

Infant Sleep Patterns: Implications for Future Development and Sleep Problems

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**List of Abbreviations**

SES	Socioeconomic status
GEE	Generalized Estimating Equations
SD	Standard deviation
M	Mean
REM	Rapid eye movement
NREM	Non-rapid eye movement
AS	Active sleep
QS	Quiet sleep
PSG	Polysomnography
EEG	Electroencephalography
EOG	Electrooculogram
EMG	Electromyography
CNS	Central nervous system
ADHD	Attention-deficit/hyperactivity disorder



## Abstract

Infants spend most of their first year of life in a sleeping state. Sleep is an important factor involved in child development and affects many areas of daily life. However, there is a lack of understanding of the association between *early* sleep patterns in infancy and general development, including future sleep problems. The aim of the present dissertation is to identify which early infant sleep variables, if any, are associated with future development and sleep problems.

Study 1 consisted of a systematic review of original studies investigating the association between sleep patterns during infancy (0 to 18 months) and cognitive/psychomotor development (two to 48 months), specifically in infants born at full term. A quality assessment of the articles was conducted and post-hoc power analyses were performed. A qualitative approach was used to synthesize findings as well as a secondary analysis of individual sleep variables.

A total of 7136 articles were identified and screened for inclusion and 22 articles met the criteria. Findings from Study 1 did not show a clear association between infant sleep-wake patterns at 0 to 18 months and cognitive/psychomotor development at two to 48 months. Seventeen of 22 studies (77%) reported mixed results regarding this association with only two studies (9%) finding exclusively significant results and three studies (14%) reporting no significant associations. Mixed results included associations which were (1) significant at one timepoint but not at another, (2) at the same timepoint, certain outcome variables were significant, but others were non-significant, or (3) an outcome variable was significant at one time point and significant at an additional timepoint but with opposite directionality (positive vs. negative associations). When considering specific sleep variables, most studies assessing nocturnal sleep duration and total sleep duration (defined as total sleep within a 24-hour period)

found no significant associations with developmental outcomes. However, it should be noted that all but one (21 of 22) included studies were found to have estimated power of under 0.80, which may increase the risk of type 2 errors (false negatives). Overall, 63% of studies were rated as having good methodological quality. The results of this review highlight the need for additional studies with larger sample sizes to better identify which sleep variables, at what age, and in what context might predict developmental outcomes.

Study 2 aimed at examining if there was an association between sleep patterns at six months of age and future sleep problems at 48 and 60 months. At six months, total sleep duration (including both nocturnal and daytime sleep) and consecutive sleep duration (longest period of consecutive sleep, without interruption) were assessed with parental reports, by using an adapted version of the Questionnaire about Sleep Habits (QASH). At 48 and 60 months, sleep problems were assessed using the Sleep Subscale of the Child Behavior Checklist (CBCL/1.5-5). Generalized estimating equations (GEE) for repeated measures were conducted to assess the associations between six-month total sleep duration and consecutive sleep duration and parental reported sleep problems at 48 and 60 months.

Findings revealed that infants who had shorter total sleep durations at six months had higher sleep problem scores at 48 and 60 months, after adjusting for maternal depression, breastfeeding status, sleeping arrangements, and socioeconomic status (SES). However, consecutive sleep duration at six months was not significantly associated with the presence of sleep problems at 48 or 60 months. These results suggest that insufficient total sleep duration, or sleep over 24 hours in infancy is more closely associated with future development of sleep problems than sleep consolidation.

Taken together, these studies revealed that the association between early infant sleep patterns and their future correlates are nuanced and complex. While the associations between sleep patterns, cognitive/psychomotor development, and sleep problems may be more consistently significant in children and adults, they are less clear during infancy. However, certain sleep variables seem to be more associated with specific future outcomes than others. Particularly, total sleep duration in infancy appears to be associated with future sleep problems but not with cognitive/psychomotor development. On the other hand, consecutive sleep duration, often referred to as sleeping through the night, did not seem to be consistently associated with neither sleep problems nor cognitive/psychomotor development. These results expand our understanding of the intricate association between infant sleep, future cognitive/psychomotor development, and future sleep problems and highlight which sleep variables may and may not be associated with later outcomes.

## Résumé

Les nourrissons passent la majeure partie de leur première année de vie à dormir. Le sommeil joue un rôle crucial dans le développement de l'enfant et il influence de nombreuses facettes de la vie quotidienne. Cependant, le lien entre les patrons de sommeil au cours de la petite enfance et le développement de l'enfant, y compris les futurs problèmes de sommeil, demeure mal compris. Cette thèse vise à identifier quelles variables liées au sommeil au cours de la petite enfance sont associées au développement de l'enfant et aux futurs problèmes de sommeil.

La première étude consistait en une revue systématique d'études originales explorant l'association entre les patrons de sommeil des nourrissons (0 à 18 mois) et le développement cognitif et psychomoteur (deux à 48 mois), spécifiquement chez les enfants nés à terme. Une évaluation de la qualité des articles a été réalisée et des analyses de puissance post-hoc ont été effectuées. Une approche qualitative a été utilisée pour synthétiser les résultats, ainsi qu'une analyse secondaire des variables individuelles du sommeil.

Au total, 7136 articles ont été identifiés et sélectionnés, et 22 articles répondaient aux critères d'inclusion. Les résultats de la première étude n'ont pas démontré d'association claire entre les patrons de sommeil de 0 à 18 mois et le développement cognitif et psychomoteur chez les enfants de deux à 48 mois. Dix-sept études sur 22 (77 %) ont rapporté des résultats mitigés concernant cette association, seulement deux études (9 %) ont rapporté des résultats exclusivement significatifs et trois études (14 %) n'ont trouvé aucune association significative. Les résultats mitigés comprenaient des associations qui étaient (1) significatives à un temps de mesure, mais pas à un autre (2) significatives pour certaines variables mais pas pour d'autres, ou (3) significatives à deux temps de mesure, mais dans des directions opposées (associations

positives versus négatives). En ce qui concerne les variables spécifiques du sommeil, la plupart des études qui ont mesuré la durée du sommeil nocturne et la durée totale du sommeil (définie comme la durée totale de sommeil sur une période de 24 heures) n'ont trouvé aucune association significative avec le développement futur des enfants. Cependant, il est important de considérer que toutes les études sauf une (21 sur 22) avaient une puissance estimée inférieure à 0,80, ce qui peut augmenter le risque d'erreurs de type 2 (faux négatifs). Dans l'ensemble, 63% des études ont été jugées de bonne qualité méthodologique. Les résultats de cette revue systématique soulignent la nécessité de réaliser des études supplémentaires avec des échantillons de plus grande taille, afin de mieux identifier quelles variables de sommeil, à quel âge, et dans quels contextes peuvent mieux prédire les répercussions sur le développement futur.

La deuxième étude avait pour objectif d'examiner s'il existait un lien entre les habitudes de sommeil à l'âge de six mois et les futurs problèmes de sommeil à 48 et 60 mois. À six mois, les parents ont rapporté la durée totale du sommeil de leur enfant (incluant le sommeil nocturne et diurne) et la durée consécutive du sommeil (la plus longue période consécutive de sommeil, sans interruption), à l'aide d'une version adaptée du Questionnaire sur les habitudes de sommeil (QASH). À 48 et 60 mois, les problèmes de sommeil ont été mesurés à l'aide de la sous-échelle « *sommeil* » du Child Behavior Checklist (CBCL/1.5-5). Des équations d'estimation généralisées (GEE) pour mesures répétées ont été réalisées pour évaluer les associations entre la durée totale du sommeil à six mois, la durée consécutive du sommeil et les problèmes de sommeil rapportés par les parents à 48 et 60 mois.

Les résultats ont révélé que les nourrissons ayant une durée totale de sommeil plus courte à six mois présentaient des scores plus élevés pour les problèmes de sommeil à 48 et 60 mois, après avoir contrôlé la dépression maternelle, la présence d'allaitement, les arrangements de

sommeil et le statut socioéconomique (SSE). Cependant, la durée consécutive du sommeil à six mois n'était pas associée de manière significative à la présence de problèmes de sommeil à 48 ou 60 mois. Ces résultats suggèrent qu'une durée totale de sommeil insuffisante sur une période de 24 heures pendant la petite enfance est plus étroitement associée au développement de problèmes de sommeil futurs que la consolidation du sommeil.

Dans l'ensemble, ces études ont révélé que l'association entre les habitudes de sommeil du nourrisson et leurs corrélats futurs est complexe et nuancée. Alors que les associations entre les habitudes de sommeil, le développement cognitif/psychomoteur et les problèmes de sommeil peuvent être plus significatives chez les enfants et les adultes, elles semblent moins claires pendant la petite enfance. Cependant, certaines variables de sommeil particulières semblent être associées à des répercussions futures spécifiques, mais pas à d'autres. En particulier, la durée totale du sommeil pendant la petite enfance semble être associée aux futurs problèmes de sommeil, mais pas au développement cognitif/psychomoteur. En revanche, la durée consécutive de sommeil, communément appelée « faire ses nuits », ne semble pas être associée systématiquement ni aux problèmes de sommeil, ni au développement cognitif/psychomoteur. Ces résultats nous permettent de mieux comprendre l'association complexe entre le sommeil du nourrisson, le développement cognitif/psychomoteur et les problèmes de sommeil pendant l'enfance, et mettent en évidence les variables de sommeil spécifiques qui peuvent ou non être associées à des répercussions futures.

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the birth of our incredible son, Bennett, who has afforded me invaluable insight into the world of infant sleep (or lack thereof). While I am honoured to be joining the ranks of my esteemed doctoral colleagues, I am most proud to be your husband and father to Bennett. I love you both more than anything in this world and forever grateful for you both.



### **Contribution to Original Knowledge**

The present dissertation contributes original knowledge to the field of pediatric sleep by furthering our understanding of the association between sleep patterns in healthy full-term infants and future developmental and sleep outcomes.

Sleep during infancy is a vital factor for child development and daily functioning. However, the association between infant sleep and cognitive and psychomotor development, as well as future sleep problems, is not well understood. The current body of research endeavors to clarify these associations through the two studies which comprise this dissertation.

Study 1 is a systematic review accepted for publication in the journal *SLEEP*, which examines the association between infant sleep patterns and cognitive and psychomotor development. A previous review published in 2009 helped to clarify this association but did not employ a systematic search strategy nor a quality assessment of included studies. Study 1 contributes original knowledge by using a systematic search strategy to synthesize the available literature concerning the sleep of healthy full-term infants and cognitive/psychomotor development and by employing a quality assessment to help with the interpretation of findings from the included studies.

Study 2 is an original study being prepared for submission to the journal *Sleep Health* and examines the association between infant sleep patterns and future perceived sleep problems. A previous study examined the association between sleep patterns and perceived sleep problems at concurrent timepoints but did not investigate this association longitudinally. Additionally, a validated measure to assess sleep problems was not used. Study 2 contributes original knowledge by utilizing a longitudinal design and a validated measure to investigate the association between specific sleep variables in infancy and perceived sleep problems during the preschool period.

### **Contributions of the Authors**

This dissertation is composed of original scholarship and contains two studies (Chapters 3 and 5). These studies contribute novel knowledge to the field of pediatric sleep. I am the principal author of each manuscript included and participated in study design, search strategy design, data extraction, statistical analysis, interpretation of results, and drafting and revising of manuscripts. All of these components were done under the guidance of my research supervisor, Dr. Marie-Hélène Pennestri.

The manuscripts are co-authored by Rebecca Burdayron, Clara Lewis, Gil Mazor-Goder, Christine Laganière, Malka Hershon, Adrienne Vandenberg, Charlène Thauvin, Dr. Hélène Gaudreau, Dr. Bassam Khoury, Dr. Mélanie Vendette, and Dr. Marie-Hélène Pennestri.

Rebecca Burdayron is a PhD student and Clara Lewis and Gil Mazor-Goder are research assistants in our research laboratory. They participated in data extraction, quality assessment, and critically reviewing manuscripts for intellectual content.

Christine Laganière is a PhD student in our research laboratory. She contributed assistance with data analysis and interpretation and critical review of manuscripts for intellectual content. Malka Hershon is a master's student in our lab and assisted with data analysis.

Adrienne Vandenberg and Charlène Thauvin are also master's students in our lab and assisted with critically reviewing manuscripts for intellectual content.

Dr. Hélène Gaudreau is a collaborator with our research team and assisted via data curation, critical review, and editing.

Dr. Bassam Khoury is an associate professor in our department and contributed to study conceptualization and design. He also contributed to the interpretation of data and critical review of manuscripts for important intellectual content.

Dr. Mélanie Vendette is a collaborator on the research team. She provided comments and suggestions on the manuscripts and approved final versions for submission to peer-reviewed journals.

Dr. Marie-Hélène Pennestri is my doctoral research supervisor and co-author on both manuscripts. She provided supervision, data curation, was involved with study design and conceptualization, interpretation of data, and critical review of manuscripts for important intellectual content. Dr. Marie-Hélène Pennestri also approved the final manuscripts and the current dissertation.

## **Chapter 1: General Introduction**

Sleep is a fundamental process that is extensively involved in child development. It has an influence on brain maturation, as well as multiple domains of daily functioning (Astill et al., 2012; Chaput et al., 2016; Fatima et al., 2015; Kurth et al., 2013; Quach et al., 2018; Quist et al., 2016; Simola et al., 2014). Sleep is also a developmental process which undergoes considerable changes from infancy into childhood (Astill et al., 2012; Pennestri et al., 2018; Touchette et al., 2007; Williamson et al., 2020). As a result, sleep during infancy is clearly different than sleep in children and adults as it undergoes rapid change even in the first year of life. For instance, it is highly fragmented, and generally consists of two-to-three-hour periods during the first months (Giganti et al., 2001; Rivkees, 2003; Seegers et al., 2016). This sleep fragmentation may result in parents feeling compelled to actively help their infant consolidate sleep early on with the goal of fostering good sleep habits and preventing future sleep problems (Burdayron et al., 2020; Porter & Ispa, 2013). Moreover, infant sleep patterns influence their parents' sleep who also experience more fragmented sleep during the post-partum period (Meltzer & Montgomery-Downs, 2011). As a result, parents may experience challenges while adjusting during this period and may be highly motivated to attempt helping their child consolidate their sleep (Medina et al., 2009; Meltzer & Montgomery-Downs, 2011). However, despite these common concerns, the association between sleep patterns during infancy and future sleep problems or later developmental outcomes is not well known.

Indeed, despite the well-researched association between sleep and cognitive/psychomotor development in children and adults, this association during infancy is far from established and is not well understood. Specifically, in children, adolescents, and adults, shorter sleep duration has been associated with poorer cognitive outcomes (Astill et al., 2012; Lowe et al., 2017; Reynaud

et al., 2018; Shochat et al., 2014). While a few studies have investigated this association in infancy, a systematic review and synthesis of studies in normally developing full-term infants has not been conducted. Identifying relevant articles and synthesizing the available literature would provide a much-needed overview of this association in infancy, especially given elevated parental concern regarding their child's sleep.

Additionally, previous studies have showed some persistence in the presence of perceived sleep problems in infants and reported sleep problems during childhood, however, few studies have examined this association using specific sleep variables such as consecutive sleep or total sleep duration (Byars et al., 2012; Williamson et al., 2019). Recognizing early indicators of future problematic sleep is of great importance as it is estimated that 20-35% of children experience sleep problems at some point during childhood (Byars et al., 2012; Gaylor et al., 2005; Owens, 2008). Additionally, identifying which sleep variables are associated with future cognitive/psychomotor functioning and sleep problems early on might help to prevent negative outcomes through early intervention. Indeed, many sleep variables exist relating to duration (e.g., nocturnal, daytime, and total sleep duration) and continuity/fragmentation (e.g., consecutive sleep duration, number of awakenings, sleep efficiency) and these need to be examined further. Consequently, elucidating these associations may help healthcare professionals as well as parents determine which early sleep patterns should be prioritized.

The main objective of this dissertation is to determine which sleep variables in early infancy are or are not associated with cognitive/psychomotor functioning and future sleep problems. Consequently, the studies included in this dissertation endeavour to highlight which sleep patterns should or should not be prioritize by parents and healthcare professionals in infancy. These objectives will be achieved through a systematic review and original research

which are contained in this dissertation. In chapter 2, a thorough review of the literature is provided and includes commentary on gaps in the literature as well as ways in which the present dissertation has attempted to address these gaps and contributed to the existing literature. Subsequently, chapter 3 is comprised of Study 1 which has been accepted for publication in the journal *SLEEP*. Study 1 investigated infant sleep patterns and their association with cognitive/psychomotor functioning. A systematic review was undertaken to synthesize the available literature concerning the association between infant (0-18 months) sleep patterns and cognitive/psychomotor development. Chapter 4 contains a transition statement, which outlines the primary findings of Study 1 and provides a bridge to Study 2. Chapter 5 presents Study 2, which is being prepared for submission to the journal *Sleep Health*. This is an original study investigating the association between total sleep duration, consecutive sleep duration, and future parent reported sleep problems in their children. Finally, in Chapter 6, a general discussion which presents a summary of key findings, clinical implications for infant and preschooler sleep, strengths and limitations of the studies, and future directions.

## **Chapter 2: Review of the Literature**

### **What is Sleep?**

Sleep is a vital function in humans and adequate sleep promotes optimal well-being and health (Carskadon & Dement, 2017). Poor sleep and sleep deprivation impact negatively several spheres such as physiological, emotional, and cognitive functioning (Ferrie et al., 2011; Holliday et al., 2013; Patel & Hu, 2008; Prather et al., 2012; Shankar et al., 2008; Short et al., 2018; Simola et al., 2014). Indeed, sleep is considered the third pillar of health and should be prioritized equally alongside nutrition and physical activity (Castillo, 2015).

In adults, sleep is organized in cycles lasting approximately 90 minutes in length alternating between non-rapid eye movement sleep (NREM) and rapid eye movement sleep (REM) (Kryger, Roth, Goldstein, & Dement, 2022). Adults have approximately four or five cycles of sleep during the night. NREM sleep contains three stages through which sleep progressively deepens. Stage N1 represents the transition between sleep and wakefulness and during this stage, a person may be easily awoken by external stimuli such as touch or sound. During stage N2, sleep deepens, and a more intense external stimulus is required to be awoken. This stage is marked by the appearance of sleep spindles and k-complexes (Kryger, Roth, Goldstein, & Dement, 2022). Stage N3 is also called slow-wave sleep or deep sleep and is considered the deepest stage of sleep. Rapid eye movement (REM) sleep is marked by rapid eye movements and similar brain activation as wakefulness, with muscle atonia. It is typically associated with the dreaming process. Typically, NREM sleep occurs in a larger proportion during the first cycles of the night, while REM sleep typically occurs in a larger proportion in the last cycles of the night (Kryger, Roth, Goldstein, & Dement, 2022).

Sleep is regulated by two complementary physiological processes, the homeostatic and circadian processes (Achermann & Borbély, 2017; Deboer, 2018). The circadian process is a 24-hour clocklike biological system which is mainly regulated by the suprachiasmatic nucleus (SCN) in the hypothalamus in all mammals. However, this system is also influenced by environmental factors such as temperature and light exposure (Dijk & Skeldon, 2022). The SCN is responsible for two primary functions regarding circadian rhythms: (1) the internal generation of circadian rhythms and (2) synchronizing circadian rhythms with the light-dark cycle (Vitaletta, Takahashi, & Turek, 2001). The alignment of these internal rhythms with external environmental cycles is crucial for maintaining optimal biological functions, as they help regulate key physiological processes like hormone release (e.g., melatonin), body temperature, and metabolism (Dijk & Edgar, 1999; Vitaletta, Takahashi, & Turek, 2001).

The other process involved in sleep regulation is the homeostatic process. This process can be thought of as a debt-based system which increases in response to longer periods of wakefulness. As a result, the longer the period of wakefulness, the stronger the drive to sleep will become and greater amounts of sleep debt will be accumulated (Achermann & Borbély, 2017). Ultimately it is the interaction of these two processes that underlies and regulates sleep behaviors, as well as sleepiness and alertness over a 24-h period. However, it is important to acknowledge that sleep is not a static process. It is rather a developmental process that changes and evolves significantly during the lifespan.

### **Infant Sleep**

Unlike sleep in adults, sleep during infancy is still under development and is rapidly evolving over the first year of life. During the first weeks of life, infants spend a very high proportion of their time sleeping. Total sleep duration (within 24 hours) in newborns is



approximately 16-18 hours per day during the first few weeks (Louis et al., 1997; Parmelee Jr et al., 1964). This gradually decreases over the first two years of life from approximately 14 to 17 hours at 0 to three months, to 12 to 15 hours at four to 11 months, and 11 to 14 hours between one and two years old (Hirshkowitz et al., 2015).

The circadian rhythm in infants is also developing during the first year of life. As a result, during the first weeks of life, infants lack the ability to differentiate between day and night (Rivkees, 2003). As the circadian rhythm develops, the percentage of sleep obtained at night will gradually increase over time and the amount of daytime sleep will gradually decrease (Louis et al., 1997; Parmelee Jr et al., 1964).

Sleep during infancy will also begin to consolidate during the first year with infants gradually awakening less frequently during the night and having fewer and shorter daytime naps (Mirmiran et al., 2003). This process or milestone of consolidation is often referred to as “sleeping through the night” and can be defined as six or eight hours of consecutive sleep (Henderson et al., 2011; Pennestri et al., 2018). While all infants eventually reach this milestone, there is great interindividual variability in this process. Indeed, not all infants sleep through the night at the same timepoint, and significant variability has been found regarding when it is achieved (Goodlin-Jones et al., 2001; Henderson et al., 2011; Pennestri et al., 2020).

In addition to interindividual differences, intra-individual differences also exist. While an infant may sleep uninterrupted for a six- or eight-hour period for one night, they may not consistently sleep through the night on a night-to-night basis (Pennestri et al., 2020). This variability is often a concern for first-time parents who are adapting to their new schedules.

## **Infant Sleep and Transition to Parenthood**

The transition to parenthood while a joyful time can be a challenging period for new parents (Teubert & Pinquart, 2010). The sleep of their infant is one of the most common complaints or worries parents share when consulting their physician during the postpartum period (Sheldon, 2014). This is a significant source of stress for parents, particularly so for parents with unrealistic expectations regarding their infant's sleep (Burdayron et al., 2020; Porter & Ispa, 2013). Indeed, sleep-related expectations vary a lot between parents. In a previous study, the expectation of when an infant should be able to "sleep through the night" (six consecutive hours) ranged from one to 15 months across parents (Burdayron et al., 2020). Given that approximately one third of infants do not consistently "sleep through the night" at six months, parents with an infant who does not "sleep through the night" at this timepoint may experience increased stress if they have high or unrealistic expectations (Pennestri et al., 2018). Additionally, parents may feel pressured to help their infant consolidate their sleep more quickly in an attempt to meet these expectations and avoid future sleep problems (Porter & Ispa, 2013). Some parents may decide to implement interventions to help their infant consolidate their sleep, which are being delivered at increasing younger ages (Crichton & Symon, 2016; Gradisar et al., 2016). However, it is unclear as to whether these interventions are necessary for infant sleep consolidation or are also implemented to help address sleep fragmentation in parents.

Parents are not only concerned about their infant's sleep but experience challenges regarding their own sleep as well. Due to the highly fragmented nature of early infant sleep, mothers and fathers also experience fragmented sleep during the post-partum period (Da Costa et al., 2021; Meltzer & Montgomery-Downs, 2011). Moreover, sleep deprivation in parents during this period has been associated with decreased mood and cognitive challenges (Medina et al.,

2009). While some parents experience increased stress and mild mood disturbances, some mothers and fathers will develop postpartum depression during this challenging transitional period (Dørheim et al., 2009; Kalogeropoulos et al., 2021; Kim et al., 2020). A better understanding regarding the implications of infant sleep patterns may help parents to have more realistic expectations regarding their infant's sleep, which may help to improve parental mood and decrease anxiety and worry.

### **Internal Factors and External Factors Influencing Infant Sleep**

However, investigating the complex nature of infant sleep requires a thorough understanding of the various influencing factors. Grandner (2019) proposed a model of sleep, which is informed by Bronfenbrenner's Social-Ecological Model (Bronfenbrenner, 1977). He conceptualizes these factors as occurring on the individual level, social level, and societal level. The complete model will not be utilized but factors relevant to the current dissertation will be presented and discussed. When adapting this model to the current dissertation, these factors will be described as internal or external to the infant.

#### ***Internal Factors***

An internal factor which cannot be overlooked is brain maturation, especially during early life. The sleep-wake cycle is controlled by the central nervous system (CNS) and undergoes accelerated development during infancy (Sheldon, 1996). The development of the CNS continues into adulthood, but the most pronounced changes occur within the first years of life and coincide with the most significant changes in the sleep-wake cycle (Kinney et al., 1988).

Neurodevelopmental and genetic conditions can also impact sleep. Children born with developmental disabilities and genetic syndromes or disorders have more sleep problems and higher prevalence of sleep disorders than neurotypical children (Esbensen & Schwichtenberg,

2016; Mainieri et al., 2021). For example, children with developmental disorders have been reported to have higher perceived sleep problems, which may include insomnia, difficulty falling asleep, more fragmented sleep, and shorter total sleep duration than children without developmental disorders (Esbensen & Schwichtenberg, 2016). Additionally, sleep state organization (i.e., sleep stages) has been shown to differ in children with neurodevelopmental disorders when compared to children without neurodevelopmental disorders (Whitney & Thoman, 1993).

Moreover, gestational age has also been found to impact sleep patterns in infancy. Differences have been reported between the sleep patterns of full-term ( $\geq 37$  weeks) and preterm ( $< 37$  weeks) infants. Premature infants have been found to have more variability regarding sleep patterns at various ages than those born at full-term (Akkus & Bahtiyar-Saygan, 2022; Anders & Keener, 1985; Guyer et al., 2015). Specifically, preterm infants have been found to have shorter nocturnal and total sleep durations and have more difficulty falling asleep than infants born at full term (Akkus & Bahtiyar-Saygan, 2022). For this reason, it is important to distinguish between full-term and premature infants.

Temperament has also been found to be associated with sleep patterns in infancy via parental report (Bouvette-Turcot et al., 2015; Wolke et al., 1995). For example, infants rated as having an *easy* temperament by parents were more likely to sleep through the night and had fewer reports of night awakenings (Keener et al., 1988; Schaefer, 1990). Correspondingly, infants with difficult temperaments have been found to have more night awakenings (Weinraub et al., 2012). It is important to note that previous studies used observational designs and thus the proposed causal relationship between sleep and temperament remains to be clarified.

### ***External Factors***

External factors also play a role in influencing infant sleep. Factors such as light exposure, the child-parent relationship, and parental practices have been found to interact with infant sleep patterns (Field et al., 2007; Pennestri et al., 2015; Rivkees, 2003; Sadeh et al., 2010).

Sleep arrangements are one of these factors and can be divided into co-sleeping and solitary sleeping (sleeping in a separate room). Co-sleeping can be further divided into room sharing (bassinet or crib in the same room as the parent) or bedsharing (infant in the same bed as the parent). For example, the literature reports associations between sharing a room with an infant and increased parental reports of night awakenings in their children (Volkovich et al., 2015). However, when these parental reports of night awakenings were compared to objective measures of night awakenings, there was no difference between room sharing and non-room sharing infants. This suggests that parents were potentially more aware of the night awakenings. Yet, the literature appears to be inconsistent as other studies have found that infants who bedshare or room share have similar sleep durations as solitary infant sleepers (Yu et al., 2017). Indeed, in a recent review by Andre et al. (2021) the authors found that the association between bedsharing and infant sleep (measured objectively and subjectively) was inconclusive.

Parenting behaviors related to bedtime routines such as soothing infants to sleep, and other parenting behaviors have also been associated with increased night awakenings in children (Larsen et al., 2023; Sadeh et al., 2010). However, there have been contradictions reported in the literature (Samdan et al., 2020). For example, although the literature has shown an association between nocturnal awakenings and certain parenting behaviors (having a parent in the bedroom, falling asleep outside of their bedroom) it is difficult to determine whether these parental behaviors are negatively impacting sleep or if these parents are simply more sensitive to their

infant's waking behaviors, as a bidirectional association has been posited (Fehlings et al., 2001; Sadeh et al., 2010; Simard et al., 2008; Tétreault et al., 2017).

Feeding method has also been associated with infant sleep patterns. For example, breastfed infants have been found to have shorter consecutive sleep durations than formula-fed infants likely due to the more rapid digestion of breastmilk (Abdul Jafar et al., 2021). However, other studies have found that while breastfed infants may have shorter consecutive sleep durations during early infancy, this does not persist into later infancy and toddlerhood (Hershon et al., 2024). Additionally, other studies have found that while exclusively breastfed infants have more night awakenings and shorter consecutive sleep durations than non- and partially-breastfed infants, there is no difference in total sleep duration (Figueiredo et al., 2017). Overall, the association between sleep patterns and breastfeeding status are complex and multifaceted.

Lastly, socioeconomic status (SES) is a known factor which can affect the sleep of children and thus is another important variable to consider while investigating sleep (Buckhalt, 2011). In a recent review, lower SES was associated with shorter total sleep duration and more fragmented sleep (Sosso et al., 2021). Additionally, higher parental education has been associated with longer childhood sleep duration and better sleep quality, while higher household income is associated with fewer sleep problems (Cameron et al., 2022).

Ultimately, both internal and external factors exert their influence on sleep patterns during infancy and both need to be taken into consideration when investigating sleep during this period of rapid development. Additionally, a bidirectional association exists between sleep patterns and most of these factors, thus, determining the precise direction is challenging.

## **Measurement of Sleep**

Sleep can be measured in various ways and researchers or clinicians investigating infant sleep patterns may choose to use subjective measures, objective measures, or a combination of both. Each category of measurement has advantages and limitations, but they are ultimately complementary. Importantly, the choice of measure may impact the results of studies and probably contributes to the discrepancies that were reported in the previous section. Below, these categories of measures are listed and described in the context of measuring sleep, more specifically in infancy.

### ***Objective Measures***

The primary objective measures used to assess infant sleep are polysomnography (PSG), actigraphy, Motility Monitoring System (MMS; a pressure sensitive pad), and time-lapse video recording (Thoman & Glazier, 1987; Thoman & Whitney, 1989). PSG is the gold standard and allows to measure sleep-wake patterns such as sleep duration and number of awakenings, as well as sleep architecture to be able to determine sleep stages (light sleep, slow wave sleep, rapid-eye movement-REM sleep). This is accomplished by placing electrodes on the scalp (electroencephalography; EEG), eyes (Electrooculogram; EOG) and chin (Electromyography; EMG). Other measures can also be included, such as breathing activity, motor activity and heart rate activity. PSG is usually performed in laboratory settings (but sometimes in-home) and used less frequently with infants in research settings, due to its invasiveness (Bennet et al., 2016).

Pressure-sensitive pads or motility monitoring measures infant breathing as well as motor movement to determine specific sleep states, with results that correlate well with direct observation (Thoman & Glazier, 1987; Thoman & Whitney, 1989). This is a non-intrusive method as the pad can be placed underneath the infant, but as a limitation, can only measure

sleep while the infant is in their crib. This method was commonly used to measure infant sleep during the first 48 hours of life; however, it appears to have been replaced by newer sleep assessment tools and has been seldomly used in more recent research.

Another objective measure is time-lapse video recording which is used to determine specific sleep-wake states such as active sleep, quiet sleep, and wakefulness via behavioral observation (Anders & Sostek, 1976). However, like the motility monitoring system, sleep is only measured while the infant is in their crib due to camera placement. This can be completed in-home but is usually employed in a laboratory setting as it involves extensive recording equipment. Time-lapse video is in high agreement with PSG concerning sleep-wake states (Anders & Sostek, 1976).

Actigraphy is another objective measure which can be used to assess in-home sleep for longer periods of time. Actigraphy uses small watch-like devices attached to the infant's ankle to measure and detect motor movements. These devices do not measure sleep directly but operate under the assumption that less motor movement is present while in a sleeping versus awake state. While actigraphy cannot measure specific sleep stages, it does correlate favorably with PSG and sleep diaries in determining whether an infant is awake or asleep (Bélanger et al., 2014; Sadeh et al., 1991; Sadeh et al., 1994). Actigraphy also has the advantage of estimating infant sleep in their natural environments thus having high external validity.

### ***Subjective Measures***

Subjective sleep measures are based on data collected from parents which can take the form of sleep diaries or questionnaires. Sleep diaries usually use 24-hour periods divided into 15- to 30-minute intervals (or a series of questions) where the parent will indicate when their infant goes to bed, falls asleep, awakes during the night, wakes up the next day, and the frequency and



duration of daytime naps (Acebo et al., 2005). This is repeated each day and data are collected anywhere from one day to multiple weeks, depending on the goal of the study.

Sleep questionnaires are used to gather similar data and may include additional questions related to perception of sleep problems, sleep location, and breastfeeding practices. Given that subjective measures are self-reported and require few resources, it is easier to assess larger samples of infants using this method. Some examples of validated measures include: The Brief Infant Sleep Questionnaire (BISQ) (Sadeh, 2004), the Children's Sleep Habits Questionnaire (CHSQ) (Owens et al., 2000), Pediatric Sleep Questionnaire (PSQ) (Chervin et al., 2000), Sleep Disturbance Scale for Children (SDSC) (Bruni et al., 1996), Self-Administered Questionnaire for the Mother (Questionnaire about Sleep Habits; QASH) (Petit et al., 2002) and the Sleep Problems Scale (Bélanger et al., 2014) of the Child Behavior Checklist (CBCL) (Achenbach & Rescorla, 2000).

However, parents may not remember or be aware of all their infant's nighttime behaviors and this may affect the accuracy of reporting of these subjective assessment measures. Additionally, most of these measures may use means to measure sleep variables not taking into account night-to-night variability which is particularly high in infancy (Pennestri et al., 2020).

While these measures provide insight into the parental perception of infant sleep, subjective reports can also be influenced by mood (Dørheim et al., 2009; Hiscock & Wake, 2001; Morrell, 1999; Orhon et al., 2007). According to a meta-analysis, the prevalence rate of depressive symptoms in mothers during the period from the first trimester to the first year postpartum is approximately 24% (Paulson & Bazemore, 2010). Thus, given the prevalence and its potential to influence maternal report, it is important to consider mood and depressive symptoms when investigating subjective reports of sleep problems. Two commonly used

measures to assess mood during this period are the Center for Epidemiological Studies Depression Scale (CES-D) (Radloff, 1977) and the Edinburgh Postnatal Depression Scale (EPDS) (Cox et al., 1987).

Overall, none of these assessment methods will be ideal for all applications and are mostly complementary. However, it is important to consider each measure's strengths and limitations when interpreting study results.

### **Sleep Variables**

Different sleep variables can be derived from both objective and subjective methods. It is important to note that sleep stages in early infancy differ from those at later ages, as sleep stages are not categorized as rapid eye movement (REM) or non-rapid eye movement (NREM) until approximately two months of age (Lenehan et al., 2023; Sankupellay et al., 2011). Prior to this point, infant sleep stages are comprised of active sleep (AS; equivalent to REM), quiet sleep (QS; equivalent to NREM), and indeterminant or transitional sleep. These sleep variables are common when using sleep assessment methods such as PSG, pressure sensitive pads, and time lapsed video recordings. For example, researchers will measure AS or QS quantity, as well as bout length, which refers to length of the sleep stage before transitioning to the next. Another derivative investigated is the duration or frequency of arousals during AS or QS, which is determined by heightened continuous movement during one of these stages for approximately three minutes (Freudigman & Thoman, 1993). Microarousals, unlike the previous variable, can also occur in QS or AS but are more subtle and transient and last for seconds versus minutes (Franco et al., 2019). Another variable cyclicity refers to the consistency and predictability of QS bouts and is used to detect the development of sleep-wake rhythms in infants (Minard et al., 1999). Additional variables which appear less frequently and require further explanation (e.g.,

cyclicality) will be further defined and expanded upon in the Study 1 and Study 2 to avoid repetition.

Other variables can be obtained from both objective and subjective sleep measures. A commonly used variable, nocturnal sleep duration is defined as the total sleep time occurring from the late evening until morning (different studies use different time cutoffs) minus the duration of any night awakenings. Daytime sleep duration is usually defined as any sleep which occurs outside of the above window and is used to assess naps throughout the day. Total sleep duration is defined as the total number of hours spent in a sleeping state within a 24-hour period and is the sum of nocturnal and daytime sleep duration. This variable is presented either as a continuous or dichotomous variable as some studies will use a cutoff (e.g., > 12 hours at six months) to determine a sufficient quantity of sleep as recommended by the National Sleep Foundation guidelines (Hirshkowitz et al., 2015). Another variable which is often used is consecutive sleep duration and is also known as longest sleep period. This variable refers to the duration of the longest consecutive uninterrupted sleep within a 24-hour period and in common language, often referred to as sleeping through the night. Another subjective variable commonly used in infancy is the ratio of nocturnal sleep duration to total sleep duration. This variable is an additional measure of sleep consolidation which assesses the percentage of sleep occurring during the night as a percentage of total sleep duration, which is related to sleeping through the night (Bernier et al., 2013; Bernier et al., 2010). Sleep efficiency is generally expressed as a percentage and is calculated by dividing the total amount of time spent asleep by the total amount of time spent in bed (i.e., crib or bassinet).

Finally, sleep onset latency refers to the amount of time taken for an infant to fall asleep once they have been placed in a bed. Longer latency is associated with a difficulty in initiating sleep.

### **Defining Sleep Problems**

Sleep patterns and correspondingly what is considered to be a sleep problem may differ as a function of age. For example, a prolonged awakening at one month of age might be considered normative given the development of the circadian rhythm, yet this same awakening at four years old may be considered problematic.

Sleep problems in childhood may present in various ways which affect sleep quality, duration, and patterns of sleep. One of the most common of these behavioral sleep problems involves initiating and maintaining sleep (Kraenz et al., 2004). Insomnia occurs in approximately 20-30% of children and is the most prevalent sleep problem in childhood (Medalie & Gozal, 2018; Mindell et al., 2006). It may present as having difficulty falling asleep, staying asleep, resisting going to bed, or having problems with sleeping when not in the presence of parents or without parental support such as feeding and rocking (American Psychiatric Association, 2013). It is quite common for children to experience transient awakenings during the night and to fall back asleep shortly after without parental intervention (Touchette et al., 2007; Zuckerman et al., 1987). However, it is when these awakenings become prolonged, frequent, and require help from a caregiver that they become problematic.

Another behavioral sleep problem which may present during childhood are reoccurring nightmares. Nightmares are a commonly reported phenomenon with prevalence ranges of 10-50% during childhood (Singh et al., 2018). However, these do not generally cause significant disturbance to sleep unless they are recurrent. The prevalence rate for frequent nightmares in

children is estimated to be approximately 5% and approximately 20% of children with frequent nightmares also experience comorbid frequent insomnia (Lin et al., 2020).

Parasomnias are other types of sleep problems which may occur during childhood. These are defined as unwanted behaviors which occur while a child is sleeping or during the transition in or out of sleep (AASM, 2023). The most common of these parasomnias include sleep talking, sleepwalking, sleep terrors, and confusional arousals. Somniloquy (sleep talking), is talking that occurs during sleep which the child is unaware of and can range from simple sounds to complex and full sentences of speech (AASM, 2023). Somnambulism (sleepwalking) typically involves walking that occurs in a state of partial arousal where the child has little or no awareness of their surroundings (AASM, 2023). Sleep terrors or night terrors often present with screaming, intense fear, rapid heartbeat, sweating, complex motor behaviors, and also involve partial arousal (AASM, 2023). Confusional arousals involve partial awakening from sleep and often result in disorientation and confusion, and may be accompanied by disoriented behavior (AASM, 2023). A shared feature amongst these parasomnias is that the child has little or no memory of the event. These episodes are often brief, however, confusional arousals (partial awakening non-REM sleep) may last between 30 and 45 minutes (Mason & Pack, 2007). Confusional arousals and sleepwalking may occur in 17% to 40% of children, while sleep terrors occur in only 1% to 7% (AASM, 2023). Most parasomnias are more prevalent in children than adults and tend to decrease across development.

### **Assessing Sleep Problems**

Unlike assessing sleep problems in adults, sleep problems in children are often assessed through subjective *parental* report, meaning that most of the time, it is not the individual themselves reporting the problem, but it is rather the perception of the parent. Some behaviors

considered to be problematic by parents are night awakenings, not returning to sleep independently, and nighttime crying (Blunden et al., 2011). Additionally, some assessments consider any sleep behavior which does not align with parental expectations or disrupts parental sleep to be a sleep problem (Barry, 2021). Thus, many assessments of sleep problems in children are indirect and may be influenced by parental perceptions and expectations. Another challenge when surveying the pediatric sleep problem literature is that many studies do not use validated measures to assess problematic sleep. Many studies simply assess the presence or absence of sleep problems (yes/no responses) versus assessing the severity of individual sleep problems (night awakenings, nightmares, etc.) (Lam et al., 2003; Williamson et al., 2019).

An example of a validated measure used to assess individual sleep problems is the Sleep Problem subscale of the Child Behavior Checklist (CBCL) (Achenbach & Rescorla, 2000). The CBCL Sleep Problems subscale (maternal report) has been shown to have good correspondence with sleep efficiency and total sleep duration via maternal sleep diaries ( $r = -0.39$  to  $-0.25$ ,  $p < 0.05$ ) (Bélanger et al., 2014). Additionally, CBCL Sleep Problem subscale items (particularly questions assessing trouble sleeping, sleep duration, and waking up during the night) correlate moderately with other validated sleep measures such as The Children's Sleep Habits Questionnaire (CSHQ), PSG, and sleep disorder diagnoses (e.g., insomnia, sleep-disordered breathing, and circadian rhythm disorders) (Becker et al., 2015). Other examples of validated tools for assessing sleep problems in children include: The Children's Sleep Habits Questionnaire (CSHQ) (Owens et al., 2000), and The Children's Report of Sleep Patterns (CRSP) (Meltzer et al., 2013).

## **Cognitive and Psychomotor Development in Infancy**

While sleep problems during infancy have been shown to be associated with future sleep problems, the association between sleep patterns in infancy and cognitive and psychomotor development are not well known. Shorter sleep durations and poorer sleep quality have been associated with poorer cognitive outcomes in children and adults, however, while it might be assumed that this would also be true for this association in infancy, it is unclear as to whether this is the case (Lowe et al., 2017; Shochat et al. 2014; Reynaud et al., 2018).

Additionally, similar to the sleep-wake cycle, cognitive and psychomotor functions in infancy are in constant development (Cioni & Sgandurra, 2013). Their development also involves a complex interaction between brain maturation (internal) and environmental (external) factors as described in Bronfenbrenner's Social-Ecological model (Bronfenbrenner, 1977). In addition to the broad cognitive and psychomotor domains, other sub-domains such as language, executive function, fine and gross motor skills, memory, and sensory functioning are also a product of this interaction (Cioni & Sgandurra, 2013; Eickmann et al., 2012). It is uncommon to assess psychomotor functioning in older populations, as the most rapid psychomotor development occurs by early childhood (Cioni & Sgandurra, 2013). However, during infancy and early childhood, psychomotor functioning is an important indicator of development.

Cognitive and psychomotor developmental assessment in infancy is accomplished through behavioral observation. One of the most common tools used to assess cognitive and psychomotor functioning in infants are the Bayley Scales of Infant Development (BSID) (Bayley, 1969; Bayley, 1993). These consist primarily of the mental development (MDI) and psychomotor development (PDI) indices, in addition to other indices which assess social-emotional and adaptive behavioral functioning. The BSID were first developed to identify

developmental delay, formulate interventional strategies, as well as monitor developmental outcomes (Harris & Langkamp, 1994). While the BSID are useful, some studies have found the ability of the MDI to predict future cognitive ability to be poor (Hack et al., 2005). Yet, this may still be a valuable tool for understanding and comparing the developmental outcomes of groups of infants (Gauthier et al., 1999). Additional standardized measures such as the Weschler Preschool and Primary Scale of Intelligence (WPPSI) (Wechsler, 2002), Ages and Stages Questionnaire (ASQ) (Schonhaut et al., 2013; Squires et al., 1997), and others are also used in clinical and research settings. In addition to tests assessing broad cognitive and psychomotor development, other tests can be administered to evaluate specific domains such as executive functioning, working memory, expressive language, gross motor skills, and problem-solving abilities.

## **Association Between Early Sleep and Future Development**

### ***Association Between Sleep and Cognitive/Psychomotor Development***

One of the known correlates of sleep patterns in older populations is cognitive functioning. This association has been well explored in preschool-aged children, children, and adults. For example, shorter sleep duration in older adults (>55 years old) has been found to be a risk factor for cognitive decline and associated with poorer performance on tests of executive function and working and verbal memory (Lo et al., 2016). Another, a meta-analysis conducted by Lowe et al. (2017) examining 61 studies with a mean age of 28 found a significant negative effect of sleep restriction on several domains such as executive functioning, sustained attention, and long-term memory. Moreover, sleep problems have also been associated with cognitive impairment and Alzheimer's disease. One systematic review and meta-analysis found that individuals with sleep problems had an almost four times higher risk of developing Alzheimer's



disease or a cognitive impairment (Bubu et al., 2017). In addition, in another review individuals with insomnia were found to have performance deficits related to working memory, episodic memory, and executive functioning (Fortier-Brochu et al., 2012).

The association between sleep and cognitive functioning can also be observed during adolescence. A systematic review by Shochat et al. (2014) found that poor sleep during teenage years was associated with decreased academic performance. Another study focusing on adolescents manipulated sleep duration over the course of a week by creating two groups (sleep deprived versus non-sleep deprived) (Lo et al., 2016). Compared to the non-sleep deprived group, the sleep deprived group were found to have deteriorated sustained attention, working memory, executive functioning (Lo et al., 2016).

This association has also been observed in younger populations. Another meta-analysis of 86 studies and 35,936 children aged five to 12 years old found that longer sleep duration was also associated with higher cognitive performance (Astill et al., 2012). More recently, Reynaud et al. (2018) conducted a meta-analysis of 13 studies investigating the association between sleep and cognition in 37, 935 preschool-aged children (two to six years old). Findings suggested an association between higher sleep quantity and better cognitive outcomes in this age group. In another meta-analysis consisting of 19 studies of the same age group, similar findings showed that children with longer sleep durations had better cognitive outcomes (Short et al., 2018). Lastly, sleep problems have also been associated with inattention and ADHD symptoms in children (Gruber, 2014).

In general, the association between sleep and cognitive function in children and adults is well studied and the negative impact of reduced sleep duration or disturbed sleep on cognitive function is well established the literature. However, the association between sleep and

cognitive/psychomotor functioning during infancy is not yet fully understood. As sleep during infancy is dynamic and in a state of perpetual change, it differs from sleep in children and adults. Thus, its association with cognitive/psychomotor and other developmental outcomes may also evolve over time. It is also possible that the developmental nature of cognitive/psychomotor functioning in infancy may influence this association.

### ***Association Between Early Sleep and Future Sleep Problems***

Despite the high prevalence rate of sleep problems in children, it is not clear whether or not sleep problems during childhood are related to earlier sleep patterns (Byars et al., 2012; Gaylor et al., 2005; Owens, 2008). Few studies have investigated this association, as most of them have examined the association between perceived sleep problems (parental endorsement) during infancy with reported sleep problems at later ages. For example, Lam et al. (2003) assessed sleep problems (eight to 48 months) by asking parents if their child had a sleep problem (yes/no) and did not assess specific sleep pattern variables (e.g., total sleep duration, longest period of consecutive sleep, sleep efficiency, etc.) However, approximately 12% of parents reported that their infant's sleep problem had persisted into childhood and another 19% of parents reported that their infant's sleep problem had reoccurred at a later timepoint. Another study by Williamson et al. (2019) asked parents "How much is your child's sleeping pattern or habits a problem for you?" and selected "a large problem, a moderate problem, a small problem, no problem at all or not sure/don't know". The authors found that 17% of children had minimal sleep problem, 14.4% had mild sleep problems which were maintained across timepoints, and 7.7% had persistent sleep problems throughout the study. However, while both these studies documented the persistence of sleep problems from one timepoint to a future timepoint, these were not compared to infant sleep pattern variables.

Lastly, Byars et al. (2012) did assess early sleep patterns and their association with parentally reported sleep problems at six, 12, 24, and 36 months. However, the authors only compared these variables concurrently at the same timepoint (e.g., six-month sleep problems with six-month total sleep duration) and not with future reports of sleep problems. While the authors found that shorter sleep duration and more frequent night awakenings were significantly associated concurrently at six, 12, and 24 months, analyses were not conducted to examine the association between early sleep patterns and future sleep problems. Additionally, the authors did not use a validated tool to measure sleep problems and instead used a single question “Do you think (child’s name) has problems sleeping?” However, the authors found that infants with parentally reported sleep problems (yes/no) during this period were three-to-five times more likely to have a sleep problem at a later timepoint.

### **Summary of Key Limitations in the Existing Literature**

There are several limitations and gaps in the existing literature, which the current dissertation aims to address. Primarily we do not have a thorough understanding of the correlates of early sleep patterns in healthy full-term infants, particularly their associations with cognitive/psychomotor development and future sleep problems.

### ***Infant Sleep and Cognitive/Psychomotor Development in Full-Term Infants***

First, as discussed in this literature review, while the association between sleep and cognitive functioning has been further explored in children, adolescents, and adults, this remains to be thoroughly investigated in infants, as one cannot presume that similar associations will be observed. Indeed, while several studies examining this association have been published, a comprehensive synthesis of the literature is required to gain a more complete understanding of this topic. While a previous (non-systematic) review examining the association between infant

sleep patterns and cognitive/psychomotor development was published by Ednick et al. (2009), this review had several limitations. The authors included infants born prematurely (under 37-weeks' gestation) and infants born with neurological, physiological, and/or genetic disorders were not explicitly included. An updated systematic review utilizing a quality assessment of articles in an attempt to reduce bias would be helpful to contribute to a more comprehensive understanding of this association. Conducting a systematic review would also help as it would include the most recently published articles as 15 years have passed since the previous review was undertaken. Additionally, focusing on only full-term healthy infants may aid in clarifying this complex association. Lastly, extracting and classifying specific sleep pattern variables from studies to determine whether they are associated with cognitive/psychomotor development would help to identify which sleep variables should be prioritized.

### ***Early Sleep Patterns and Future Sleep Problems***

Few studies have examined early sleep variables and their association with sleep problems into later childhood. While previous studies have investigated the association between perceived sleep problems in early infancy and childhood, these studies have focused on perceived sleep problems and their persistence into childhood (Byars et al., 2012; Lam et al., 2003; Williamson et al., 2019). One study assessed sleep patterns and their association with concurrent sleep problems but did not investigate the association between early sleep patterns and *future* sleep problems (Byars et al., 2012). The use of a validated tool to assess sleep problems such as the CBCL Sleep Problems subscale is also needed and may be more accurate and may provide more specificity than the measures used in previous studies (Achenbach & Rescorla, 2000). Identifying early sleep variables and disentangle the contribution of *total* sleep duration versus *consecutive* sleep duration) during early infancy are associated with future

reports of sleep problems using a validated measure would allow parents and health professionals to determine which sleep patterns should be emphasized.

The present dissertation will address the previously mentioned limitations by: (1) Implementing a systematic search strategy to synthesize the available literature concerning the sleep of healthy, full-term infants and cognitive/psychomotor development (Study 1), (2) Using a quality assessment tool to aid in the interpretation of the reviewed research evidence (Study 1), (3) Using a longitudinal design to examine the association between specific sleep variables in early infancy and sleep problems in preschool-aged children, (Study 2) and (4) Employing a validated measure to assess sleep problems (Study 2) and (5) Increase the understanding that specific sleep variables in infancy play for later developmental and sleep outcomes (Study 1 and Study 2).

### Chapter 3: Study 1

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## The Association Between Infant Sleep, Cognitive, and Psychomotor Development: A Systematic Review

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## **Abstract**

### **Study Objectives:**

To synthesize findings of original articles examining the association between sleep-wake patterns of typically developing infants aged 0-to-18 months and cognitive and psychomotor development.

### **Methods:**

A systematic search strategy was used to identify articles assessing the association between infant sleep (0-to-18 months) and cognitive/psychomotor development (Medline, PsycINFO, SCOPUS). Of 7,136 articles screened, 22 articles met inclusion criteria, and the results were subsequently synthesized. A quality assessment was conducted, and studies were categorized as “poor”, “fair”, or “good”.

### **Results:**

Out of 22 studies, two found exclusively significant associations between infant sleep and cognitive/psychomotor development, three found no significant associations and 17 found mixed results. Studies with exclusively significant results used a single sleep variable and single timepoint designs. Studies finding mixed results or no significant associations used multiple sleep, developmental variables, or multi-timepoint designs. Eight out of 10 studies and seven out of eight studies investigating nocturnal and total sleep duration, respectively, found no significant association with developmental outcomes. While 63% of studies were rated as having good methodological quality, all studies but one had estimated power of less than 0.80.

### **Conclusion:**

Findings of this review do not support conclusive associations between sleep-wake patterns in infancy and cognitive/psychomotor development. This conclusion contrasts with the literature in

older populations, questioning if the association between sleep and development is of a different nature in infancy, potentially because of brain maturation. More studies including larger samples will be needed to clarify the presence or absence of such an association.

**Keywords:** Pediatrics – Infants, Cognitive Development, Sleep/Wake Cognition



## Introduction

Sleep is a key factor involved in child development and brain maturation<sup>1-5</sup>. The association between sleep and cognitive functioning has been well investigated in adults and in children<sup>6-8</sup>. For instance, shorter sleep duration has been associated with lower cognitive performance in preschool and school-aged children<sup>6,9,10</sup>. Sleep problems in early childhood have also been associated with poorer school achievement scores<sup>11,12</sup>.

While the association between sleep and cognitive functioning has been explored further in adults and children, less is known about this association in infancy. Indeed, sleep is a developmental process and sleep-wake patterns in infancy are markedly different than those in older children<sup>9,11,13-16</sup>. During the first several weeks of life, infants spend approximately 16 to 18 hours per day in a sleeping state<sup>17-19</sup>. During this period, sleep is highly fragmented with infants sleeping for short periods of two to three hours<sup>17,20,21</sup>. Moreover, infants' ability to differentiate between day and night during early infancy is limited as their circadian rhythm is in a developmental state<sup>16</sup>. Sleep stages during early infancy differ from those in older populations. During infancy, rapid eye movement (REM) and non-REM sleep are classified as quiet sleep (QS), active sleep (AS), and indeterminate (IS) sleep, until approximately two months of age<sup>22,23</sup>.

Gradually, the percentage of nocturnal sleep will increase over time, while nocturnal awakenings will decrease as well as the number and duration of daytime naps<sup>24</sup>. The development and consolidation of the sleep-wake cycle in infancy is influenced by multiple internal (e.g., circadian timing system, temperament) and external (e.g., light exposure, parental behaviors) factors<sup>16,25-30</sup>. One of the main challenges related to the investigation of sleep during infancy is that it is a developmental process, whose maturation varies significantly from infant to infant<sup>15,17,20</sup>. Moreover, major variations also occur from night to night within the same

infant<sup>31,32</sup>. Considering the inter- and intra-individual variability and the developmental aspect, it is unclear whether infant sleep patterns are associated with measures of cognitive and psychomotor functioning in early development. The associations observed in older children and adults may not be applicable during earlier developmental periods.

To that effect, a previous review conducted by Ednick et al<sup>33</sup> in 2009 examined the association between sleep and development during the first year of life. Based on the 18 studies included in the review, the authors concluded that a causal relationship could not be drawn concerning infant sleep and its association with cognitive and psychomotor development during this period of development. Moreover, the authors observed that findings from one developmental period (e.g., first 48 hours of life; six months) could not be generalized to another, emphasizing the developmental nature of sleep during infancy. While this review paper clearly expanded the understanding of the association between infant sleep and development during early infancy, it did not employ a systematic search process nor include a quality assessment. Moreover, both full-term and preterm infants were included in the review, introducing an important confounding variable, since sleep in preterm infants exhibits distinct characteristics and developmental trajectories<sup>20,34,35</sup>. Likewise, the methodology was also not explicit regarding the inclusion or exclusion of other comorbidities, such as neurological, physiological, or genetic disorders. Finally, new studies may have been published since Ednick et al's review<sup>33</sup> and could help to refine and update our understanding of the association between infant sleep and developmental outcomes.

Building on this previous review, the present systematic review focuses on the associations between infant sleep and cognitive and psychomotor development in full-term healthy infants aged 0 to 18 months. Specifically, the aims of this review are to 1) synthesize the

available research assessing the association between infant sleep (< 18 months) and cognitive and psychomotor development in typically developing infants and 2) assess and critique the current quality of knowledge. This systematic review will provide updated knowledge and offer an overview of the current available research. Given the developmental nature of infant sleep, results will be organized and presented as a function of age.

### **Methods**

This systematic review was registered with the International Prospective Register of Systematic Reviews (CRD42019123272) and conducted according to the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA)<sup>36</sup>.

#### **Article Search and Selection**

The search strategy was designed by the research team in collaboration with a research librarian. All databases were searched on August 17<sup>th</sup>, 2018, and included MEDLINE (1946 to Present), PsycINFO (1806 to Present), and SCOPUS. The keyword *Sleep* was solely used while searching MEDLINE and PsycINFO, as the addition of *Cognitive* and *Psychomotor* produced limited results. The search strategy can be accessed in the supplementary material (appendix A). All results were imported into EndNote and duplicate records were discarded using the de-duplicate method developed by Bramer and colleagues<sup>37</sup>. Updated searches were conducted on February 14<sup>th</sup>, 2020, September 7<sup>th</sup>, 2022, and March 10<sup>th</sup>, 2024, and yielded eight additional studies, which met our inclusion criteria.

The subsequent stages of the review were conducted independently by two researchers (B.B. and G.M-G. or R.B.) to minimize bias. A third member of the research team was available to consult in the event of a disagreement. The search yielded a total of 8336 articles. After the removal of duplicates, 7136 abstracts were screened in Rayyan—a web and mobile app for

systematic reviews<sup>38</sup> for the following inclusion criteria: (1)  $\leq 18$  months of age at time of sleep assessment, (2) born at full-term ( $\geq 37$  weeks), (3) absence of neurological, physiological, and/or genetic disorders (autism spectrum disorder Down's syndrome, epilepsy, asthma, etc.), (4) assessed participant sleep, either with an objective or subjective measure (e.g. polysomnography, actigraphy, observation, time-lapse video, motility monitoring system, sleep diary, sleep questionnaire, etc.), (5) assessed participant cognitive and/or psychomotor functioning (e.g. Bayley Scales of Infant Development, Ages and Stages Questionnaire, Wechsler Preschool & Primary Scale of Intelligence, etc.), (6) full-text available in English or French, and (7) only empirical quantitative studies were included (i.e., case studies and review articles were excluded), and (8) experimental studies in which diurnal sleep (napping) was manipulated to assess its immediate impact on a specific cognitive test. Based on these criteria, the full texts of 98 articles were examined in further detail and 22 studies were included in the final sample. See Fig. 1 for the PRISMA flow diagram.

### **Data Extraction**

The following data were extracted from papers that met the inclusion criteria: (1) number of study participants; (2) mode of sleep assessment (i.e., actigraphy, sleep diary, etc.), 3. sleep variables (i.e., number of awakenings, longest sleep period, etc.), (4) age at time of sleep assessment, (5) duration of sleep assessment, (6) type of cognitive and/or psychomotor assessment, (7) age at time of cognitive and/or psychomotor assessment, and (8) study outcomes comparing sleep variables and cognitive and/or psychomotor variables, (9) confounding variables (e.g., socioeconomic status, maternal age at conception, breastfeeding status, etc.). The extracted data were organized and grouped as a function of infant age at the time of the sleep assessment.

## Secondary Classification of Sleep Variables

In an attempt to further document the associations between specific sleep variables and developmental outcomes, a secondary classification was performed. Therefore, 101 associations were extracted from the 22 included studies and classified into three categories: (1) significant association (SA), (2) non-significant association (NS), or (3) mixed results (MR). Mixed results were defined as follows: an association was significant at one timepoint but non-significant at an additional timepoint; at the same timepoint, certain outcome variables were significant, but others were non-significant; or an outcome variable was significant at one time point and significant at an additional timepoint, but with opposite directionality (positive vs. negative associations).

## Quality Assessment

The methodological quality of included studies was assessed using the National Institutes of Health (NIH) Quality Assessment Tool for Observational Cohort and Cross-Sectional Studies<sup>39</sup>. This tool contains 14 items (Yes, No, and Not Applicable options) related to sampling procedure, sample sized justification (i.e., power analysis), study design, psychometric properties, and the presence of covariates used in the analyses. As per the tool's instruction, each study was assigned a rating of *good*, *fair*, or *poor*, as shown in Table 1. The quality assessment was conducted independently by two researchers (B.B. and C.L.) after which discrepancies were discussed and ratings were combined. A third researcher was available in the event that a consensus could not be reached.

## Power Estimates

Post hoc power analyses were conducted to assess the adequacy of sample sizes in the included studies using G\*Power Version 3.1.9.6<sup>40</sup>. Two relevant meta-analyses<sup>9,10</sup> investigating

the associations between sleep and cognitive outcomes in children aged five to 13 years old reported mean average effect sizes of  $r = 0.08$  and  $r = 0.06$ . As a result, conventional cut-offs corresponding with small effect sizes<sup>41</sup> (Pearson's  $r = 0.1$ , Cohen's  $d = 0.3$ , Cohen's  $f^2 = 0.02$ ) were used to mitigate the risk of calculation errors. A significance level of  $\alpha = 0.05$  was used for power calculation estimates.

## Results

Table 1 provides a detailed summary of the papers selected for inclusion and highlights associations found between infant sleep-wake patterns and developmental outcomes. Six studies (27.3%) used objective measures to assess sleep, 11 studies (50%) used subjective measures, and the remaining five studies (22.7%) employed a combination of both. The results have been summarized below and grouped as a function of infant age at the time of the sleep assessment: (1) First 48 hours of life ( $n = 2$ ); (2) four to 11 months ( $n = 8$ ); (3) 12 months or greater ( $n = 2$ ); and (4) longitudinal studies using several timepoints ( $n = 10$ ). Studies in which infants' age overlapped with these groups were assigned to the group of best fit. Studies were additionally organized into subcategories based on whether they used subjective or objective measures or a combination of both.

### Power Calculation Estimates

Power analyses were conducted for 19 of 22 studies. There were three<sup>42-44</sup> for which power analyses could not be performed in G\*Power as these studies used linear mixed effects, generalized linear, or multilevel modelling. However, given the sample sizes of these three studies and the very small effect sizes being used for the calculations, it is likely that power would have fallen below 0.80. Overall, power ranged from 0.063 to 0.876, with only one study<sup>45</sup> meeting the power requirements to detect a small effect size. Due to the low power of the

included studies, there exists an increased risk that type II errors were made and thus the following non-significant results should be interpreted with caution.

### **First 48 hours of life ( $n = 2$ )**

#### *Objective measures*

Two studies investigated the association between sleep and development in the first 48 hours of life. Freudigman and Thoman<sup>46</sup> assessed the sleep of infants ( $n = 36$ ; power = 0.089; quality = fair) using the Motility Monitoring System<sup>47,48</sup> (MMS; a pressure sensitive pad) and found that higher sleep-wake transition percentage and shorter mean sleep periods on the first day of life were associated with better cognitive and psychomotor scores at six months (BSID). Moreover, greater longest-sleep period and more arousals in quiet sleep were associated with poorer cognitive scores, but not with psychomotor outcomes. While these associations were not significant on the second day, longer quiet sleep bout length and greater quiet sleep quantity were associated with poorer cognitive scores (day 2).

Another study<sup>49</sup> also assessed infant sleep ( $n = 27$ ; power = 0.078; quality = good) on the first two days of life using the MMS and found that higher sleep-wake transition and shorter mean active sleep bout length on days one and two were associated with poorer facial recognition (FTII) scores at 6 months, but not at 9 months. Higher sleep-wake transition (day two), as well as higher frequency of arousals in active sleep (day one) were associated with poorer problem-solving scores at nine months (WIPT). Additionally, longer mean sleep periods on the second day were associated with better facial recognition (FTII) scores at six months.

Taken together, these two studies show some negative and positive significant associations between sleep in the first two days of life and early cognitive or psychomotor development, but results seem inconsistent as a function of the day and sleep variable.

Specifically, findings concerning the association between the variables sleep-wake transition percentage and mean sleep period with developmental outcomes appear to be contradictory.

### **Five to 11 Months ( $n = 8$ )**

#### *Subjective measures*

In a study<sup>45</sup> conducted in infants ( $n = 590$ ; power = 0.876; quality = good) ranging from two to 11 months old, a lower number of nocturnal awakenings (BISQ; Brief Infant Sleep Questionnaire<sup>50</sup>) was associated with better cognitive (MDI) but not psychomotor (PDI) scores, after controlling for birth weight, maternal education, breast-feeding, and bed-sharing practices. Total sleep duration was not associated with cognitive or psychomotor outcomes. In another study<sup>25</sup>, less variability in circadian sleep-wake cycles across four nights of sleep ( $n = 62$ ; power = 0.148; quality = fair) at seven months (telephone interviews) was associated with better cognitive ability (MDI) at 24 months and better language development (RDLS) at 36 months, while controlling for sex, temperament, maternal sensitivity, parenting values, and education.

Tham et al<sup>51</sup> divided ( $n = 267$ ; power = 0.648; quality = good) six-month-old infants into two groups based on parental-report (BISQ) of total 24-hour sleep duration: (1) Short Sleepers (less than 10 hours each day) and (2) Typical Sleepers (10 to 18 hours each day). Typical sleepers performed better than short sleepers (six months) on a memory task which involved the infant imitating three actions of an experimenter. Longer sleep latency was associated with poorer ability to relate an object to a location at six months. No associations were found between the duration or frequency of nighttime awakenings and developmental outcomes. Another study from Scher et al<sup>52</sup> using the Sleep Questionnaire<sup>53</sup> showed that higher sleep problem scores ( $n = 83$ ; power = 0.147; quality = poor) at nine months (composite sleep problem score) were associated with poorer object permanence ability (nine months). Lukowski and Milojevich<sup>54</sup> used



the BISQ to assess sleep ( $n = 21$ ; power = 0.072; quality = good) at 10 months of age and found that shorter duration of daytime napping and higher percentages of sleep occurring at night were associated with poorer immediate recall (imitation task). Furthermore, shorter duration of daytime napping and more night awakenings were associated with poorer generalization ability, while nighttime sleep duration was not associated with any developmental outcome.

### *Objective and subjective measures*

Other studies assessed the association between sleep and development between 5 and 11 months using both objective and subjective sleep measures. Scher<sup>55</sup> assessed infant sleep ( $n = 59$ ; power = 0.084; quality = fair) using actigraphy and the Sleep Questionnaire<sup>56</sup> at eight months of age and found that infants who had greater nocturnal sleep fragmentation (composite score of number of interrupted nights, number of awakenings per night, and average time spent awake), and longer sleep duration had better gross motor ability (GMCL). More nocturnal awakenings were also associated with better gross motor ability, while longest continuous sleep period and sleep efficiency were not associated with gross motor ability. In another study using actigraphy and a sleep questionnaire ( $n = 50$ ; power = 0.106; quality = fair), which appears to use the same sample, Scher<sup>57</sup> found that more motor activity during sleep, more episodes of night-waking, and lower sleep efficiency were associated with poorer mental ability (MDI) in infants aged 10 months. Nocturnal sleep duration was not associated with developmental outcomes and no significant associations were found between sleep variables and psychomotor ability (PDI). Gossé et al<sup>42</sup> ( $n = 76$ ; power could not be determined; quality = fair) assessed sleep (four to 14 months of age) using actigraphy, the BISQ, and a sleep diary and observed that longer nocturnal sleep duration was associated with better gross motor (actigraphy) and communication ability (sleep diary) (ASQ) in younger infants, while the opposite was found in older infants. Longer

daytime sleep duration throughout the age range was associated with poorer problem-solving (actigraphy, BISQ, sleep diary), communication (actigraphy), and fine motor abilities (actigraphy). Increased frequency of night awakenings (sleep diary) was associated with poorer fine motor ability in younger infants and better ability in older infants. Lastly, higher wake after sleep onset (WASO) was associated with better communication and poorer gross motor abilities at all ages (sleep diary).

In summary, the results from the above studies investigating sleep between five and 11 months old show some negative and positive significant associations between sleep patterns and developmental outcomes. Interestingly, while some studies showed that poorer sleep was associated with better development, others showed the contrary. Again, an important number of associations were not significant, and others were inconsistent or even contradictory.

## **12 Months or Greater ( $n = 2$ )**

### *Subjective measures*

Bernier et al<sup>58</sup> ( $n = 65$ ; power = 0.202; quality = good) employed sleep diaries to assess the sleep of infants at 12 months and found that while not associated with general cognitive ability (MDI), higher ratios of night-to-total sleep were associated with better executive functioning and reasoning abilities (WPPSI) after controlling for socioeconomic status (SES) and previous developmental outcomes. Total sleep duration was not associated with any developmental outcome.

### *Objective measures*

In the same age range, Montgomery-Downs and Gozal<sup>59</sup> ( $n = 20$ ; power = 0.070; quality = good) did not find any association between any sleep variables measured with actigraphy at 14 months (total sleep time, sleep efficiency, mean sleep bout time, immobile percentage, mean

activity in sleep, movement and fragmentation index) and infant mental (MDI) or psychomotor ability (PDI).

In this age and study design category, only ratio of nighttime sleep-to-total sleep was associated with better cognitive development.

### **Longitudinal Studies ( $n = 10$ )**

#### *Subjective measures*

Sleep was assessed at multiple timepoints in ten studies. Liang et al<sup>60</sup> ( $n = 182$ ; power = 0.270; quality = good) assessed the sleep of infants at six and 12 months using the BISQ and found that longer daytime sleep duration was associated with poorer gross motor (six months) and fine motor ability (12 months) (PDI). Additionally, greater frequency and longer duration of awakenings were associated with poorer gross motor ability at 12 months. Onset of nocturnal sleep, duration of nocturnal sleep, and sleep onset latency were not associated with motor ability (PDI). Another study ( $n = 156$ ; power = 0.222; quality = good) used the BISQ to assess sleep at four and eight months of age and found that higher quantities of daytime sleep at four and eight months were associated with better language ability (MCDI-SF) and developmental scores (DP3), respectively, while adjusting for temperament, breastfeeding, physical activity, siblings, gender, maternal education, and pacifier use<sup>61</sup>. Night sleep duration, number of night awakenings, and duration of night awakenings were not associated with language or developmental outcomes.

Pennestri et al<sup>15</sup> assessed sleep consolidation of infants at six ( $n = 388$ ; power = 0.490; quality = good) and 12 ( $n = 369$ ; power = 0.460; quality = good) months using the question, “During the night, how many consecutive hours does your child sleep without waking up?”. Sleeping through the night was operationalized as six or eight hours of consecutive sleep and

was not associated with cognitive or psychomotor functioning at six, 12, and 36 months. Another multi-timepoint study<sup>62</sup> ( $n = 194$ ; power = 0.254; quality = good) assessed sleep (BISQ) at six, 12, and 18 months of age and found that more frequent night awakenings were associated with poorer developmental scores (WPPSI) at six months while controlling for potential confounding variables (e.g., SES, maternal BMI, smoking status). However, at 12 and 18 months this association ceased to be significant. Moreover, lower day-to-nighttime sleep ratio at 12 and 18 months, but not six months, was also associated with poorer developmental scores (WPPSI). Longer daytime sleep duration at 18 months was associated with better development scores. Lastly, total sleep and nighttime sleep duration were not associated with cognitive functioning.

Bernier et al<sup>63</sup> ( $n = 60$ ; power = 0.145; quality = fair) assessed sleep using sleep diaries and found that infants with higher ratios of night-to-total sleep at 12 and 18 months had better impulse control, controlling for SES, working memory, and cognitive functioning (MDI). Higher ratios of night-to-total sleep at 18 months were also associated with better working memory and cognitive ability (MDI) scores. Total sleep duration and sleep fragmentation (number of awakenings) were not associated with any developmental outcome.

### *Objective measures*

Minard et al<sup>64</sup> ( $n = 58$ ; power = 0.120; quality = good) used the MMS to assess sleep on the first and second postnatal days, as well as at six months of age and divided infants into two groups (significant periodicity and non-significant periodicity) based on a cyclicity score (defined as periodicity and predictability of quiet-sleep bouts). Infants in the significant versus non-significant groups were infants whose sleep cycles were deemed to be regular versus irregular, respectively. Infants with more developed cyclicity at six months were found to have better cognitive abilities (MDI) than the non-significant group (second day was not significant).

Another study<sup>43</sup> ( $n = 78$ ; power could not be determined; quality = good) using polysomnography on the first and second postnatal days and at six months found that infants with more daytime sleep, higher daytime sleep efficiency, fewer microarousal during the day, and more arousals at night had poorer cognitive ability (WPPSI) at 36 months. Night sleep duration, night sleep efficiency, and microarousals were unrelated to cognitive outcomes.

Becker and Thoman<sup>65</sup> (group 1:  $n = 15$ ; power = 0.064; group 2:  $n = 14$ ; power = 0.063; quality = fair) assessed infant sleep (two, three, four, five weeks; three, six, 12 months) and found that a greater number of rapid eye movement (REM) storms (short intense bursts including eye movement during active sleep) at six and 12 months were associated with poorer cognitive ability (BSID) at 12 months. The amounts of active sleep and quiet sleep were not related to cognitive ability at any timepoint.

#### *Objective and subjective measures*

Pisch et al<sup>44</sup> ( $n = 40$ ; power could not be determined; quality = good) used actigraphy and the BISQ to assess infant sleep at four, six, eight, and 10 months of age and found that infants who spent less time awake during the night displayed earlier signs of memory maturation (working memory task). However, no association was found between average night waking frequency, nighttime sleep duration, and daytime sleep duration, and cognitive outcomes.

Another longitudinal study<sup>66</sup> ( $n = 20$ ; power = 0.070; quality = good) measured sleep using a sleep diary and actigraphy at three, six, 11, and 12 months of age and found no significant relationships between any sleep variable (nighttime, daytime, and total sleep) and developmental ability assessed at 12 months (BSID).

Overall, the studies investigating infant sleep and developmental outcomes using multiple timepoints found few significant associations between sleep and developmental outcomes. Yet,

like in previous studies using a single timepoint, these results appear to be inconsistent and vary considerably by sleep variable and developmental outcome. For instance, out of the nine longitudinal studies, six studies measured nocturnal sleep duration and none of these studies found significant associations with any developmental outcome. Moreover, contradictory associations were also observed for the same sleep variables.

### **Classification of Specific Sleep Variables**

To better identify which sleep-wake variables were or were not associated with developmental outcomes a secondary analysis was conducted. A total of 101 associations between sleep and cognitive or psychomotor development were extracted from the 22 included studies and classified as significant, non-significant, or mixed results (significant at one timepoint but non-significant at an additional timepoint; at the same timepoint, certain outcome variables were significant, but others were non-significant; or an outcome variable was significant at one time point and significant at an additional timepoint, but with opposite directionality [positive vs. negative associations]). The vast majority were either non-significant ( $n = 54$ , 53.5%) or mixed ( $n = 40$ , 39.6%) and only six associations ( $n = 6$ , 5.9%) were categorized as consistently significant. These six significant associations were extracted from four different studies<sup>25,44,52,55</sup>. The power of these four studies ranged from 0.058 to 0.097 and the power of one study<sup>44</sup> was not estimated due to employing a multilevel modelling analysis. The quality of two studies<sup>25,55</sup> were rated as fair, one<sup>52</sup> as poor, and the other<sup>44</sup> as good. The following six sleep-wake variables were associated with better developmental outcomes: more developed circadian sleep regulation, lower sleep problem scores, less time awake after sleep onset, more night awakenings, and poorer sleep continuity.

## Summary of the Risk of Bias Appraisals

Fourteen of the 22 studies (63%) included in this review were rated overall as having good methodological quality, seven were rated as fair (32%), and one was rated as poor (5%). In terms of strengths, most studies (72%) measured infant sleep prior to measuring developmental outcomes, and 86% used valid and clearly defined developmental outcome measures. All studies measured varying levels of sleep using multiple variables such as total sleep duration, number of awakenings, and duration of night awakenings. More than half (59%) of the studies measured infant sleep at more than one timepoint and half of the included studies attempted to control for confounding variables such as SES, maternal age, and breastfeeding status.

Additionally, loss to follow-up was reported as less than 20% in approximately half of the studies (45%) and the dropout rates were either not reported or could not be determined for half of the studies (45%). The remaining 9% of studies had losses higher than 20% at follow up. Rates of participation of eligible persons were not reported by more than three quarters of the studies (77%), with four explicitly reporting rates over 50%, and one study reporting a participation rate below 50%. Finally, almost all studies (95%) did not provide a sample size justification (i.e., power analysis).

## Discussion

The aim of this review was to survey and synthesize the existing literature on the association between sleep in infants aged 0 to 18 months and cognitive and psychomotor development. While the association between sleep and daytime functioning is more established in older populations, the current review suggests that this association is not as clear and more complex in infancy. Among the 22 studies included in this review, only two<sup>25,52</sup> reported exclusively significant results, three<sup>15,59,66</sup> found no significant associations, and the remaining

17 found a mix of significant and non-significant results between sleep and cognitive/psychomotor outcomes. Examples of mixed results include associations being significant at one timepoint but non-significant at an additional timepoint<sup>43,46,49,61,62,67</sup>; at the same timepoint, certain outcome variables were significant, but others were non-significant<sup>42,43,45,46,49,51,54,57,58,61,63,67</sup>; or an outcome variable was significant at one time point and significant at an additional timepoint<sup>42</sup>, but with opposite directionality (positive vs. negative associations). The observed lack of clarity may be attributed, in part, to the low power in the included studies, potentially contributing to the non-detection of significant associations and increasing the likelihood of type II errors.

While approximately two thirds of studies ( $n = 14$ ) were rated as having “good” methodological quality, the other one third of studies were rated as fair ( $n = 7$ ) and poor ( $n = 1$ ) and thus the results from these studies should be interpreted while taking this into account. The majority of studies rated as “good” were found to have mixed results ( $n = 11$ ) and the remaining did not find any significant associations ( $n = 3$ ). In contrast, as it will be further developed below, the single study<sup>52</sup> rated as “poor” was one of two studies which found exclusively significant associations. Studies rated as fair also found mostly mixed results ( $n = 6$ ) and one found exclusively significant results.

One ( $n = 83$ ; power = 0.147; quality rating = poor)<sup>52</sup> of the two studies reporting exclusively significant associations showed that a higher sleep problem composite score at nine months was associated with poorer object permanence ability. This study only used a single sleep variable (although based on a sleep composite score) as well as a single developmental outcome variable (object permanence). Not including covariates in the analysis mainly contributed to the “poor” rating of this study. The second study ( $n = 62$ ; power = 0.148; quality rating = fair)<sup>25</sup>



reporting exclusively significant associations demonstrated that more developed circadian sleep regulation at seven months was associated with better mental and language development. However, it is important to note that this study relied on a single sleep variable (circadian sleep regulation).

In contrast, the three studies<sup>15,59,66</sup> that did not find any significant associations all employed multiple sleep variables and developmental variables and were rated as having “good” methodological quality. Notably, two<sup>15,66</sup> out of three studies employed multi-timepoint designs. Among the 18 studies with mixed results, eight<sup>43,44,61-65,67</sup> utilized a multiple timepoint design, while nine<sup>42,45,46,49,51,54,55,57,58</sup> relied on a single timepoint. Of these 18 studies, 17 utilized multiple sleep and/or developmental variables. The one exception<sup>25</sup>, which used a single sleep and developmental variable, employed a multi-timepoint design.

Studies using a single time point or single sleep outcome variable were of course more likely to yield to exclusively significant results without the presence of additional inconsistencies. Thus, it is possible that these findings would have been categorized as mixed or inconsistent results if multiple timepoints and/or outcome variables had been employed. It is also possible that more sleep variables were tested but not reported given the non-significant results, although it is not possible to verify this hypothesis.

In order to better identify which sleep variables were associated or not with developmental outcomes, a secondary classification was performed among the 101 total associations included in the 22 studies. Once again, these results should be interpreted cautiously given the low power estimates of all but one included study. Most of these associations were either non-significant ( $n = 54$ , 53.5%) or mixed ( $n = 40$ , 39.6%) with only six associations ( $n = 6$ , 5.9%) being categorized as consistently significant. These six significant associations were

extracted from four<sup>25,44,52,55</sup> studies, one with a “good” quality rating and two being rated as “fair” and one as “poor”. Dearing et al<sup>25</sup> ( $n = 62$ ; power = 0.148; quality rating = fair) showed that more developed circadian sleep regulation at seven months was associated with better mental and language development. Scher et al<sup>52</sup> ( $n = 83$ ; 0.147; quality rating = poor) found that a higher sleep problem composite score at nine months was associated with poorer object permanence ability. Another article reporting a significant association was from Pisch et al<sup>44</sup> ( $n = 40$ ; power could not be determined; quality rating = good) showing that infants aged four-to-10 months who spent less time awake after sleep onset demonstrated better memory maturation. Lastly, while the fourth study showed two other significant associations, these were in the opposite direction of what might be expected. Indeed, Scher<sup>55</sup> ( $n = 59$ ; power = 0.084; quality rating = fair) found that infants at eight months with poorer sleep continuity and more night awakenings had better gross motor outcomes. Specifically, infants who had achieved the milestone of “crawling” were found to have more night awakenings (>5 minutes) than “non-crawlers”. The authors posited that this may have been a result of brief arousals turning into longer episodes due to the infants’ newly developed locomotion skills. It should be noted that this study appeared to use the same sample as another study included in the review and thus the results of this study should be interpreted with this in mind<sup>57</sup>.

The high variability in sleep measures, sleep variables, as well as developmental outcome variables clearly limited the ability to compare the association between sleep and development across studies. Indeed, nine different sleep assessment measures using objective or subjective tools were employed across the included studies, ranging from polysomnography to telephone-based interviews. Additionally, studies also varied widely regarding developmental variables with a total of 11 different instruments being used. Harmonizing sleep and

developmental measures would be highly beneficial to this field of research and could improve comparisons between studies.

However, despite the observed variability, some specific sleep variables consistently failed to yield significant results. For instance, among 10 studies measuring nocturnal sleep duration, eight studies<sup>43,44,54,57,61,62,66,67</sup> found no significant association with development outcomes, one study<sup>55</sup> found a significant association and one study<sup>42</sup> showed mixed results. A similar pattern was observed for total sleep duration over 24 hours, where six<sup>45,58,59,62,63,66</sup> out of seven studies failed to find a significant association with developmental outcomes, and one<sup>51</sup> found mixed results. Daytime sleep duration exhibited a more variable pattern, with six<sup>42,43,54,61,62,67</sup> studies yielding mixed results and two<sup>44,66</sup> studies finding no significant associations with developmental outcomes. Lastly, the ratio of night-to-total sleep also displayed variability with three studies<sup>54,58,63</sup> reporting mixed results.

While a previous review investigating infant sleep and development by Ednick et al<sup>33</sup> did not utilize categorizations as those employed in the present review and did not exclude preterm births or neurodevelopmental conditions, the authors arrived at similar conclusions. The authors noted inconsistencies such as changes in directionality of relationships depending on the timing of sleep assessment in included studies. They also observed unexpected associations, such as less total sleep being linked to higher developmental scores in some studies. In line with these findings, the present review arrives at a similar conclusion. Despite employing a systematic search strategy and focusing exclusively on typically developing infants born at term, an association between infant sleep-wake patterns and cognitive and/or psychomotor development could not be conclusively established.

There is a greater amount of literature available investigating this association in children over 18 months of age. A systematic review comprised of 19 studies by Short et al<sup>10</sup> found that shorter sleep duration patterns between five and 13 years old were generally associated with poorer cognitive functioning. Additionally, another systematic review<sup>6</sup> of 26 studies also found that longer sleep durations in two- to six-year-old children were associated with better cognitive outcomes, although effect sizes were observed to be small. Moreover, similar results were found in a systematic review<sup>9</sup> of 86 studies showing that school-aged children aged five to 12, with longer sleep duration associated with better executive functioning, cognitive tasks performance, and school performance.

Various factors could explain this differing pattern in infancy, in addition to the fact that both sleep and cognitive functioning are still in development during this period of time. The National Sleep Foundation Guidelines<sup>19</sup> publishes sleep guidelines for infants, children, and adults, and recommend that infants (four to 12 months) sleep for 12 to 16 hours (including naps) each day, resulting in a large range of “normal” sleep duration in infancy. However, the majority of the studies included in this review measure sleep duration using continuous variables. Perhaps one way to better evaluate the consequences of sub-optimal sleep during infancy on later development would be to target infants who sleep less than these normative ranges using thresholds versus the continuous variables, allowing for these important interindividual differences. Finally, while 14 studies (64%) incorporated meaningful covariates known to be associated with sleep and development, the remaining eight (36%) studies did not include any covariates, potentially influencing the reported results. A good example is found in the study of Dearing et al<sup>25</sup> where the addition of covariates (sex, temperament, maternal education, maternal

sensitivity, and parenting values) strengthened the relationship between circadian sleep regulation and developmental outcomes.

## **Limitations**

The findings of the current review should be considered in the context of various limitations. As discussed previously, the main limitation is the consistent underpowered status across almost all included studies, which may have contributed to the notable proportion of non-significant results. However, the power calculation should also be nuanced as a function of the specificity of the sleep measure (for example a questionnaire versus actigraphy or polysomnography). Smaller sample sizes are probably less concerning in studies using objective sleep measures than subjective ones. Moreover, assessing sleep with objective sleep measures, such as polysomnography, in infants may also raise concerns related to participant burden, ethical considerations, and resource intensiveness, therefore limiting the possibility of recruiting larger sample sizes. An objective measure such as actigraphy may be a better choice given its ability to track infant sleep over several days in their home environment.

Despite the utilization of a systematic search strategy, it is conceivable that some eligible studies may have been inadvertently omitted. Additionally, publication bias may have influenced the results due to excluding non-peer-reviewed studies and grey literature. Indeed, one can wonder if the number of non-significant associations could be even higher due to publication bias regarding the publication of significant results. This review also excluded studies which were not published in English or French and those lacking available translations in these languages. The diversity of sleep assessment tools, sleep variables, and timepoints, precluded the feasibility of conducting a meta-analysis. While the majority of included studies

demonstrated good methodological quality, it is noteworthy that almost all of them lacked sample size justifications.

## **Conclusion**

Results from this systematic review showed that most studies assessing the association between infant sleep and psychomotor or cognitive development yielded mixed or inconsistent findings in this specific developmental period. Results from this review should not be interpreted as a conclusion that infant sleep is not important for healthy development. Instead, it underscores the need for the field of pediatric sleep to refine which sleep variables, at what age, and in what context could more accurately predict optimal or suboptimal indices of development. Due to the low power of the majority of included studies, future investigations should consider increasing sample sizes where feasible to account for non-detection of significant findings. Additionally, perhaps looking at persistent poor sleep patterns and considering interindividual variability could also constitute interesting avenues. Overall, clinicians working with families and infants should be aware that as of today, we do not have sufficient data to describe a causal relationship between sleep patterns and development in early infancy. These two developmental processes are in a constant evolution during early development, are highly variable and are also largely influenced by both biological determinants and the environment.

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**Table 1.** Extracted data from included studies

Author (Year)	Sample size and quality appraisal	Sleep assessment (duration) and timing	Sleep variables	Cognitive and/or psychomotor assessment	Outcome(s)
First 48 hours of life ( $n = 2$ )					
Freudigma n et al (1993) <sup>46</sup>	N = 36 (20f, 16m)  Quality appraisal :  Fair	MMS (24 hours)  Age: Postnatal days 1 & 2	Active sleep %, Quiet sleep %, Active-quiet transitional sleep %, Sleep-wake transition %, Wake %, Active sleep bout length, Quiet sleep bout length, Mean sleep period, Longest sleep period, Arousals in quiet sleep	BSID (MDI and PDI)  Age: 6 months	<b>Mixed results</b>  Significant: Day 1: Higher sleep-wake transition percentage and shorter mean sleep periods associated with higher cognitive and psychomotor scores. Higher longest sleep period and more arousals in quiet sleep associated with lower cognitive scores.  Day 2: Longer quiet sleep bout length and greater amount of quiet sleep were both associated with lower cognitive scores.  Non-significant: Quantity of active sleep, active-quiet transitional sleep, wake time, and active sleep bout length not associated with cognitive or motor scores.  Co-variates: Sex, mode of delivery (vaginal caesarean section)

Judge et al (2015) <sup>49</sup>	N = 27	MMS (24 hours)	Active sleep, Quiet sleep, Sleep-wake transition, Wakefulness, Arousals in quiet sleep, Arousals in active sleep, Mean bout length of quiet sleep, Mean bout length of active sleep, Mean sleep period, Longest sleep period, Active sleep to quiet sleep ratio	WIPT (Problem-solving):  Age: 9 months  FTII (Facial recognition)  Age: 6 & 9 months	<p><b>Mixed results</b></p> <p>Significant: Higher sleep-wake transition and shorter mean active sleep bout length on days 1 and 2 were associated with poorer facial recognition memory at 6 months.</p> <p>Higher sleep-wake transition (day 2) and frequency of arousals in active sleep (day 1) were associated with lower problem-solving abilities at 9 months.</p> <p>Longer mean sleep period on day 2 was associated with better facial recognition at 6 months.</p> <p>Non-significant: Sleep-wake transition and number of arousals in active sleep on day 2 were not related to problem-solving. No association between sleep and facial recognition at 9 months. No association between wakefulness, longest sleep period, and cognitive outcomes</p> <p>Co-variates: None</p>
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Sun et al (2018) <sup>45</sup>	N = 590 (269f, 321m)  Quality appraisal :  Good	BISQ (7 days)  Age: 2 to 11 months ( $M = 5.23$ )	Sleep duration, Nighttime awakenings	BSID (MDI and PDI)  Age: 2-11 months ( $M = 5.23$ )	<p><b>Mixed results</b></p> <p>Significant: Infants who awoke twice during the night had higher cognitive scores than those who awoke once and <math>\geq 3</math>x times per night, after controlling for birth weight, maternal education, breast-feeding, and bed-sharing practices.</p> <p>Non-significant: Awakenings were not associated with psychomotor scores Total sleep duration was not associated with cognitive or psychomotor scores No sleep variables associated with psychomotor scores.</p> <p>Co-variates: Infant age and sex, birth weight, maternal education level, bedroom sharing, current feeding pattern.</p>
Dearing et al (2001) <sup>25</sup>	N = 62 (29f, 33m)  Quality appraisal :  Fair	Telephone interview (4 days)  Age: 7 months	Circadian sleep regulation (via periodogram analysis)	BSID (MDI)  Age: 24 months  RDLS  Age: 36 months	<p><b>Significant</b></p> <p>Significant: More advanced circadian sleep regulation at 7 months predicted better cognitive outcomes at 24 months and language abilities at 36 months.</p> <p>Co-variates: Sex, temperament, maternal education, maternal sensitivity, and parenting values.</p>

Tham et al (2019) <sup>51</sup>	N = 267 (117f, 150m)	BISQ (7 days)	Sleep latency, Total sleep duration	Relational memory: deferred imitation, relational binding (eye-tracking).  Recognition memory: recognition/novel ty preference.  Attentional orienting: visual expectation.  Age: 6 months	<b>Mixed results</b>  Significant: Typical sleepers (10-18 hours/day) were better at imitating an action than short sleepers (< 10 hours/day). Longer sleep latency was associated with poorer ability to relate an object to a location.  Non-significant: No associations between duration or frequency of nighttime awakenings, and imitating an action, ability to relate an object to a location, or recognition memory. No association between sleep onset latency and deferred imitation  Co-variates: Breast-feeding exposure, napping, maternal anxiety and sensitivity, maternal education
	Quality appraisal :				
	Good				
		Age: 6 months			
Scher et al (2000) <sup>52</sup>	N = 83 (N/A)	Sleep question- naire (duration = N/A)	Sleep composite score	Object permanence test  Age: 9 months	<b>Significant</b>  Significant: Higher composite sleep problem scores were associated with poorer object permanence ability.  Co-variates: None
	Quality appraisal :				
	Poor	Age: 9 months			

Lukowski et al (2013) <sup>54</sup>	N = 21 (N/A)	BISQ (7 days)	Nighttime sleep duration, Frequency of night awakenings, Daytime nap duration, % of sleep obtained at night	Elicited imitation task  Age: 10 months
	Quality appraisal :	Age: 10 months		
	Good			

**Mixed results**

## Significant:

Shorter duration of daytime napping and higher percentages of sleep occurring at night were associated with poorer immediate imitation. Shorter duration of daytime napping and more night awakenings were associated with poorer delayed recall generalization.

## Non-significant:

Nighttime sleep duration and night awakenings unrelated to encoding.

Nighttime sleep duration and percentages of sleep occurring at night were unrelated to generalization.

All sleep variables unrelated to recall memory

## Co-variates:

None

Scher (2005a) <sup>55</sup>	N = 55 (N/A)	Actigraphy (3 days); Sleep diary (7 days)	Nightwaking index, Schedule index, Sleep onset time, Sleep duration, % of activity per minutes of sleep, # of transitions from sleep to wake,	Gross Motor Checklist  Age: 8 months
*same sample as Scher 2005b	Quality appraisal :	Age: 8 months		
	Fair			

**Mixed results**

## Significant:

Infants who had achieved the crawling milestone had higher night-waking index scores, more awakenings, and longer sleep durations than non-crawlers.

## Non-significant:

No differences for sleep onset time, activity percentage, sleep-wake transition, longest sleep segment, sleep efficiency, or schedule index.

			Longest sleep period, Sleep efficiency, # of awakenings	
Scher (2005b) <sup>57</sup>	N = 50 (24f, 26m)	Sleep questionnaire (7 days); Actigraphy (3 days)	Sleep onset time, Sleep duration, % of activity per minutes of sleep, Sleep efficiency, Number of awakenings, Nightwaking index, Schedule index	BSID  Age: 10 months
*same sample as Scher 2005a	Quality appraisal :  Fair	Age: 10 months		
Gosse et al (2022) <sup>42</sup>	N = 76 (42f, 34m)	Actigraphy (7 days); BISQ (7 days); Sleep diary (7 days)	Day sleep duration, Night sleep duration, Night awakenings, Wake after sleep onset	ASQ  Age:
	Quality appraisal :  Fair	Age 4-14 months		

Co-variates:  
Infant sex

### Mixed results

Significant:  
Higher motor activity during sleep, more episodes of night-waking, and lower sleep efficiency were associated with lower cognitive scores.

Non-significant:  
No association between sleep variables and psychomotor scores  
Schedule index and night-waking index (sleep diary), and sleep onset and duration (Actigraphy) were not associated with cognitive scores.

Co-variates:  
None

### Mixed results

Significant:  
Longer nocturnal sleep duration associated with higher gross motor scores (actigraphy) and communication scores (sleep diary) at younger ages. Opposite association in older infants.  
Longer day sleep duration associated with lower problem-solving scores (actigraphy, BISQ,

4-14 months

sleep diary), association became stronger with age.

Longer day sleep duration associated with lower communication and lower fine motor scores (actigraphy).

More night awakenings were associated with lower fine motor score (sleep diary) in younger infants. Opposite relationship found in older infants

More wake after sleep onset (sleep diary) was associated with higher communication scores in younger infants. Opposite relationship found in older infants.

More wake after sleep onset (sleep diary) was associated with lower gross motor scores.

Non-significant

Night sleep duration not associated with problem-solving skills, or fine motor scores.

Day sleep duration not associated with gross motor scores

Night awakenings not associated with communication, gross motor, or problem solving

WASO was not associated with problem-solving skills or fine motor scores.

Co-variates:

Infant age

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12 Months or Greater ( $n = 2$ )

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Bernier et al (2013) <sup>58</sup>	N = 65 (38f, 27m)	Maternal sleep diary (3 days)	Sleep duration, % of total sleep occurring at night	BSID (MDI)  Age: 12 months  Spin the pots; delay of gratification; shape stroop; baby stroop  Age: 2 years  WPPSI-III (Information and Matrix Reasoning subscales)  Age: 4 years	<b>Mixed results</b>  Significant: Higher ratio of nighttime/total sleep duration at 1 year old predicted higher cognitive scores at four years after controlling for socioeconomic status and cognitive scores at 1 and 2 years.  Non-significant: Sleep duration was not predictive of cognitive scores.  Co-variates: Child sex and birth weight, maternal and paternal age and education, family income
Montgomery-Downs et al (2006) <sup>59</sup>	N = 20 (8f, 12m)	Actigraphy (5 days); PSG (1 day)	Sleep start time, Sleep end time, Total sleep time, Sleep efficiency %, Mean sleep bout time, Immobile %, Mean activity in sleep,	BSID  Age: 14 months	<b>Non-Significant</b>  No significant associations  Co-variates: None
	Quality appraisal :  Good	Age: 12 months			



Movement and  
fragmentation  
index

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Longitudinal Studies ( $n = 10$ )

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Liang et al (2022) <sup>60</sup>	N = 182 (89f, 93m)  Quality appraisal :  Good	BISQ (7 days)  Age: 6 & 12 months	Timing of nocturnal sleep onset, Nocturnal sleep duration, Daytime sleep duration, Sleep onset latency, Frequency of awakenings, Duration of awakenings	BSID III (PDI)  Age: 6 & 12 months	<p><b>Mixed results</b></p> <p>Significant Longer daytime sleep duration associated with lower gross motor scores at 6 months Longer daytime sleep duration associated with lower fine motor scores at 12 months Greater frequency and longer duration of awakenings associated with lower gross motor scores at 12 months</p> <p>Non-significant Onset of nocturnal sleep, duration of nocturnal sleep and sleep onset latency not associated with BSID scores.</p> <p>Co-variates: None</p>
Pecora et al (2022) <sup>61</sup>	N = 156 (75f, 81m)  Quality appraisal :	BISQ  Age:	Night sleep duration, Day sleep duration, Night awakening #, Night awakening	DP3 (cognitive and communication subscales)  MCDI-SF  Age:	<p><b>Mixed results</b></p> <p>Significant: Longer daytime sleep duration at 4 months was associated with higher MCDI-SF scores at 8 months Longer daytime sleep duration at 8 months was associated with higher DP3 cognitive scores</p>

	Good	4 & 8 months	time	4 (DP3) & 8 (DP3 & MCDI-SF) months	<p>Non-significant: Night sleep duration, night awakening # or time not associated with DP3 or MCDI-SF scores</p> <p>Co-variates: Temperament, breastfeeding, physical activity, siblings, gender, maternal education, and pacifier use</p>
Pennestri et al (2018) <sup>15</sup>	<p>6 months: N = 388 (182f, 206m); 12 months: N = 369 (176f, 193m)</p> <p>Quality appraisal :</p> <p>Good</p>	<p>SAQM-Adapted. (2 weeks)</p> <p>Age: 6 &amp; 12 months</p>	<p>Sleeping through the night (6-hour window), Sleeping through the night (8-hour window)</p>	<p>BSID</p> <p>Age: 6, 12, &amp; 36 months</p>	<p><b>Non-significant</b></p> <p>No significant associations</p> <p>Co-variates: Infant sex, SES, breastfeeding status, co-sleeping status, total sleep duration</p>
Plancoulaine et al (2017) <sup>62</sup>	<p>N = 194 (N/A)</p> <p>Quality appraisal :</p>	<p>BISQ (duration = 7 days)</p> <p>Age:</p>	<p>Night awakenings, Day-to-night sleep ratio</p>	<p>WPPSI-III</p> <p>Age: 36 months</p>	<p><b>Mixed results</b></p> <p>Significant: Answering “Yes” vs. “No” to night awakenings at 6 months, and lower day-to-night sleep</p>

	Good	6, 12, & 18 months	Total sleep duration, Daytime sleep duration, Night sleep duration		duration ratio at 12 and 18 months was associated with lower IQ scores. Longer day time sleep duration at 18 months was associated with higher IQ scores  Non-significant: Total sleep duration, night sleep duration  Co-variables: SES, maternal BMI, maternal smoking status, infant birth order, TV watching
Bernier et al (2010) <sup>63</sup>	N = 60 (36f, 24m)  Quality appraisal :  Fair	Maternal sleep diary (3 days)  Age: 12 & 18 months	Sleep duration, % of total sleep occurring at night, Sleep fragmentation	BSID (MDI)  Age: 12 months  Hide the pots  Age: 18 months  Spin the pots; delay of gratification; shape stroop; baby stroop  Age: 26 months  MCDI	<b>Mixed results</b>  Significant: Higher ratio of night/total sleep at 18 months predicted better working memory at 18 months while controlling for SES and cognitive functioning (MDI) at 12 months. Higher ratio of night/total sleep at 12 and 18 months predicted better impulse control at 26 months after controlling for SES, working memory, and cognitive functioning (BSID; MDI) at 12 months.  Non-significant: Ratio of nighttime/total sleep at 12 and 18 months did not predict better working memory, set shifting, or inhibitory control at 26 months Total sleep duration and sleep fragmentation (number of awakenings) were not associated with any developmental outcomes

				Age: 18 & 26 months	Co-variates: Child age, sex, number of siblings, maternal and paternal age and education, family income
Minard et al (1999) <sup>64</sup>	N = 58 (26f, 32m)	MMS (24 hours)	Cyclicity (significant vs. non-significant)	BSID (MDI)	<b>Mixed results</b>
	Quality appraisal :  Good	Age: Postnatal days 1 & 2; 6 months (2 days)		Age: 6 & 12 months	Significant: Significant cyclicity on day 1 associated with lower 6-month cognitive scores. Significant cyclicity at 6 months associated with higher 12-month cognitive scores.  Non-significant: Cyclicity assessed on day 2 was not associated with cognitive outcomes. 6-month cyclicity was not associated with 6-month cognitive outcomes.
					Co-variates: Birth weight, maternal age
Franco et al (2019) <sup>43</sup>	N = 78 (37f, 41m)	PSG (24 hours)	Sleep duration, Sleep efficiency, Active sleep %, Quiet sleep %, Micro-arousals, Micro-arousals in active sleep, Micro-arousals in quiet sleep	WPPSI-III (FSIQ, VIQ, PIQ, GLC)	<b>Mixed results</b>
	Quality appraisal :  good	Age: Postnatal day 1 & 2; 6 months		Age: 36 months	Significant: Infants with longer daytime sleep duration and fewer microarousals during the day had lower FSIQ, VIQ, and GLC scores. Higher daytime sleep efficiency was associated with lower FSIQ, PIQ, and GLC scores More nighttime micro arousals were associated with lower VIQ and GLC scores  Non-significant:

Becker et al (1981) <sup>65</sup>	Two sub-groups: N = 15 (7f, 8m) and N = 14 (8f, 6m)  Quality appraisal :  Fair	In-laboratory observation (2-5 weeks = 7 hours; 3-12 months = 2 hours)  Age: 2, 3, 4, 5 weeks, 3, 6, 12 months	REM storms, Active sleep quantity, Quiet sleep quantity, REM sleep quantity	BSID     Age: 12 months
Pisch et al (2019) <sup>44</sup>	N = 40 (21f, 19m)  Quality appraisal :	Actigraphy & BISQ (7 days)  Age: 4, 6, 8, & 10 months	Wake after sleep onset, Night sleep duration, Night waking frequency, Daytime sleep duration	Eye tracking   Age: 4, 6, 8, & 10 months

Night sleep duration, night sleep efficiency, percent active sleep, percent quiet sleep, microarousals during active sleep, and microarousals during quiet sleep unrelated to cognitive outcomes.

Co-variables:

Maternal age at delivery, tobacco consumption and anxiety score during pregnancy, SES, sex, gestational age, breastfeeding duration

### Mixed results

Significant:

In two samples, more REM storms at 6 and 12 months were associated with poorer cognitive outcomes.

Non-significant:

Active sleep, quiet sleep, and total REM at all ages not correlated with cognitive outcomes.

Co-variables:

None

### Mixed results

Significant:

Infants who were awake for less time during the night showed earlier signs of memory maturation.

Non-significant:

Good

Night Sleep Duration, Night Waking Frequency,  
Daytime sleep duration

Co-variates:

Sex, number of siblings, sleeping arrangement,  
and maternal educationSpruyt et  
al (2008)<sup>66</sup>N = 20  
(7f, 13m)Actigraphy &  
Sleep diary  
(3 days)Diurnal sleep,  
Nocturnal sleep,  
Total sleep

BSID

**Non-Significant**

No significant associations

Quality  
appraisal  
:Age:  
3, 6, 11, & 12  
monthsAge:  
12 monthsCo-variates:  
None

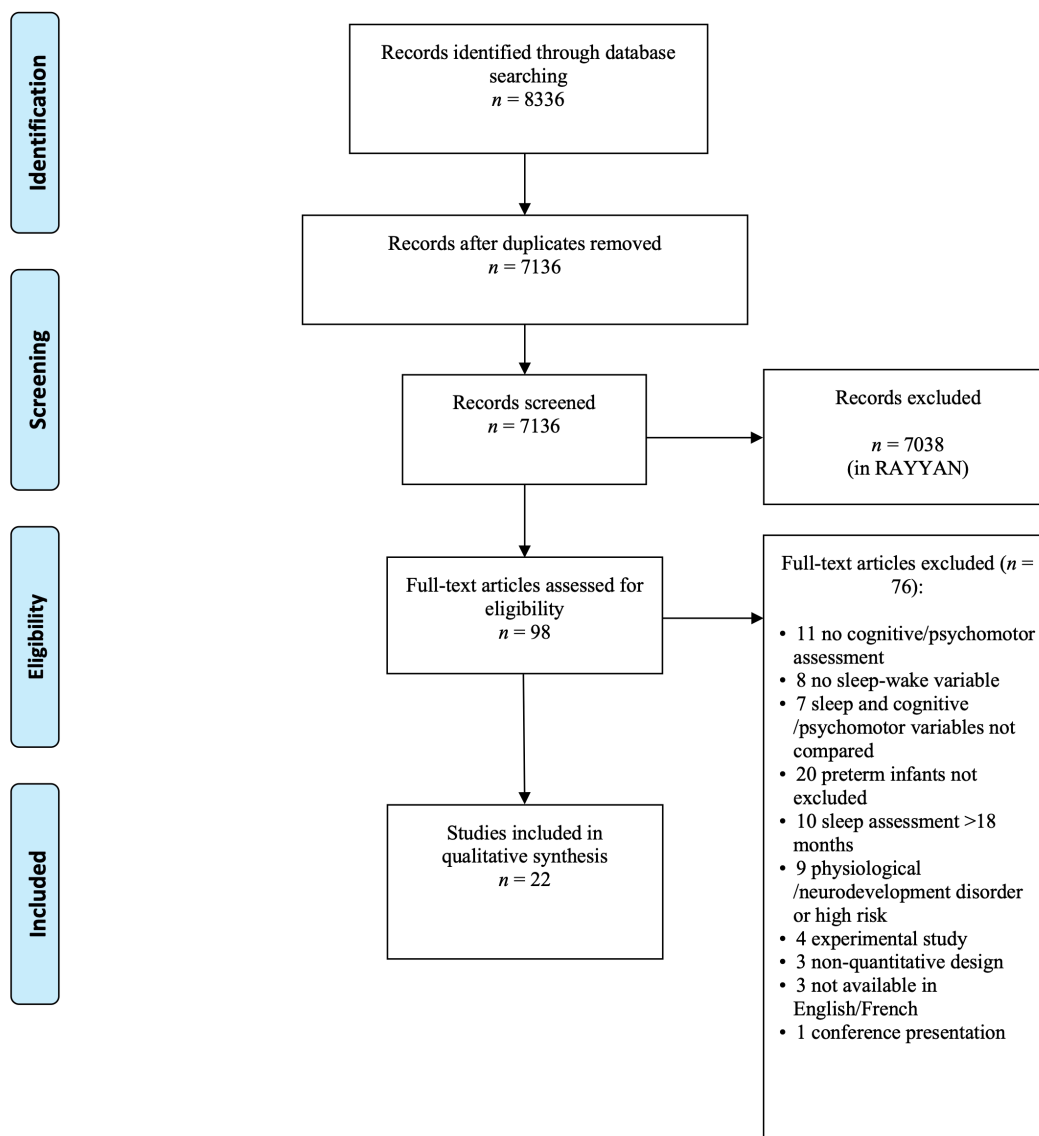
Good

N/A = not available; m = male; f = female; SES = socioeconomic status; REM = rapid eye movement; BSID = Bayley Scales of Infant Development; MDI = Mental Development Index; PDI = Psychomotor Development Index; FTII = Fagan Test of Infant Intelligence; MMS = Motility Monitoring System; WIPT = Willatts Infant Planning Test; PSG = polysomnography; WPPSI-III = Wechsler Preschool & Primary Scale of Intelligence – Third Edition; FSIQ = full-scale intelligence quotient; VIQ = verbal intelligence quotient; PIQ = performance intelligence quotient; GLC = general language composite score; RDLS = Reynell Developmental Language Scales; BISQ = Brief Infant Sleep Questionnaire; SAQM = Self-Administered Questionnaire for the Mother – Adapted; ASQ = Ages and Stages Questionnaire; DP3 = Developmental Profile; MCDI-SF = MacArthur-Bates Communicative Development Inventory - Short Form; MCDI = MacArthur-Bates Communicative Development Inventory

**Table 2.** List of cognitive and psychomotor measures

Cognitive/Psychomotor Measure	Abbreviation
Ages and Stages Questionnaire <sup>68,69</sup>	ASQ
Bayley Scales of Infant Development <sup>70-72</sup> (Mental Development Index)	BSID (MDI)
(Psychomotor Development Index)	(PDI)
Developmental Profile-3 <sup>73</sup>	DP3
Fagan Test of Infant Intelligence <sup>74</sup>	FTII
Gross Motor Checklist <sup>75</sup>	GMCL
MacArthur-Bates Communicative Developmental Inventory <sup>76</sup>	MCDI-SF
Reynell Developmental Language Scales <sup>77</sup>	RDLS
Wechsler Preschool and Primary Scale of Intelligence <sup>78</sup>	WPPSI
Willatts Infant Planning Test <sup>79</sup>	WIPT

Figure 1. PRISMA flowchart





## Appendix

### Supplementary Material

#### Search Strategy

##### *Medline*

This version of the database was utilized for the search: MEDLINE® and Epub Ahead of Print, In-Process & Other Non-Indexed Citations, Ovid MEDL®(R) Daily 1946 to Present. The following search terms were used: SLEEP (subject heading), limited by age group (All infants).

##### *PsycINFO*

PsycINFO 1806 to Present. The following search terms were used: SLEEP (subject heading), exploded, and limited by age group (120 neonatal <birth to age 1 mo> or 140 infancy <2 to 23 mo>).

##### *SCOPUS*

The default version of SCOPUS was used for the purpose of this search. The following terms were used: (TITLE-ABS-KEY (sleep) AND TITLE-ABS-KEY (infant OR baby OR babies OR infants) AND TITLE-ABS-KEY (cogni"\* OR "psycho"moto"" OR "psych"moto"" OR "psycho"motor")). Due to the interdisciplinary nature of SCOPUS, the additional search terms 'cognitive' and 'psychomotor' were added to limit search results.

#### **Chapter 4: Transition Statement from Study 1 to Study 2**

Study 1 employed a systematic review to examine the developmental correlates of early infant sleep patterns. Specifically, this review identified and synthesized the available research concerning infant sleep (0 to 18 months) and cognitive/psychomotor development. Overall, the majority of the studies included in the review found mixed results and thus the association between sleep and cognitive/psychomotor functioning during infancy appears to be less clear than in older populations. However, despite most studies finding mixed results, associations with certain individual sleep variables appeared to be more conclusive. For example, total sleep duration and nocturnal sleep duration were both found to be mostly unassociated with cognitive/psychomotor development. The association with daytime sleep duration, consecutive sleep, and ratio of nocturnal-to-total sleep duration was more variable. Overall, the findings from Study 1 indicate that the association between early sleep patterns and cognitive/psychomotor development is complex, the currently available research evidence is far from conclusive, and findings differ substantially from that of older populations where suboptimal sleep has been shown to be associated with poorer cognitive outcomes. These results indicate that further investigation of the future correlates of early sleep patterns is warranted, as the associations between these specific sleep variables and other correlates may differ.

Additional future correlates of early infant sleep patterns which are not well understood are future sleep problems. Studies examining the association between infant sleep patterns and future sleep problems are not as prevalent as those investigating infant sleep patterns and cognitive/psychomotor development. Thus, additional original studies are needed to better understand this association. The association between early sleep problems and later perceived sleep problems has been investigated in the literature and it has been found that these sleep

problems can persist into later childhood. However, it is not known whether early infant total sleep duration or consecutive sleep duration are also associated with future perceived sleep problems. Building on Study 1, Study 2 will continue to investigate the future correlates of early sleep patterns by further examining these associations. Overall, these insights will help both parents and health professionals determine if total sleep duration or consecutive sleep duration should be prioritized.

## Chapter 5: Study 2

(Prepared for submission to the journal: *Sleep Health*)

### **Shorter Total Sleep Duration but not Consecutive Sleep Duration in Infancy is Associated with Future Sleep Problems in Preschoolers**

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### **Abstract**

*Objectives:* Poor sleep during childhood can affect multiple domains of functioning. However, the association between early infant sleep patterns and future sleep problems is not well understood. This study aims to assess whether total sleep duration over the course of 24 hours and consecutive sleep duration in infancy are associated with later parental reports of sleep problems in preschoolers.

*Methods:* This longitudinal study included a community sample of 286 mother-child dyads. Infant sleep was measured using the Questionnaire about Sleep Habits (QASH) at six months. Sleep problems were assessed using the Sleep Problems subscale of the Child Behavior Checklist (CBCL/1.5-5) at 48 and 60 months.

*Results:* Generalized Estimating Equations models revealed that infant shorter total sleep duration, but not consecutive sleep duration was associated with more sleep problems in preschoolers (higher total CBCL Sleep Problems subscale scores), after adjusting for socioeconomic status (SES), maternal depression, breastfeeding status, sleeping arrangements, time, and biological sex. Regarding specific sleep dimensions, infant shorter total sleep duration was associated with more bedtime resistance, difficulty falling asleep, the presence of nightmares, more nocturnal awakenings, and shorter sleep duration, while consecutive sleep duration was not associated with any specific dimensions.

*Conclusions:* Shorter total sleep duration but not consecutive sleep duration at six months was a marker of parental reports of sleep problems during preschool years. Results suggest that total sleep duration over 24 hours and sufficient opportunity to sleep during the night and day should be prioritized over sleep consolidation (often referred to as sleeping through the night) during early infancy.

## Introduction

The importance of sleep during child development cannot be overstated as it affects multiple areas of functioning encompassing physiological<sup>1-3</sup>, cognitive<sup>4</sup>, and mental health<sup>5,6</sup> domains. As a result, parents may experience pressure to help their infant consolidate sleep quickly with the goal of promoting good sleep habits and mitigating future sleep problems<sup>7,8</sup>. Indeed, interventions targeting infant sleep<sup>9,10</sup> are being developed and delivered at progressively younger ages.

In the first few weeks of life, infants are unable to distinguish between day and night as their circadian rhythm is still maturing<sup>11,12</sup> resulting in highly fragmented sleep across 24 hours. Initially, infants sleep in periods of two to three hours of consecutive sleep, which will gradually increase throughout development<sup>12-15</sup>. When describing nocturnal infant sleep, parents and health professionals often refer to “sleeping through the night”, which is often defined as a six- or eight-hour period<sup>16-18</sup> of consecutive sleep. Although the duration of this consecutive period is not always consistent in the literature, sleeping through the night is generally used as a measure of consecutive sleep<sup>18,19</sup> duration, without interruption, which is different from total sleep duration. As sleep consolidates, daytime sleep duration begins to decrease and infants have fewer naps each day<sup>20</sup> until eventually almost all sleep occurs during the night later in childhood<sup>21,22</sup>. Prior to this point, according to the National Sleep Foundation<sup>23</sup>, it is important to consider total sleep duration (defined as total sleep within a 24-hour period), which includes both nocturnal sleep and daytime sleep (naps).

It is estimated that approximately 20-35% of children will experience sleep problems during childhood<sup>24-26</sup>. These are alarming numbers, given that sleep problems are associated with behavioral<sup>27,28</sup>, cognitive<sup>29,30</sup>, and emotional<sup>31,32</sup> issues. Whether or not sleep problems have their

origin in early sleep patterns is not fully understood and was not much explored. Indeed, very few studies have looked at the association between sleep patterns in infancy and the development of later sleep problems, as most of them rather focused on the *persistence* of sleep problems from infants to preschool aged children. For instance, one study<sup>33</sup> assessing parentally reported sleep problems (yes/no) at six timepoints from infancy to 11 years old found that 14.4% of children had mild sleep problems over time, 7.7% had persistent sleep problems throughout middle childhood, and 17% of children had minimal sleep problems in early childhood. Another study<sup>34</sup> found that 12% of infants with a sleep problem at eight to 10 months still had sleep problems at three to four years old. In contrast, a study<sup>35</sup> assessed parental reports of sleep problems at four, 12, 24 months, and six years found that sleep problems at earlier timepoints did not predict problems at six years. While these studies assessed the evolution of parental perception of sleep problems from infancy to childhood, often using simple dichotomous variables (presence or absence of sleep problems) or Likert scales of sleep problem severity, they did not assess the association between infant sleep patterns such as total sleep duration or consecutive sleep duration during early infancy and later sleep problems.

One study<sup>24</sup> by Byars et al. did assess early sleep patterns and found that shorter total sleep duration and more frequent night awakenings were associated with more perceived sleep problems at six, 12, and 24 months. However, these variables were compared concurrently (e.g., at the same timepoint: six-month total sleep duration with six-month sleep problems) so the association between early sleep patterns and future sleep problems was not examined. Additionally, no validated tool was used to measure sleep problems but rather a dichotomous variable (“yes” or “no” endorsement) similar to the previous studies. Moreover, only a general sleep problems measure was used, and it was thus not possible to specify what type of sleep

problems were associated with sleep patterns (i.e., difficulty falling asleep, not sleeping enough, presence of nightmares).

While these studies suggest that parental perception of early sleep problems may be associated with sleep problems at older ages, it is not clear whether early sleep patterns are also associated with later sleep problems, particularly when using a validated measure.

Building on these previous studies, the aim of the current study was to disentangle the association between total sleep duration and consecutive sleep duration at six months and later sleep problems at 48 and 60 months, using a validated measure of sleep problems. Moreover, analyses will be adjusted for maternal depression<sup>36,37</sup>, breastfeeding status<sup>38</sup>, sleeping arrangements<sup>39</sup>, socioeconomic status (SES)<sup>40-43</sup>, and biological sex<sup>44</sup>, considering their well-known influence on infant/child sleep as well as with parental perception of sleep problems.

## **Participants and Methods**

### *Participant recruitment*

Participants were part of the longitudinal Maternal Adversity, Vulnerability, and Neurodevelopment birth cohort study<sup>45</sup> (MAVAN), which received approval from the Douglas Mental Health University Institute (Montreal, Quebec) and St. Joseph Healthcare/McMaster University (Hamilton, Ontario) ethics boards. Expecting mothers were recruited between 13- and 20-weeks' gestation via obstetric clinics in the Montreal and Hamilton areas and written consent was obtained by all participants ( $n = 629$ ). Inclusion criteria specified that expecting mothers were at least 18 years old, fluent in English or French, and free from severe health conditions, and chronic illness. Exclusion criteria included APGAR score  $< 7$ , premature birth ( $< 37$ -week gestation), serious obstetric complications, and severe infant health conditions. A total of 286 mother-child dyads were included in the present study (53.5% male children).



## *Measures and Procedures*

### *Infant Sleep Patterns*

When infants were six months of age, mothers completed the Questionnaire about Sleep Habits (QASH), an adapted version from the Self-Administered Questionnaire from the Mother used in the *Étude Longitudinale du Développement des Enfants du Québec*<sup>46</sup> (ELDEQ), which assessed the previous two weeks of sleep. Consecutive sleep duration was assessed with the question “During the night, how many consecutive hours does your child sleep without waking up?” Total sleep duration was assessed using the questions, “What is the total length of your child's sleep during the night?” and “What is the total length of sleep of your child during the day?” Answers to these two questions were summed to obtain the total sleep duration within a 24-hour period.

### *Sleep Problems in Preschoolers*

At 48 and 60 months, mothers completed the Child Behavior Checklist<sup>47,48</sup> for Ages 1.5-5 (CBCL/1.5-5). The Sleep Problems subscale of the CBCL is validated to assess reported sleep problems<sup>49</sup> in children aged one-and-a-half to six years old. Specifically, maternal reports on the CBCL Sleep Problems subscale have been shown to have good correspondence with sleep efficiency and total sleep duration derived from maternal sleep diaries<sup>49</sup>. Mothers rated descriptive items related to their child on a three-point Likert scale according to how true each statement was (0 = not true; 1 = somewhat or sometimes true; 2 = very true or often true) based on the past two weeks. Seven items are included in the Sleep Problems subscale: (1) item 22 “Does not want to sleep alone,” (2) item 38 “Has trouble getting to sleep,” (3) item 48 “Nightmares,” (4) item 64 “Resists going to bed at night,” (5) item 74 “Sleeps less than most children during day and/or night,” (6) item 84 “Talks or cries out in sleep,” and (7) item 94

“Wakes up often at night.” Item values were summed to obtain the total sleep problems subscale score ranging from 0 to 14. A higher score indicated a higher endorsement of sleep problems.

### *Covariates*

SES was assessed at six-months post-partum using education level and family income as reported by the mother. Mothers reported family income and education level, and both were dichotomized into high and low groups. Statistics Canada's low-income cut-off<sup>50</sup> was used to determine income group membership. Maternal education was considered high if the mother attended college or university. Overall SES was determined as followed: high SES (high income and high maternal education) and low SES (low on at least one category).

Maternal depressive symptoms were used as a covariate. The Center for Epidemiologic Studies Depression scale<sup>51</sup> (CES-D) was completed by mothers when their child was 48 and 60 months old. A higher score on this measure indicates a greater frequency of depressive symptoms during the past week.

Breastfeeding status was determined retrospectively at six months by asking mothers if they had breastfed or not and if so until what age. Breastfeeding status was dichotomized as breastfed or not based on whether they were still breastfeeding at six months.

Sleeping arrangements were assessed at six months using the question, “does your baby sleep” (a) “alone in her/his room,” (b) “in her/his room, with someone else,” (c) “in your room, but alone in her/his bed,” or (d) “in your room, in your bed.” Sleep arrangements were dichotomized into two groups: (1) solitary sleeping, defined as not sleeping in the same room with the mother (answers a or b), and (2) co-sleeping, defined as sleeping in the same room as the mother (answers c or d).

### *Statistical Analysis*

SPSS 29.0.1.1 (IBM Corporation, Armonk, NY) was used to perform Generalized estimating equations (GEE) for repeated measures assessing the associations between consecutive sleep duration and total sleep duration during early infancy (six months), and sleep problems in toddlerhood (48 and 60 months). Covariates included maternal depression (48 and 60 months), breastfeeding status (six months), sleeping arrangements (six months), socioeconomic status (SES) (six months), and child's biological sex. To analyze longitudinal data, a linear GEE model was used to estimate population effects. The total score for the CBCL Sleep Problems subscale at 48 and 60 months were used as outcome measures. A p-value of  $< 0.05$  was used to determine statistical significance. Subsequently, multiple GEE models were used to assess whether consecutive sleep duration or total sleep duration were associated with individual items of the CBCL Sleep Problems subscale to clarify the association with specific sleep dimensions.

## **Results**

### *Descriptive results*

The mean 24-hour sleep duration for infants at six months was 13.2 hours (SD = 1.83). The mean consecutive sleep duration for infants at six months was 6.97 hours (SD = 2.70). High SES families accounted for 68.2% of the sample and the remaining 31.8% fell into the low SES group. The mean maternal depression score was 10.2 (SD = 9.0). Biological males comprised 53.1% of the sample. Breastfed infants and infants who co-slept comprised 60.1% and 29.4% of the sample, respectively.

*Association between infant sleep patterns and preschooler sleep problems*

The results of the first GEE model showed that shorter total sleep duration in infancy was associated with higher total sleep problem scores in the preschool period ( $B = -8.30$ , 95% CI [0.00 to -3.82],  $p < 0.001$ ; Table 1) while adjusting for maternal depression, breastfeeding status, sleeping arrangements, SES, and biological sex. However, consecutive sleep duration in infancy was not associated with total sleep problem scores in the preschool period ( $B = -0.06$ , 95% CI [-0.19 to 0.07],  $p = 0.363$ ; Table 1).

To further specify the association between early sleep patterns and future sleep problems, distinct GEE models were used to assess the association between total sleep duration and consecutive sleep duration (six months) and each dimension included in the Sleep Problems subscale (48 and 60 months), while adjusting for the same covariates. Infants with a shorter total sleep duration at six months had a higher score on the following items at 48-60 months: “has trouble getting to sleep” (table 2;  $B = -1.72$ , 95% CI [-2.92 to -5.24],  $p = 0.005$ ), “nightmares” (table 3;  $B = -1.01$ , 95% CI [-1.90 to -1.10],  $p = 0.028$ ), “resists going to bed at night” (table 4;  $B = -1.50$ , 95% CI [-2.72 to -2.89],  $p = 0.015$ ), “sleeps less than most kids during the day and/or night” (table 5;  $B = -1.09$ , 95% CI [-1.97 to -2.17],  $p = 0.014$ ), and “wakes up often at night” (table 6;  $B = -1.15$ , 95% CI [-2.26 to -4.78],  $p = 0.041$ ). Infant consecutive sleep duration was not associated with any individual item of the Sleep Problems subscale in preschoolers ( $p > 0.05$ ). Neither infant total sleep duration nor consecutive sleep duration were significantly associated with item “doesn’t want to sleep alone” (table 7) or item “talks or cries out in sleep” (table 8) during the preschool period.

## Discussion

The aim of this study was to investigate if total sleep duration and consecutive sleep duration at six months old were associated with parentally reported sleep problems in the preschool period (48 and 60 months), while adjusting for confounding variables (maternal depression, breastfeeding status, sleeping arrangements, SES, and biological sex). Present results showed that shorter total sleep duration at six months was significantly associated with more sleep problems at 48 and 60 months and more specifically, more bedtime resistance, difficulty falling asleep, the presence of nightmares, more nocturnal awakenings, and shorter sleep duration. However, shorter consecutive sleep duration in infancy was not significantly associated with total sleep problems scores nor any individual items of the sleep problems subscale in preschoolers. Difficulty sleeping alone and talking and crying during sleep in the preschool period were not associated with neither total sleep duration nor consecutive sleep duration in infancy.

Overall, shorter sleep duration in infancy was associated with more parental reports of sleep problems in their preschoolers, which is consistent with findings of other studies. Indeed, a previous study<sup>24</sup> found that shorter sleep duration was associated with concurrent higher parent-reported sleep problems at six, 12, and 24 months. Therefore, the present results build on these previous findings by showing that shorter total sleep duration in infancy is also associated with *later* sleep problems. This is also consistent with results of other studies<sup>52-55</sup> that have examined the association between total sleep duration at different timepoints in development and identified a subset of children having persistent short sleep durations from childhood to adolescence. Taken together, these results suggest that shorter sleep duration might persist over time and that it can be a marker of concurrent or later sleep problems.

Shorter infant total sleep duration was also significantly associated with some specific items of the CBCL Sleep Problems subscale in preschoolers, such as bedtime resistance and difficulty falling asleep. Bedtime resistance is often a component of pediatric insomnia where child passively or actively refuses to go to sleep at night<sup>56,57</sup> and can be associated with both internalized and externalized difficulties<sup>27,58,59</sup>. In addition, bidirectional associations have been shown between bedtime resistance, inconsistent bedtime routines and parenting stress<sup>60</sup>. Irregular bedtimes have been associated with both bedtime resistance and difficulty falling asleep, often resulting in shorter sleep duration<sup>61</sup>. Indeed, bedtime resistance and difficulty falling asleep are commonly delaying sleep onset which may in turn reduce the sleep window, resulting in shorter overall sleep duration<sup>62-64</sup>.

The current results also show an association between shorter sleep duration at six months and higher parental report of nightmares in their preschoolers, which may potentially lead to increased night awakenings and increased bedtime resistance<sup>65</sup>. Indeed, a bidirectional association has been suggested between sleep deprivation and nightmares<sup>66</sup>. In the current study, shorter total sleep duration at six months was significantly associated with parental reports of “sleeping less than others” at 48 and 60 months which is congruent with results from previous studies. As described earlier, findings from multiple previous studies<sup>52,54,55</sup> have suggested that shorter sleep duration may persist from infants to preschool-aged children. Lastly, shorter total sleep duration at six months was also associated with increased reports of nocturnal awakenings. This finding is congruent with the literature<sup>67,68</sup> where shorter sleep duration has been associated with more nocturnal awakenings.

Unlike total sleep duration, infant consecutive sleep duration at six months, often referred as not “sleeping through the night”, was not associated with more sleep problems in

preschoolers. Although very few studies assessed the association between early consecutive sleep duration and future sleep problems, a previous study<sup>17</sup> found that approximately one third of infants who were not “sleeping through the night” (six consecutive hours) at five or 17 months were also not sleeping through the night at 29 months, suggesting that a portion of children had persistent shorter consecutive sleep duration across development. Authors also identified that at 17 and 29 months not sleeping six hours consecutively was strongly associated with parental presence until sleep onset and concluded that this association was likely bidirectional. In another study<sup>69</sup>, one measure of sleep fragmentation (nocturnal awakenings) at 12 months, was not associated with nighttime sleep efficiency and duration at six years old.

The dimensions related “not wanting to sleep alone” and “talking and crying during sleep” were not related to either total sleep duration or consecutive sleep duration in early infancy. This is consistent with a previous study<sup>70</sup> that found that children who sleep in the same room or bedshared with parents had later bedtimes but had similar sleep duration to children who slept in a separate sleep location. Yet, the literature appears to be inconsistent as other studies<sup>71</sup> have found that bedsharing may result in shorter nocturnal sleep duration, more frequent night awakenings, and a greater awareness of these awakenings in parents. Indeed, the literature appears to be inconsistent as in a recent review<sup>39</sup> of studies investigating bedsharing and infant sleep using objective and subjective measures, inconclusive results were reported. The literature<sup>72,73</sup> describing difficulty sleeping alone and co-sleeping is also complicated by the fact that most instruments and studies do not account for planned co-sleeping versus reactive co-sleeping. Moreover, the specific CBCL item used in the present study “not wanting to sleep alone” does not assess if parents simply report that their child does not want to sleep alone or if they actually let their child sleep in their room or bed as a result.

The item “talking or crying during sleep”, probably captures parasomnias<sup>62</sup> such as somniloquy, sleepwalking and sleep terrors, although it is not highly specific to one or the other. Somniloquy itself does not appear to be associated with poor sleep<sup>74</sup> but if it begins to affect subjective sleep quality intervention might be indicated. However, sleep terrors, which are highly prevalent in childhood, have been associated with increased sleep fragmentation in other studies<sup>75,76</sup>.

Overall, the findings of the current study suggest that early shorter sleep duration may be a marker of later sleep problems, including several sleep dimensions. It might be that some individuals are more prone to developing sleep problems than others based on a biological sensitivity that is already observed in infancy. Indeed, adolescents and adults with insomnia often report life-long problematic sleep<sup>77-79</sup>. Environmental practices which do not allow for sufficient opportunity to sleep may also contribute to the current findings. Indeed, later bedtimes as well as having inconsistent bedtime routines have been associated with shorter nocturnal sleep duration<sup>80,81</sup> as early as six months. Increased nocturnal parental presence in families, especially when parents aim at solitary sleep may also reduce the sleep window and the total duration of sleep<sup>17</sup>.

In contrast, shorter consecutive sleep duration was not a marker of later parental perceived sleep problems in preschoolers. It might be that the consolidation of the sleep-wake patterns in infancy is a developmental process, reflecting both brain development and specific environmental conditions linked to sleep fragmentation during infancy. For instance, studies<sup>38,82,83</sup> have found that breastfed infants have shorter consecutive sleep durations and more night awakenings than formula fed infants, but that this association is no longer significant at 24 and 36 months once breastfeeding is discontinued. Finally, major variability both between



different infants and across several nights for the same infant have been described regarding consecutive sleep duration or sleeping through the night<sup>15,18</sup>.

### *Strengths and limitations*

The main strengths of this article are the longitudinal design and the use of a validated measure to assess sleep problems in a relatively high sample size of typically developing children. Additionally, using both total sleep duration and consecutive sleep duration allowed to disentangle which of these early sleep variables were associated with later sleep problems. Similarly, different sleep problems dimensions were assessed in preschoolers, allowing for greater specificity.

However, several limitations should also be taken into consideration when interpreting the results of the current study. Sleep variables and sleep problems were measured with subjective parental report and are subject to bias. It cannot be ruled out that some parents may be more sensitive than others to their child sleep-wake patterns, especially if they themselves have insomnia. While controlling for maternal depression scores minimizes this bias, it still needs to be considered. The use of objective measures, such as actigraphy or polysomnography could have presented a complementary perspective, although the use of subjective measurements gives us the parental perspective which should not be neglected. Another limitation is the large proportion of higher SES participants were part of in the sample and thus caution should be used when generalizing the results to the wider population. Moreover, given that the sample was comprised of a North American population, the results may not be generalizable to other populations.

## **Conclusion**

In summary, the current study contributes to the current literature by showing that shorter sleep duration during infancy, but not consecutive sleep duration, is associated with more sleep problems, as perceived by parents, during preschool years using a validated measure. These findings suggest that offering more opportunities to sleep throughout the day and night to promote a longer total sleep duration over 24 hours should be prioritized over the consolidation of sleep (sleeping through the night) during early infancy. The results may help to inform advice given to parents by clinicians concerning their infant's sleep development.

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## Tables

**Table 1**

Association between total sleep duration and consecutive sleep duration at 6 months, and sleep problems at 48 and 60 months (CBCL Total Sleep Subscale score).

Factors (Unit or Reference)	B	Standard Error	95% CI	P-Value
Child's sex (male)	-0.63	0.32	-1.26 to 0.002	0.051
Time (in months)	0.10	0.69	-1.25 to 1.45	0.886
Socioeconomic status (low)	0.53	0.36	-0.16 to 1.23	0.134
Maternal depressive symptoms (CES-D total score)	0.03	0.02	-0.01 to 0.06	0.124
Sleeping arrangement (solitary)	-0.05	0.36	-0.76 to 0.66	0.88
Breastfeeding status (no)	-0.14	0.35	-0.82 to 0.55	0.69
Total sleep duration (in hours)	-8.30	2.29	0.00 to -3.82	<b>&lt; 0.001</b>
Consecutive sleep duration (in hours)	-0.06	0.07	-0.19 to 0.07	0.363

CES-D, Center for Epidemiological Studies Depression

**Table 2**

Association between total sleep duration and consecutive sleep duration at 6 months, and CBCL Sleep Subscale item, "has trouble getting to sleep" at 48 and 60 months.

Factors (Unit or Reference)	B	Standard Error	95% CI	P-Value
Child's sex (male)	-0.10	0.08	-0.25 to 0.06	0.224
Time (in months)	0.16	0.15	-0.14 to 0.46	0.282
Socioeconomic status (low)	0.09	0.10	-0.10 to 0.28	0.352
Maternal depressive symptoms (CES-D total score)	0.002	0.004	-0.01 to 0.01	0.677
Sleeping arrangement (solitary)	0.11	0.09	-0.07 to 0.29	0.234
Breastfeeding status (no)	-0.01	0.09	-0.19 to 0.17	0.922
Total sleep duration (in hours)	-1.72	6.10	-2.92 to -5.24	<b>0.005</b>
Consecutive sleep duration (in hours)	-0.01	0.02	-0.04 to 0.02	0.462

CBCL, Child Behavior Checklist

CES-D, Center for Epidemiological Studies Depression

**Table 3**

Association between total sleep duration and consecutive sleep duration at 6 months, and CBCL Sleep Subscale item, “nightmares” at 48 and 60 months.

Factors (Unit or Reference)	B	Standard Error	95% CI	P-Value
Child’s sex (male)	0.03	0.06	-0.09 to 0.15	0.630
Time (in months)	-0.07	0.12	-0.28 to 0.14	0.525
Socioeconomic status (low)	0.03	0.07	-0.11 to 0.18	0.650
Maternal depressive symptoms (CES-D total score)	0.01	0.004	0.003 to 0.02	<b>0.007</b>
Sleeping arrangement (solitary)	-0.04	0.08	-0.19 to 0.11	0.573
Breastfeeding status (no)	0.06	0.07	-0.07 to 0.20	0.346
Total sleep duration (in hours)	-1.01	4.57	-1.90 to -1.10	<b>0.028</b>
Consecutive sleep duration (in hours)	0.01	0.01	-0.02 to 0.03	0.679

CBCL, Child Behavior Checklist

CES-D, Center for Epidemiological Studies Depression

**Table 4**

Association between total sleep duration and consecutive sleep duration at 6 months, and CBCL Sleep Subscale item, “resists going to bed at night” at 48 and 60 months.

Factors (Unit or Reference)	B	Standard Error	95% CI	P-Value
Child’s sex (male)	-0.07	0.08	-0.23 to 0.09	0.364
Time (in months)	0.04	0.19	-0.03 to 0.41	0.847
Socioeconomic status (low)	0.21	0.09	0.03 to 0.40	<b>0.020</b>
Maternal depressive symptoms (CES-D total score)	0.004	0.004	-0.004 to 0.01	0.300
Sleeping arrangement (solitary)	0.106	0.090	-0.07 to 0.28	0.241
Breastfeeding status (no)	-0.02	0.90	-0.19 to 0.15	0.825
Total sleep duration (in hours)	-1.50	6.20	-2.72 to -2.89	<b>0.015</b>
Consecutive sleep duration (in hours)	-0.01	0.02	-0.04 to 0.02	0.584

CBCL, Child Behavior Checklist

CES-D, Center for Epidemiological Studies Depression

**Table 5**

Association between total sleep duration and consecutive sleep duration at 6 months, and CBCL Sleep Subscale item, “sleeps less than most kids during the day and/or night” at 48 and 60 months.

Factors (Unit or Reference)	B	Standard Error	95% CI	P-Value
Child’s sex (male)	-0.14	0.06	-0.27 to -0.02	<b>0.024</b>
Time (in months)	0.23	0.05	0.13 to 0.33	<b>&lt;0.001</b>
Socioeconomic status (low)	-0.001	0.07	-0.15 to 0.15	0.993
Maternal depressive symptoms (CES-D total score)	0.003	0.003	-0.003 to 0.01	0.369
Sleeping arrangement (solitary)	0.03	0.07	-0.11 to 0.17	0.658
Breastfeeding status (no)	-0.02	0.07	-0.15 to 0.12	0.806
Total sleep duration (in hours)	-1.09	4.46	-1.97 to -2.17	<b>0.014</b>
Consecutive sleep duration (in hours)	0.00	0.01	-0.03 to 0.03	0.970

CBCL, Child Behavior Checklist

CES-D, Center for Epidemiological Studies Depression

**Table 6**

Association between total sleep duration and consecutive sleep duration at 6 months, and CBCL Sleep Subscale item, “wakes up often at night” at 48 and 60 months.

Factors (Unit or Reference)	B	Standard Error	95% CI	P-Value
Child’s sex (male)	-0.15	0.08	-0.30 to -0.01	<b>0.040</b>
Time (in Months)	-0.06	0.17	-0.40 to 0.28	0.733
Socioeconomic status (low)	0.04	0.08	-0.12 to 0.19	0.640
Maternal depressive symptoms (CES-D total score)	0.003	0.004	-0.004 to 0.01	0.682
Sleeping arrangement (solitary)	0.02	0.08	-0.14 to 0.18	0.821
Breastfeeding status (no)	-0.05	0.08	-0.20 to 0.11	0.544
Total sleep duration (in hours)	-1.15	5.64	-2.26 to -4.78	<b>0.041</b>
Consecutive sleep duration (in hours)	-0.02	0.02	-0.05 to 0.01	0.230

CBCL, Child Behavior Checklist

CES-D, Center for Epidemiological Studies Depression



**Table 7**

Association between total sleep duration and consecutive sleep duration at 6 months, and CBCL Sleep Subscale item, “doesn’t want to sleep alone” at 48 and 60 months.

Factors (Unit or Reference)	B	Standard Error	95% CI	P-Value
Child’s sex (male)	-0.18	0.09	-0.36 to -0.003	<b>0.047</b>
Time (in months)	-0.13	0.17	-0.46 to 0.20	0.429
Socioeconomic status (low)	0.12	0.10	-0.09 to 0.33	0.252
Maternal depressive symptoms (CES-D total score)	0.001	0.005	-0.01 to 0.01	0.907
Sleeping arrangement (solitary)	-0.26	0.12	-0.47 to -0.05	<b>0.017</b>
Breastfeeding status (no)	-0.08	0.10	-0.27 to 0.12	0.437
Total sleep duration (in hours)	-1.18	7.07	-2.56 to 2.07	0.095
Consecutive sleep duration (in hours)	-0.02	0.02	-0.06 to 0.18	0.296

CBCL, Child Behavior Checklist

CES-D, Center for Epidemiological Studies Depression

**Table 8**

Association between total sleep duration and consecutive sleep duration at 6 months, and CBCL Sleep Subscale item, “talks or cries out in sleep” at 48 and 60 months.

Factors (Unit or Reference)	B	Standard Error	95% CI	P-Value
Child’s sex (male)	-0.02	0.07	-0.15 to 0.12	0.791
Time (in months)	0.01	0.14	-0.27 to 0.28	0.967
Socioeconomic status (low)	0.06	0.08	-0.10 to 0.22	0.452
Maternal depressive symptoms (CES-D total score)	0.004	0.004	-0.003 to 0.01	0.277
Sleeping arrangement (solitary)	-0.01	0.08	-0.16 to 0.14	0.860
Breastfeeding status (no)	-0.04	0.07	-0.17 to 0.10	0.596
Total sleep duration (in hours)	-7.27	5.16	-1.74 to 2.85	0.159
Consecutive sleep duration (in hours)	5.03	0.01	-0.03 to 0.03	0.997

CBCL, Child Behavior Checklist

CES-D, Center for Epidemiological Studies Depression

## **Chapter 6: General Discussion**

### **General Summary**

The objective of the current dissertation was to advance knowledge regarding the association between early infant sleep patterns and developmental outcomes, namely cognitive/psychomotor functioning, as well as sleep problems, in healthy full-term infants. Overall, the conclusions of a systematic review (Study 1), and results of an original longitudinal article (Study 2) included in this dissertation highlight that early sleep patterns are not clearly correlated with later cognitive/psychomotor functioning and sleep problems. Moreover, this dissertation underscores the importance of considering the potential contribution of individual sleep variables, since different patterns of associations are sometimes observed as a function of the variable of interest.

### ***Infant Sleep Patterns and Subsequent Cognitive/Psychomotor Functioning***

Findings from Study 1's systematic review showed mixed findings regarding associations between infant sleep patterns and cognitive or psychomotor development. The primary analysis revealed that individual sleep variables were not consistently associated with developmental outcomes. Most studies included in the review found a mix of significant and non-significant associations, with three reporting no significant associations and only two studies finding exclusively significant associations. In terms of specific sleep variables, both total sleep duration (over 24 hours) and nocturnal sleep duration were largely not associated with cognitive or psychomotor development. As for daytime sleep and the ratio of nocturnal to total sleep, the studies found mainly mixed results regarding their association with cognitive and psychomotor development.

The secondary analysis, which extracted a total of 101 individual associations from the included studies, revealed that the majority of these associations were non-significant or mixed. Only six associations between early sleep variables (more developed circadian sleep regulation, lower sleep problem scores, less time awake after sleep onset, more night awakenings, and poorer sleep continuity) and developmental outcomes were consistently significant. However, these significant associations were extracted from only four studies with quality ratings ranging from good to poor.

Study 1 contributed to the literature by employing a systematic strategy to identify and synthesize studies investigating the association between sleep patterns in infancy and cognitive and psychomotor development. Overall, associations were inconsistent, and results highlight the complexity of this association during infancy. Thus, practitioners working with families and infants should be cautious not to apply findings from older populations (children, adolescents and adults) to this early period of sleep-wake cycle development that is infancy.

### ***Infant Sleep Patterns and Subsequent Sleep Problems***

The results from Study 2 indicated that shorter total sleep duration at six months was significantly associated with more sleep problems in preschoolers. Importantly, these results remained significant after controlling for confounding variables such as socioeconomic status (SES), maternal depression, breastfeeding status, sleeping arrangements, time, and biological sex. In contrast, consecutive sleep duration at six months was not associated with sleep problems in preschoolers.

Again, the secondary aim of Study 2 showed the specificity of different sleep variables both in terms of infant sleep patterns and different types of sleep problems in preschoolers. In one respect, shorter total sleep duration in infancy was specifically associated with higher scores

on the individual sleep problem dimensions “has trouble getting to sleep”, “resists going to bed at night”, “sleeps less than most kids during the day and/or night”, “wakes up often at night”, and “nightmares” in preschoolers, but not with “doesn’t want to sleep alone” or “talks or cries out in sleep”. In contrast, shorter consecutive sleep duration was not associated with any individual sleep problem dimensions. Study 2 made important contributions to the literature, as to the best of our knowledge is among few studies that have investigated the association between early infant sleep patterns and perceived sleep problems during the preschool period. It also contributed to disentangling the importance of total sleep duration in infancy as opposed to consecutive sleep duration and provided further specificity regarding different sleep problem outcomes using individual items.

### **Are Early Sleep Patterns Markers of Developmental Difficulties or Sleep Problems?**

#### ***Consecutive Sleep Duration***

While the two studies included in the present dissertation examined different outcomes associated with early infant sleep patterns, neither study found consistent associations with early consecutive sleep duration. In Study 1 (systematic review), three out of five studies using measures related to consecutive sleep duration (sleeping through the night or longest sleep period), did not find a significant association with cognitive and psychomotor outcomes and two found inconsistent results (significant at one timepoint but not at another). Interestingly, the studies which found inconsistent results contrasted in their directionality, with one study finding that longer consecutive sleep was associated with better cognitive outcomes and the other finding the opposite. However, neither of these studies commented on feeding practices, which is an important consideration given that breastfeeding is known to be associated with more frequent night awakenings and shorter consecutive sleep duration in infancy (Abdul Jafar et al., 2021;

Figueiredo et al., 2017; Volkovich et al., 2015). Consistent with these results, Study 2 did not show significant associations between consecutive sleep duration in infancy and total or individual sleep problems scores as reported by parents in the preschool period, this time, while controlling for both feeding method and sleeping arrangements.

Overall, findings of this dissertation do not support consecutive sleep duration as an important marker of later outcomes, namely cognitive/psychomotor development and sleep problems. This is an important contribution to the field of pediatric sleep, considering the importance of the messaging around “sleeping through the night” in new parents both by health professionals and in media in general. Indeed, new parents often report feeling pressured to help their infant consolidate sleep rapidly, assuming that this will help to develop good sleep habits and mitigate future sleep problems (Burdayron et al., 2020; Porter & Ispa, 2013). Moreover, interventions targeting infant sleep problems are being delivered at an increasingly younger age (Crichton & Symon, 2016; Gradisar et al., 2016; Honaker et al., 2018). The results of the current dissertation do not support the need for such interventions and suggest that an over importance has been placed on early infant sleep consolidation.

### ***Total Sleep Duration***

In contrast, results related to total sleep duration differ more between the two studies. Indeed, Study 2 found that shorter total sleep duration at six months was associated with higher parent reported total sleep problems during preschool years. Additionally, shorter total sleep duration was also associated with higher severity of multiple individual sleep problem dimensions later in the preschool period. These results are similar and build on results of a previous study which found that shorter sleep duration was associated with concurrent sleep problems from six to 24 months of age (Byars et al., 2012).

Conversely, results from Study 1 largely suggest that total sleep duration between the ages of 0 to 18 months is generally not associated with cognitive and psychomotor outcomes. Specifically, out of seven studies examining this association, six failed to find a significant association between total sleep duration and developmental outcomes and one study found mixed results. Therefore, it is possible that while shorter sleep duration in infancy is associated with later sleep problems, it does not necessarily predict later cognitive or psychomotor development. Alternatively, the timing of the measures could also contribute to these contrasting results as four out of the seven studies that assessed total sleep duration were single timepoint (non-longitudinal) designs (Bernier et al., 2013; Montgomery-Downs & Gozal, 2006; Sun et al., 2018; Tham et al., 2019). The remaining three studies that employed longitudinal designs, assessed total sleep duration at each timepoint independently of each other (Bernier et al., 2010; Plancoulaine et al., 2017; Spruyt et al., 2008). As a result, individual infant sleep trajectories could not be determined, and the persistence of short total sleep duration could not be assessed. Previous longitudinal studies have looked at individual sleep trajectories of children from *infancy into childhood and adolescence* and found that short sleep durations do persist in some children and are associated with several negative outcomes, such as poorer physical, emotional, and social health (Magee et al., 2014; Manitsa et al., 2024; Petit et al., 2023; Touchette et al., 2024). Additionally, a study by Touchette et al. (2007) found that persistent shorter sleep duration in children from two and a half to six years old was associated with lower cognitive performance and higher hyperactivity/impulsivity scores. Taken together, these studies suggest that shorter sleep duration specifically in infancy is not necessarily associated with later negative outcomes. However, in some of these infants, short sleep duration persists over development and thus in specific cases, later development seems impacted. Future studies could therefore focus on the

persistence of short sleep duration and aim to identify children who might fall into this category early in development.

## **Methodological Considerations**

### ***What Should be the Duration of a Sleep Assessment During Early Infancy?***

An important factor which could have influenced the results of the systematic review (Study 1) is the number of days included in the sleep assessment of each individual study. Unlike Study 2 of the present dissertation which assessed sleep over a two-week period, the longest assessment period for total sleep duration in Study 1 (systematic review) was seven days. Additionally, three of the seven studies only assessed total sleep duration over a three-day period (Bernier et al., 2013; Bernier et al., 2010; Spruyt et al., 2008). Although it is not uncommon to use three nights of sleep in pediatric sleep studies, it is possible that total sleep duration was not assessed over a long enough period of time to be representative, specifically in an infant population.

Relatedly, night-to-night variation of total sleep duration would have also been better captured if a longer assessment period had been used. Additionally, often studies have an assessment window which may vary by plus or minus one to two weeks (i.e., a six-month assessment could be at five and a half months or six and a half months). Due to the rapid development of sleep during this period, it is possible that these margins might influence the associations with developmental outcomes and that two or three weeks later or earlier, the developmental period could be significantly different.

### ***The Role of Parental Perception When Measuring Infant Sleep***

While it is possible that certain parenting behaviors may maintain night awakenings from infancy into preschool years, it is also possible that other factors may contribute to parents

overreporting night awakening in their children. For example, sleeping arrangements such as co-sleeping have been found to influence parental report of sleep patterns. One study found that mothers sharing a bedroom with their child reported more night awakenings than mothers who did not co-sleep (Volkovich et al., 2015). However, no significant differences were found in the same study using objective measures. Additionally, a review by Andre et al. (2021) who examined both subjective and objective measures of sleep in infants who were bed or room sharing found mixed results. Specifically, the authors reported that some studies using objective measures found that bedsharing was associated with increased sleep fragmentation, while other studies found no significant differences. However, in other studies, co-sleeping mothers were also found to have more awakenings and longer wakefulness during the night than solitary sleeping mothers. Thus, it is possible that parents who bed or room share with their infant are more aware of their night awakenings due to proximity.

Another factor which may influence parental report of their child's sleep is parental sleep problems. One study by Urfer-Maurer et al. (2017) used objective and subjective sleep measures and found that insomnia symptoms in mothers and fathers were associated with higher reports of night awakenings in their children from seven to 12 years of age while controlling for objective sleep measures. Another study found similar results, where increased maternal insomnia scores were significantly associated with increased nighttime wakefulness and higher perceived sleep problems from three to 36 months after adjusting for maternal depression scores (Zreik et al., 2022). Thus, parents who are awake more often during the night as a results of elevated insomnia scores may witness and report more frequent awakening of their child.

Parental perception may also affect reporting of their child's sleep problems. A study by Burdayron et al. (2021) found that mothers with higher levels of depression reported more sleep



problems in infants with higher negative affectivity, after adjusting for nocturnal sleep duration and number of awakenings. Furthermore, a previous study also found that mothers with perinatal depression were more likely than non-depressed mother to reported more infant night awakenings, despite no significant differences being found via actigraphy (Halal et al., 2021). Taken together, it is possible that these factors may contribute to the overreporting of night awakenings in children. The perception of what represents a “problem” can also vary as a function of personal experience and culture (Mindell et al., 2013; Sadeh et al., 2011).

One could therefore think that using an objective measure such as actigraphy in addition to parental report might be the ideal solution. However, while actigraphy is a valuable tool, it also has its challenges. For example, using actigraphy to measure sleep duration in infancy may sometimes result in data artifacts and inaccuracies and it is generally recommended that parentally completed sleep diaries are used to corroborate the data (Tétreault et al., 2018). However, some families may find completing additional sleep diaries to be time-consuming and challenging. Additionally, parents may forget to reattach actigraphy devices after bathing and older children may remove the devices themselves. Another technical limitation of actigraphy, specifically in children, is that it often overestimates awakenings in children who have increased motor activity during their sleep (Bélanger et al., 2013; Simard et al., 2013).

Overall, the use of subjective and objective measures simultaneously to assess sleep in infancy may help to mitigate the shortcomings of one another but will still not provide an ideal assessment of sleep patterns. Taking this into account, it is important to consider the strengths and limitations of each assessment method when interpreting results.

## Importance of Sleep in Infancy

One question which the findings of the studies included in this dissertation poses is: is early sleep important? While Study 1 found mostly mixed results, total sleep duration and nocturnal sleep duration were largely unassociated with cognitive development. Study 2 found that although total sleep duration was associated with future sleep problems, consecutive sleep duration was not. These results clearly contrast with the literature in older children and in adults. Should we therefore conclude that sleep is not important in infancy? Certainly not. We rather believe that the association between early sleep and later outcomes is complex.

The common absence of associations between early sleep patterns and later outcomes in this dissertation might be explained by the presence of a wide continuum of sleep needs among different individuals. While we often address this variability regarding individual requirements in adults, it is less often discussed in infants. Indeed, the National Sleep Foundation guidelines recommended that infants between the ages of four and 11 months sleep for *between* 12 to 15 hours within a 24-hour period and state that infants below 10 hours are at particular risk (Hirshkowitz et al., 2015). Thus, a total sleep duration of 12 hours might be sufficient for one specific infant but be inadequate for another one. When performing a correlation or a regression between total sleep duration and any outcomes, we do not know which individual has a sufficient or insufficient sleep duration as a function of their own needs. Perhaps a future approach could be to use a minimum cutoff for total sleep duration as opposed to a continuous measure. This approach is often used in adults' epidemiological studies, where a cutoff of 6, 6.5 or 7 hours is often used (Chaput et al., 2020).

Overall, while the association between early sleep patterns and future outcomes is complex and nuanced, sleep is a fundamental pillar of health and should be further investigated

during this early period. However, assuming that longer sleep durations, either consecutive or total, will be associated with specific domains of functioning in a dose-response manner may not be applicable if they fall into normative ranges.

### **Clinical Implications**

The results of this dissertation provide valuable insights, which may be used to inform advice and consultation given by healthcare professionals. The current findings suggest that overall, total sleep duration and giving enough opportunity to sleep over 24 hours should be prioritized over focusing on consecutive sleep duration (or sleeping through the night) during early infancy. Specifically, neither study found that consecutive sleep duration was associated with cognitive development nor future sleep problems. This is particularly important given that approximately 35-40% of typically developing infants do not sleep through the night (six consecutive hours) at six months (Henderson et al., 2011; Pennestri et al., 2018). Healthcare professionals working with families with young infants ought to communicate that evidence of negative consequences of shorter consecutive sleep durations during this period are limited. Given that many parents are focused on their infant's ability to consolidate their sleep in an attempt to mitigate future sleep problems, this reassurance might also help to quell parental worry (Burdayron et al., 2020; Porter & Ispa, 2013). It is important to reassure parents during this period of time, given that it is a vulnerable moment, and parents are prone to anxiety and depressive symptoms, even in the absence of a postpartum depression diagnosis. It might also be communicated to parents that sleep training during early infancy does not appear to be necessary. While some parents are comfortable with this practice and should be supported if they wish to use it, they should also be supported if they prefer not to use behavioral techniques (Blunden et al., 2011).

The present research also has important implications regarding reduced total sleep duration during early infancy and increased report of sleep problems during preschool years. Given the results of Study 2, increasing total sleep duration might be addressed by focusing on opportunity to sleep. For example, past studies have shown that earlier bedtimes, providing more opportunity to sleep, as well as bedtime routines and regular sleep schedules have been associated with significantly higher sleep durations in infants (Adams et al., 2020; Tsai et al., 2022). Perhaps by addressing these influential behaviors the probability of short sleep duration persisting into preschool years might be mitigated.

Healthcare professionals should also consider addressing behaviors and environmental factors that have been associated with shorter sleep duration during infancy. For example, exposure to screens and TV viewing as early as six months has also been associated with shorter sleep duration during infancy (Cheung et al., 2017; Nevarez et al., 2010). Given the ubiquity of screen use in the present day, it would be prudent for healthcare workers to inform parents of this association.

Sleep-related parental practices associated with infant sleep patterns could also be assessed and addressed by health professionals. However, while considering the clinical need of the parent and child, it is important to take into account both familial and cultural contexts.

Lastly, reported sleep problems are largely a product of parental perceptions and what is considered a sleep problem by one parent or family may not be considered a sleep problem for another. This is particularly important to consider from a cultural perspective, as the perception of a problematic sleep has also been shown to vary culture to culture (Mindell et al., 2013; Sadeh et al., 2011). For example, perception of sleep problems in children may be higher in predominantly Caucasian versus Asian countries/regions (Mindell et al., 2015). Additionally,

children from predominantly Asian countries/regions have also been found to have later bedtimes and shorter nocturnal sleep durations compared to predominantly Caucasian countries/regions (Mindell et al., 2013). Thus, this is an important factor to consider when interpreting the sleep patterns of individual infants.

### **Strengths, Limitations and Future Directions**

However, while the current dissertation has added valuable contributions to the literature, several limitations should also be considered when interpreting the findings of this research. Firstly, while some significant associations were found concerning sleep patterns during infancy, cognitive outcomes, and report of future sleep problems, causality cannot be established even with a longitudinal design. There are many environmental and biological factors which may mediate and moderate the associations between sleep patterns during infancy, developmental, and sleep problem outcomes and this must be considered when interpreting the findings. While several confounding variables were included in our original study and used in the studies included in the systematic review (mainly SES, breastfeeding status, maternal depression) others such as diet/nutrition, sleep environment (light exposure, temperature), and physical activity may also influence sleep patterns. While experimental studies could help to clarify the causality of these associations, performing sleep deprivation in infancy clearly pose ethical issues and remains difficult to do.

Next, the findings of this dissertation should be generalized with caution due to multiple factors. For example, a higher proportion of high SES participants were included in the sample for Study 2. Future studies should endeavour to recruit and include participants from diverse SES background to aid in generalizability. Study 2 also would also be enriched if more non-traditional families would have been recruited such as lesbian, gay, bisexual, transgender, queer, and other

(LGBTQ+) parents or single-parent families. A recent review suggested that sleep disturbances affect LGBTQ+ subgroups differently and that minority stress contributes to sleep disparities in these individuals (Butler et al., 2020). Another important factor which may influence the ability to generalize these findings is cultural differences regarding sleep patterns and sleep practices. The participants in Study 2 were comprised of a North American sample who additionally were required to be fluent in English or French to enroll in the study. This may have excluded newly arrived immigrants from non-English- or French-speaking countries who could have enriched the study by including a more diverse sample. Similarly, almost half of the studies included in Study 1 (systematic review) were comprised of North American samples. The search strategy used for Study 1 also excluded studies that were not available in English or French (original or translated text) and thus it is possible that studies available in other languages were excluded which could have decreased the cultural diversity of included studies. These are important limitations to consider as sleep patterns as well as sleep practices have been found to differ transculturally and thus could impact generalizability (Ash et al., 2019; Sadeh et al., 2010). In general, future studies should attempt to be more inclusive of participants from diverse backgrounds, especially considering the association between culture and parental practices in general and specifically related to sleep. Given the increasing prevalence and ability of artificial intelligence, perhaps new technologies or programs could be developed to aid in the systematic search process, particularly in identifying and translating studies published in various languages.

Along with these general considerations, Study 1 and Study 2 are impacted by their own unique limitations. Study 2 employed subjective sleep assessment measures and thus, although maternal depression scores were adjusted for, bias may still impact the reports. While the CBCL has been employed frequently to assess sleep in children and overall good correspondence with

sleep dairies, its validity is limited when compared with actigraphy (Bélanger et al., 2014).

Additionally, as discussed previously, parental sleep problems such as insomnia symptoms may also have increased the reports of their children's sleep problems as it increases the chances of witnessing an awakening which was not measured. Future studies should consider assessing problematic sleep in parents in addition to children and adjust for this confounding variable. It is also recommended that future studies also employ objective measures to verify the accuracy of parent reports.

Study 1 also has its own distinct limitations. While Study 1 employed a robust systematic search strategy with clearly defined inclusion and exclusion criteria, it is always possible that certain papers may have been missed and as a result not included in the analysis. Secondly, due to the variability of sleep assessment tools, variables, and timepoints using a meta-analytic synthesis approach was not practical.

However, there are several strengths of the current dissertation which should be highlighted. First, to the best of our knowledge, Study 1 was the first review paper to use a systematic search strategy and quality assessment of studies to synthesize the literature investigating the association between sleep patterns in health full-term infants and cognitive functioning. Second, Study 2 is one of few studies to examine the association between early infant sleep patterns and future sleep problems and the only study to our knowledge that used a validated tool to assess future sleep problems. Both studies disentangled the role of specific sleep variables and-or different sleep problem dimensions.

## **Chapter 7: Conclusion**

Sleep is essential for child development that interacts with various physiological, behavioral, psychological, and environmental factors. It is also a developmental process that undergoes significant changes from early infancy into childhood and beyond. The aim of this dissertation was to determine whether certain sleep patterns in infancy were associated with developmental outcomes and future sleep problems. Correspondingly, the findings of the current dissertation have made substantial contributions in addressing these important questions. Specifically, results from the current research found that consecutive sleep duration during infancy is largely not associated with parent reports of sleep problems nor with cognitive and psychomotor development during preschool years. Moreover, the presented research demonstrates that while total sleep duration during early infancy is associated with future reports of sleep problems, its association with cognitive development was virtually non-significant. An important finding of this dissertation is that associations between infant sleep patterns and cognitive development were inconsistent, which differs from findings in older populations, and warrant further investigation.

Overall, the findings of the current dissertation further our understanding of the complex relationship between infant sleep, future sleep problems, and cognitive/psychomotor development and suggest practical considerations to promote optimal sleep patterns in infants and preschool-aged children. However, there is a need for further investigation in this area of the pediatric sleep literature, examining the association between early sleep patterns and later outcomes. This is particularly salient given the importance of sleep for parents during their transition of parenthood. This field of research is important to continue exploring, both for a



better understanding of infant development and for parental well-being during this crucial and sensitive period of life.

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## Appendix

## Center for Epidemiologic Studies – Depression Scale (CES-D)

**Center for Epidemiologic Studies Depression Scale (CES-D), NIMH**

Below is a list of the ways you might have felt or behaved. Please tell me how often you have felt this way during the past week.

Week	During the Past			
	Rarely or none of the time (less than 1 day )	Some or a little of the time (1-2 days)	Occasionally or a moderate amount of time (3-4 days)	Most or all of the time (5-7 days)
1. I was bothered by things that usually don't bother me.	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
2. I did not feel like eating; my appetite was poor.	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
3. I felt that I could not shake off the blues even with help from my family or friends.	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
4. I felt I was just as good as other people.	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
5. I had trouble keeping my mind on what I was doing.	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
6. I felt depressed.	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
7. I felt that everything I did was an effort.	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
8. I felt hopeful about the future.	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
9. I thought my life had been a failure.	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
10. I felt fearful.	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
11. My sleep was restless.	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
12. I was happy.	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
13. I talked less than usual.	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
14. I felt lonely.	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
15. People were unfriendly.	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
16. I enjoyed life.	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
17. I had crying spells.	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
18. I felt sad.	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
19. I felt that people dislike me.	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
20. I could not get "going."	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>

**SCORING:** zero for answers in the first column, 1 for answers in the second column, 2 for answers in the third column, 3 for answers in the fourth column. The scoring of positive items is reversed. Possible range of scores is zero to 60, with the higher scores indicating the presence of more symptomatology.

## Questionnaire About Sleep Habits

Sujet no: \_\_\_\_\_

DCC ID : \_\_\_\_\_

Nom AR: \_\_\_\_\_

Date : \_\_\_\_\_

### QUESTIONS ABOUT SLEEPING HABITS

**For the questions involving references to time, please indicate the length of time in hours and minutes. Remember to make reference to your child's behaviour and to your habits during the last weeks.**

1. At what time do you put your child to bed for the night? \_\_\_\_\_
  
2. How much time does your child take to fall asleep at night? \_\_\_\_\_
  
3. When you put you child to sleep in the evening :
  - a) S/he is awake and falls asleep by her/himself.
  - b) S/he is awake and you stay in the room until s/he falls asleep
  - c) You rock her/him until s/he falls asleep
  - d) S/he falls asleep while drinking her/his bottle
  - e) Other, please specify: \_\_\_\_\_
  
4. What is the total length of your child's sleep during the night? \_\_\_\_\_
  
5. During the night, how many hours does your child sleep without waking up (in a row)?  
\_\_\_\_\_
  
6. At what time does s/he wake up in the morning? \_\_\_\_\_
  
7. Does your baby wake up at night to drink?
  - a) yes, how many times: \_\_\_\_\_
  - b) no (Go to question 9)

8. Does s/he go back to sleep by herself/himself in general?
- a) yes
  - b) no
  - c) s/he falls asleep while drinking
9. Does your baby wake up at night (for another reason than to drink)?
- a) yes, how many times: \_\_\_\_\_
  - b) no (Go to question 13)
10. What is the total duration of those awakenings? \_\_\_\_\_
11. Does s/he fall back to sleep by herself/himself in general?
- a) yes
  - b) no
12. If no, what do you do to make her/him fall asleep?
- a) You put her/him to bed and you stay in the room until s/he falls asleep
  - b) You speak to her/him or sing until s/he falls asleep
  - c) You rock her/him until s/he falls asleep
  - d) You let her/him cry
  - e) You bring her/him in your bed
13. Does your child sleep:
- a) Alone in her/his room
  - b) In her/his room, with someone else
  - c) In your room, but alone in her/his bed
  - d) In your room, in your bed
14. How many naps a day does your child take?
- a) 1
  - b) 2
  - c) 3
  - d) 4
  - e) more than 4
  - f) Does not take naps (Go to question 16)

15. What is the total length (amount) of sleep of your child during the day? \_\_\_\_\_
16. Does your child sleep with a pacifier?
- a) yes
  - b) no
17. Does your child suck his thumb?
- a) yes
  - b) no
18. Does your child sleep with a transition object like a teddy bear or a security blanket?
- a) yes
  - b) no
19. Does your child sleep in a darkened room at night (with curtains or shades in the windows)?
- a) yes
  - b) no
20. Does your child walk or talk during her/his sleep?
- a) No
  - b) Sometimes
  - c) Often
  - d) Every night
  - e) Don't know
21. Does your child breathe loudly during her/his sleep?
- a) No
  - b) Sometimes
  - c) Often
  - d) Every night
  - e) Don't know