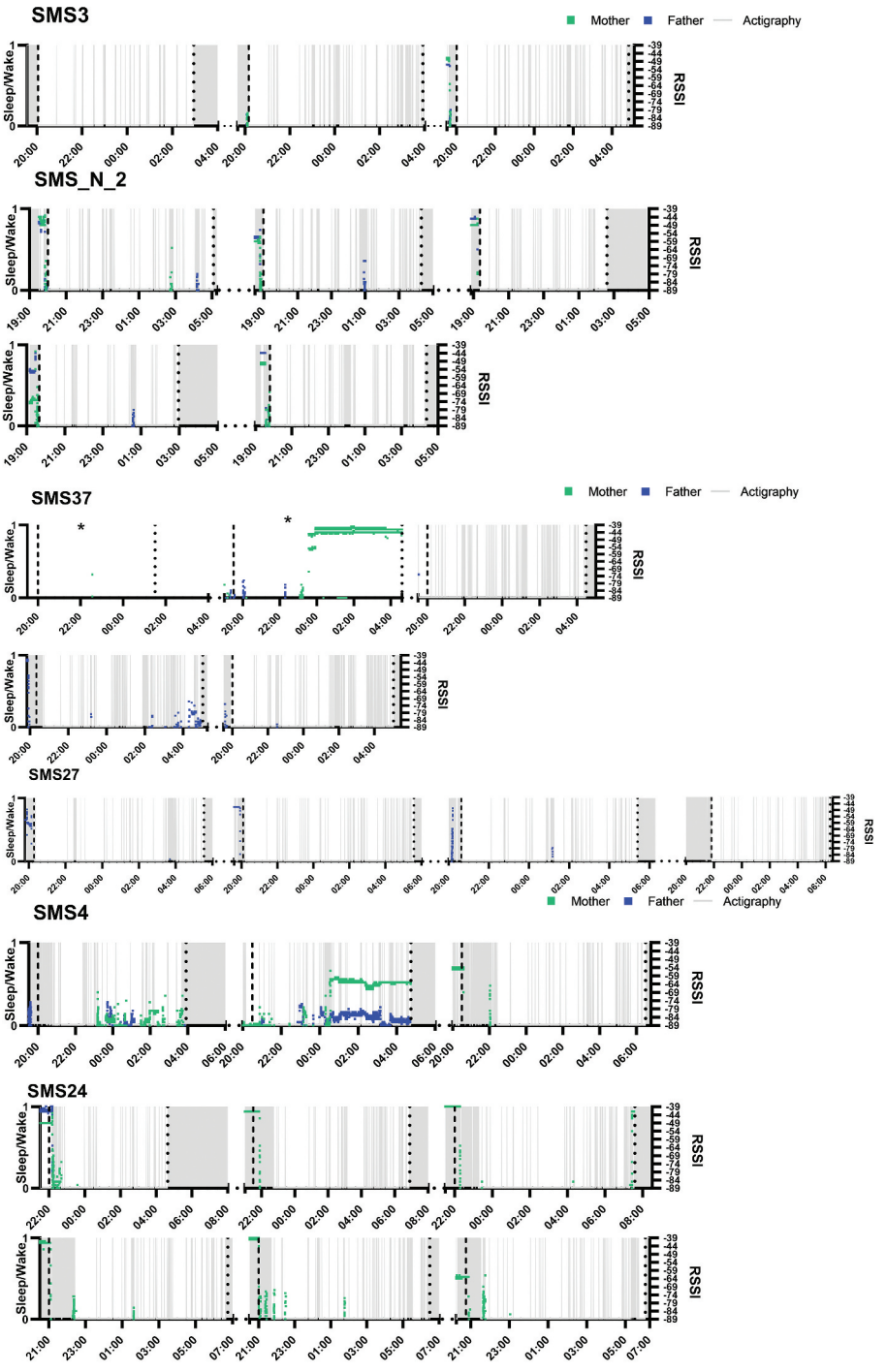

Figure 3. Continued.

have proximal episodes at waking and non-waking. Thus the data do not support the hypothesis that children who interact with their parents at settling and sleep onset will be less able to self-soothe and therefore more likely to interact with their parents during night waking.

Discussion

We evaluated a novel approach to quantifying parent-child proximity overnight in children with poor sleep and preference for adult interaction. Through combining the novel RFID data with actigraphy-defined periods of settling, sleep onset, waking and non-waking, predictions based on the behavioral model of insomnia were examined. The application of RFID technology was feasible, with 81% of the sample able to tolerate the sensors overnight, despite known sensory difficulties in both groups (Heald et al., 2020; Hildenbrand & Smith, 2012). Critically, the novel use of RFID technology overcomes the limitations of previous approaches that used videosomnography (Agar et al., 2020; Kim et al., 2014; Teti et al., 2010), providing the first description of parent-child proximal episodes across the entire night, unconstrained by location. The use of actigraphy to define periods of interest strengthens the validity of the study while reducing parent burden. Additionally, it was possible to consider the role of parental involvement at the exact time of sleep onset, a period of time crucial to understanding sleep associations and behavioral insomnia.

Overall, the findings demonstrate a variable profile of proximity between participants, despite objectively characterized poor sleep. As expected, children in both syndrome groups experienced reduced sleep efficiency and night waking (Trickett et al., 2019, 2020) with particular difficulties with early morning waking in the SMS group (De Leersnyder et al., 2001; Trickett et al., 2020). Some children

Table 2. Radio frequency identification and sleep hygiene data for each participant in each classification group.

	Syndrome	Mean Number of Checks Per Night	Mean Time between checks (mins)	Mean Length of Co-Sleep (mins)	Total Sleep Hygiene Score (FISH)	Returned child to own bed after waking? (FISH Item 22)*	Child insists on sleeping with someone else? (MSPSQ Item 52)
Checking group							
AS_N_11	AS	2.5	62.63	-	53	N/A	Never
ANG17	AS	67	6.07	-	53	Sometimes	Many times a week/daily
AS_N_1	AS	15	25.88	-	44	Occasionally	Monthly
ANG26	AS	7.5	20.32	-	51	N/A	Never
AS_N_7	AS	3.5	253.2	-	51	N/A	Never
ANG23	AS	28.75	10.33	-	47	Always	Never
SMS14	SMS	36	5.63	-	38	Sometimes	Many times a week/daily
SMS15	SMS	0.75	N/A	-	47	N/A	Never
SMS_N_2	SMS	3.6	64.43	-	48	Always	Never
SMS27	SMS	3.25	55.38	-	50	Usually	Monthly
SMS24	SMS	2.6	59.33	-	45	N/A	Never
Mean	-	15.5	56.32	-	47.91	-	-
(SD)		(19.8)	(69.5)		(4.46)		
Co-sleeping group							
SMS41	SMS	-	-	198.28	38	Sometimes	Many times a week/daily
SMS11	SMS	-	-	213.4	44	Sometimes	Many times a week/daily
SMS37	SMS	-	-	302.22	44	Sometimes	Never
SMS4	SMS	-	-	266.72	45	Sometimes	Monthly
Mean	-	-	-	245.15	42.75	-	-
(SD)				(37.22)	(3.20)		
No proximity group							
AS_N_10	AS	-	-	-	55	N/A	Never
ANG11	AS	-	-	-	46	N/A	Never
SMS_N_3	SMS	-	-	-	53	Usually	Monthly
SMS3	SMS	-	-	-	53	N/A	Never
Mean	-	-	-	-	51.75	-	-
(SD)					(3.95)		

* N/A indicates that the parent reported their child does not get out of bed during the night.

with SMS also experienced early bed times, which is likely due to their shifted circadian rhythm (De Leersnyder et al., 2001) resulting in excessive daytime sleepiness and a propensity to fall asleep earlier than typically developing peers (Smith et al., 2019). More broadly, early bed times may reflect parental desire to maximize children's sleep duration and improve the overall caregiving experience.

Two patterns of proximity were identified, encompassing briefer "checking" interactions and extended "co-sleeping" interactions. However, four children did not interact with their caregivers at all overnight and relative risk analyses suggested parental involvement at settling and sleep onset did not make parental involvement at waking or non-waking more likely in the overall sample. These findings have significant implications for our understanding of poor sleep in AS and SMS, indicating the need to examine potential causal mechanisms beyond the associative and operant conditioning accounts of insomnia. In addition, the lack of relationship between parent involvement at settling and parent involvement at waking is worthy of further investigation in children with neurodevelopmental conditions more broadly, and typically developing children, using the novel RFID approach developed in the current study.

Eleven of nineteen parent-child triads were classified as showing "checking", with parents and children interacting briefly at settling and throughout the night. Sometimes these episodes occurred when actigraphy suggested the child was asleep, possibly because of the need to adjust children's medical equipment or check that they were still asleep (Trickett et al., 2017). The time between episodes was very brief, with some parents checking on their children every hour. This suggests families may benefit from interventions designed to encourage parents not to enter their child's bedroom until an agreed checking time, and then to gradually extend the time between checks, such as graduated extinction (Rigney et al., 2018). Checking remotely via video monitoring equipment may also be a suitable approach especially when there are health concerns such as seizures. In the AS group there were significantly more brief "checking" episodes when children were asleep than awake, which may be due to the elevated rates of epilepsy reported in individuals with AS (Dan, 2009), the need to assist children with toileting or medical equipment overnight or concerns around children being in pain at waking (Agar et al., 2020). In an interview study conducted by Trickett et al. (2017) these were common parent perceptions for the cause of waking in children with AS, more common than the perception that children were waking due to a desire for parent attention or play. These concerns should therefore be considered before implementing any extinction-based sleep interventions.

Four of nineteen parent-child triads were classified as "co-sleeping", indicated by extended periods at closer proximity, often with both parents. This typically began following a period of actigraphy-defined wake and continued into a period of sleep. Children in this group had a trend toward poorer sleep parameters, and significantly poorer sleep hygiene, than children in the "no proximity" group. Most parents classified as "co-sleeping" reported that their child insists on sleeping with someone else "a few times a month – daily", and that they only return their child to their own bed after waking "sometimes". This supports the pattern identified through visual inspection where co-sleeping occurred on just one night in each child's study week, and suggests a different intervention approach may be needed. Though parents in this classification group may benefit from using extinction techniques to return their child to their own bed (Weiskop et al., 2005), given that all the children in this group have SMS, co-sleeping may have been deemed necessary by parents to keep their children safe (Agar et al., 2022). Therefore, other strategies to monitor children's safety such as a camera in the child's bedroom, or alarm systems to notify parents when their child leaves the room, may be more beneficial to families in this classification group.

Importantly, four children did not interact with parents at all overnight, despite the majority of children experiencing an average wake after sleep onset of over an hour. This is somewhat surprising given the established phenotypes of preference for adult interaction and poor sleep noted in both groups (Adams et al., 2011; Agar et al., 2021; Wilde et al., 2016) and the reliance on operant theory to explain the maintenance of insomnia in childhood more broadly (Vriend & Corkum, 2011). This "no proximity" group also had the highest sleep hygiene scores, suggesting parents were already following typical good sleep habits which would be recommended as a first-line sleep intervention. Commensurate with findings from the "no proximity" group, less than half

Table 3. The number (N) and mean length of parent-child proximal episodes which co-occurred with actigraphy-defined periods of settling, waking and non-waking for each child, presented from greatest WASO to least WASO. The Mother:Father ratio indicates the number of episodes which involved each parent. [a] indicates a single-parent family, [b] indicates a family where data could only be retrieved from one of two participating parents.

		Settling				Waking					Non-waking		
Participant	N of Nights of Data	N of proximal episodes at Settling	Mean length of episode (mm:ss) (SD)	Mother: Father Ratio	N of proximal episodes at Sleep Onset	N of Wakes	N of proximal episodes at Waking	N of wakes involving at least one proximal episode	Mean length of episode (mm:ss) (SD)	Mother: Father Ratio	N of proximal episodes	Mean length of episode (mm:ss) (SD)	Mother: Father Ratio
AS_N_11	4	3 (1†)	26:37 (01:09)	3:0	0	8	3	2	01:49 (33)	3:0[a]	0	-	-
AS_N_10	4	4	46:37 (31:11)	4:2	2	20	1	1	08:52(-)	1:1	0	-	-
ANG17	4	10	13:13 (23:19)	6:10	1	8	18	6	03:33 (03:07)	13:10	51	03:18 (02:51)	14:41
AS_N_1	3	3	18:12 (15:16)	3:3	0	5	2	2	08:48 (09:29)	2:2	10	05:55 (06:49)	4:6
ANG26	4	0	-	–	0	8	4	1	03:34 (03:26)	4:0[a]	3	04:02 (02:32)	3:0[a]
AS_N_7	4	1	01:16(-)	1:1	0	5	1	1	09:18(-)	1:0	0	-	-
ANG11	4	2 (1†)	20:02 (06:39)	2:0	0	0	-	-	-	-	0	-	-
ANG23	4	2 (2†)	26:56 (02:17)	2:2	0	1	6	1	03:08 (02:23)	6:0	12	03:16 (02:37)	6:6
Total	31	25	-	21:18	3	55	35	14	-	30:13	76	-	27:53
SMS14	4	1	02:29(-)	1:0	0	8	37	8	07:23 (14:00)	35:5	9	12:13 (13:58)	9:1
SMS15	4	0	-	-	0	9	0	0	-	-	3	35:21 (15:29)	3:0[a]
SMS41	4	12	02:38 (02:29)	10:6	1	10	26	7	12:40 (38:58)	20:6	12	16:36 (18:58)	7:6
SMS11	3	2	54:00 (59:23)	2:2	1	7	3	3	39:33 (66:11)	2:1	2	03:57 (02:31)	2:0
SMS_N_3	5	5	22:34 (09:40)	5:0[b]	0	3	0	0	-	-	0	-	-
SMS3	3	1	11:11(-)	1:1	0	2	0	0	-	-	0	-	-
SMS_N_2	5	9	12:47 (08:56)	9:8	1	10	5	4	01:28 (51)	1:4	0	-	-
SMS37	5	0	-	-	0	5	5	1	02:08 (49)	0:5	7	47:54 (112:09)	3:4
SMS27	4	4	30:33 (40:18)	0:4[b]	1	3	0	0	-	-	4	06:09 (06:33)	0:4[b]
SMS4	3	2	31:40 (38:49)	1:2	1	2	2	1	132:50 (181:39)	2:2	19	09:07 (11:33)	16:7

(*Continued*)

Table 3. (Continued).

	Settling						Waking				Non-waking		
Participant	N of Nights of Data	N of proximal episodes at Settling	Mean length of episode (mm:ss) (SD)	Mother: Father Ratio	N of proximal episodes at Sleep Onset	N of Wakes	N of proximal episodes at Waking	N of wakes involving at least one proximal episode	Mean length of episode (mm:ss) (SD)	Mother: Father Ratio	N of proximal episodes	Mean length of episode (mm:ss) (SD)	Mother: Father Ratio
SMS24	6	3	02:34 (01:13)	3:0	0	2	0	0	-	-	1	09:22(-)	1:0
Total	46	39	-	32:23	5	61	75	24	-	59:19	57	-	41:22

† Settling periods were identified using parent diary due to missing actigraphy data.

Table 4. Relative risk of parent-child interactions at waking and non-waking given parent-child interaction at settle, sleep onset and wake (n = 18).

Relative Risk	Given		
	Parent at Settle	Parent at Sleep Onset	Parent at Wake
Parent at Wake	1 (0.42–2.40)	1.57 (0.85–2.92)	/
Parent at Non-Wake	-	1.31 (0.64–2.68)	1.38 (0.58–3.33)

- Denotes incalculable statistic due to an empty cell.

of the total sample had a parent present at the time they fell asleep (five with SMS and two with AS). For those who did have a parent present, this only occurred on one or two nights of the assessment, suggesting children were not solely reliant on parent presence to fall asleep, though there may be other associative cues that were influential. Together, these findings suggest that a behavioral intervention, with the aim of correcting inappropriate sleep associations and reducing parent-child interactions at settling and waking, might be successful in extinguishing signaling behaviors and responses, but will not necessarily improve children's sleep. This interpretation is further supported by the relative risk analyses which found that children were not at significantly higher risk of having a parent at wake or non-wake if they had a parent present at settling or even sleep onset specifically. Furthermore, in 75% of wakings in the AS group and 60% in the SMS group children were able to re-settle themselves to sleep without parental support, suggesting there are likely broader mechanisms underpinning poor sleep in these groups. It is plausible that signaling behaviors are reinforced on a lean and/or variable schedule, but it is also plausible that biological factors such as pain or natural circadian rhythms are contributing to behavioral insomnia in these groups (Agar et al., 2020; De Leersnyder et al., 2001). This requires further empirical evaluation.

The results of this study show that RFID technology can be used alongside actigraphy to assess proximity between children and their parents at settling and overnight as part of an assessment package to identify specific intervention targets for poor sleep. However, the challenges of applying this technology to a group of children with sensory difficulties must be acknowledged, as seven of the thirty-six children were not able to tolerate the RFID watches. Future research may therefore benefit from implementing a systematic desensitization procedure with "dummy" watches and reward systems, as in Thackeray and Richdale (2002). In addition, while feasible, the extraction and matching of actigraphy and proximity was effortful, with data processing time per participant often exceeding eight hours, depending on the number of proximal episodes. However, it may be possible to automate this process through further research and future studies may benefit from using machine learning approaches to classify the data. Given that cameras were not used to corroborate findings from the RFID sensors, it is possible that the results were impacted by undetected measurement errors. For example, the "no proximity" group may have experienced a loss of signal overnight rather than a lack of parent-child interaction. However, given that the sensors recorded interactions between children and parents up to the point of sleep onset, and overnight interactions between two parents in the majority of this group, this seems unlikely. The sample size was modest and limited to individuals recruited for a "sleep problem" in order to maximize evaluation of feasibility. It is therefore not possible to draw conclusions about the frequency, timing or duration of nighttime parent-child proximity in all individuals with AS and SMS based on these data. However, the study highlights that it is possible to examine parent-child proximity at objectively-defined periods of settle, sleep onset, wake and non-wake using wearable, tolerable RFID technology, and identifies some families for whom these episodes could be problematic and some for whom episodes are brief and/or non-existent whilst reducing the assessment burden on parents. The classification groups identified are meaningful and appear to map onto clinical differences in the

child and parent data at a trend level. Future larger scale studies should incorporate proximity sensors and extend this classification approach to explore differences in caregiver wellbeing and child sleep more robustly.

Taken together, the findings provide an initial challenge to an exclusively operant understanding of behavioral insomnia in these groups by identifying a group of children, with objectively defined poor sleep, who do not interact with their caregivers at all overnight. The use of RFID data to classify overnight parent-child proximity highlights the different approaches taken by caregivers and may allude to specific concerns around safety and/or pain which should be addressed in the assessment and intervention process. The data also highlight the level of demand experienced by some caregivers and the variability in approaches taken by parents in interacting with their children overnight, from hourly checking to long periods of co-sleeping. These results have significant implications for our understanding of the etiology of poor sleep in children with AS and SMS and the application of behavioral sleep interventions in neurodevelopmental conditions more broadly.

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