



Sleep in Children with Autism Spectrum Disorder: A Narrative Review and Systematic Update

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Abstract

Purpose of Review Sleep problems are a common comorbidity for children with autism spectrum disorder (ASD), and research in this area has a relatively long history. Within this review, we first outline historic patterns in the field of sleep and ASD. Second, we conducted a systematic update and coded these studies based on their alignment with historic patterns. Research on ASD and sleep over the past two decades has primarily focused on four principal areas: (1) documenting the prevalence and types of sleep problems; (2) sleep problem treatment options and efficacy; (3) how sleep problems are associated with other behavioral, contextual, or biological elements; and (4) the impact of child sleep problems on families and care providers. The systematic update in this paper includes empirical studies published between 2018 and 2021 with terms for sleep and ASD within the title, keywords, or abstract.

Recent Findings In sum, 60 studies fit the inclusion/exclusion criteria and most fit within the historic patterns noted above. Notable differences included more global representation in study samples, studies on the impacts of COVID-19, and a growing body of work on sleep problems as an early marker of ASD. The majority of studies focus on correlates of sleep problems noting less optimal behavioral, contextual, and biological elements are associated with sleep problems across development for children with ASD.

Summary Recommendations for future directions include continued expansion of global and age representation across samples, a shift toward more treatment and implementation science, and studies that inform our mechanistic understanding of how sleep and ASD are connected.

Keywords Sleep · Autism · Actigraphy · Review · Circadian

Introduction

Autism spectrum disorder (ASD) is a common neurodevelopmental disorder (NDD) characterized by social/communication difficulties with the presence of restrictive or repetitive behaviors (American Psychiatric Association, 2013). With a

worldwide prevalence of roughly 1–2% [1], ASD is a well-documented NDD that impacts millions of individuals, their families, and our community care systems. Although sleep problems are not a part of the core ASD diagnostic criteria, they are among the most common comorbid symptoms reported by families and medical care providers [2–5], even before children receive a diagnosis [6]. Sleep problems are reported for up to 80% of individuals with ASD, including young children, adolescents, and adults [7]. Treatment for sleep problems is often a standard part of medical screening and care for children with ASD [8, 9]. Therefore, to provide medical care practitioners and families with the most up-to-date research information, this two-part review provides (1) a narrative summary of ASD and sleep research over the past twenty years and (2) a systematic review of ASD and sleep advances in the past 3 years (2018–2021).

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Narrative Summary of ASD and Sleep Research

Research on ASD and sleep over the past two decades has primarily focused on four principal areas: (1) documenting the prevalence and types of sleep problems present in ASD, (2) treatment options for sleep problems in ASD, (3) how sleep problems are associated with behavioral and other biological elements of ASD, and (4) the impact of sleep problems on families and care providers.

Autism Sleep Problem Prevalence and Type

It is estimated that 40 to 83% of individuals with ASD have some form of sleep disturbance [4, 5, 10–14]. Sleep problems are among the first concerns that parents report in young children later diagnosed with ASD and are similarly common in adolescence and adulthood [15–17]. It is important to note that sleep problems are not present for all individuals with ASD, though they are common and relatively stable across age groups [12, 18, 19].

Autism is a diverse condition with noted heterogeneity—similarly, sleep problems in ASD are just as diverse. Sleep problems in ASD can include insomnias, parasomnias, sleep-disordered breathing, circadian rhythm disorders, daytime sleepiness, and/or sleep movement-related disorders [4, 5, 13, 15, 20–23]. Like in the general population, insomnia is the most common type of sleep problem in individuals with ASD and often includes prolonged sleep onset latency, extended night awakenings, and early morning rise times [4, 24–26].

Treatment Options for Sleep Problems in Autism

Education is often the first step in the treatment of sleep problems in ASD. Studies of parent/caregiver education programs focus on providing developmentally appropriate sleep information in various forms along with basic behavioral approaches for families to support optimal sleep [27]. For example, sleep-related information provided via pamphlets did not improve child sleep problems [28], but small group information sessions have been effective in improving parental sleep perceptions and child sleep onset delay [27, 29].

Currently, a behavioral treatment approach to sleep problems in ASD is considered a best practice. Behavioral treatment programs often start when children are young, with age-appropriate modifications as sleep needs and/or behaviors change [30]. Behavioral treatments frequently include cognitive-behavioral approaches, extinction protocols, sleep hygiene, chronotherapy, scheduled awakenings, sleep restriction, visual supports, stimulus fading, and reward programs. Given the diverse nature of individuals with ASD and their sleep problems, current field

recommendations include the use of an individualized treatment approach that starts with education and behavioral sleep treatments [27]. If these approaches are not successful, then a combined behavioral and pharmacological treatment plan is often recommended [31].

Currently, there are no Food and Drug Administration (FDA) approved medications for sleep problems in children with ASD, but off-label medication use is common. Up to 40% of children with ASD receive medication to assist with sleep difficulties [32]. Pharmacological treatments for sleep problems in ASD most commonly include melatonin and may also include alpha-agonists, anticonvulsants, antidepressants, atypical antipsychotics, and benzodiazepines [33]. The use of melatonin to improve (i.e., shorten) sleep onset times for individuals with ASD has the most empirical support [34] but still does not meet clinical recommendation standards.

Sleep Problem Correlates

Given the diverse array of sleep problems in individuals with ASD, sleep problems may function as a co-occurring symptom, a consequence rising from neurobiological abnormalities in ASD, or a clinical phenotype [35]. A large body of research documents how sleep problems in ASD are associated with core ASD symptom severity, challenging behaviors, attention problems, adaptive functioning, and gastrointestinal symptoms [7, 36–38]. For example, sleep-disordered breathing and parasomnias were associated with elevated ASD symptom severity [4]. Additionally, children with ASD and comorbid clinical anxiety or depression concerns are more likely to present with sleep concerns [39–43]. Similarly, children with ASD and elevated internalizing and externalizing problems are also more likely to present with clinical sleep problems [44]. Sleep problems are, at times, considered a phenotypic marker [7, 37, 45, 46] as they tend to cluster in individuals who are more impacted by ASD and/or other comorbidities [42, 46, 47].

Family and Caregiver Impact of Sleep Problems

In early childhood, successful sleep is often conceptualized as a dyadic process that reflects child, caregiver, and contextual elements. Sleep problems in children with ASD are frequently associated with reports of elevated parental stress and depression symptoms across cultures and countries [2, 48]. Studies document an overall decline in well-being as parents find themselves incurring sleep debt as a consequence of attending to their child's sleep problems and associated health issues [49]. This caregiving burden can have cascading effects on parenting behaviors and broader family functioning, potentially implicating the entire family system [50]. In families raising children with ASD, elevated

rates of sleep problems are reported for parents, children with ASD, and younger siblings in the home [5, 11, 12, 51, 52]. With sleep problems present in young children, school-age children, and adolescents with ASD [53–56], supporting families in the sleep process often follows a life course perspective. Although research readily documents sleep concerns for families raising children with autism, treatment, or evidence-based practices designed to support the family unit (i.e., beyond the child with ASD) are scarce.

Systematic Update

To further inform the field, this review includes a systematic update of studies conducted within the past 3 years that assessed sleep and ASD. Through this review, we will compare past patterns (summarized above) to those present in the most recent literature.

Methods

To identify articles for this update, we built and executed comprehensive searches in the bibliographic databases PubMed, PsycINFO (EBSCOhost platform), and Web

of Science Core Collection on July 22, 2021. The search strategy combined terms related to autism and sleep. The “autism” keywords searched included *autism*, *autistic*, *Asperger’s*, and *pervasive developmental disorder not otherwise specified (PDD-NOS)*. The “sleep” keywords included *sleep*, *sleeping*, *circadian*, *waking*, *night*, *melatonin*, *clock gene*, and *rest*. Controlled vocabulary terms for these keywords were included for each database, as appropriate. Each search was then limited to those published in English, between 2018 and 2021, and related to pediatric patients (birth to 18 years). A breakdown of the retained studies is provided in Fig. 1. For full-search details, please see supplemental Appendix A.

Each study selected for full review was coded for (1) study design; (2) sample size, age, and location; (3) sleep assessment method; (4) ASD diagnosis and ASD assessment method; (5) alignment with four historic research areas; and (6) included a summative note of key findings. Following reviewer feedback, additional information was extracted from the included studies: the percentage of each sample with endorsed sleep problems, type of sleep problem, and for those with the most common sleep measure (CSHQ) mean scale scores were also extracted. All coded data are provided at <https://osf.io/4aq8p/>.

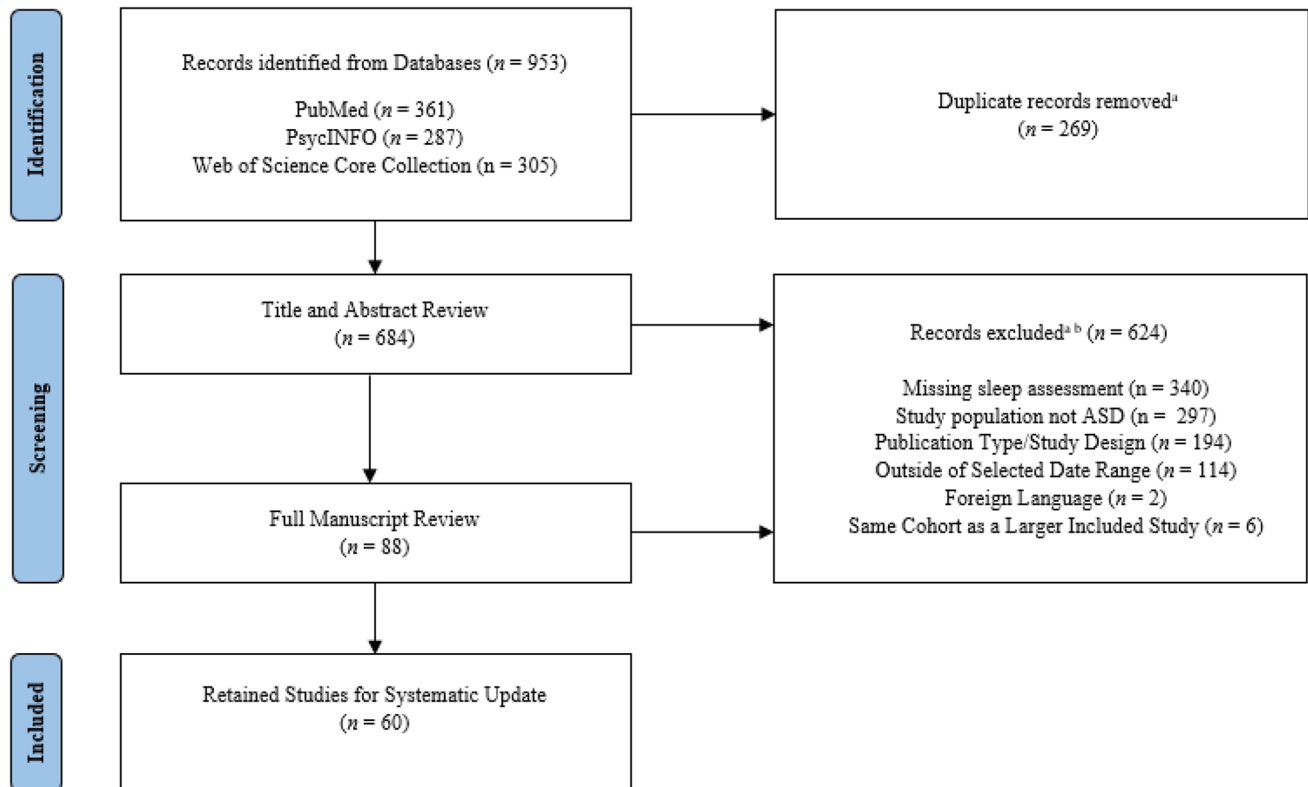


Fig. 1 PRISMA flow diagram for the systematic update from June 2018 to June 2021. ^aRayyan systematic review tool used to exclude duplicates and to track reasons for exclusion. ^bStudies could be

excluded for more than one reason; therefore, the number excluded does not add up to the total excluded records

Results

Sixty studies met our final inclusion criteria for the systematic review. Overall, 6 studies evaluated interventions, and a majority of the remaining studies (52 out of 54 studies) were correlational in nature (i.e., assessing how sleep is associated with various aspects of autism, behaviors, and context). Three studies aimed to improve our measures and/or protocols for collecting sleep information in ASD populations. In total, 24,229 children with average ages ranging from 1.81 to 16.90 years (Fig. 2). Sixteen studies assessed children in infancy/early childhood (< 2 years of age). The majority of studies assessed children in the preschool period (2–5 years of age) and at school age (6–12 years of age) with 42 and 52 studies, respectively. Twenty-eight studies assessed adolescents aged 13 to 18 years. Studies could include more than one age bracket.

Over 90% of studies confirmed each child's ASD diagnosis with the Diagnostic and Statistical Manual of Mental Disorders (DSM-5) criteria, and/or Autism Diagnostic Observation Schedule (ADOS-2) scores, and/or a parent-report interview or questionnaire. All studies in this review reported samples that included children with ASD, and some (11 studies) included samples with ASD and other comorbidities (e.g., gastrointestinal problems, intellectual

disability). Several also included a typically developing comparison group (25 studies). Sleep assessments included (from least common to most common) diary (13%), polysomnography (15%), actigraphy (18%), and questionnaires (91%). The most common questionnaire was the Children's Sleep Habits Questionnaire [57] used by 48% of studies.

Seventeen questionnaire studies (28%) reported the percentage of their ASD sample with endorsed general sleep problems—estimates ranged from 37 to 93% of the included sample, $M(SD) = 71.41(15.92)$. These reports of general sleep problems did not systematically vary by average sample age ($r(17) = -0.03, p > 0.05$). Given the wide range of sleep problems reported across studies, it is difficult to summarize across all studies; however, Table 1 provides the percentage of samples with each sleep problem type. In sum, sleep onset insomnia was (on average) the most common (~65%), followed by other types of insomnia and symptoms of obstructive sleep apnea were (on average) the lowest at ~23%. Seven studies also noted daytime sleepiness or hypersomnolence although sample percentages could not be consistently extracted. Similarly, four studies noted hyperhidrosis (excessive sweating) at night and one study noted restless leg symptoms. Studies did not consistently note if these symptoms (hypersomnolence, hyperhidrosis, restless legs) were consistent with medication side effects or if they were “clean” sleep concerns.

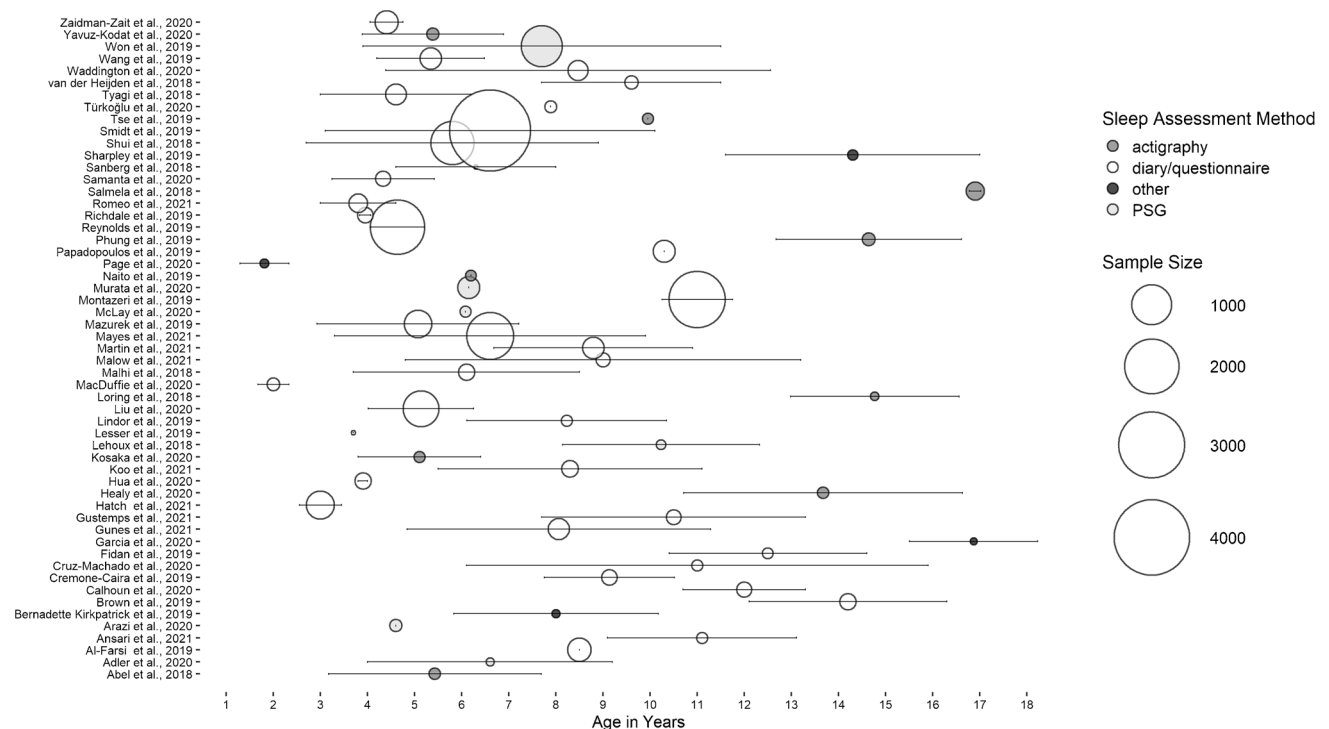


Fig. 2 Child age, sample size, and sleep assessment measure by the study in the systematic update. *Note.* There were 60 studies included in the systematic review (total $n = 24,229$). The age range of participants in each study is represented by horizontal bars

Table 1 Percentage of samples with endorsed sleep problems by type

Sleep problem type	<i>n</i> (reporting studies)	Min	Max	<i>M</i> (<i>SD</i>)
General	17	37	93	71.41 (15.92)
Insomnia				
Unspecified/general	4	25	62	42.67 (20.50)
Sleep onset	7	20	97	64.61 (27.44)
Night waking	8	5	85	47.65 (32.41)
Early morning rise	6	6	81	40.37 (26.37)
Parasomnia				
Unspecified/general	7	13	70	46.75 (21.73)
Obstructive sleep apnea	7	4	45	22.87 (18.09)

Min and Max reflect the minimum and maximum percentages reported across the reporting studies. For example, 37 reflects that 37% of a given sample had an endorsed sleep problem. The *M*(*SD*) reflects the mean and standard deviation of all the reported percentages (with equal weight across studies).

Table 2 Children's sleep habit questionnaire raw score averages

CSHQ scale	<i>n</i> (reporting studies)	Min	Max	<i>M</i> (<i>SD</i>)
General	23	43.29	66.86	50.39 (6.36)
Bedtime resistance	16	8.33	16.05	10.70 (1.99)
Sleep onset delay	11	1.47	2.80	1.94 (0.47)
Sleep duration	14	3.23	7.11	5.07 (1.32)
Sleep anxiety	15	5.99	9.80	7.13 (1.21)
Night waking	16	3.15	7.80	4.89 (1.26)
Parasomnia	14	7.27	12.96	9.35 (1.32)
Sleep breathing disorder	15	1.17	5.06	3.39 (0.76)
Daytime sleepiness	15	9.97	15.87	12.33 (1.60)

Min and Max reflect the minimum and maximum means reported across the reporting studies. The *M*(*SD*) reflect the mean and standard deviation of all the reported means (with equal weight across studies).

For the most common measure, CSHQ, 23 studies (38%) reported scale and/or subscale scores that could be extracted. In all reporting studies, the average CSHQ general problems total score was above the commonly used cutoff of 41 (see Table 2). Subscale ranges, means, and standard deviations are provided in Table 1 to help guide future studies but given the lack of established cutoffs for these subscales, scores were not interpreted further.

The four common historic themes (as noted above) also emerged across the systematic update studies. Over half of the studies, 35 studies (58%), documented that sleep problems were common in children with ASD. For example, Samanta and colleagues [58] documented that parent-reported sleep problems were extremely common (up to 95% of 2–6-year-old children with ASD). Similarly, 27 studies

(45%) documented less optimal sleep behaviors in children with ASD when compared to other groups. For example, Arazi et al. [59] documented decreased sleep pressure in children with ASD (as indexed by weaker slow-wave activity power, shallower slow-wave activity slopes, and less overall slow-wave sleep in comparison to controls). Roughly one third (18 studies) reported that elements of the family context influenced child to sleep or vice versa. Finally, 25 studies (42%) endorsed that sleep problems were associated with a more impacted phenotype (i.e., sleep problems or less optimal sleep were associated with increased symptom severity, other comorbidities, or lower IQ estimates).

Discussion

Sleep problems for children with autism remain a consistent concern for families, but our research approach to these problems historically and recently (as indexed by the provided systematic update) focuses heavily on the correlates of these sleep problems rather than treatment or mechanistic understanding. With over 20 years of clearly documented sleep problems with several documented negative correlates, the field is ready for a shift in focus. The following sections will outline how recent research aligns with historic patterns and highlight new/emerging areas in sleep and autism research.

Autism Sleep Problem Prevalence and Type

Over the past 3 years, research efforts continue to document the elevated rates of sleep problems in children with autism [60, 61] and studies have aimed to improve our measures of sleep problems in individuals with ASD [62–66]. For example, Hatch and colleagues [63] examined a commonly used parent-report measure (CSHQ) and identified a two-factor structure (rather than one that aligns with the established subscales); thus, for children with ASD, the CSHQ may work well to capture sleep initiation and awaking difficulties but not all types of sleep problems. Similarly, actigraphy is a commonly applied sleep assessment method and Alder et al. [62] developed a novel actigraph scoring algorithm for children with ASD.

In this systematic update, the percentage of samples with endorsed sleep problems varied greatly and this variability likely reflects differences in reporters (e.g., parent, caregiver, medical records), assessment types (e.g., questionnaire, actigraphy, polysomnography), and other sample characteristics (e.g., comorbidity). Despite this variability, the overall message was clear, and sleep problems in ASD are more common than in typically developing (age-matched) comparison groups [59, 100] and in most cases are more common than in other pathology-based comparison groups [89, 94].

Treatment Options for Sleep Problems in Autism

The intervention studies in the past 3 years included: (1) three studies that highlight the beneficial impact of physical activity on sleep [67, 68, 69•], (2) two trials noting improved sleep with extended-release melatonin [70•, 71] and (3) three studies of how individualized functional behavior intervention strategies can be implemented to improve sleep [72–74]. The behavioral intervention studies build on a robust line of work documenting how individualized behavioral intervention can be efficacious. The extended-release melatonin studies represent a relatively new area and offer families another relatively low-risk option to improve sleep onset (and to some extent) sleep maintenance difficulties. Importantly, standard preparations of melatonin and extended-release preparation are not efficacious in ameliorating all types of sleep problems. The three studies on physical activity represent a growing body of work that note the benefits of physical activity across several health/developmental domains including sleep.

Sleep Problem Correlates

Following the historic pattern present in ASD and sleep research, the majority of studies in the past 3 years document how sleep is associated with other aspects of context, autism, and/or development. Sleep in autism is associated with (1) demographic factors [75–77], (2) challenging behaviors [36, 78–87], (3) core ASD symptoms [38, 65, 77], and (4) other psychiatric symptoms [84, 88, 89]. Studies with a developmental focus also document correlations with executive functioning [88, 90], memory [91–93], attention [79, 94], and phonological learning [95].

The correlational studies pulled in this systematic update include several biological markers (e.g., pro-inflammatory cytokines, gut microbiome, cortisol, and melatonin present in urine). These studies represent a growing area in the field wherein biological indicators of atypicality are now employed more often (with or instead of more traditional behavioral metrics).

Family and Caregiver Impact of Sleep Problems

Recent studies continue to document the impact sleep problems have on families. Families raising children with ASD and sleep problems report more maternal mental health difficulties, greater family conflict, and less optimal caregiver physical health [2, 83, 84]. One study in the systematic update noted how improving sleep with treatment (via extended-release melatonin) improved self-reports of quality of life for caregivers [71].

New/Emerging Areas

Increased Global Representation

One notable change in the studies completed over the past 3 years is the increase in global representation across studies. Historically, studies were predominantly from the USA, Australia, the UK, and Canada. Though informative, these countries' cultural norms and federal guidelines surrounding sleep and bedtime routines are fairly comparable (e.g., share a room with separate sleep spaces for the first 6 months of life) and may not be generalizable to other cultural norms/preferences. In our recent assessment of sleep and autism studies, it was refreshing to see studies with samples from Malaysia, India, Pakistan, Bangladesh, and Oman [58, 96–100]. Collectively, the aforementioned studies highlight similar parent-reported concerns in sleep disturbances for children with ASD.

COVID-19

Another historically unique pattern in the systematic update included the emergence of studies assessing sleep in autism within the context of COVID-19 [75, 98, 101]. Similar to the general population, the stresses of COVID-19 were associated with high rates of sleep problems for families and children.

Sleep as an Early Behavioral Marker of Autism

In infancy, sleep problems can precede an ASD diagnosis and some researchers hypothesize sleep may be a part of a developmental cascade for children who later receive an ASD diagnosis [102•]. Several large cohort studies document sleep problems before the onset of the full behavioral manifestation of ASD [103, 104•, 105]. Similarly, a few prospective infant sibling studies [6] document sleep as parent's concerns early in development (before children have a diagnosis). Additionally, work by Shen and colleagues [106] noted how atypical cerebral spinal fluid (CSF) disbursement (excess CSF in the subarachnoid space) is an early brain biomarker of ASD and is associated with sleep problems. In our systematic update, two studies built on this work to highlight topographical differences during sleep [107] and parent reports of sleep problems [81] in infants/toddlers before their ASD diagnosis.

Conclusion

The field is ready for a shift toward more (1) intervention-based and implementation science studies, (2) global and age representation, and (3) mechanistic understanding. A long line of work documents elevated rates of sleep problems and their association with several aspects of development

and context. Developing and implementing efficacious interventions can potentially improve developmental trajectories across several domains. Although samples representing a more global ASD population are on the rise, the larger field will benefit from continued growth in this area. The majority of studies assessing sleep and autism focus on early childhood (as illustrated in Fig. 2), with relatively few studies in adolescents and emerging adulthood. Future studies should aim to fill this gap as sleep problems are still quite prevalent in older ASD populations. Finally, the plethora of work connecting sleep and autism is not an accident and likely reflects a robust biological link between sleep processes and autism processes. Future work should aim to understand not only how they are connected but why (i.e., improve our mechanistic understanding of sleep–autism relations).

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Authors’ Contributions Schwichtenberg led the manuscript effort and contributed to the text generation and editing. Lindsay led the systematic review coding and contributed to the text generation and editing. Janis led the data visualization efforts and contributed to the text. Desai assisted with study coding and contributed to the text. Sahu organized the references, drafted the PRISMA figure, and drafted the sections of the discussion. Kellerman, Chong, and Abel mentored the manuscript text and provided feedback and revision support on the manuscript. Yacilla completed the systematic data pull, supported the coding team efforts, and contributed to the text/content to the “Methods” section of the manuscript. All authors read the penultimate draft of the manuscript and provided edits as needed. The authors read and approved the final manuscript.

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Data Availability All systematically pulled data will be available at <https://osf.io/4aq8p/> upon release of the manuscript. Both figures are original and generated for this manuscript.

Code Availability Not applicable.

Declarations

Ethics Approval This study did not include direct data collection from human subjects, not applicable.

Consent to Participate Not applicable.

Consent for Publication All authors give their consent for the publication of this manuscript in its current form.

Conflict of Interest The authors declare no competing interests.

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