SLEEP DISTURBANCE IN FAMILY CAREGIVERS
OF CHILDREN WHO DEPEND ON MEDICAL TECHNOLOGY COMPARED TO
FAMILY CAREGIVERS OF HEALTHY CHILDREN:
The CARE to SLEEP STUDY

by

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A thesis submitted in conformity with the requirements
for the degree of Doctor of Philosophy
Graduate Department of Nursing Science
University of Toronto
2015
Abstract

Sleep disturbance in family caregivers of children who depend on medical technology compared to family caregivers of healthy children: 
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Background/Rationale: Society relies on family caregivers of children who depend on medical technology (e.g., home ventilation) to provide highly skilled and vigilant care 24 hours per day. Few studies exist that have measured sleep in family caregivers, and those that do have relied entirely on subjective measures. These data suggest that sleep disruption places family caregivers at risk for poor health and related outcomes that may impair their daytime function and long-term capacity for caregiving.

Objectives/Research Questions: The primary aim of this study was to compare sleep and related outcomes in family caregivers of children who depend on medical technology to outcomes in family caregivers of healthy children.

Methods: In a prospective cohort study (balanced per child’s age), 42 family caregivers of children who depend on medical technology from a tertiary-level paediatric hospital and 43 controls from community-based paediatric clinics were recruited. Actigraphy was used for 6 days and 7 nights, and the Pittsburgh Sleep Quality Index (PSQI) was applied to collect sleep data. At home visits, sleep diaries were collected and measures of depression (CES-D), daytime sleepiness (ESS), fatigue (MAF), quality of life (SF-12\textsuperscript{TM}), sleep hygiene (SHI), and child’s sleep habits (CSHQ) were administered.
**Results:** Family caregivers of children who depend on medical technology achieved 40 minutes less sleep per night (6.56 [1.4h] vs. 7.21 [0.6], \( p = .02 \)), had more nocturnal awakenings (8.00 [3.9] vs. 6.01 [3.7], \( p = .02 \)), and had more sleep deprived (< 6 h) nights (2.18 [2.4] vs. 0.60 [.8], \( p < .01 \)) than controls. Scores on sleep quality (PSQI) also differed (7.75 [2.9] vs. 5.45 [2.8], \( p < .01 \)) with family caregivers of children who depend on medical technology getting ‘poorer’ sleep. Other statistically significant differences were found in outcomes of depression (11.70 [8.7] vs. 6.95 [6.0], \( p = .01 \)), daytime sleepiness (8.35 [4.2] vs. 5.07 [3.3], \( p < .01 \)), and fatigue (22.12 [9.1] vs. 17.44 [9.0], \( p = .02 \)).

**Significance/Implications:** This study confirms, using objective measurement, that family caregivers of children who depend on medical technology experience sleep deprivation and its negative consequences. The results of the study inform clinical practice (need for screening/teaching), policy (enhanced respite), and future research studies (targeted sleep intervention) with this vulnerable group.
ACKNOWLEDGEMENTS

I am extremely grateful to my PhD supervisor, Robyn Stremler, for her unwavering commitment to my learning and the quality of this thesis. It has been a privilege to be her doctoral student and benefit from her vast knowledge, deep integrity, and balanced perspective on becoming a scientist. I thank her for modeling what was required and conveying what was expected of me with absolute clarity; thus ensuring I had the requisites that I needed to stay focused on my goals and achieve my vision. I also wish to thank my committee members Eyal Cohen and Karen Spalding who brought forward critical insights that challenged my thinking from beginning to end. Together they helped to clearly situate the relevance of this research and provided instrumental and encouraging support for which I am very grateful.

I am enormously appreciative for the partnership of Michelle Ho, Nicole Sidhu and Lyndsey McCrae who engaged so enthusiastically in the conduct of this study. I offer tremendous thanks also to Eleanor Pullenagyam, who with warm reassurance, provided oversight of the study’s statistical analyses, and Stephanie Gee who patiently sat with me and enabled that I become more confident in my ability to work with numbers. Thanks also to Elizabeth Uleryk, health science librarian, and Heather Gough, whose expertise enabled this to be a polished final copy.

Big thanks to my colleagues and mentors: Allan Coates, Pam Hubley, Karima Karmali, Margaret Keatings, Mary McAllister, Sioban Nelson, and David Nicholas for helping open doors and pave a path for me and my aspirations. To Sherri Adams and Tanya Smith I thank you- your professional practice and family values inspired me and made me think- I liked that. I am also thankful to Louise Rose and Sandi Blitz who provided instrumental guidance and feedback even when I wasn’t sure I wanted it. I want also to express appreciation for Efrosini Papaconstantinou who told me ‘you can do it’ showed me what ‘it’ looked like.
I’m hugely thankful for the open arms and weary smiles of the families who participated so enthusiastically in this study. I am also thankful for the sources of funding that I received that made the timely completion of this research possible: the Ontario Respiratory Care Society (Lung Association of Ontario), the Canadian Respiratory Health Professionals (Lung Association of Canada) and the Norman Saunders Innovation in Complex Care Grant (SickKids Foundation). Also, instrumentally supportive were the staffs in the Respiratory Medicine and Paediatric Specialty clinics at The Hospital for Sick Children, Village Park Paediatrics and Maple Kidz Clinic who communicated and acted on their belief in the value of this study.

While I pursued doctoral studies, a community of unbelievably caring and supportive friends surrounded me including: Sharon, Karen & Dave, Bob & Jen, Jackie & Ross, Bert & Crocetta, Midge & Steve, Vicky & Ian, Susan, Kate and many others near and far. It is true that ‘it takes a village’. I am so thankful for your friendship and for never hesitating to lend a hand or an encouraging word. To the ‘Poncho Girls’ who kept me KALMRR- I ‘raise a glass’. And special recognition goes to Rachel, who read copies of this thesis and told me it was ‘fabulous’ long before it deserved any big accolades.

I am very appreciative to those closest to home for their care and encouragement over the course of my PhD studies. Thank you to David and Allison Lau, my mother and father in-law who were a constant source of support and never asked when I would finish my PhD. Their gracious and reliable presence helped me to abate my guilt and allowed to feel secure- knowing that our family, and especially my children, were always well taken care of. To Enriqueta Manalo, I am so thankful for your prayers and for making every day together safe and full of sustenance- you’ve nurtured my family and my soul.
I am deeply grateful to my biggest source of inspiration to pursue these studies, my parents. The integrity and caring values of my father, G.C. (Pete) Keilty, provided the exemplar of what it looks like to commit wholeheartedly to what really matters in life, and never, ever, take a good education for granted. I thank him for showing me the way. My mother, Patricia Keilty (nee McCarthy), always conveyed a never-ending belief and pride in my abilities. I thank her for making such a lasting impression and ensuring that I ‘had it in me’ to achieve this goal. Also, thanks to the best cheerleaders anyone could ask for- my brother Peter, sister Jill, and their families- while over recent years you have usually been heard from a distance, I have always felt you close to my heart.

Finally, I am forever grateful to my daughters who provided the light and laughter that pulled me out of the darkest shadows on this journey. Gabrielle you are so strong, empathic, wise, and spirited – amazing qualities that I know will keep you grounded and take you far. Phoebe, I am so thankful for the sunshine you bring to every situation, and for your faith in general, and especially in me that ‘everything will fine’. You exemplify what it is to be talented, determined, brave and capable. Girls, I am so proud of you. And finally, to my husband, Eddy Lau, there are no words for what your strength of character, sense of purpose, and ‘fun-loving’ presence mean to me. I am forever grateful for your partnership in achieving my dreams. It is my desire that this thesis provides evidence for the value of hard work and our abiding commitment to health, happiness, love and family.
# Table of Contents

List of Tables ..................................................................................................................... xi  
List of Figures ................................................................................................................... xii  
List of Appendices ........................................................................................................... xiii  
Chapter 1: Introduction ....................................................................................................... 1  
  Problem Statement .......................................................................................................... 6  
  Purposes of the Study ...................................................................................................... 7  
Chapter 2: Review of the Literature and Conceptual Framework ...................................... 8  
  Sleep ................................................................................................................................ 8  
    Regulation of sleep. .................................................................................................... 8  
    Patterns of sleep ....................................................................................................... 10  
    Amount of sleep. ....................................................................................................... 11  
  Common Sleep Disturbances ........................................................................................ 11  
  Consequences of Poor Sleep ......................................................................................... 12  
    Negative health outcomes. ....................................................................................... 12  
    Other outcomes. ........................................................................................................ 13  
  Sleep in Family Caregivers ........................................................................................... 14  
    Sleep in family caregivers of older adults ............................................................ 14  
    Sleep in family caregivers of children ................................................................... 16  
      Family caregivers of healthy children ............................................................... 16  
      Family caregivers of children with chronic illness .......................................... 17  
      Sleep in family caregivers of children with disabilities .................................... 19  
      Sleep in family caregivers of children who depend on medical technology ...... 22  
    Quantity and quality of sleep in family caregivers of CMT. .................................... 25  
      Sleep quantity........................................................................................................ 25  
      Sleep quality.......................................................................................................... 26  
  Correlates of Sleep Disruption in Family Caregivers ................................................... 27  
    Health-related quality of life ..................................................................................... 27  
    Depression ................................................................................................................ 29  
    Sleepiness ................................................................................................................ 29  
    Fatigue ..................................................................................................................... 30  
    Family function ....................................................................................................... 31  
  Factors Influencing Sleep in Family Caregivers ........................................................... 33  
    Caregiver factors. ...................................................................................................... 33  
      Sociodemographics ............................................................................................... 33  
      Sleep hygiene ....................................................................................................... 34  
      Vigilance ............................................................................................................... 34  
    Child factors ............................................................................................................. 35  
      Technology use. .................................................................................................... 35  
      Nocturnal care needs. ........................................................................................... 36  
      Child sleep ............................................................................................................ 36  
    Environmental factors .............................................................................................. 38  
      Use of homecare personnel. .................................................................................. 38  
      Use of monitoring. ............................................................................................... 40  
      Sleeping arrangements. ....................................................................................... 41
<table>
<thead>
<tr>
<th>Chapter 4: Results</th>
<th>Chapter 5: Discussion</th>
</tr>
</thead>
<tbody>
<tr>
<td>Sample Acquisition</td>
<td>Sample characteristics</td>
</tr>
<tr>
<td>Feasibility</td>
<td>Recruitment to strata</td>
</tr>
<tr>
<td>Procedure</td>
<td>Research ethics</td>
</tr>
<tr>
<td>Data Management</td>
<td>Pilot testing</td>
</tr>
<tr>
<td>Data Analysis</td>
<td>Recruitment and consent</td>
</tr>
<tr>
<td>Analysis of Sleep Outcomes in Family Caregivers</td>
<td></td>
</tr>
<tr>
<td>Primary Research Question: Difference in Sleep</td>
<td></td>
</tr>
<tr>
<td>Question 1</td>
<td>Question 2</td>
</tr>
<tr>
<td>Question 3</td>
<td>Question 4</td>
</tr>
<tr>
<td>Secondary Research Question</td>
<td></td>
</tr>
<tr>
<td>Question 5</td>
<td>Other group differences</td>
</tr>
<tr>
<td>Secondary Analysis of Primary Question</td>
<td></td>
</tr>
<tr>
<td>Question 6</td>
<td>Caregiver factors</td>
</tr>
<tr>
<td>Question 7</td>
<td>Child factors</td>
</tr>
<tr>
<td>Model 1</td>
<td>Environmental factors</td>
</tr>
<tr>
<td>Model 2</td>
<td>Study feasibility and acceptability</td>
</tr>
<tr>
<td>Study feasibility and acceptability</td>
<td></td>
</tr>
<tr>
<td>Chapter 5: Discussion</td>
<td>Primary Research Question</td>
</tr>
<tr>
<td>Differences in sleep quantity.</td>
<td>Nighttime sleep</td>
</tr>
</tbody>
</table>
|                    | }
List of Tables

<table>
<thead>
<tr>
<th>Table</th>
<th>Page</th>
</tr>
</thead>
<tbody>
<tr>
<td>Table 1 – Study variables and data sources for primary and secondary questions</td>
<td>52</td>
</tr>
<tr>
<td>Table 2 – Study variables and data sources for exploratory questions</td>
<td>52</td>
</tr>
<tr>
<td>Table 3 – Distribution of family caregiver participants by index child’s age and gender</td>
<td>82</td>
</tr>
<tr>
<td>Table 4 – Baseline demographic characteristics of family caregivers</td>
<td>82-83</td>
</tr>
<tr>
<td>Table 5 – Clinical characteristics: Children dependent on medical technology</td>
<td>86</td>
</tr>
<tr>
<td>Table 6 – Use of homecare among family caregivers of children who depend on medical technology</td>
<td>88</td>
</tr>
<tr>
<td>Table 7 – Between-group differences on objective sleep outcomes</td>
<td>90</td>
</tr>
<tr>
<td>Table 8 – Sleep onset latency subjectively reported in repeated entries in sleep diary</td>
<td>93</td>
</tr>
<tr>
<td>Table 9 – Group comparisons on measures of quality of life, depression, sleepiness, and fatigue</td>
<td>94</td>
</tr>
<tr>
<td>Table 10 – Linear regression between caregiver factors and TST-N</td>
<td>97</td>
</tr>
<tr>
<td>Table 11 – Linear regression between caregiver factors and NNA-O</td>
<td>98</td>
</tr>
<tr>
<td>Table 12 – Linear regression between child factors and TST-N</td>
<td>99</td>
</tr>
<tr>
<td>Table 13 – Linear regression between child factors and NNA-O</td>
<td>99</td>
</tr>
<tr>
<td>Table 14 – Linear regression between environmental factors and TST-N</td>
<td>100</td>
</tr>
<tr>
<td>Table 15 – Linear regression between environmental factors and NNA-O</td>
<td>100</td>
</tr>
<tr>
<td>Table 16 – Regression: TST-N on multiple modifiable and non-modifiable factors</td>
<td>101</td>
</tr>
<tr>
<td>Table 17 – Regression: NNA-O on multiple modifiable and non-modifiable factors</td>
<td>102</td>
</tr>
</tbody>
</table>
# List of Figures

<table>
<thead>
<tr>
<th>Figures</th>
<th>Page</th>
</tr>
</thead>
<tbody>
<tr>
<td>Figure 1 – Flow diagram- review of records in systematic review</td>
<td>24</td>
</tr>
<tr>
<td>Figure 2 – Conceptual Model of Sleep in Family Caregivers</td>
<td>47</td>
</tr>
<tr>
<td>Figure 3 – Planned study schema using flow diagram</td>
<td>71</td>
</tr>
<tr>
<td>Figure 4– Schema achieved for cohort study recruitment</td>
<td>81</td>
</tr>
<tr>
<td>Figure 5 – Distribution of average TST-N in FC CMT versus FC controls from 9 p.m.– 9 a.m. in minutes</td>
<td>91</td>
</tr>
</tbody>
</table>
# List of Appendices

<table>
<thead>
<tr>
<th>Appendix</th>
<th>Page</th>
</tr>
</thead>
<tbody>
<tr>
<td>A – Summary of studies of sleep in family caregivers of older adults</td>
<td>185</td>
</tr>
<tr>
<td>B – Summary of studies of sleep in family caregivers of children with chronic illness</td>
<td>190</td>
</tr>
<tr>
<td>C – Search terms and strategy for systematic review</td>
<td>200</td>
</tr>
<tr>
<td>D – Summary of studies in systematic review</td>
<td>203</td>
</tr>
<tr>
<td>E – Characteristics of sample participants in systematic review</td>
<td>206</td>
</tr>
<tr>
<td>F – Letter of information to study participants</td>
<td>210</td>
</tr>
<tr>
<td>G – Letter of information: Parents of healthy adolescent children</td>
<td>213</td>
</tr>
<tr>
<td>H – Study eligibility screening forms</td>
<td>216</td>
</tr>
<tr>
<td>I – Study consent forms</td>
<td>219</td>
</tr>
<tr>
<td>J – Baseline sociodemographic data forms</td>
<td>240</td>
</tr>
<tr>
<td>K – Daily sleep diaries</td>
<td>249</td>
</tr>
<tr>
<td>L – Home visit questionnaires</td>
<td>271</td>
</tr>
</tbody>
</table>
Chapter 1: Introduction

Across Canada, family caregivers are estimated to provide over two billion hours of caregiving annually (Fast, Niehaus, Eales, & Keating, 2002). Among these family caregivers, society relies upon those with children who depend on medical technology (CMT) to provide highly skilled and vigilant care in their homes on a 24-hour per day basis. This extraordinary responsibility, lasting potentially years to decades, has been linked to chronic sleep disturbances that place family caregivers of CMT at risk of negative health and related outcomes that may threaten their ability to provide long-term quality care for their child and family.

Sleep is recognized as a complex biobehavioral process. Sleep of adequate quantity and of good quality is considered necessary for health (Borbely, 1982) and optimal performance (Durmer & Dinges, 2005). Sleep disturbances are prevalent in the general public but appear to be even more so in family caregivers of CMT, with rates of up to 100% reported in descriptive studies of this group.

CMT include those with substantial family-identified health care service needs, complex medical conditions, severe functional limitations, and high utilization of resources (Cohen et al., 2011b). They are classically categorized according to their use of specific technologies (Wagner, Power, & Fox, 1987) and defined as a group of children with chronic health conditions requiring homecare nursing services and for whom an equipment failure or malfunction is not unlikely and would result in a negative health outcome for the child. This group of children includes those who are reliant on the daily use of one or more technologies, such as tracheotomies, mechanical ventilators, oxygen delivery systems, and enteral feeding devices. CMT often require around-the-
clock observation and are considered among the most complex and resource intensive of all community-dwelling individuals (Glendinning, Kirk, Guiffrida, & Lawton, 2001).

The number of CMT in Canada is not clear. The Paediatric Complex Care Coordination Expert Panel (Rosenbaum, 2008) has reported that the rates for medically fragile children (many of whom are technology-dependent) in Ontario is approximately 55/100,000. Child health experts concur, however, that with advances in health care and technology, the global numbers of children with complex care needs, including those who are CMT, is steadily on the rise. In a recent population-based study in the United States, 22% of all children enrolled in Medicaid were found to be medically fragile (Buescher, Whitmire, Brunssen, & Kluttz-Hile, 2006). Moreover, among children with specialized health care needs, 3.2% ($n = 324,323$) met criteria for more complex children, representing 0.4% of all children in the United States (Kuo, Cohen, Agrawal, Berry, & Casey, 2011). In Canada, the prevalence of CMT has been estimated in a recent study that examined hospital administrative data from 2005–2007. In this retrospective cohort study, 6.2% ($n = 1,584$) of all children ($n = 25,662$) who had been identified with neurological impairments and complex chronic conditions required technological assistance (Cohen et al., 2011). This cohort was estimated to represent approximately 0.9% of all children in the province of Ontario, but given that these data represent only those children who have been hospitalized, they may not have fully captured the number of CMT in the community or homecare who did not present to hospital during the study period. Given this, and based on the exponential growth of specialized hospital and homecare programs for children with complex care and technology dependence, these data likely underestimate the real prevalence of CMT and their caregivers.

The Canadian Caregiving Coalition (2001) defines *caregiver* as those who provide care
and assistance for their family members and friends who are in need of support because of physical, cognitive, or mental health conditions. The work of family caregivers in Ontario contributes to significant public savings (Premier’s Council on Health, Wellbeing and Social Justice, 1994). Family caregivers of CMT are responsible for care that is demanding and includes constant monitoring and frequent therapeutic interventions (Keilty, Nicholas, & Selkirk, 2008; Kirk, Glendinning, & Callery, 2005; Wagner et al., 1987). This responsibility may include the constant surveillance and management of artificial airways, mechanical ventilators, oxygen delivery systems, and enteral feeding devices, and it has been linked to chronic sleep disturbances that place family caregivers at risk of negative health outcomes with daytime consequences. The risk of a bad outcome, including unexpected death at home, is a constant source of concern that may contribute to hypervigilance and constant worry that intrudes on sleep for family caregivers of CMT (Buescher et al., 2006; Chye & Gray, 1995; Mentro, 2003).

It is well understood that specialized paediatric nurses would otherwise assume the care provided by family caregivers of CMT if the CMT remained in hospital.

Although many family caregivers describe benefits of caregiving, qualitative studies of family caregivers of children with chronic illnesses have documented compelling descriptions of their sleep disturbances and caregiver burdens (Andrews & Nielson, 1988; Ferrell, Rhiner, Shapiro, & Dierkes, 1994; Heaton, Noyes, Sloper, & Shaw, 2006; Horner, 1997; Sullivan-Bolyai, Deatrick, Gruppuso, Tamborlane, & Grey, 2003). Also, in descriptive studies using subjective sleep measures, variable rates of sleep disturbance, from 15%–86%, have been reported in family caregivers of children with chronic illnesses (Meltzer & Moore, 2008) and from 51%–100% in studies of those with CMT (Heaton et al., 2006; Hopkins, Whetstone, Foster, Blaney, & Morrison, 2009). It is also reported that family caregivers of CMT requiring
mechanical ventilation have self-reported poorer sleep quality and significantly less sleep quantity per night (6.31 hrs ± 1.2 hrs vs. 7.34 hrs ± 0.9 hr, \( p = < .001 \)) than family caregivers of healthy controls (Meltzer & Mindell, 2006).

The negative health and related consequences of sleep disturbances in family caregivers appear to be numerous. In laboratory experiments, a sleep duration of < 7 hours per night has been associated with poor performance on simulated tasks among new parents of healthy infants (Plessow, Kiesel, Petzold, & Kirschbaum, 2010). A poll by the National Sleep Foundation (2004) reported that 29% of all parents surveyed experienced insomnia a few nights per week, 53% were awakened by their child at least once per week, and 19% experienced daytime sleepiness severe enough to interfere with their daily activities. In clinical studies, self-reported sleep disturbances in family caregivers of children with chronic illnesses are associated with caregiver depression (Meltzer & Moore, 2008), and depression is a known risk factor for poor parenting abilities and negative effects on children’s physical, psychological, behavioral, and social development (National Academies Press, 2009). Additionally, in a study of family caregivers of children with asthma, poor sleep quality combined with depression was reported to impede family caregiver motivation and performance, with resultant increased emergency room use (Yuksel et al., 2007). Together, these data provide beginning evidence for a link between sleep disturbance and negative health and related outcomes in family caregivers that may hinder their executive function and daytime performance.

The health-related quality of life (HRQoL) of family caregivers of CMT has been reported to have a positive relationship with self-reported sleep quality (Read, Simonds, Kinlai, Muntoni, & Garralda, 2010). Elevated rates of fatigue associated with objectively measured (Gedaly-Duff, Lee, Nail, Nicholson, & Johnson, 2006) and self-reported (Zupanec, Jones, &
shorter sleep durations have been described in childhood cancer caregivers. In a study of family caregivers of CMT, it was reported that subjective sleep quality mediated the relationship between caregiver fatigue, and depression (Meltzer & Mindell, 2006). Moreover, in studies of family caregivers of older adults, sleep disruption has been associated with excessive daytime sleepiness, caregiver burden, and higher rates of institutionalization (Pollak & Perlick, 1991). Thus, the potential contributors to and consequences of sleep disturbance in family caregivers of CMT, including associations with HRQoL, depression, daytime sleepiness, and fatigue, merit careful investigation.

In descriptive studies, family caregivers of CMT have described caregiver, child, and environmental factors that influence their sleep (Andrews & Nielson, 1988; Meltzer & Moore, 2008). It has been reported that family caregivers of CMT experience stress and intrusive thoughts that impede their ability to sleep (Meltzer & Mindell, 2006). Some family caregivers have reported staying up multiple nights in a row to monitor and care for their children (Heaton et al., 2006; Horner, 1997), and others have reported staying awake to let homecare nurses into the house at the beginning of a night shift.

Paradoxically, family caregivers have also reported that they may be woken at night by homecare nurses and technology that are intended to provide respite, promote sleep, and relieve family caregiver burden (Andrews & Nielson, 1988; Heaton et al., 2006). The daytime consequences of these sleep disturbances may be seen in rates of daytime sleepiness and fatigue along with heightened depression and poor HRQoL in family caregivers of CMT.

To summarize, studies of sleep in family caregivers of CMT are few, and those that exist have relied entirely on self-reported sleep measures. Broad variation in methods, measurement, and minimal details about sample clinical characteristics limit their applicability to clinical
practice and research with this group. Hence, the quality and quantity of sleep experienced by family caregivers of CMT are not clearly known, and it is unclear how their experiences compare to that of parents of otherwise healthy children for whom there are not good estimates of sleep. Evidence exists for the presence of sleep disturbances in family caregivers of new infants (Stremler et al., 2006), but no studies have reported the results of objectively measured sleep patterns of parents of children across developmental ages and stages. Moreover, little is known about what role the caregiver, child, and the environment play in promoting or inhibiting sleep in family caregivers. Finally, the influence of sleep on the health and related daytime consequences in family caregivers remains unclear. Some potential sources of sleep disturbances in family caregivers may be modifiable. Researchers, expert clinicians, and family caregiver advocacy groups suggest that more intervention is needed to promote sleep in family caregivers; however, data are not available to inform such interventions. Thus, there is a need for further understanding of the unique features of sleep in family caregivers of CMT.

**Problem Statement**

Sleep disturbance in family caregivers of CMT is a compelling problem. Nonetheless, few studies of sleep in family caregivers exist, and those that do have used subjective measures of sleep. Epidemiological data and clinical practice suggest a rising prevalence and negative consequences of sleep disturbance in family caregivers, but the unique characteristics of sleep in this group are not known. To date, there has not been adequate investigation of the factors that contribute to sleep disturbances in family caregivers of CMT. In recent studies, sleep disturbance has emerged as a problem despite the use of supports, including homecare nursing and advanced medical technology (Meltzer, Boroughs, & Downes, 2010). Before targeted interventions may be developed, data are needed that describe the prevalence of sleep disruption, detail sleep and its
associations, and identify which individual, child, and environmental factors most influence sleep disturbance in family caregivers of CMT.

**Purposes of the Study**

Due to the shortcomings of existing evidence, the overall objective of the study outlined for this thesis was to characterize sleep in family caregivers of CMT. It employed actigraphy, which is an objective measure of sleep that has been deemed acceptable to participants in studies of new parents at home (Stremler et al., 2006) and of family caregivers of elders with dementia (Akkerman & Ostwald, 2004; Carter, 2002, 2006). A comparative cohort of parents of healthy, same-aged children served to provide a comparison for the family caregiver data.

The primary aim of this investigation was to compare sleep in family caregivers of CMT to that of family caregivers of healthy children. The secondary aim was to describe the nature of associations between sleep disturbances in family caregivers and related outcomes including HRQoL, depression, daytime sleepiness, and fatigue. Finally, this study has begun to explore factors that influence sleep disturbance in family caregivers of CMT.

The results of this investigation are expected to be of interest to family caregivers of CMT along with child health clinicians, researchers, policy makers, and advocates who seek to promote health and the capacity for long-term caregiving among family caregivers of CMT.
Chapter 2: Review of the Literature and Conceptual Framework

This chapter explores the need to better understand sleep in family caregivers of CMT, the factors that influence sleep, and the outcomes associated with sleep disturbances in this vulnerable population. Sleep disturbances and processes are described, as well as the literature on sleep in varied types of family caregivers, including those who care for older adults, healthy children and children with chronic conditions. Description of the findings of a systematic review of studies that have reported on sleep outcomes in family caregivers of CMT is also included. Overall the chapter offers a critical examination of the possible influences on and consequences of sleep disturbance in family caregivers of CMT, and draws from the larger caregiver literature to conclude that sleep disturbance in family caregivers of CMT is influenced by multiple caregiver, child, and environmental factors that merit further investigation. Finally, this chapter presents an evidence-informed conceptual model that guided this prospective cohort investigation of sleep in family caregivers of CMT and family caregiver controls. The chapter ends with an outline of the specific research questions answered in this study.

Sleep

While the purpose of sleep is not fully understood, it is generally accepted that sleep is required by all living beings and that it plays a role in restoration and recovery of cells, tissues, and organs (Eidelman, 2002). Thus, sleep is considered essential for the promotion and maintenance of overall health (Borbely, 1982) and daytime function (Stepanski, 2002).

Regulation of sleep. Sleep is a complex biobehavioral condition that adjusts to the body’s everyday needs by the integration of three related processes (Borbely & Achermann, 1999). The first is a homeostatic process, referred to as Process S, mediated by neurotransmitters and influenced by the duration since the last sleep episode. Process S influences the timing and
amount of slow-wave sleep, considered an index of sleep intensity, as evidenced on a sleep
electroencephalogram. Second, a circadian process called Process C is linked to an internal 24-
hour periodicity. This process is independent of the amount of time since or duration of the
previous sleep episode. Rather, the propensity for sleep regulated by Process C is influenced by
zeitgebers, which are cues to circadian timing. Light exposure is the most influential zeitgeber,
with social interactions, medication administration, exercise, and eating/drinking patterns among
other 24-hour cues that help establish this periodic process.

Under normal circumstances, Process S and Process C are synchronized, thus promoting
good sleep regulation and related outcomes. However, under many conditions, these sleep
processes may be disrupted. Process S is sensitive to the effects of shortened sleep duration over
a single night or multiple nights, resulting in a powerful accumulation of sleep pressure and the
subjective experience of excessive daytime sleepiness and fatigue. Daytime naps can influence
Process S by diminishing the pressure to fall asleep; however, naps can also disrupt sleep
regulation by causing the timing of preferred sleep onset, influenced by Process S, to be out of
synch with the more entrenched circadian influences. Process C may also be negatively
influenced by poor sleep hygiene practices, resulting in irregular sleep–wake patterns and
frequent nocturnal arousals. Process C is sensitive to the influences of sleeping in uncomfortable
spaces, thinking about emotionally charged events/activities while in bed, and the intrusion of
noise, lights, and social interactions during the night. “Variations in the natural and artificial
rhythms of the technology-dependent child” (Heaton, Noyes, Sloper, & Shaw, 2005, p. 4447)
may impose similar irregularities in the biological clock of family caregivers of CMT and
influence their circadian regulation and Process C. Intrinsic biological features, including the
presence of select chronic health conditions and the effects of aging, may also influence Process C and overall sleep regulation in adults.

The third mechanism in sleep regulation is an ultradian process, which is one that repeats itself over a period of less than 24 hours, such as the cycles of appetite and thermoregulation. An ultradian cycle is also responsible for two basic sleep states: rapid eye movement (REM) and non-rapid eye movement (NREM; Borbely & Achermann, 1999). The distribution of NREM and REM sleep is an important feature of this process, which is responsible for sleep that is organized, efficient, and of good quality. Disruption of the ultradian process may be inferred by the presence of frequent nocturnal awakenings or shortened length of sleep.

All three sleep processes are sensitive to biobehavioral influences that may affect their synchrony and regulation. Together, the three processes of sleep are summarily influenced by a combination of an individual’s sleep pattern and amount of sleep.

**Patterns of sleep.** Adults typically experience up to six sleep cycles of approximately 90–120 minutes in length per night (Carskadon & Dement, 2005). A healthy and well-rested adult will pass through a sleep cycle in predictable stages as follows: wakefulness, Stage 1, Stage 2, and Stages 3 and 4, which are referred to together as slow-wave sleep (Hirshkowitz, Sarwar, & Sharafkhaneh, 2010). As a function of Process S, in most adults, slow-wave sleep dominates the sleep cycle towards the beginning of the night. This homeostatic drive is highest at sleep onset and diminishes across the night as sleep pressure wanes. In contrast, episodes of REM sleep are longest in the last third of the night and are linked to the circadian oscillator and body temperatures, which are associated with Process C. NREM sleep most prominently contributes to the restorative function of sleep, whereas REM sleep influences mood regulation and cognition (Carskadon & Dement, 2005). For those who experience shortened sleep times and frequent
awakenings, it is difficult to achieve the right balance of NREM and REM sleep; thus, they may not benefit from the restorative features of normal sleep–wake patterns.

**Amount of sleep.** The total sleep time (TST) or length of sleep in adults depends on a number of physiological and behavioral factors. Weekday to weekend sleep typically differs, with adults reporting total sleep times (TST) of 7.5 hours a night during the work week and 8.5 hours on weekends (Carskadon & Dement, 2005). Men and women in Canada (Hurst, 2005) and the United States (Hale, 2005) have reported clinically insignificant differences in the amounts of sleep they get per night, with men in Canada, for example, sleeping on average only about 11 minutes less than women.

**Common Sleep Disturbances**

Sleep disturbances are prevalent and on the rise in the general population. Up to 75% of healthy adults reported at least one sleep problem a few nights a week within the previous year (National Sleep Foundation, 2005).

Sleep fragmentation is caused by frequent and sustained arousals that disturb sleep patterns and shorten the overall amount of time spent asleep. Sleep fragmentation is common among individuals with intrinsic sleep disorders. Sleep fragmentation may also result when sleep is disrupted due to external cues such as noise and other alerts in the environment. Complaints of poor subjective sleep quality and impaired daytime function are frequently cited among those with frequent nocturnal awakenings, even when sleep architecture (the cyclical pattern of sleep as it shifts between the different sleep stages) and total sleep duration is preserved (Stepanski, 2002).

Sleep restriction refers to when an individual, willfully or not, limits the length of sleep or prolongs a period of wakefulness on either an acute or chronic basis. Population-based data
suggest that North Americans get an average of two hours less sleep per night than they did two decades ago (National Sleep Foundation, 2005). Thus, sleep restriction is being hallmarked as one of the great public health concerns of this generation and is considered to be a consequence of shifting societal norms for the length of the workday and the intrusiveness of technologies (e.g. handheld devices, tablets) on sleep.

Sleep deprivation is the result of getting less sleep than is needed. It can be either acute, following a single period of extended wakefulness, or chronic, with the accumulation of sleep debt over multiple nights of sleep restriction. Sleep deprivation may occur due to short or absent sleep phases, resulting in shortened length of sleep, but it may also occur as a consequence of disrupted sleep cycles, with resultant sleep fragmentation. Sleep fragmentation results when the sleep–wake cycle is repeatedly disrupted, and it can have the same effect on waking behavior as shortened sleep, including deficits in task attendance and excessive daytime sleepiness (Durmer & Dinges, 2005).

In summary, sleep disturbances may emerge as a consequence of sleep fragmentation and sleep restriction. These sleep disturbances may mimic some of the features of select sleep disorders, including insomnia and circadian rhythm disorders, that, if left untreated, may lead to chronic sleep deprivation and the debilitating long-term effects of sleep deprivation, including negative health and related outcomes.

**Consequences of Poor Sleep**

**Negative health outcomes.** HRQoL is reported to be poorer in those with chronic non-restorative sleep (Ohayon, 2005). Elevated levels of cholesterol, insulin, and cardiovascular disease are associated with poor sleep quality in the general public (Ohayon & Vecchierini, 2005). Obesity rates are much higher among poor sleepers than the general public (Gangwisch,
Malaspina, Boden-Albala, & Heymsfield, 2005). Moreover, immune function is negatively affected by shorter sleep duration in longitudinal population-based studies (Bryant, Trinder, & Curtis, 2004), and higher rates of select cancers are found in women who are night-shift workers (Schernhammer, Kroenke, Laden, & Hankinson, 2006).

Both in practice and research, chronic sleep disruption has been associated with negative mental health, including low mood (Banks & Dinges, 2010; Ohayon, 2005), depression (Gerber et al., 1992), psychiatric illness (Breslau, Roth, Rosenthal, & Andreski, 1996; Peterson & Benca, 2011), and overall poorer quality of life (Ancoli-Israel, Moore, & Jones, 2001).

**Other outcomes.** It is reported that sleep-deprived individuals experience negative daytime consequences. Cognitive deficits have been demonstrated as a result of testing subjects’ ability to pay attention (Van Cauter et al., 2003). Reaction times and performance are lowered in laboratory studies of those experiencing mild (6–7 hrs of sleep), moderate (4–6 hrs), and severe (0–4 hrs) sleep deprivation, even after a single night of disrupted sleep (Dinges et al., 1997; Durmer & Durmer, 2005). Sleep loss in shift workers and health care providers results in an increase in errors that is often associated with subjective sleepiness on the job (Balas, Scott, & Rogers, 2006; Richardson et al., 1996; Scott, Hwang, & Rogers, 2006).

Beyond the negative effects on health and the resultant daytime consequences, there are also large financial costs associated with chronic sleep problems, including both elevated rates of absenteeism (missed work) and presenteeism (reduced productivity at work). In a randomly selected sample of 948 adults with a mean age of 43.7 years old (60% female) from the province of Québec, the economic burden of insomnia was described to be very high, with the largest proportion of all expenses (76%) attributable to insomnia-related work absences and reduced productivity. Annual indirect costs associated with insomnia-related absenteeism were estimated
at $970.6 million, with insomnia-related productivity losses estimated at $5.0 billion (Daley, Morin, LeBlanc, Grégoire, & Savard, 2009).

**Sleep in Family Caregivers**

Increasingly, society relies on a smaller number of family caregivers to care for an expanding number of care recipients. This is a result of a number of trends. First, medical intervention is often the preferred choice among family caregivers of those with complex and life-limiting illnesses, and with advances in health care, those with chronic health needs are living longer (Dumas & Péron, 1992). Second, family units are smaller than in the past, and marriage breakdown has increased (Aneshensel, Pearlin, Mullan, Zarit, & Whitlatch, 1995). Third, family caregivers may choose to move from their local communities to urban centers within closer reach of specialized hospitals, thus leaving behind the support of extended family members. Together, these factors typically leave the around-the-clock responsibility for family caregiving, potentially spanning years to decades, to a small number of family caregivers. Among these family caregivers, sleep disturbances and related outcomes emerge as a common complaint regardless of the age of the recipient of care.

**Sleep in family caregivers of older adults.** Despite efforts to support family caregivers, it is generally accepted that the health of family caregivers of older adults is of poorer quality than that of their non-caregiver counterparts. Findings of a recent metasynthesis of differences between caregivers and non-caregivers in psychological health and physical health support this assumption. In their review of 84 studies of family caregivers of older adults ($M \pm SD$; 62.5 [8.6] yrs old), most of whom had dementia (63.3%) patients, Pinquart and Sorensen (2003) reported that family caregivers had poorer physical health, higher stress levels, more depressive symptoms, and lower levels of subjective well-being and self-efficacy than non-caregivers.
There is a growing body of literature describing sleep in family caregivers of older adults. In comparative studies, self-reported sleep quality in family caregivers of older adults is reported to be poorer than that of non-caregivers (Fletcher, Dodd, Schumacher, & Miaskowski, 2008; Rowe, McCrae, Campbell, Pa Benito, & Cheng, 2008; Sato, Kanda, Anan, & Watanuki, 2002; Wilcox and King, 1999). In one observational study, family caregiver nighttime sleep was reported to be less than 6 hours per night over multiple nights (5.6 [1.7] hrs), thus signaling risk for chronic sleep deprivation. In studies using objective measures of sleep, similar differences between sleep in family caregivers and non-caregivers have also been described. Sato et al. (2002) described differences in the proportion of Stage 1 and Stage 2 sleep on overnight electroencephalograms that were suggestive of more sleep deprivation in family caregivers (Sato et al., 2002) when compared to that of non-caregivers. Using actigraphy, which is also an objective measure, Rowe et al. (2008) found that family caregivers ($n = 102$) had shorter sleep times (7.1 [6.6] hrs, $t = 3.19, p = 0.002$) and took longer to fall asleep (22.8 mins vs. 12.5 mins, $t = 2.87, p = .007$) compared to older non-caregiving adults ($n = 31$). It has also been asserted that sleep disturbance in family caregivers of older adults is a significant risk factor for caregiver fatigue (Rowe et al., 2008; Sato et al., 2002), depression (Carter & Chang, 2000; Rowe et al., 2008), and care recipient institutionalization (Hope, Keene, Gedling, Fairburn, & Jacoby, 1999; Pollak & Perllick, 1991).

The evidence surrounding sleep disturbance in family caregivers of older adults also includes a small group of studies that have tested a variety of sleep interventions, including cognitive behavioral therapy (Carter, 2006; Carter, Mikan, & Simpson, 2009; McCurry, Logsdon, Vitiello, & Teri, 1998), exercise programs (King, Baumann, O’Sullivan, Wilcox, & Castro, 2002), sleep education (McCurry, Lodgson, & Teri, 1996), pharmacological therapy
(Ancoli-Israel, Amatniek, Ascher, Sadik, & Ramaswamy, 2005), institutional respite care (Lee, Morgan, & Lindesay, 2007), and cranial electrical stimulation (Rose, Taylor, & Bourguignon, 2009). Results of these studies are summarized (see Appendix A). These studies show promise, as they each effected small improvements in at least one sleep-related variable, but several limitations were noted by each of the researchers. The most common limitation acknowledged in these studies was that the majority had small homogeneous sample sizes, again involving mainly female dementia caregivers, limiting the generalizability of the findings.

**Sleep in family caregivers of children.**

**Family caregivers of healthy children.** Highly variable rates of sleep disruption, from 25%–40%, have been reported in clinical studies of family caregivers of healthy children (Mindell & Owens, 2010). These studies however have used varied study designs and methods and have relied solely on subjective sleep measurement. In contrast, in two recent population-based studies of sleep in North America, family caregivers of healthy children described little difference in their sleep related to their childcare responsibilities (Hurst, 2005; National Sleep Foundation, 2005). In the United States, family caregivers of children under 11 years of age self-reported a sleep duration of 12 minutes less than the 7 hours per night reported for the population as a whole (National Sleep Foundation, 2005). Canadians without children reported 25 minutes more sleep per night than those with two or more children and 17 minutes more sleep than those with only one child (Hurst, 2005). As well, in a nation-wide poll that asked about average sleep durations in healthy respondents, adult Canadians \( n = 9,500 \) with one and two or more children \( (8.01 \text{ hrs and } 7.88 \text{ hrs respectively}; \text{Statistics Canada, 2005}) \). These reported differences in TST are small, and their clinical significance is uncertain. Thus, it is thought that parents of normally developing healthy children experience sleep disturbance owing to their childcare
responsibilities, but the magnitude and related significance of this association remains unclear.

**Family caregivers of children with chronic illness.** Sleep in family caregivers of children with chronic illness has been described in a systematic review of 19 descriptive studies (Meltzer & Moore, 2008). The rates of sleep disruption across these qualitative and cross-sectional investigations were reported to range from 15%–86%. The authors attributed this wide variation to different study methods, time of assessment, or presence of acute illness in the child. The authors summarized that there were four potential causes of sleep disruption in these studies: stress, false monitor alarms, childcare (of the chronically ill child), and nocturnal monitoring needs (Meltzer & Moore, 2008). The consequences of sleep disruption in family caregivers of children with highly varied types of chronic illness included negative outcomes with respect to depression, anxiety, marital satisfaction, sleep location, poor health, fatigue, frustration, and exhaustion.

Building on this review since its publication date in 2007, nine more studies have been identified by this investigator that describe sleep in family caregivers of chronically ill children, and a total of 31 studies are summarized in (see Appendix B). The sleep-related findings of the nine descriptive studies of family caregivers of chronically ill children that were identified by this investigator and not previously cited by Meltzer & Moore (2008) are described immediately below. Studies that investigated sleep in family caregivers of CMT are the subject of a systematic review completed by this investigator and will be detailed in this thesis in the section that follows this one.

The descriptions of sleep and related outcomes across the nine studies of sleep in family caregivers of children with chronic illness (but not technology dependent) were informed by varied methods and measures. None reported on testing interventions. Three studies described
sleep in family caregivers based on cross-sectional investigations, three used mixed-methods combining qualitative interviews and cross-sectional data collection, two described comparative cohort designs of cross-sectional data, and one was a prospective cohort-controlled investigation. Validated self-report sleep measures were used, including the Pittsburgh Sleep Quality Index (PSQI; Read et al., 2010; Yilmaz et al., 2008; Yuksel, et al., 2007), the General Sleep Disturbances Scale (Zupanec et al., 2010), and the 24-Hour Sleep Patterns Inventory (Meltzer et al., 2010). Three studies used qualitative thematic analysis of family caregiver interviews to describe sleep in family caregivers (Hopkins et al., 2009; Montagnino & Mauricio, 2004; Nolan, Camfield, & Camfield, 2006). One study used a validated questionnaire, meant to assess the impact of dermatitis that included a single embedded sleep item (Ricci, Bendandi, Bellini, Patrizi, & Masi, 2007). The children’s conditions in these studies differed in terms of diagnosis, prognosis, and level of care. Most studies reported on family caregivers of children with common and non-life-threatening chronic illnesses, such as asthma and atopic dermatitis, while others reported on very rare conditions (e.g., Dravet syndrome), and a few reported on family caregivers of children with complex conditions. However, sleep did emerge as a consistent complaint across all nine studies.

Scores on the PSQI were higher, indicating poorer sleep quality, in 62 family caregivers of children with asthma (4.8 [2.8]) and in 21 family caregivers of children with cystic fibrosis (4.6 [2.4]) compared to 35 family caregivers of healthy children (3.2 [1.6], \(p = .04\), Yilmaz et al., 2008). In contrast, and despite a modest sample size, Yuksel et al. (2007) did not identify significant differences between self-reported sleep quality on the PSQI in family caregivers of children with asthma (\(n = 75\)) and healthy controls (\(n = 46\)). Using a different measure, the General Sleep Disturbances Scale, Zupanec et al. (2010) reported that approximately half of the
family caregivers of children with childhood leukemia \((n = 77)\) had scores above the cut-off, which suggested significant sleep disturbance. Sleep disturbance was reported in family caregivers of children with atopic dermatitis \((n = 45)\) as the greatest single problem among the items measured by using the Dermatitis Family Impact Questionnaire (Ricci et al., 2007). Thematic analyses of qualitative data that were derived from open semi-structured interviews (Nolan et al., 2006) and open-ended survey questions (Zupanec et al., 2010) have also shown that sleep disturbances are a problem in the population of family caregivers of children with chronic illnesses. The findings of these nine studies extend those described by Meltzer & Moore (2008) and, overall, suggest that sleep disturbance is an important consideration in family caregivers of children with chronic illnesses.

A number of health and related outcomes appear to be correlated with sleep disturbance in family caregivers of children with chronic illness. Elevated rates of depression and anxiety have been reported in family caregivers of children with chronic respiratory conditions who experience sleep disturbances (Yilmaz et al., 2008; Yuksel et al., 2007). Fatigue was correlated with sleep disturbances in family caregivers of children with leukemia \((r = .56, p < .001;\) Zupanec et al., 2010). It is challenging, however, to draw conclusions from these studies. Beyond the problems of the many varied study designs and reliance on subjective sleep measures, this literature relies mainly on samples defined by specific diseases, many of which are not considered complex in terms of impact on family and resource utilization, making the generalizability of these findings to family caregivers of CMT questionable.

**Sleep in family caregivers of children with disabilities.** In a small body of literature, the sleep in family caregivers of children with physical and developmental disabilities has been described. This group of children may share many of the same diagnoses and clinical
characteristics with CMT, including limited mobility and neurocognitive deficits, but differ in that they are not reliant on medical technology to sustain life and achieve optimal health.

Clinically and statistically significant differences were reported for TST in a cross-sectional cohort study of family caregivers of children with physical disabilities ($n = 178$) compared to family caregivers of typically developing children ($n = 69$; Wright, Tancredi, Yundt, & Larin, 2006). The weekday TST was reported to be 42 minutes less (6.5 [1.3] vs. 7.2 [1.0] hrs, $p < .001$) and average weekend TST a full hour less (6.9 [1.5] vs. 7.9 [1.1] hrs, $p < .001$) in family caregivers of children with physical disabilities compared to those of typically developing children. In the same study, more family caregivers of children with physical disabilities reported sleep disturbances due to the child’s sleep patterns than family caregivers of typically developing children (65% vs. 43%, $p = .002$). Specifically, children’s night awakenings were prevalent, with 66% of the family caregivers of children with physical disabilities compared to 41% ($p < 0.001$) of controls reporting this as a cause of their sleep disturbance. One mother in this study stated, “The sleep pattern for 17 years it to be awakened every 2 hours, all night long (to turn and position our child)” (Wright et al, 2006, pp. 64). This investigation, however, was limited by the use of subjective measures and a low response rate in the family caregivers of both children with physical disabilities (25%) and typically developing children (35%). It was also notable that the participants differed from non-participants. Those with more cognitive impairment experienced by the child were more likely to participate, contributing to a potential non-respondent bias.

The quality of sleep and psychological health reported by family caregivers of children with developmental disabilities was also reported to be poor. Chu and Richdale (2009) described that children’s sleep quality predicted 22% of the variance in mothers’ ($n = 46$) sleep quality as
measured by the PSQI, \( p = < 0.05 \) and that the mother’s sleep quality predicted 34% of the variance in their own depression scores \( p = .001 \). The authors acknowledged, however, that their choices of measurement tools, including a researcher-generated child sleep questionnaire, were major limitations of this study. Missing data was also cited as a limitation, and a return rate was not calculated. Gallagher, Phillips, and Carroll (2010) also investigated sleep and parenting stress in family caregivers of children with developmental disabilities. In this cross-sectional investigation, 67 family caregivers of children with developmental disabilities reported sleep quality to be twice as high (where higher scores are suggestive of poorer sleep quality; 9.9 [3.5] vs. 4.9[2.0], \( p < .001 \)) than 42 family caregivers of typically developing children. In this study, the majority (78% vs. 22%) of family caregivers of children with developmental disabilities met the criterion for poor sleeper (score> 5) on the PSQI. Parenting stress was the most robust predictor of poor sleep quality in parents of children with developmental disabilities, accounting for 30% of the variation even after controlling for both age and body mass index (BMI).

Gallagher et al. (2010) also reported that parents of children with developmental disabilities were more likely to be classified as overweight (BMI \( \geq 25 \)) compared to controls which is notable, given that obesity is a known risk factor for poor sleep quality in laboratory subjects and the population at large (Van Cauter & Knutson, 2008).

Since these studies collectively do not provide adequate detail in terms of the characteristics of the children, who are a heterogeneous group, and their caregiving needs, interpretations of their findings will be limited. Family caregivers of children with disabilities have described similar challenges in terms of some aspects of caregiving that may influence their sleep, but the care they deliver differs from that of family caregivers of CMT in that it may not be as intensive, complex, and around the clock (Kirk & Glendenning, 2004).
Sleep in family caregivers of children who depend on medical technology. It has been reported that the care of a CMT may complicate every aspect of a parent’s life (Carnevale, F., Alexander, E., Davis, M., Rennick, J., & Troini, R., 2006). One of the unique features of life for family caregivers of CMT is the need to be on constant alert for a crisis (McKeever, 1992). In a recent published abstract, sleep in family caregivers of CMT was characterized as a state of what the authors described as “chronobiotic chaos,” with extended napping in the day and short, fragmented sleep at night (Meltzer & Avis, 2011). It has been identified that there are no consistently applied assessments and few interventions targeting caregiver stress, health, and daytime function. Closing the gap between what is known and what may influence the sleep and respite experiences of family caregivers of CMT represents a great opportunity for future practice and research with family caregivers of CMT.

Sleep in family caregivers of CMT was the focus of a systematic review of the peer-reviewed literature completed by this investigator (Keilty, Cohen, Ho, Spalding, & Stremler, 2014, submitted for publication). The main aim of the systematic review was to determine what was known about the quality and quantity of sleep in family caregivers of CMT.

The systematic review process followed recommendations for evidence based practice reviews (Moher, Liberati, Tetzlaff, & Altman, 2009) and included the a priori establishment of eligibility criteria for the review:

(a) full-text reports published in peer-reviewed English-language journals;

(b) study sample included family caregivers (e.g. mother, father, grandparent, foster parent);

(c) study included report of a CMT;

(d) study reported on sleep or sleep disturbance as an outcome; and,
(e) study design was any type other than a case report.

The review was conducted by the study investigator under the guidance of an expert librarian located at a paediatric academic health science center. Databases were searched for articles indexed as of December 19th, 2013 and included: OvidSP MEDLINE (from 1946), EMBASE (from 1980), and PsycINFO (from 1986), and EBSCOHost CINAHL (from 1983). A combination of controlled vocabulary (MeSH, EMBASE, PsycINFO, and CINAHL descriptors) and free text terms were used to generate a high sensitivity search that would locate any study reporting on sleep outcomes in family caregivers of children. In addition, the search strategy included a hand search of relevant citations from article bibliographies was completed (see Appendix C).

Each database was searched to perform initial screening of articles for relevance using study titles and abstracts. Two independent reviewers (study investigator and Research Assistant [RA]) reviewed full texts for all studies that reported sleep outcomes in family caregivers of technology dependent children. Upon retrieval, the reference lists of individual studies that met study criteria were examined for additional relevant references. Experts in the field were contacted to locate additional published studies (see Figure 1).
Data were extracted from each of the study reports by two independent reviewers (study investigator and Research Assistant) and placed in standardized evidence tables. Characteristics of the studies documented in the table included: author, year, target population including number of family caregivers and nature of child’s technology dependence, study design, methods, measurement and results related to sleep and/or sleep deprivation. Tabled studies were compared between the two reviewers and any discrepancies analyzed, errors or omissions corrected, and
consensus achieved among the authorship team where there was disagreement between the reviewers.

Thirteen studies with varied designs were included in the review (see Appendix D). All of the studies used observational designs, and neither randomized controlled trials nor intervention studies were located. The majority of the studies used cross-sectional designs (Andrews & Nielson, 1988; Evans, Holden, & MacDonald, 2006; Evans, MacDonald, Daly, Hopkins, & Holden, 2007; Evans et al., 2010; Feeley et al., 2013; Hopkins et al., 2009; Meltzer, Boroughs, & Downes, 2010; Meltzer & Mindell, 2006), employing mainly surveys, with the remaining studies using mixed methods that combined the use of qualitative interviews and surveys (Heaton, Noyes, Sloper, & Shah, 2006; Mednick, Cogen, & Streisand, 2004; Montagnino & Mauricio, 2004; Read et al., 2010). A single study used a distinctly qualitative design that employed in-depth interviews (Sullivan-Bolyai, Knafl, Tamborlane, & Grey, 2004). Only one study included a control group for comparative analysis (Meltzer & Mindell, 2006). Sample characteristics of both the family caregivers and CMT were variably described (see Appendix E).

**Quantity and quality of sleep in family caregivers of CMT.**

The studies in this review relied entirely on self-report measures of both sleep quantity and quality. The related findings are described in the next section.

**Sleep quantity.** Participants in all of the studies described general sleep disturbances, with reported rates ranging from 51% (Hopkins et al., 2009) to 100% (Evans et al., 2007; Heaton et al., 2006). The quantity of sleep in hours achieved in a 24-hour period, or TST for the family caregivers was reported in only three of the studies. In a cross-sectional study, mothers of ventilator-dependent children ($n = 29$) were found on self-report (i.e., 24-Hour Sleep Patterns...
Inventory) to get one hour less sleep per night and wake up 45 minutes earlier than mothers of otherwise healthy children (6.31 [-1.2] vs. 7.34 [0.9] hrs, \( p = < .007 \); Meltzer & Mindell, 2006). In two more studies, family caregivers of CMT reported getting just below six hours of sleep on average per night (Feeley et al., 2014; Meltzer et al., 2010). It is notable that in laboratory studies with healthy adults, fewer than six hours of sleep has been found to be the critical cut-off for demonstrating negative effects on daytime performance (Durmer & Dinges, 2005).

**Sleep quality.** In family caregivers of children with neuromuscular disorders receiving noninvasive ventilation at home, sleep quality (measured by the PSQI) was poor in 70% of the participants (Read et al., 2010). Similar results were found in family caregivers of infants on supplemental home oxygen, with 78% scoring above the cut-off for criteria (>5) for poor sleepers (7.6 [3.7]; Feeley et al., 2014). The remaining studies used open-ended interviews or single questions to elicit the perceptions of the family caregivers about their quality of sleep. In a series of three observational studies, family caregivers of children using enteral feeding devices reported frequent sleep disturbances (e.g., getting up to check on child) on a weekly basis (58–100% of the time), with most participants reporting nightly disturbances (Evans et al., 2007; Evans et al., 2010; Feeley et al., 2014). Heaton et al. (2006) conducted in-depth qualitative interviews that detailed the responses to questions targeting sleep quality in 36 families of CMT. Four themes emerged that characterized sleep disruption (a component of sleep quality that generally refers to perceived inadequacy of sleep because it is too short and/or overly interrupted) in these family caregivers: 1) incidence of sleep disruption; 2) reasons for sleep disruption; 3) help with care overnight; and 4) effects of sleep disruption. Sixty-one percent of these families described poor sleep quality at least two nights per week, and one mother reported staying awake for multiple nights in a row to suction her daughter’s airway when there was no
night nursing available (Heaton et al., 2006). In another qualitative account, Read et al. (2010) described that family caregivers experienced poor sleep quality associated with negative emotional symptoms and burden on family function and relationships. In family caregivers of children with tracheostomy and gastrostomy, fatigue was identified as a theme owing to poor sleep quality (Montagnino & Mauricio, 2004).

Interestingly, and in contrast to other studies, family caregivers of children with Type 1 Diabetes have reported that the use of home insulin pumps has had a positive influence on their sleep quality (Mednick et al., 2004; Sullivan-Bolyai et al., 2004). Mednick et al. (2004) used the Insulin Pump Therapy Satisfaction Questionnaire (including items with a five-point Likert scale on which higher scores indicate greater satisfaction) to solicit responses from family caregivers (n=22) subsequent to starting their child on home insulin therapy, and they reported that the overall greatest improvement experienced by the family caregivers in the pilot study was "flexibility of sleep schedules" (4.68[0.57]). In a qualitative study, parents described that with the introduction of an insulin pump to manage their child’s diabetes, their “sleep returned to normal” (Sullivan-Bolyai et al., 2004, p. 321).

Correlates of Sleep Disruption in Family Caregivers

A number of debilitating conditions have been found to be associated with sleep disturbance in the general population and among new mothers and fathers (n = 47; Gay, Lee, & Lee, 2004). This section describes in detail what has been reported about correlates of sleep disruption in family caregivers of CMT (i.e., health-related quality of life, depression, daytime sleepiness, and fatigue) and also provides an overview of what is known about these relationships in other groups of family caregivers.

Health-related quality of life. Family caregivers, including those of children with
medical complexity, are widely described as at risk for poor HRQoL (Canam & Acorn, 1999; Rosenbaum, 2011). In the broad sleep literature, it is suggested that HRQoL may negatively influence sleep or vice versa (Szentkiralyi, Madarasz & Novak, 2009). In family caregivers of older adults with dementia ($n = 39$), significant relationships have been reported between sleep quality and quality of life outcomes, particularly with caregiver physical and emotional role limitations (Lee, 2008). Similar findings have been reported in studies of family caregivers of adults with multiple sclerosis (McKeown, Porter-Armstrong, & Baxter, 2003), schizophrenia (Caqueo-Urízar, Gutiérrez-Maldonado, & Miranda-Castillo, 2009), and other chronic health conditions.

In studies of children with medical complexity, interventions have been aimed at improving upon the HRQoL of family caregivers deemed at risk for caregiver burden and negative health outcomes (Cohen et al., 2010). Cottrell and Khan (2005) described the relationship between sleep and health in family caregivers of children with epilepsy. In this cross-sectional investigation, the RAND Health Survey (Stewart, Sherbourne, & Hays, 1992) was used to detect parental health status at the time of the assessment, and overall, the majority of parents (i.e., 60%) perceived their health to be related to their subjective sleep quality.

Children’s sleep quality was associated with HRQoL in family caregivers of CMT, as measured using the Medical Outcomes Study 36-Item Short Form (SF-36™; Read et al., 2010). In a small sample of family caregivers of youth with neuromuscular conditions using non-invasive ventilation at home ($n = 10$), subjective sleep quality was positively related to the domains of energy and vitality ($r = .68, p = .03$), emotional well being ($r = .82, p = .007$), and general health ($r = .69, p < .02$; Read et al., 2010). No intervention studies have examined HRQoL and sleep disturbances in family caregivers of children with chronic illness or among
those with CMT.

**Depression.** The prevalence rate of depression in family caregivers is not known, but it is thought to be close to 10% (Mahoney, Regan, Katona, & Livingston, 2005), exceeding the one-year prevalence rate of major depressive disorder in Canada of 3.2%–4.6% (Canadian Psychiatric Society, 2001). Regarded as one of the most common downsides of caregiving, depression has been the focus of numerous systematic reviews of psychological outcomes in family caregivers of older adults (Han & Haley, 1999; Selwood, Johnston, Katona, Lykestos, & Livingston, 2007).

In studies of family caregivers of children with asthma, sleep disturbances were associated with elevated rates of depression (Yilmaz et al., 2008), and when combined, poor sleep quality and depression have impeded family caregiver motivation and performance with resultant increased emergency room use for the care recipient (Yuksel et al., 2007) and lost days of paid work for the caregiver (Diette et al., 2000).

In family caregivers of CMT, increased depressive symptoms were associated with poorer sleep quality, as measured using the Centre for Epidemiological Studies-Depression Scale (CES-D; Meltzer et al., 2010; Meltzer & Mindell, 2006) and the Hospital Anxiety and Depression Scale (Read et al., 2010). It is compelling to think that if appropriate interventions were targeted to treat sleep disturbance in family caregivers, their risk for depression may be significantly lessened. Moreover, there are promising interventions for depression in family caregiver adults (one-on-one counseling) and other at-risk populations (e.g., cognitive behavioral therapy, light therapy) that, if trialed in family caregivers with depressive symptomatology, may reduce sleep disturbances in this vulnerable group.

**Sleepiness.** Excessive daytime sleepiness is reported by approximately 20% of the adult
population (Johnson, 1991; National Sleep Foundation, 2005). Sleepiness is a basic physiological need state that is directly related to the quality and quantity of nocturnal sleep (Carskadon & Dement, 2005). Its presence and intensity can be inferred by how rapidly sleep onset occurs (sleep latency), how easily or often sleep is disrupted (sleep fragmentation), and how long sleep lasts (total sleep time). In laboratory subjects, sleepiness has been described as one of the consequences of modest to severe sleep deprivation and is linked to a decline in daytime performance on psychomotor and other simulated tasks (Durmer & Dinges, 2005).

Elevated rates of daytime sleepiness were measured among family caregivers of CMT with fewer hours of night nursing per week (Meltzer et al., 2010). It is worrisome that family caregivers may find it difficult to consistently and accurately perform routine care tasks, many of which are described by family caregivers as mundane and repetitive, as a result of excessive daytime sleepiness. Concerns about caregiver health and child safety emerge in discussions about this relationship among clinicians and parents alike. Shift-work literature and other sources that have examined the effects of sleep deprivation suggest that excessive sleepiness may also influence decrements in performance, impede the ability to attend to routine and complex tasks, and lessen HRQoL. It is not known if these factors negatively impact the capacity of family caregivers to carry out their important roles and responsibilities on a day-in-day-out basis.

**Fatigue.** When sleep deprived, the daytime function of caregivers is characterized by complaints of fatigue (Sato et al., 2002), frustration, anger, and exhaustion (Lawson, Lewis-Jones, Finlay, Reid, & Owens, 1998). In family caregivers of children with atopic dermatitis, improved nocturnal symptoms (i.e., child less itchy) were associated with lower rates of self-reported fatigue (from 50% to 44%; Arvola, Tahvanainen, & Isolauri, 2000). Moreover, in parents of children with cancer ($n = 64$), sleep disturbance and fatigue are reported as highly
correlated \( r = .56, p < .001; \) Zupanec et al., 2010).

Among the studies of family caregivers of CMT, Meltzer and Mindell (2006) reported that increased fatigue, measured using the Iowa Fatigue Scale, was associated with heightened depression and poorer sleep quality. These findings were not replicated in a second study by Meltzer et al. (2010), in which the association between sleep quality and fatigue in family caregivers of CMT did not reach statistical or clinical significance. Based on these data, it is thus unclear to what extent family caregivers of CMT experience fatigue and how it relates to sleep disturbance and/or other features of their caregiving role (e.g., total number of homecare hours per week).

**Family function.** Family function has also been assessed in studies of sleep in family caregivers of children with medical complexity. Marital strain was measured using the Dyadic Adjustment Scale (Spanier & Thompson, 1982) to assess the quality of adjustment to marriage or similar adult relationships in a study of 50 parents of children with epilepsy (Cottrell & Khan, 2005). In this cross-sectional study, increased nocturnal awakening was inversely related to sleep quality \( r = -0.38, p = .01 \) and marital satisfaction \( r = -0.28, p < .05 \).

Among studies of family caregivers of CMT, Read et al. (2010) investigated the burden and impact of neuromuscular conditions on family caregivers of youth requiring non-invasive ventilation. In this study, the Family Burden Scale (Pai & Kapur, 1981) was used as a global subjective measure of burden; financial impact; effect on family routines, leisure, and interactions; and effect on the physical and mental health of family members. This study also used the Family Assessment Device (Miller, Epstein, Bishop, & Keitner, 1985) to assess individual perceptions of several dimensions of family functioning, including problem solving, communication, and general functioning. In this small sample of 10 family caregivers sleep
quality was measured using the PSQI, and findings suggested an association between it, scores on the Family Burden Scale ($r = .60, p = .04$), and items on the Family Assessment Device, including disruption to family interactions ($r = .73, p = .01$) and impact on mental health ($r = .70, p = .02$). Finally, in a cross-sectional study using different measures, Montagnino and Mauricio (2004) also examined family function in relationship to sleep in family caregivers. In this modest sample size, and in contrast to the findings of Read et al. (2010), they did not find significance in the association between sleep quality and family function in 50 family caregivers of children requiring tracheostomy and gastrostomy. The authors of each of the studies in family caregivers of CMT acknowledged a number of limitations, importantly including those related to measurement using instruments not validated for use with family caregivers of CMT.

Overall, findings from the systemic review of studies of sleep in family caregivers of CMT and the related literature suggest that more evidence is needed to fully understand which health and related outcomes are most closely linked to sleep disruption in family caregivers of CMT. For the purposes of this investigation, the HRQoL, depression, daytime sleepiness, and fatigue of family caregivers will be examined, but not family function, due to a lack of an appropriate measure. These correlates of sleep disturbance have been chosen, as they have been examined the most frequently in previous studies, can be measured using validated tools with strong psychometric properties, and have demonstrated significance in terms of the magnitude of their association with subjective sleep measures. Knowledge of these relationships in the context of objective sleep data is expected to inform practice, policy, and development of sleep interventions in future studies of family caregivers.
Factors Influencing Sleep in Family Caregivers

Sleep disturbance in family caregivers may be influenced by multiple factors. The literature describing the relationship between sleep and caregiver, child, and environmental factors in family caregivers (across the ages and continuum of care) is summarized below. Owing to study methods, the directionality between sleep and these factors cannot be confirmed, but the nature of their relevance and relationship to sleep disturbance in family caregivers of CMT merits consideration.

Caregiver factors.

Sociodemographics. In population-based studies, it has been described that advancing age and female gender are risk factors for sleep disturbances (Wilcox & King, 1999). Women generally report poorer sleep quality compared to their male counterparts (Vitello, Larsen, & Moe, 2004), and the effects of sleep disruption on female caregivers of adults with chronic disease and disability appear to be more pronounced than in men (Atienza, Henderson, Wilcox, & King, 2001; Yee & Schulz, 2000). This is notable, as mothers provide the vast majority of care for children with chronic illness and technology dependence (McKeever, 1991).

Physical health, including the presence of chronic conditions, and risk factors for sleep disturbance, such as high BMI (Krueger & Friedman, 2009), have not been consistently reported in studies of family caregivers of CMT. However, as described by Gallagher et al. (2010), this group may share the propensity for poor health, with other family caregivers, owing to a lack of time for self-care and exercise. Population-based data (Krueger & Friedman, 2009) suggests that other sociodemographic features, including marital status, employment status, socioeconomic status, level of education, race/ethnicity, number of members in a household, and parity influence sleep and thus are important features to explore in family caregivers of CMT.
**Sleep hygiene.** Sleep hygiene refers to the habits and practices that influence sleep quantity and quality (Stepanski & Wyatt, 2003). In family caregivers, irregular sleep/wake cycles and longer periods of wakefulness may be commonplace (Berger et al., 2005). Practices related to bedtime, nighttime rituals, and disruptions in sleep have not been prospectively evaluated in family caregivers of CMT. In meeting the needs of their CMT, family caregivers may experience similar risk factors as shift-workers, who often experience sleep disturbances and are known to acquire sleep disorders due to their lifestyle and occupational demands that may negatively influence sleep hygiene practices (Committee on Sleep Medicine and Research, 2006). This factor merits investigation in family caregivers of CMT, as there are well-developed (i.e., feasible and acceptable in other caregiver populations) behaviorally based interventions designed to improve sleep hygiene that may be tested in future studies and adopted in clinical practice with family caregivers of CMT.

**Vigilance.** CMT rely on the use of medical technologies with the potential to fail during the night and, if so, would likely result in a negative consequence for the child’s health (e.g., mechanical ventilator disconnect or tracheostomy tube decannulation). Family caregivers of CMT have described difficulty getting to sleep and staying asleep, owing to worry and being unable to “turn off the switch” (i.e., their minds) at night, even in the presence of homecare personnel (Heaton, et al., 2006; Horner, 1997; Reid & Lewis-Jones, 1995; Sullivan-Bolyai et al., 2003). “Constant vigilance” that impairs sleep quality has emerged as a construct in a naturalistic inquiry of family caregivers of children with chronic illness (Sullivan-Bolyai et al., 2003). In cross-sectional studies of family caregivers of children with chronic illnesses (Boman, Lindahl, & Bjork, 2003; Cottrell & Khan, 2005; Ferrell et al., 1994) and technology dependence (Meltzer & Mindell, 2006), it is reported that intrusive thoughts and stress about the child’s illness have
interfered with caregiver sleep. The reasons for awakenings in family caregivers of CMT have not been explored prospectively, although they are frequently discussed in clinical encounters. The relationship between vigilance, worry, and arousal (i.e., cognitive and somatic) for sleep disruption merits investigation in future studies, as knowledge of these contributors to sleep disruption may help inform targeted cognitive behavioral interventions (e.g., progressive muscle relaxation, negative thought stopping, reframing; Nicole et al., 2012; Morin, 2002).

**Child factors.**

**Technology use.** The influence of medical technology use on sleep in family caregivers of CMT has been described in qualitative accounts. Heaton et al. (2006) reported that the need to check devices and manage equipment problems were reasons for sleep disturbance in 62% ($n = 34$) of the mothers of CMT. Nocturnal awakenings, due to the need to check devices and manage equipment problems, have also been reported in family caregivers of CMT (Evans et al., 2006; Evans et al., 2007; Heaton et al., 2006). Read et al. (2010) have reported that family caregivers of children requiring long-term mechanical ventilation cited the need to re-apply face-masks as a reason for getting out of bed, thus contributing to their self-reported poor sleep quality.

Most existing studies have categorized CMT according to a 4-point scale, first proposed by the Office of Technology (Wagner et al., 1987) and since revised by Feudtner et al. (2005), that delineates different types of technology use. This typology, however, is based on outdated assumptions, including the relative ranking of technology dependence based on the type of medical technology or device used, and its use has not adequately accounted for many other important features in determining a child’s level of technology dependence. In their investigation of families’ experiences of caring for a CMT from a temporal perspective, Heaton et al. (2005) concluded that the patterns of use of the technology, including the amount in hours and time of
day, were also important features of the family caregiver experience. In this qualitative study, 46 parents of CMT described that in particular, nocturnal use of medical technology influenced their daily rhythms and overall sleep quality. Homecare nursing provider agencies and policy makers indicate that improved understanding of the relationship between family caregiver sleep disturbance and specific features of a child’s technology may help inform policy and practices in allocation of homecare nursing and related resources (A. Cooper, personal communication, September 9, 2011).

**Nocturnal care needs.** The childcare needs of CMT are extraordinary. Throughout the night, family caregivers may be required to observe, assess, and intervene to ensure the comfort and safety of their CMT. In qualitative reports, it has been described that family caregivers of CMT have stayed up multiple nights in a row to observe their children, carry out routines (e.g.; turning and positioning), and tend to exceptional or unscheduled health care needs (e.g., management of symptoms; Heaton et al., 2005; Heaton et al., 2006; Horner, 1997). Family caregivers of ventilator-dependent children have reported nighttime awakenings related to caregiving tasks more often than family caregivers of normally developing children (1.39 [0.09] vs 0.85 [0.7] \( p = .04 \); Meltzer & Mindell, 2006). There is a need to understand the influence of routine and exceptional nocturnal care needs of CMT related to reasons for awakenings and other features of sleep disruption in family caregivers of CMT. Some of these nocturnal care needs may be predictable and/or preventable (e.g., timing of treatments) and/or delegated to others (e.g., if the right personnel is available) in order to limit their influence on sleep disruption in family caregivers of CMT.

**Child sleep.** In a population-based investigation of typical adults, it was reported that parents associated the quality of their sleep with that of their child’s sleep (Johnson, 1991). This
relationship is reinforced by anecdotal accounts from parents of both healthy and chronically ill children. Nonetheless, there are few studies that have investigated this relationship and none that have used objective sleep measurement in parents of children beyond infancy. In studies employing self-report sleep measures in family caregivers of chronically ill children, the child’s sleep quality, including number of nocturnal awakenings, was a predictor of maternal sleep quality and daytime sleepiness (Boergers, Hart, Owens, Streisand, & Spirito, 2007; Meltzer & Mindell, 2007). Chu and Richdale (2009) reported that subjective sleep problems in children with developmental disabilities significantly predicted maternal sleep quality ($r = .45, p < .01$). Similarly, in childhood leukemia research, Zupanec et al. (2010) have reported that parent self-reported sleep quality and child sleep quality were highly correlated ($r = .41, p < .001$). These family caregivers cited children’s nighttime awakenings, due most commonly to the need for help with bathroom and other activities of living, as the reason for their sleep disturbances.

Qualitative themes have emerged related to the influence of the child’s sleep quality on sleep in family caregivers of CMT (Heaton et al., 2006; Hopkins et al., 2009), but this potentially modifiable risk factor has not been investigated using a validated measure in any studies of sleep in this population.

Nonetheless, Meltzer & Mindell (2007) have suggested that, based on the results of their cross sectional study, there is a need for future research examining the relationship between child sleep disturbances and family caregiver sleep disturbances and daytime functioning. They highlight the importance of screening for and treating paediatric sleep disruptions in paediatric practice and family caregiver research. Clinical experience with normally developing and sleep-disordered children suggests that select interventions (e.g., behaviorally based interventions and pharmacological treatments) may offer some benefits in promoting sleep quality in CMT.
Environmental factors. Given that the classic definition of sleep is “a state of unresponsiveness to the environment” (Carskadon, 2010, p. 7 in Kryger, 2011), it is not difficult for clinicians and researchers alike to recognize that the home environment may be an influential factor on sleep in family caregivers of CMT. Family caregivers of CMT often make changes to their homes in order to accommodate the needs of their children, such as placing their child’s hospital bed and related medical equipment in the main living/dining room (Bradley, Parette, & Van Bierliet, 1995). Along with changes to the physical layout of the home, the intrusion of homecare technologies and personnel may result in the home feeling more like an ICU (Smith, 1991) than a private place of comfort and security (Arras & Dubler, 1995).

Use of homecare personnel. Use of the term homecare in this study refers to the presence of nursing care services delivered either in the home, out of the home, or both, targeting the care of children with medical complexity, including those who depend on medical technologies. Routine care practices in Ontario and most geographical jurisdictions in Western Canada include the allocation of publicly funded homecare personnel for CMT. This support is widely thought to promote respite in family caregivers of CMT, but the evidence for the utility of homecare help is equivocal. In qualitative studies, family caregivers of CMT have described that they “can’t live with or without” these supports (Heaton et al., 2006) and strongly advocated for more homecare supports in aid of their sleep and quality of life. However, in a cross-sectional examination of the influence of homecare nursing on family caregiver sleep, it was described that family caregivers of CMT with regular homecare nursing coverage experienced significant improvements in only one sleep variable (Meltzer et al., 2010). By post hoc analysis (Tukey’s HSD), the time it took to fall asleep was prolonged in family caregivers with no night nursing (>20 mins) compared to those with 16-48 hours per week of homecare nursing (15 mins, p <.05).
but associations between homecare use with other important sleep variables, namely TST-Nighttime, did not reach significance. Moreover, greater than 50% of the family caregivers in the study conducted by Meltzer et al. (2010) reported being disturbed by noises made by the night nurse that were not related to the child’s care. Findings from this investigation, carried out in the United States, may have limited generalizability to the Canadian health care context, as the number of homecare hours allocated to family caregivers in this study (> 48 hrs and up to 90 hrs on average per week) far surpasses the typical allocation of homecare nursing for family caregivers of children with similar care needs in Canada.

In Ontario, family caregivers of CMT may receive from public sources not more than 43 hours of in-home nursing care from a registered nurse (RN) per week, which is funded by the Ministry of Health and Long-Term Care (MOHLTC; Community Care Access Centre, Home Care Services, 2006). Some families supplement this homecare with funds provided by private insurance companies or small funds provided for enhanced respite through the Ministry of Community and Social Services and administered by the Community Care Access Centres, but hours per family caregiver would seldom total more than 50 hours of nursing care per week (A. Cooper, personal communication, November 13, 2010).

Variable night-nursing hours are reported widely in the literature and locally by family caregivers of CMT, with reports ranging from 0–24 hours of homecare nursing per day. Parents report sleep disruptions owing to a lack of sufficient night nursing due to funding limitations or unfilled shifts (Meltzer et al., 2010). Constant threats of losing night nursing results in self-reported caregiver stress and sleep loss (Kirk, Kahn, & Brouillette, 1998). No studies to date in Canada, however, have investigated the influence of the time of day and the number of funded and filled hours of homecare per week on the sleep, health, or daytime consequences in family
caregivers of CMT. This is despite the recommendation by health policy researchers (Peter et al., 2007) that studies need to capture the differences across jurisdictions regarding paediatric respite use and identify related associations with caregiver outcomes.

Homecare provider agencies are required to provide personnel with specialized training, certification, knowledge, and the ability to meet the needs of CMT. Paediatric homecare nurses have expressed a substantial amount of knowledge about homecare technologies and how to communicate with parents and assess their strengths and needs (McGillis-Hall, Spalding, Booth, Lacroix, & Acton, 2004). Nonetheless, it is reported that family caregivers experience sleep disturbances due to their lack of confidence in the skills and abilities of their night nurses (Andrews & Neilson, 1988; Heaton et al., 2006; Meltzer et al., 2010). While parents desire the support, they also describe that homecare nurses may not have adequate knowledge of their child’s unique care needs and, therefore, may wake them for assistance and clarification (Reeves, Timmons, & Dampier, 2006). No studies have evaluated the influence of level of preparation of homecare personnel on sleep and related outcomes in family caregivers of CMT and yet, in the face of economic constraints, there has been a shift in most jurisdictions towards the use of less expensive, lesser-educated providers.

**Use of monitoring.** In many jurisdictions, the standard of care is to distribute monitors for use in homecare by family caregivers of CMT with chronic and complex respiratory diseases. Since 2007, over 250 oxygen saturation monitors have been funded for paediatric homecare in Ontario (M. Williams, personal communication, March 23, 2010). Cardiorespiratory and apnea monitors are also funded by the MOHLTC and prescribed for homecare in select circumstances. Moreover, family caregivers often purchase commercial monitors (i.e., baby monitors) for use at home to supplement their children’s medical devices. These technologies are intended to relieve
family caregivers of the need to constantly observe their children. Oxygen saturation monitors will alert family caregivers of CMT by alarming in the event of a drop in oxygen saturation associated with a need for intervention and/or to avert a life-threatening cardiorespiratory event. Alerts and alarms are also built into other homecare technologies to signal the true need for routine assessment or the need for trouble-shooting and immediate intervention (e.g., enteral feeding pumps or ventilator alarms). However, if any of these devices fail, the CMT may suffer an adverse health consequence, resulting in hospitalization (Feudtner et al., 2005) or even death (R. Amin, personal communication, June 18, 2011).

On the other hand, the use of medical technology in the home has also been described as problematic. The routine use of apnea monitors has fallen out of favor, since evidence now suggests that they lack efficacy and are negatively associated with sleep disturbance and fatigue in new mothers of children at risk for apnea (Williams et al., 1999). In early studies of children on home ventilation, false monitor alarms were cited by 62% of mothers (Andrews & Nielson, 1988) as keeping them awake at night. These complaints have persisted in caregiver reports even with recent advances in homecare technologies (Heaton et al., 2006; Meltzer & Mindell, 2006). More and more, modern homecare technologies come with the capacity to set alarm limits, thought to enhance patient safety, potentially resulting in more frequent alerts and both “true” and “false” alarms, which may intrude on family caregivers’ sleep.

**Sleeping arrangements.** Sleep location and sleep partners, meaning where (e.g., in one’s own bedroom vs. other rooms) and with whom one sleeps (e.g., sharing a sleep surface with a spouse, children, or extended family), can influence sleep quality and quantity. The most accepted recommendation for sleep location and partners is one that is in a consistent location, on a proper sleep surface, and either alone or with a spouse (Troxel, Robles, Hall, & Buysse,
2007), but this recommendation precludes family caregivers routinely sleeping with their children, a notion often met with resistance among family caregivers who do not share this belief. Family caregivers of children with epilepsy have been reported to room share (68% vs. 29%) and bed share (37% vs. 10%, \( p < .001 \)) more with their children compared to family caregivers of healthy children (Sullivan-Bolyai et al., 2003). Moreover, family caregivers of CMT have described making changes to where they slept, usually towards sleeping in their child’s room and often in their ill child’s bed, in order to better respond to them at night, especially when their children were unwell (Andrews & Neilson, 1998; Evans et al., 2010).

Anecdotal reports from family caregivers and homecare personnel include descriptions of sleeping in various locations and with various partners to accommodate the needs of their CMT and related homecare technology and personnel in the home. In one study, a family caregiver of a CMT gave an account of sleeping on the floor of their child’s bedroom (Andrews & Neilson, 1998). This loss of privacy in the home, especially at night, has been described as a stressor associated with the extraordinary responsibilities of family caregivers of CMT (Kirk, 1998).

**Studies of Sleep Interventions with Family Caregivers**

A review of studies that described sleep interventions in family caregivers of those with exceptional care needs (across the ages, excluding parents of newborns) was carried out by this study investigator (Keilty & Stremler, 2011). After an exhaustive electronic search of the relevant databases, only one paediatric sleep intervention study in family caregivers was located, along with 13 other studies of sleep in the older adult literature, mainly of family caregivers of those with dementia and cancer.

Wiggs and Stores (1998) reported on a behavioral treatment for sleep problems in children with severe learning disabilities. Sleep duration was measured based on self-
sleep onset and wake times (actigraphy data was used to verify the accuracy of the subjective measure). No differences were reported for sleep duration between the intervention and control groups. At baseline mothers in the treatment group (n=15) averaged a sleep time of 6.9 hrs [.9] thus approaching that recommended for healthy adults. Post-intervention, sleep improved for the treatment group at the four-week mark (7.6 [.7] hrs, $p= .012$) but these improvements were not sustained (at 6 weeks). Thus, to date, the intervention literature does not yield results that can be readily applied to clinical care and research with family caregivers of CMT, and it points to the need for a better understanding of the characteristics of sleep disturbance and related factors in family caregivers of CMT.

**Summary of Literature Review**

Although few in number, studies of sleep in family caregivers of CMT have demonstrated consistently that subjective sleep quality in this group is poor. Specifically, family caregivers have described that their sleep is often interrupted and short in duration. These sleep disturbances are suggested to be, at least in part, a consequence of the 24-hour-per-day caregiving responsibility assumed by family caregivers of CMT. Studies have concluded that this may place them at risk of sleep deprivation and fragmentation, which together may threaten their capacity for sustained long-term care of their CMT at home. Moreover, subjective sleep disturbances in family caregivers of CMT have been associated with negative health and related outcomes, including poor HRQoL, depression, excessive daytime sleepiness, and fatigue. Other factors that may influence sleep disturbance in family caregivers of CMT, derived from the caregiver (e.g., sleep hygiene), child (e.g., technology use), and environment (e.g., presence of homecare), have received very little, if any, attention in the related literature.
While beginning to offer some understanding, conclusions from these studies of sleep disturbance and its associations in family caregivers of CMT are somewhat tentative. This is mainly due to limitations in the studies’ choice of measurement, design, and sample selection. The studies have relied solely on subjective measures of sleep that have left them open to threats of social desirability and recall bias. Study designs have lacked comparator groups, in particular other family caregivers (e.g., parents of healthy children), thereby limiting the level of analysis. Descriptions of the samples lacked detail about recruitment and did not describe key sociodemographic and clinical features of the family caregivers and their CMT, thus leaving the studies open to selection bias. Finally, the generalizability of these findings is limited, as studies have only been reported on outside of Canada, in both the United States and the United Kingdom. All of these limitations mean that results of these studies cannot be interpreted with confidence and cannot dependably inform the development of sleep interventions in family caregivers of CMT.

In future studies of sleep disturbance in family caregivers of CMT, the use of objective sleep measurement, specifically actigraphy, supplemented by data from a customized sleep diary, will enhance the reliability of study findings and help inform understanding of both sleep quality and quantity. Prospective evaluation of sleep over multiple nights will ensure that day-to-day variability in sleep routines and rhythms is captured and sleep disturbances resulting from multiple influences can be adequately described. The use of a control group of family caregivers of healthy children will enable analysis that limits influence of the age of the index child, described as a confounder in studies of sleep in adults with healthy children. Detailed information about the sample characteristics will ensure that findings may be critiqued for their relevance in other jurisdictions.
The systematic review of sleep in family caregivers of children with chronic illnesses expanded upon by this author, and the systematic review in family caregivers of CMT, now in publication, suggested that sleep disturbance was due to a number of factors. This assertion merits further investigation, as knowledge of factors that influence sleep disruption may help inform the development and testing of targeted interventions in future research. Moreover, the exploration of modifiable risk factors, using standardized measures when available, and considering the multiple influences of the caregiver, child, and environment may generate knowledge that can be immediately put to use. Health care providers may be able to use this knowledge to screen for known risk factors and, borrowing from the success of other intervention research with family caregivers of older adults, may be able to develop and test strategies to promote sleep and enhance caregiver respite (e.g., behaviorally based interventions). Finally, policy makers may be able to use these data to inform changes to respite services or other child/family health systems that seek to support the capacity of family caregivers and promote sustainable homecare for CMT.

Overall, this review of the literature suggests that (a) subjective sleep quality is poor among family caregivers, including those of CMT; (b) sleep disturbance in family caregivers is associated with negative health and related outcomes; (c) multiple factors derived from the caregiver, child, and environment may influence sleep in family caregivers; and (d) due to a gap in the literature, there is not sufficient evidence to inform the development of a sleep intervention aimed at improving sleep quantity and quality in family caregivers of CMT. However, there is a need for reliable evidence that describes the quantity as well as the quality of sleep in family caregivers of CMT. This characterization of sleep and sleep disturbance in family caregivers of CMT would be enhanced by comparison to family caregivers of healthy same-age children, the
use of a theoretically justified conceptual framework, and measures that have well-established reliability and validity.

**Conceptual Framework**

To compare sleep and its correlates in family caregivers, this study proposed a conceptual framework that examined the influence of factors on sleep and the relationships between sleep, health, and related outcomes in family caregivers of CMT. This model was informed by the theory and understanding of sleep regulation and restoration. It situated sleep as an interaction between both a homeostatic and circadian process (Borbely, 1982) and as one that is necessary for the repair and restoration of the brain and body (Oswald, 1970). Another important tenet of this model was that it suggested that sleep in family caregivers of CMT may be influenced by multiple promoting or inhibiting factors, including (a) caregiver (e.g., sociodemographics or sleep hygiene); (b) child (e.g., sleep quality, technology use); and (c) environmental (e.g., alarms or presence of homecare nurses). The relationships between daytime consequences of sleep disturbances, including depression and HRQoL, were situated as bi-directional, with daytime sleepiness and fatigue positioned as direct consequences of sleep disturbances. This conceptual framework guided this investigation with an aim to characterize the quality and quantity of sleep in family caregivers and identify which factors influenced and were associated with sleep disruption in family caregivers of CMT (see Figure 2).
Figure 2. Conceptual model of sleep disturbance in family caregivers of CMT

**Purposes of Cohort Study**

The primary aim of this prospective cohort study was to compare sleep disturbance in family caregivers of CMT to family caregivers of healthy same-age children. A secondary aim was to describe health and related outcomes of sleep disruption in family caregivers. Other research questions aimed to explore which individual, child, and environmental factors influenced sleep in family caregivers of CMT.

**Research Questions**

The following research questions were answered, drawing from multiple data sources employed across one week (i.e. six days, seven nights).
Primary research questions. $H_A$: Sleep in family caregivers of CMT is of lesser quality and quantity than sleep in family caregivers of controls.

Sources of data for Questions 1–3. The following questions were answered based on analyses of actigraphy data.

1. Across one week, was there a difference between the mean (minutes) in
   a) total sleep time—nocturnal (TST-N), *
   b) total sleep time—daytime (TST-D),
   c) longest stretch of continuous sleep—nocturnal (LSS-N), and
   d) longest stretch of continuous sleep—daytime (LSS-D)

in family caregivers of CMT compared to family caregiver controls?

*denotes primary outcome of interest

2. Was there a difference in the number of sleep deprived nights (< 6 hours per night) across one week in family caregivers of CMT compared to family caregiver controls?

3. Across one week, was there a difference between the mean number of
   a) nocturnal awakenings—objectively measured (NNA-O) and
   b) nocturnal awakenings—self-reported (NNA-S)

in family caregivers of CMT compared to family caregiver controls?

Sources of data for Question 4: The following questions were answered based on analyses of the Pittsburgh Sleep Quality Index (PSQI).

4. Was there a difference in the proportion of family caregivers of CMT that report poor sleep quality ($> 5$ as a cut-off on the PSQI) compared to family caregiver controls?
Secondary research question. $H_a$: Family caregivers of CMT experience lessened health-related quality of life (HRQoL) and heightened depression, daytime sleepiness, and fatigue than family caregivers of controls.

Sources of data for Question 5. The following questions were answered based on analyses of actigraphy, daily sleep diary, and the scores on the Centre for Epidemiological Studies—Depression Scale (CES-D), the Epworth Sleepiness Scale (ESS), the Multidimensional Assessment of Fatigue (MAF), and the SF-12$^{TM}$ Health Survey (SF-12$^{TM}$):

5. Was there a difference in
   a) HRQoL;
   b) depression;
   c) daytime sleepiness; and
   d) fatigue
reported in family caregivers of CMT compared to family caregiver controls?

Secondary analysis of primary question.

Sources of data for Questions 6 and 7. The following questions were answered based on analyses of multiple sources of data collected, including participant case records for all sociodemographic and clinical variables, the Sleep Hygiene Index (SHI), and the Children’s Sleep Habits Questionnaire (CSHQ), along with actigraphy data.

6. Across 1 week, was there a significant relationship with mean
   (a) sleep quantity (TST-N ) and
   (b) sleep fragmentation (NNA-O)
in family caregivers of CMT and the following risk factors?
Caregiver risk factors.

Nominal/ordinal variables. Gender, race, marital status, level of education, employment status, diagnosed chronic health conditions, sociodemographic status (e.g., born in Canada, income level), and vigilance (kept awake or awakened due to intrusive thoughts).

Interval/ratio variables. Age, body mass index (BMI), sleep hygiene (SHI)*, number of children in household, number of adults in household, and duration of caregiving.

Child risk factors.

Nominal/ordinal variables. Gender, neurocognitive delay or impairment.

Interval/ratio variables. Age, sleep problems (CSHQ)*, and amount of technology use (daytime, nighttime).

Environmental risk factors.

Nominal/ordinal variables. Use of monitoring, sleep ecology (i.e., ideal location, surface, partner).

Interval/ratio variables. The number of total homecare hours allocated/funded per week, the total of number of homecare hours used/filled per week, the number of nocturnal (9 p.m.–9 a.m.) homecare hours per week*.

* denotes modifiable risk factors

7. Which combination of modifiable caregiver (i.e., sleep hygiene), child (i.e., sleep problems), and environmental (i.e., nocturnal homecare use) factors and sociodemographic risk factors (i.e., caregiver age, gender, marital status) best predicted

(a) sleep quantity (TST-N) and

(b) sleep fragmentation (NNA-O)

in family caregivers of CMT?
Chapter 3: Methods

This chapter outlines the methods used to conduct this study, including study design, definition of outcomes, variables, measures, study sample, and study procedures, including methods of data collection, data management, data analysis, and risks and benefits of participation. Research ethics approvals were acquired from the Hospital for Sick Children and the University of Toronto to conduct this study.

Study Design

A prospective observational cohort design was used for this study. This study design allowed for characterization of sleep disturbances in family caregivers of CMT and avoided the recall bias inherent in retrospective designs. The inclusion of a control group allowed for comparisons of the family caregivers of CMT to family caregivers of healthy, normally developing children (Hulley, Cummings, Browner, Grady, & Newman, 2007). The study also explored factors associated with sleep disturbance in family caregivers.

Study Outcomes, Variables, and Measurement

The sleep, health, and daytime consequences of sleep disturbance in family caregivers were investigated. The primary outcome of interest in this study was the difference in total sleep time in minutes (TST-N) averaged across a week in family caregivers of CMT compared to family caregivers of healthy children. Conceptual and operational definitions for this and other outcomes of interest that were examined in this group and family caregivers of healthy children follow below.

The measurement of variables in this study was completed using a combination of objective and subjective measures. A description follows of the variables and data sources for the
primary questions, secondary questions (see Table 1), and exploratory research questions (see Table 2).

Table 1

**Study Variables and Data Sources for Primary and Secondary Questions**

<table>
<thead>
<tr>
<th>Construct Variable(s)</th>
<th>Sleep quantity</th>
<th>Sleep quality</th>
<th>Health and related outcomes</th>
</tr>
</thead>
<tbody>
<tr>
<td>TST-N*</td>
<td>Sleep deprivation</td>
<td>Subjective sleep quality</td>
<td>NNA-O</td>
</tr>
<tr>
<td>TST-D</td>
<td>&lt; 6 h TST-N</td>
<td>NNA-self</td>
<td></td>
</tr>
<tr>
<td>LSS-N</td>
<td>N</td>
<td>HRQoL</td>
<td>Depression</td>
</tr>
<tr>
<td>LSS-D</td>
<td></td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

**Data source(s)**

| Actigraph | Actigraph | PSQI | Actigraph | SF-12® | CES-D | MAF | ESS |

*primary outcome

Table 2

**Study Variables and Data Sources for Exploratory Questions**

<table>
<thead>
<tr>
<th>Construct Variable(s)</th>
<th>Caregiver</th>
<th>Child</th>
<th>Environmental</th>
</tr>
</thead>
<tbody>
<tr>
<td>Gender, marital status, race, level of education, employment status, income, health conditions, BMI, children in household, adults in household, duration of caregiving</td>
<td>Sleep hygiene</td>
<td>Child sleep problems</td>
<td>Use of monitoring, hrs homecare approved/wk, hrs homecare used/wk, nighttime homecare use</td>
</tr>
<tr>
<td>Vigilance</td>
<td>Gender, neurocognitive delay or impairment, hrs technology use (day/night)</td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

**Data source(s)**

| Demographics | Sleep diary | SHI | CSHQ | Demographics | Sleep diary | Demographics | Sleep diary |

*primary outcome

**Sleep outcomes.** Sleep is considered a requisite for restoration and realignment of fundamental biological processes, and it may be evaluated in terms of both quantity and quality (Oswald, 1969, 1970). In this study, sleep was characterized using a combination of objective and subjective measures.

**Sleep quantity.** Sleep quantity is the amount of time spent asleep by an individual during a 24-hour period. The amount of sleep acquired during the nocturnal sleep episode is classically
considered the best representation of sleep quantity, as it is most aligned with the natural processes of sleep (Carskadon & Dement, 2005).

Sleep quantity variables in this study were derived from multiple nights of actigraphy collected over a one-week period. Total sleep time—nocturnal (TST-N) served as the primary outcome of this investigation. TST-N was documented as the average amount of sleep in minutes from 9:00 p.m. to 9:00 a.m. experienced by the family caregivers averaged over 7 nights of data collection. Total sleep time—daytime (TST-D) was documented as the average amount of daytime sleep experienced by participants over the 6 days in the study period. The longest sleep session for night (LSS-N) and longest sleep session for day (LSS-D), measured in minutes, was averaged over 6 days and 7 nights.

**Sleep deprivation.** Sleep deprivation is the acute or chronic lack of sufficient sleep. Durmer and Dinges (2005) have described negative effects of sleep deprivation in study participants after a single night of restricted sleep. To create a sleep deprivation variable, nightly TST-N data were transformed into dichotomous data using < 6 hours of sleep per night as a cut-off (Van Dongen & Dinges, 2005). These data were used to generate the mean number of sleep deprived nights (< 6h) over 7 nights of data collection. Night-to-night variability was calculated as the number of nights during the study period per participant that were tabulated to be +/- 30% different than the preceding night.

**Sleep quality.** Sleep quality is a “complex phenomenon that is difficult to define and measure objectively” (Buysse, Reynolds, Monk, Berman, & Kupfer, 1989, p. 194). Poor sleep quality is characterized as tiredness upon waking and throughout the day, not feeling rested and restored upon waking, and frequent awakenings experienced during the night (Harvey, Stinson, Whitaker, Moskovitz, & Virk, 2008). Good sleep quality is associated with a wide range of
positive health related outcomes. In this study, subjective sleep quality was measured along with the number of nocturnal awakenings, an element known to contribute to sleep fragmentation and poor sleep quality.

A sleep quality variable was informed by data from the Pittsburgh Sleep Quality Index (PSQI; Buysse et al., 1989), which categorizes participants as either good or poor sleepers. This dichotomous variable was created by using the recommended cut off score of 5, where a total score of $< 5$ was considered a good sleeper and $\geq 5$ a poor sleeper. The number of nocturnal awakenings (NNA), or brief arousals per night, was averaged over seven nights, using objective and self-report data. These have been documented as two sleep quality variables, including the NNA—objective (NNA-O), informed by actigraphy, and the NNA—self, informed by the sleep diary that was completed on a daily basis. This study has used two data sources to strengthen measurement of the NNA, as arousals are frequently under-estimated when based on self-report alone.

**Sleep measurement.**

**Actigraphy.** Actigraphy uses a wrist-watch like device that provides an objective measurement of sleep–wake parameters derived from movements. It is used in sleep clinical practice and research as a tool to evaluate sleep patterns (defined as the circadian pattern of sleep and wakefulness over multiple sleep cycles) in ambulatory individuals up to 24 hours per day and over a number of nights in a row (Morgenthaler et al., 2007). The actigraph, considered a non-intrusive device, captures data using a piezo-electric linear accelerometer that senses motion. These data are translated into digital counts across 1-minute intervals (i.e., epochs) and stored in internal memory to be analyzed using computer software and algorithm-driven autoscoring programs (Rupp & Balkin, 2011). The use of 1-minute epochs and event markers to
note when subjects go to sleep (“lights out”) and when they wake up (“lights on”) has been found to generate reliable data in healthy subjects (Rupp & Balkin, 2011).

The Standards of Practice Committee of the American Academy of Sleep Medicine has recommended using at least four consecutive 24-hour periods of actigraph recording time to provide an accurate estimate of sleep (Littner, et al., 2003). In this study, night-to-night variability in sleep among family caregivers of CMT was expected, and thus, 6 days and 7 nights of actigraphy data were collected (Stone & Ancoli-Israel, as cited in Kryger, 2010; Sadeh & Acebo, 2002). This duration of data collection was justified since differences in daily routines and the availability of homecare supports might influence sleep differently during the week than on weekends (A. Cooper, personal communication, July 9, 2011). This period of sleep data collection was longer than that previously found to be acceptable in a study of new parents, which used 3 days and 4 nights (Stremler et al., 2006), but 7 nights has been found to be acceptable in studies of family caregivers of older adults (McCurry, LaFazia, Pike, Logdson, & Teri, 2009).

The use of actigraphy is expected to generate data that will discriminate between sleep and wake cycles and will produce accurate results (Hirshkowitz et al., 2010). Polysomnography (PSG), considered the gold standard for sleep measurement, was impractical for this study. PSG requires a sleep laboratory technologist and 12 electrodes attached to the face and scalp for continuous monitoring, precluding its use in studies of family caregivers in their home environments. Congruence between polysomnography and actigraphy indicates adequate validity and reliability when sleep is assessed in healthy young adults, including women of childbearing age (Ancoli-Israel et al., 2003), with 88% agreement between the two methodologies. Actigraphy has also been shown to be valid in clinical studies of adults with select sleep disorders, including
insomnia (Morgenthaler et al., 2007). Actigraphy has had limited use in studies of sleep in family
caregivers, but in those that exist, it has been well accepted by participants. It has been
successfully used in studies of family caregivers of older adults (Carter, 2006; McCurry,
Gibbons, Logson, Vitiello, & Teri, 1995), new mothers (Stremler et al., 2005), and in a pilot
investigation of seven parents of children with cancer living at home (Gedaly-Duff et al., 2006).

The Octagonal Basic Motionlogger® (60 grams, Ambulatory Monitoring Inc., Ardsley,
NY) actigraph was used in this study. Motionlogger® Actigraphs were the first commercially
available devices for ambulatory 24-hour monitoring of activity and sleep patterns. This
technology continues to be widely accepted for use in sleep medicine and research. Study
participants wore the actigraph on their non-dominant arm for the entire study period, removing
it only for periods of bathing. Data capture began in the clinic after completing the consent
procedures and continued until the home visit on day 7. A sleep diary was completed
concurrently to capture data about aspects of the participants’ sleep and wake patterns and
if/when the actigraph was removed. Data analysis was aided by use of the Kole-Cripke scoring
algorithm using the zero crossing mode and set to capture at 1-minute epochs. All actigraphy
data were observed directly before being analyzed, such that any artifact, including for example
episodes of wake that corresponded with removal of the actigraphy, was identified and recoded
(from sleep to wake) appropriately. Interpretation bias was limited, with over 30% of the
actigraphy files scored and comparisons made by 2 different members of the study team (study
investigator and RA). Consultation with the study investigator’s supervisor was carried out when
discrepancies or disagreements in interpretation were found between the study investigator and
RA. Missing data were very infrequent in the study, with substitutions made from reported sleep-
wake activity in the sleep diary for well under < 10% of the calculations from TST-N.
The major limitation of the use of actigraphy is that it generally underestimates sleep onset latency and may overestimate total sleep time (Sadeh & Acebo, 2002). This is due to the inability of the actigraph to consistently discriminate between quiet wakeful periods and sleep. Nonetheless, refinement of actigraph technology, in combination with carefully selected algorithms and closely set event markers, have yielded data that are considered acceptable in the evaluation of sleep–wake patterns, total sleep time, responses to treatment, and select sleep disorders in patient groups and healthy individuals (Morgenthaler et al., 2007).

**The Pittsburgh Sleep Quality Index.** The PSQI is a self-rated questionnaire used in clinical practice and sleep research (Buysse et al., 1989). The PSQI takes less than 5 minutes to complete and includes 19 individual items that are sub-grouped into seven component scores: subjective sleep quality, sleep latency, sleep duration, habitual sleep efficiency, sleep disturbances, use of sleeping medication, and daytime dysfunction. Together, these component scores are summed to provide a global score with a cut off of > 5, demonstrating a diagnostic sensitivity (89.6%) and specificity (86.5%) to distinguish between good and poor sleepers. Psychometric testing of the PSQI demonstrates good internal consistency and construct validity in men and women of various populations (Beck, Schwartz, Towsley, Dudley, & Barsevick, 2004; Buysse et al., 1989; Carpenter & Andrykowski, 1998). The PSQI was originally designed to ask participants to reflect on their sleep quality over the previous one-month interval. In this study, family caregivers were asked to reflect on sleep quality over the previous week (see Appendix D). This is similar to the use of the PSQI in studies of family caregivers of older adults (Ancoli-Israel et al., 2005; Carter, 2006; King et al., 2002; Korn et al., 2009; McCurry et al., 2005; McCurry et al., 1998; Lee, Morgan & Lindesay, 2007; Rose et al., 2009).
**Daily sleep diary.** Daily sleep diaries are used to capture the subjective responses of participants in sleep research and clinical practice. Compared to actigraphy, their use results in systematic overestimation of total sleep time in healthy subjects and underestimation of total sleep time in insomniacs. Sleep diaries are prone to missing or inaccurate data, due to the tendency of subjects to back-fill data and experience diary fatigue. It was expected that family caregivers in this study might find it challenging to consistently integrate the completion of a sleep diary into their daily routines, due to their caregiving demands and the resultant constraints on their time. Thus, the sleep diary was used to supplement and contextualize objective sleep data and support the interpretation of actigraphic data, but it was not the primary source of sleep data (see Appendix E).

Participants were asked to record the following in their sleep diaries: sleep times, wake times, and events that occurred that might have affected sleep or actigraphy recording. For example, if a family caregiver forgot to press the event marker on the actigraph at bedtime, the sleep diary was used to identify the correct sleep time. Periods of complete inactivity, such as when an actigraph was removed (e.g., for bathing), represented artifacts that were recoded to avoid being analyzed as sleep time by the analysis software. Thus, when sleep diary and actigraphy data indicated that artifacts were present, objective data were recoded as appropriate using the Action4 actigraphy analysis software.

It is recommended that sleep diaries be customized for family caregivers to gather the most relevant information and lessen participant burden (Carter et al., 2009). Therefore, the daily sleep diary created for this study was based on standardized tools available in the literature and sleep diaries previously customized for use in sleep investigations with new parents (Stremler et al., 2006). The sleep diary asked family caregivers about their choice of sleep surface, sleep
location, and bed-sharing/partners. It also asked if any nighttime help to care for their child was received and for details of the reasons for being awake and for getting up the previous night (see Appendix F). Additional information was collected from the family caregivers of CMT regarding their child’s technology use and included additional items under the response items of nighttime help (e.g., from paid homecare personnel), reasons for being kept awake (e.g., noises from homecare personnel), and reasons for getting up (e.g., responding to equipment alarms; see Appendix G). Participants completed the sleep diary for 6 consecutive days and 7 nights, and it was expected to take less than 15 minutes per day to complete.

Health outcomes and daytime consequences. This study investigated the HRQoL, depression, daytime sleepiness, and fatigue in family caregivers. A description of the measures used for these sleep disturbances and their outcomes is below.

Health related quality of life. On an individual level, HRQoL can be viewed as a psychological construct that describes the physical, mental, social, psychological, and functional aspects of well-being from the patient’s perspective. This definition stresses the multidimensionality of the health-related quality of life concept as well as the relevance of patients’ perspectives (Bullinger, 1991).

The SF-12™ Health Survey is a 12-item questionnaire adapted from the SF-36™ Health Survey (Version 1; Ware, Kosinski, & Keller, 1996). Items address physical functioning, role limitations because of physical health problems, bodily pain, general health perceptions, vitality (energy/fatigue), social functioning, role limitations because of emotional problems, and general mental health (psychological distress and psychological well-being). Scores range from 0–100, with 0 indicating the lowest level of health and 100 indicating excellent health. The SF-12™ is used in clinical studies that aim to monitor overall physical and mental health outcomes while
using a tool that is the least burdensome on participants. In instrument testing, compared to the widely accepted SF-36™, the SF-12™ has demonstrated good test–retest reliability (0.76–0.89) and construct validity (0.95–0.96; Ware et al., 1996). The SF-12™ was used in this study, with the necessary authorization for its use in place, to describe the HRQoL in family caregivers and allow for a comparison of scores between the two study groups. It was expected to take less than 5 minutes to complete.

**Depression.** The Canadian Psychiatric Society (2001) defines depression as either depressed mood or loss of interest or pleasure associated with clinically significant distress or impairment in social, occupational, or other important areas of functioning. Symptoms can range in magnitude from a mild subclinical “blue mood” state and general feelings of hopelessness to a major depressive disorder (Frank-Stromberg & Olsen, 2004).

The Centre for Epidemiologic Studies Depression Scale (CES-D Scale) is a short self-report scale with 20 items intended to measure the presence of depressive symptomatology, with scores ranging from 0–60, in which higher scores indicate the presence of more symptomatology (Radloff, 1977). The scale is not intended for clinical diagnosis but for exploring the relationship between depression and other variables across population sub-groups. The CES-D was used in this study to ask participants to reflect over the past week and indicate on a 4-point range from rarely or none of the time to most or all of the time if they had experienced any of the symptoms described. About 20% of the general population is expected to score ≥ 16, suggestive of a clinically significant level of psychological distress. The CES-D has been tested in the general population and with depressed patients and demonstrates high internal consistency ($\alpha=0.85–0.90$), moderate test–retest reliability ($\alpha=0.45–0.70$), and good convergent validity with other depression rating scales (Radloff, 1977). It has also been found to be acceptable for use by
participants in previous studies of family caregiver sleep and depression (Carter, 2006; Carter et al., 2009; Korn et al., 2009; McCurry, Logsdon, & Teri, 1996) and has consistently shown adequate internal consistency (α > .86) in studies of caregivers (Carter & Chang, 2000; McCurry & Teri, 1995). It was expected to take less than 5 minutes to complete. Per the approved ethics protocol, all participants in the study that scored above the cut-off for potentially serious depressive symptoms were called by the study investigator, who is also a clinician, informed of their score on the CES-D, and directed to follow up with their health care provider.

**Sleepiness.** Sleepiness is a subjective feeling of decreased ability to maintain wakefulness or an increased likelihood of falling asleep in inappropriate situations that may interfere with the activities of daily living (Hirshkowitz et al., 2010; Johns, 2000). Excessive daytime sleepiness can be viewed as either a state (at this moment in time) or trait (over time; Weaver, 2001). It is a subjective experience that is, in part, a consequence of inadequate sleep quantity and poor sleep quality. It has been linked to poor performance in psychomotor and neurocognitive testing with healthy subjects and with those experiencing sleep disorders in clinical populations (Schneider, Fulda, & Schultz, 2004).

The Epworth Sleepiness Scale (ESS) is a simple-to-use, self-administered eight-item questionnaire (total score range 0–24) intended to measure daytime sleepiness in adults (Johns, 1991). The subject is asked to reflect on recent times and rate, on a scale of 0–3, the chance that he/she would have dozed in eight specific situations that are commonly encountered in daily life. About 10%–20% of the general population has ESS scores > 10 (Johns & Hocking, 1997). Elevated scores (> 10) may be suggestive, but not diagnostic, of clinically significant sleep disturbances that would benefit from further assessment and intervention.
There is good evidence for the reliability and validity of total ESS scores as a measure of sleepiness (Johns, 1992). ESS scores differ between normal subjects and those with conditions known to increase sleepiness (Johns, 1991, 1993); the ESS is also sensitive to treatment changes (Johns, 1993; Hardinge, Pitson, & Stradling, 1995). ESS scores do not differ significantly between normal men and women (Johns & Hocking, 1997), nor do they change much with age. The use of the ESS has been successful in previous investigations of sleep and sleepiness in family caregivers (McCurry et al., 2005; Morgan et al., 2007) and was used in this study to measure excessive daytime sleepiness in family caregivers over the previous week. It was expected to take less than 5 minutes to complete.

**Fatigue.** The symptom of fatigue is inherently subjective and may be described as state of weariness. If chronic, the perception of fatigue increases and results in diminished energy and mental capacity (Lee, Hicks, & Nino-Murcia, 1991). Fatigue may be described as a symptom that limits participation in activities because of an imbalance in the availability, utilization, or restoration of resources (physical and psychological) needed to perform the activity (Pierce & Press, 1999).

Participants in this study completed the Multidimensional Assessment of Fatigue (MAF) scale, a 16-item measure of four dimensions of fatigue: severity, distress, degree of interference in activities of daily living, and timing. The MAF generates a Global Fatigue Score ranging from 1 (*no fatigue*) to 50 (*severe fatigue*). It has demonstrated good internal consistency and good construct validity (Belza, 1995; Belza, Henke, Yelin, Epstein, & Gilliss, 1993) and has been used with healthy men and women, as well as in populations with chronic and acute disease (Belza, 1995; Belza, Henke, Yelin, Epstein, & Gilliss, 1993, Belza et al., 2001; Fairbrother, Hutton, Stoll, Hall, & Kluka, 2008). The MAF has also been acceptable for use in a study that described
the amount of fatigue self-reported by mothers of children on home apnea monitors compared to mothers of healthy newborns at 1 week and 1 month post-discharge from hospital (Williams et al., 1999). This measure was appropriate for use in this study, as it evaluates fatigue patterns over the week prior to completion, rather than a daily measure, which would have been more burdensome. Additionally, the measure permitted participants to omit activity items that were not relevant, enhancing the measure’s ability to assess the impact of fatigue on caregiver activities of daily living. It was expected to take less than 5 minutes to complete.

**Factors that influence sleep disturbance.** This study explored factors associated with sleep disturbances in family caregivers. This study collected data on caregiver, child, and environmental factors with items embedded in the baseline demographic questionnaires (see Appendix L), the sleep diary, and standardized questionnaires.

**Caregiver factors.** For descriptive and analytical purposes, data were collected on the following caregiver variables: age, gender, health status based on presence of diagnosed chronic health conditions, body mass index (BMI) calculated from self-reported weight and height, race, marital status, highest level of education, employment status (i.e., outside of the home), annual income, number of members in household over and under 18 years, and duration of caregiving, in months, at home. The influence of vigilance was measured using a customized item on the sleep diary that asked, “Did you have trouble falling asleep or were you kept awake last night due to intrusive thoughts?”

A *sleep hygiene* variable was created and measured in the family caregivers. Sleep hygiene best practice includes avoiding behaviors that would interfere with normal sleep patterns and engaging in behaviors that would promote good sleep quality (Stepanski & Wyatt, 2003). Participants in this study completed the Sleep Hygiene Index (SHI; Mastin, Bryson, & Corwyn,
which was derived from the diagnostic criteria for inadequate sleep hygiene in the International Classification of Sleep Disorders (American Sleep Disorders Association, 1990; see Appendix M). The SHI is a 13-item index of sleep hygiene that provides a global score, in which higher scores suggest more maladaptive sleep hygiene. The SHI has good test–retest reliability ($r_{(139)} = 0.71, p < .001$; Mastin et al., 2006) and good convergent validity, with positive correlations ($p < .001$) associated with all features of inadequate sleep hygiene ($r = 0.37–0.46$), the ESS ($r = 0.24$), and the PSQI ($r = 0.48$). The SHI was used in this study to measure the sleep hygiene practices reported by family caregivers over the previous week. It was expected to take less than 10 minutes to complete.

**Child factors.** Family caregivers of CMT described their child’s technology use on the demographic data sheet by the type of technology (e.g., home oxygen, tracheostomy), the quantity of technology in hours of technology use per week, and the timing of technology use (i.e., days, nights, or a combination of both). If relevant, the presence or absence of acute illness in the child was documented by the family caregivers on the sleep diary.

Children’s sleep problems were measured using the Children’s Sleep Habits Questionnaire (CSHQ; Owens, Spirito, & McGuinn, 2000; see Appendix L). The CSHQ has been used to assess sleep in samples of community-based children (Owens et al., 2000), those with neurodevelopmental issues (Malow et al., 2006), and in one ambulatory study of sleep in family caregivers and their children with cancer (Zupanec et al., 2010). The measure contains items about the child’s usual sleep habits as well as sleep problems experienced over the most typical week immediately prior to completion. Scores on subscales (i.e., bedtime resistance, sleep onset delay, sleep anxiety, night awakenings, parasomnias, sleep disordered breathing, and daytime sleepiness) are summed for an overall total score ranging from 33–99, with a higher
score suggesting more sleep disturbance. Psychometric testing of the CSHQ indicates satisfactory test–retest reliability (0.62–0.79), with a cut-off score of 41 and a reasonable sensitivity (0.80) and specificity (0.72; Owens et al., 2000). It was expected to take less than 10 minutes to complete.

Environmental factors. Environmental factors that may have influenced sleep disturbances were studied using items collected on the demographic form at baseline and the daily sleep diary. Participants described the amount of approved homecare that they had in hours per week, their typical use of homecare in hours per week, and their nocturnal use of homecare, defined as the number of overnight hours of homecare between 9:00 p.m. and 9:00 a.m. typically used per week.

In summary, multiple data sources and measurement tools were used in this investigation to collect robust data on sleep and related variables in family caregiver participants. The administration of baseline measures at intake was expected to take less than 15 minutes to complete. Family caregivers were asked to complete a sleep diary every morning for 7 days, which was expected to take less than 15 minutes to complete.

Sample

Inclusion criteria. The family caregivers of CMT group was comprised of the self-identified primary caregiver of a child aged 12 months–18 years of age, living at home, who:

- routinely depended on medical technology* at night due to a complex chronic medical condition.

* those that are not unlikely to fail during the night and, if they did so, would likely result in a negative consequence for the child’s health
The control group was comprised of one member per family who self-identified as the primary caregiver of a child aged 12 months–18 years of age, living at home, who:

- was considered healthy, with no known developmental or chronic health conditions.

This study only recruited family caregivers of children over 12 months of age because of known developmental sleep patterns that predominate in the younger age group. Sleep in infants under a year of age is normally characterized as unpredictable and is associated with sleep fragmentation and deprivation in new parents (Mindell & Owens, 2010; Stremler et al., 2006).

Inclusion for family caregivers of healthy children was based proportionally on their match to four age-based strata of family caregivers of CMT that are defined later in this thesis.

**Exclusion criteria.** The exclusion criteria for the study were family caregivers that:

- had insufficient English language skills;
- had a diagnosed sleep disorder (e.g., sleep apnea, narcolepsy);
- lived greater than a 45 minute drive from a participating paediatric clinic;
- had an infant (i.e., index child or other sibling) under 12 months of age living at home.

A final study exclusion criterion was added after the study was originally approved requiring the research ethics boards at both host institutions to approve an amendment. Because it was not feasible to achieve balanced strata according to the ages of multiple CMT, a criterion that excluded FC of more than one child in the home who depended on medical technology at night was added. It is estimated that this additional criterion eliminated 2–3 families in total and that the experiences of these families (i.e., with multiple children dependent on homecare technology) were deemed ‘highly exceptional’ by their health care providers.
Recruitment Sites

Study participants were recruited from hospital and community clinics. Recruitment of family caregivers of CMT occurred at a paediatric academic health science centre (Hospital for Sick Children [SickKids]) in Southern Ontario. This hospital offers primary and secondary care for the paediatric population in metropolitan Toronto, as well as tertiary and quaternary care for patients from all communities in Ontario. Two ambulatory clinics located in this paediatric hospital (the Paediatric Specialties Clinic and the Respiratory Medicine Clinic) hosted the study for recruitment purposes.

Two community paediatric clinics located in the Greater Toronto Area (GTA) served as sites of recruitment for family caregiver controls. The first paediatric clinic is located within 5 km of the hospital in an urban neighborhood, that serves a diverse community in terms of its mixed ethnicity and socioeconomic status (http://www1.toronto.ca). The second paediatric community clinic, also located within the paediatric hospital catchment, is in a suburban community with growing rates of visible minorities, low-income individuals, and single-parent families. Families attending this clinic were also culturally diverse and included many first- and second-generation Canadians of Italian or Chinese descent (City of Vaughan, 2011).

Sample Size

There is very little evidence to inform what a minimal clinically meaningful difference in TST-N is in this population. In adults with insomnia, some authors have suggested that a 30-minute difference in TST-N is clinically meaningful (Okajima, Komada & Inoue, 2011). There is neither population-based nor clinical evidence however to support that a 30-minute difference is considered important in healthy adults. A difference of 1 hour was reported in a cohort study comparing sleep in family caregivers of ventilator-dependent children with family caregivers of
healthy controls using subjective sleep measures. Meltzer & Mindell (2006) stated that the difference ($6.31 \pm 1.2h$ vs. $7.34 \pm 0.9h$, $p = .007$) placed family caregivers of CMT at risk for chronic sleep deprivation. At this amount of sleep, an additional hour of sleep per night would move family caregivers CMT into the ideal 7-9 hours of sleep per night, representing a substantial reduction in sleep deprivation. Thus, using a 2-sided independent groups $t$-test with a significance level of 5% and assuming a standard deviation of 1.2 hours in TST-N, as reported in the study by Meltzer and Mindell (2006), 23 subjects per group were needed to detect a difference in sleep duration of 1 hour with 80% power. For this study, the sample was increased from 23 to 40 in each cohort to account for the potential influence of under-reported (due to self-report) variation in TST-N as described in the Meltzer and Mindell (2006) study. As well, this sample size allowed for a 20% rate of missing data due to loss to follow-up.

Sample Acquisition

**Feasibility.** In this study, recruitment of participants occurred on a weekly basis during scheduled clinic hours and according to peak clinic volumes. Based on access to actigraphs and logistics related to data management, including coordination of home visits, it was feasible to recruit up to six participants at a time.

Between the two ambulatory hospital clinics for CMT, there were an estimated 250 family caregivers that were expected to meet inclusion criteria (R. Amin & E. Cohen, personal communication, March 16, 2011). In prior research conducted with a similar population of CMT drawn from these clinical services, recruitment rates of family caregivers were between 50% (Keilty et al., 2008) and 75% (Keilty et al., 2010). The community paediatric clinics have over 3,000 registered patients each, yielding a combined pool of over 6,000 patients. Health care providers in the paediatric hospital clinics and the community paediatric clinics strongly
endorsed this study and were instrumentally supportive in facilitating participant recruitment. Thus, with a conservative estimate of consent rates at 50%, completion of study recruitment was expected to occur within six months.

**Sample characteristics.** In this study, it was anticipated that recruitment would result in a final sample of family caregivers of CMT with children living with varied diagnoses (e.g., neuromuscular, respiratory, gastroenterology, and genetic diseases) and using varied technologies (e.g., ventilators, home oxygen, enteral feeding tubes). While fathers and grandparents were eligible to participate, it was expected that mothers would predominate the final sample of family caregivers from both groups, given that they most often assume the primary caregiving role in families.

**Recruitment to strata.** The distribution in number of FC CMT to controls was planned to be proportionate on each stratum of the variable- age of the index child. The family caregiver CMT cohort was recruited first followed by the family caregivers control cohort. Recruitment of the family caregiver controls was based on the known distribution of cases on each stratum for age of the CMT (index child). A balanced data set was created according to the following strata: toddlers (12 months–2 years), pre-school children (3–5 years), school-aged children (6–12 years), and youth (13–18 years). These categories are grounded in knowledge of developmental norms and related sleep–wake patterns and habits for children in these age groups (Mindell & Owens, 2010). The most challenging stratum to fill was the youth age group (13–18 years) in the family caregiver control cohort. Because adolescents were uncommonly encountered in the primary care setting, two strategies to target recruitment of family caregivers of healthy adolescents were used: 1) a snowballing technique, with family caregiver control participants referring to others in the community with children aged 13–18 years, and 2) an online SickKids
Balanced recruitment to age strata was planned to help limit, but not eliminate, the influence of confounding by child’s age on the sleep in family caregivers. It was recognized that the presence of one or more children in the household, other than the index child, might also influence family caregiver sleep. The sampling strategy was preferred based on knowledge of patterns of primary care use, since straight consecutive sampling from community paediatric clinics would have yielded a sample heavily skewed towards the younger age group, resulting in a non-normative distribution for child’s age in the family caregiver control cohort. In this study, balanced recruitment on more variables (i.e., by age of one or more other children in the family) was not foreseen as feasible and therefore was not done.

**Procedure**

This study was carried out prospectively across 6 days and 7 nights. Participants were recruited from paediatric clinics: family caregivers of CMT from two hospital-based clinics and family caregiver controls from two community-based clinics. The only information collected at the enrollment interview was participant sociodemographics and clinical data about the child. Sleep measurement began on the night of enrollment and continued prospectively over the study period. Standardized questionnaires were administered at the final interview, conducted during a home visit. Study recruitment occurred one day per week in the hospital (March 19–September 3, 2013), coinciding with set clinic days for children with medical complexity and in the community (June 6–September 19, 2013) on the days having peak clinic volumes at the primary care provider’s office. To populate the strata with balanced groups, 40% of the family caregiver
of CMT sample was recruited before initiation of recruitment among the family caregiver controls. The study schema was planned as follows (see Figure 3).

**Paediatric Clinics**
—mail-out to inform all potential participants

- Not eligible
- Refused
- Other reasons

**FCs of CMT**
- Assessed for eligibility & consent
- Baseline clinical and demographic data collected
- Actigraph applied on Day 1
- Daily reminder phone calls
- Daily sleep diary completed for 7 days/night
- Home visit on Day 8
- Actigraph data collected
- Questionnaires completed for sleep quality, HRQol, depression, sleepiness, fatigue
- Loss to follow-up

- FCs of healthy same-age controls
- Assessed for eligibility & consent
- Baseline clinical and demographic data collected
- Actigraph applied on Day 1
- Daily reminder phone calls
- Daily sleep diary completed for 7 days/night
- Home visit on Day 8
- Actigraph data collected
- Questionnaires completed for sleep quality, HRQol, depression, sleepiness, fatigue
- Loss to follow-up

**Data analyses**

*Figure 3.* Planned study schema using flow diagram.

**Research ethics.** Research ethics approval for investigation of human subjects was obtained from both the University of Toronto’s Health Sciences Research Ethics Board and the Research Institute at the Hospital for Sick Children. All participants received standard care from their health care providers during the study period. Amendments to the protocol received
approvals as required. Relevant details regarding the ethical conduct of this study are described in the sections that follow.

Pilot testing. With ethics board approval in place, a sleep diary customized for use in this study was pretested with a group of ineligible family caregivers of CMT (i.e., those who live > 45 mins away; n = 4). Daily sleep diary data were collected for 6 days and 7 nights per participant in the trial period. The use of the actigraph was not pilot tested, as it had been found to be feasible and acceptable in other community-based studies. Pilot work for this study aided in the development and customization of the sleep diary, resulting in clarifications to the protocol, and alternate response options to the diary were made. The pilot data collection process also helped to evaluate the amount of time required to complete the measures and gauged participant burden and logistics before the full investigation began.

Recruitment and consent. During the set up phase, a brief study information session was provided to health care providers in the paediatric clinics. A letter of information (see Appendix F) was also prepared in lay language and mailed to potential participants in advance of their clinic visit to help them plan their time and identify any questions that they might have about the study, including the acceptability of wearing the actigraph at home for a week. Also to promote recruitment, flyers with brief study details and research investigator contact information were posted in the participating clinics. An amendment to the study protocol enabled study participants to identify family caregivers of adolescents through the nomination of neighbourhood friends and the distribution of letters of introduction targeting this stratum (see Appendix G).

The routine recruitment procedure included that a health care provider known to the family caregivers (e.g., registered nurse, nurse practitioner, or physician in the clinic) introduced
them to the background and purpose of the study. Once the family caregiver had granted permission for the research team to approach them, the study investigator or research assistant (RA) assessed them for eligibility in more detail (see Appendix H) and provided information about the study. Informed consent from all participating family caregivers and assent from competent children was obtained and documented in the health record (see Appendix I). Family caregivers were given the option of consenting to study participation at this initial encounter or to take additional time and, if they chose, to complete the consent process in a later encounter.

Data collection. Baseline sociodemographic and clinical data about the child was obtained directly from the family caregivers in the clinics (see Appendix J). The actigraph was applied and participants were taught about its care, use, and trouble-shooting. Participants received instruction on how to complete the daily sleep diary along with details of how to contact the study investigator and RA (see Appendix K).

Participants were asked to wear the actigraph on their non-dominant wrist 24 hours per day for a 7-day period (excluding time spent bathing or swimming). To limit missing data, a member of the investigation team was available from 8:00 a.m. to 6:00 p.m. to respond to questions about study participation or use of the actigraph.

During the data collection period, participants were asked to complete the customized sleep diary to capture detailed information about sleep–wake activity and the caregiver, child, and environmental factors thought to influence the previous night’s sleep. As per participant preferences, daily reminders to complete the sleep diary were sent in the form of brief telephone calls, emails, or texts.

Upon completion of the sleep data collection phase, the investigator scheduled a home visit of approximately 30 minutes in length at a time convenient to the family caregiver. Home
visits (Keilty et al., 2010) and daily reminders (Meltzer & Mindell, 2006) for data collection purposes have been acceptable in other studies with family caregivers of CMT and in studies of sleep in family caregivers of older adults (Carter et al., 2009). In this study, during the home visit, the actigraph was removed and study questionnaires administered, including the PSQI, CSHQ, SHI, SF-12™, CES-D, ESS, and MAF (see Appendix L). At the end of the home visit, the researcher provided the family caregivers with a thank-you note and a gift card of a nominal amount ($25) from a national chain drug store.

Data Management

To maintain participant confidentiality and study integrity, all electronic data were stored on password-protected computers and backed up on a weekly basis to a server located at SickKids. Only select data, all de-identified, were transferred to the University of Toronto (U of T), per the requirements of the SickKids Research Ethics Board. All study data forms were stored in locked filing cabinets in the investigator’s office. Access to the data was limited to the study team. Any identifying participant information, including consent forms and contact sheets, were stored in an alternate locked cabinet. A unique study ID was generated for each participant and used together with a secondary identifier (i.e., a 6 digit integer comprised of mmyyyy) to ensure anonymity of data, enable matching procedures, and prevent lost data due to transcription or other data entry errors.

The RA and study investigator were responsible for participant recruitment and the collection of baseline data. Family caregiver and CMT demographic data were obtained directly from the participants in a private location in the paediatric clinics. Administration of the sleep diary, prompted by daily reminders, was the chief responsibility of the RA who did not have a prior or present therapeutic relationship with the participants. Responsibility for the home visits
and data acquisition following the sleep data collection was shared between the study investigator and the RA. The study investigator and RA were also responsible for downloading data from the actigraph to a computer loaded with specialized software required for use with Ambulatory Monitoring Inc. Octagonal Basic Motionloggers. This computer was password protected, and the data did not have any identifying patient-related information. Before analysis, a visual inspection of the data was performed to ensure that the histogram represented a reasonable sleep–wake pattern (Morgenthaler et al., 2007).

All baseline, sleep, and outcome data were entered into REDCap, a secure web application for building and managing online surveys and databases (details of which are available at http://www.project-redcap.org), the specifics of which were developed in consultation with Research Services at SickKids. To promote accuracy, double data entry was carried out for all data entered for the primary outcome and a minimum of 30% of the data entry for all other variables. Data were cleaned, including checks for logic and range (Katz, 2006) before being downloaded for analysis.

Data Analysis

Following data entry, data were analyzed using SPSS® version 22 software. The significance level was set at 0.05 for all analyses completed in this study.

Sociodemographic and clinical data. Descriptive statistics were used to describe the sociodemographics of the family caregiver cohorts and characteristics of the CMT. Means and standard deviations or medians and interquartile ranges were reported based on data distribution for continuous variables and frequencies and percentages for categorical variables.

Comparison between family caregivers of CMT and controls. The differences between the sleep in family caregivers of CMT and control subjects were analyzed using
inferential statistics. Assuming a normative distribution, independent $t$ tests ($t$) were used to analyze the differences between the two groups in TST-N (primary outcome), TST-D, LSS-N and LSS-D, number of sleep deprived nights (< 6hrs), and NNA (objectively measured and self-reported), HRQoL, depression, daytime sleepiness, and fatigue. The difference in the proportion of family caregivers of CMT to family caregiver controls that reported poor sleep quality (> 5, as a cut-off) was analyzed using Chi-square ($X^2$). When non-normative distributions were observed, Mann Whitney U tests were completed for continuous and Fisher’s Exact for categorical variables, as indicated.

**Family caregiver sleep data.** Descriptive statistics were also used to report on continuous sleep variables in all family caregivers. Values for the following variables were averaged in minutes across the number of nights available and are reported as means, standard deviations (SD), and ranges: TST-N, TST-D, LSS-N and LSS-D, and NNA (i.e., objectively measured and self-reported). The number of sleep-deprived nights (< 6h) across one week was also reported as means (SD), and ranges.

The proportion of family caregivers that reported poor sleep quality (scores > 5, as a cut-off) was reported as frequencies. The variability in TST-N between subsequent nights was reported as the difference between the cohorts on the mean number of consecutive nights (SD) during the study period that varied in length by at least 30%. This variable was derived by calculating \(\frac{(TST-N \text{ night 2} - TST-N \text{ night 1})}{TST-N \text{ night 1}}\) x 100 = % difference; then tabulated for how many nights had a +/- 30% difference per participant in a week.

**Associations with sleep in family caregivers.** Simple linear regressions were conducted to explore which caregiver, child, and environmental variables independently predicted TST-N and NNA-O in family caregivers of CMT. Multiple regression analysis was completed to explore
associations between modifiable factors and select sociodemographic risk factors (i.e., informed by theory and practice) with TST-N and NNA-O in family caregivers of CMT. The regression model included caregiver sleep hygiene, child sleep quality, nocturnal use of homecare, family caregiver age, family caregiver gender, and marital status. Marital status (partnered = 1 and single = 0) and gender (female = 1 and male = 0) were dummy coded. All predictor variables were entered on the same step of the regression equation. These data were screened for violation of assumptions prior to analyses. An alpha level of .05 was set for all analyses.

**Monitoring the Study**

A manual that described the study design guided the study team and ensured consistency in protocol administration. A study notebook was kept to collect information about the execution of the study and keep notes about any challenges encountered or strategies employed to enhance recruitment and retention to the study.

**Loss to Follow-Up and Missing Data**

The use of actigraphy is generally well accepted in ambulatory studies of family caregivers and parents of healthy children; thus, minimal (< 20%) missing actigraphic data was anticipated. Missing data were handled by averaging all variables over the number of nights in a week. As per AASM recommendations, a minimum of 4 nights of actigraphy data was required for the participant to be included in the analysis (Littner et al., 2002) and, if available, sleep diary data was used to substitute for missing actigraphic data (i.e., additional sleep diary data complemented 4 nights of actigraphic data). Thus, we considered a case missing if there were less than 4 nights of actigraphy, even if there was sleep diary data available. To prevent missing data, the items in the standardized questionnaires were carefully chosen to minimize participant burden and required less than 30 minutes to complete. The questionnaires were designed to
require a low level of literacy. The RA or study investigator was on hand at the time of baseline and final instrument administration to provide any clarifications necessary so that the family caregivers could complete them as accurately and efficiently as possible. Finally, to enhance retention rates and minimize missing data, daily reminder phone calls and home visits were employed.

**Risks and Benefits to Participation**

**Risks.** There was minimal risk known to participants in this study. Participants were informed that they could wear the actigraph over their clothes or use a wrist-band provided to them if allergies to the metal backing of the actigraph or other irritation was present. Only one participant reported mild skin irritation, for which the recommended methods of limiting contact directly with the skin were employed successfully, and data capture continued. During the study period, participants received their usual care from their health care providers in the hospital and the community.

It was planned that if a family caregiver scored above the cut-off for clinically significant depression (i.e., > 21 on the CES-D), the family caregiver would be informed immediately and directed to follow up with a relevant health care provider (e.g., primary care provider or mental health specialist). It was also planned that if a child scored above the cut off (≥ to 7/9) on the sleep disordered breathing subscale on the CSHQ, and the child was not currently being seen by a paediatric sleep or respiratory medicine specialist, the family caregiver would be informed immediately and directed to follow up with a relevant health care provider (e.g., primary care provider or sleep specialist). Through the consent process, family caregivers were made aware that if, at any time, they conveyed concerns about harming themselves or others (e.g., child
abuse, suicidal ideation), they would be immediately informed by the investigator that the concern would also be reported to the appropriate authorities, as required by law.

**Benefits.** There were no known benefits to participating in this study for the family caregivers. In similar studies, participants have communicated that they appreciated the opportunity to speak with individuals about their experiences and potentially help others through research participation. Following the conclusion of the study, participants had the option of learning about their individual actigraphy results. Participants that have requested follow-up will be informed of study results in a brief study summary and will be provided information about resources to promote healthy sleep, including links to the Canadian Sleep Society and National Sleep Foundation websites.
Chapter 4: Results

Sample

In total, 42 family caregivers of CMT and 43 family caregiver controls were recruited from the 108 family caregivers approached and assessed for eligibility during the study period, March 19–September 19, 2013 (see Figure 4). The overall recruitment rate among eligible family caregivers of CMT was 84% \((n=42/50)\) and 76% \((n=44/58)\) among family caregiver controls. The most common reason for non-participation in the hospital-based sample (i.e., family caregivers of CMT) was time constraints either on the day of the clinic or at home \((n=3)\). In the community derived sample, family caregiver controls declined most often due to a lack of time on the day of recruitment \((n=5)\) and/or having an upset or sick child (e.g., crying after immunization, fever; \(n=3\)).

Participants were recruited first into the family caregivers of CMT cohort, followed by recruitment to the family caregiver controls arm of the study. All participants were followed for 6 days and 7 nights. Only one participant \((n=1 \text { FC Control})\) withdrew from the study. Actigraphy data were not available for analysis for five participants due to actigraph malfunction \((n=1 \text { FC CMT}, n=1 \text { FC Control})\) and non-adherence to wearing the actigraph for the required minimum 4/7 nights \((n=1 \text { FC CMT}, n=2 \text { FC Control})\) during the study period.
To achieve balanced groups, family caregivers were categorized into strata according to the index child’s age (see Table 3). Overall, the mean (SD) age of the children in the study sample was 7.4 (4.9) with a small but not significant difference in the child’s age per cohort: family caregivers of CMT 7.9 (5.2) vs. family caregiver controls 7.1 (4.6), \( p = .16 \). While there

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**Figure 4.** Schema for cohort study recruitment.

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**Note.** CMT = children who depend on medical technology, FC = family caregivers
were more male children than females represented in both cohorts, there was no difference found in proportion between the cohorts \( \chi^2 = 2.01, p = .16 \).

Table 3

*Distribution of Family Caregiver Participants by Index Child’s Age and Gender*

<table>
<thead>
<tr>
<th>Child’s age</th>
<th>FC CMT Partial data*</th>
<th>FC CMT Complete data</th>
<th>FC Controls Partial data*</th>
<th>FC Controls Complete data</th>
</tr>
</thead>
<tbody>
<tr>
<td>12 months -2 years</td>
<td>1 (2.4%)</td>
<td>8 (20%)</td>
<td>1 (2.3%)</td>
<td>8 (20%)</td>
</tr>
<tr>
<td>3–5 years</td>
<td>0 (0.0%)</td>
<td>11 (27.5%)</td>
<td>1 (2.3%)</td>
<td>11 (27.5%)</td>
</tr>
<tr>
<td>6–12 years</td>
<td>0 (0.0%)</td>
<td>13 (32.5%)</td>
<td>1 (2.3%)</td>
<td>13 (32.5%)</td>
</tr>
<tr>
<td>13–18 years</td>
<td>1 (2.4%)</td>
<td>8 (20%)</td>
<td>0 (0.0%)</td>
<td>8 (20%)</td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>Gender</th>
<th>FC CMT</th>
<th>FC Controls</th>
</tr>
</thead>
<tbody>
<tr>
<td>Male</td>
<td>22 (52.4%)</td>
<td>29 (67.4%)</td>
</tr>
<tr>
<td>Female</td>
<td>20 (47.6%)</td>
<td>14 (32.6%)</td>
</tr>
</tbody>
</table>

*Note. *refers to data obtained from FC CMT that completed only partial (subjective—surveys, diaries) not complete (subjective—surveys, diaries and objective—actigraphy) data collection. CMT = children who depend on medical technology, FC = family caregivers

Overall, the age of the family caregivers in the study was 40.2 (7.0) years, with a total of 85.9% \( (n = 73) \) of the participants’ female and 14.1% \( (n = 12) \) male (Table 2). The vast majority of the participants were mothers \( (n = 72) \), although a few fathers \( (n = 12) \) and one grandmother confirmed being the primary caregiver in the home (see Table 4).

Table 4

*Baseline Demographic Characteristics of Family Caregivers*

<table>
<thead>
<tr>
<th>Family Caregiver Characteristics</th>
<th>FC CMT Age ( (8.27) )</th>
<th>FC Controls Age ( (5.52) )</th>
<th>test ( t )</th>
<th>( p ) value</th>
</tr>
</thead>
<tbody>
<tr>
<td>Gender</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Male</td>
<td>7 (16.7%)</td>
<td>5 (11.6%)</td>
<td>( X^2 = .45 )</td>
<td>.549</td>
</tr>
<tr>
<td>Female</td>
<td>35 (83.3%)</td>
<td>38 (88.4%)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Relationship to child</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Mother</td>
<td>34 (81.0%)</td>
<td>38 (88.4%)</td>
<td>( X^2 = .15 )</td>
<td>.446</td>
</tr>
<tr>
<td>Father</td>
<td>7 (16.7%)</td>
<td>5 (11.6%)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Other (i.e., Grandparent)</td>
<td>1 (2.4%)</td>
<td>0 (0.0%)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Number in household</td>
<td>&lt; 18 years age</td>
<td>1.76 (0.9)</td>
<td>2.09 (0.8)</td>
<td>( t = -1.46 )</td>
</tr>
<tr>
<td>Variable</td>
<td>Group 1</td>
<td>Group 2</td>
<td>t</td>
<td>p</td>
</tr>
<tr>
<td>----------------------------------------------</td>
<td>-----------</td>
<td>-----------</td>
<td>-------</td>
<td>-------</td>
</tr>
<tr>
<td>&gt; 18 years age</td>
<td>2.17 (0.9)</td>
<td>2.12 (0.6)</td>
<td>.31</td>
<td>.758</td>
</tr>
<tr>
<td>Health status</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Diagnosed chronic health condition</td>
<td>12 (28.6%)</td>
<td>8 (18.6%)</td>
<td>1.17</td>
<td>.315</td>
</tr>
<tr>
<td>Medical appointments in last 12 months</td>
<td>2.43 (2.5%)</td>
<td>3.07 (2.6%)</td>
<td>-1.16</td>
<td>.249</td>
</tr>
<tr>
<td>BMI (self-report)</td>
<td>25.92 (4.9)&lt;sup&gt;a&lt;/sup&gt;</td>
<td>24.11 (3.9)&lt;sup&gt;b&lt;/sup&gt;</td>
<td>1.79</td>
<td>.078</td>
</tr>
<tr>
<td>Marital status</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Married or stable relationship</td>
<td>31 (73.8%)</td>
<td>41 (95.3%)</td>
<td>7.62</td>
<td>.008</td>
</tr>
<tr>
<td>Single</td>
<td>10 (25.0%)</td>
<td>2 (4.7%)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Other</td>
<td>1 (2.5%)</td>
<td>0 (0.0%)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Born in Canada</td>
<td>22 (52.4%)</td>
<td>30 (69.8%)</td>
<td>2.70</td>
<td>.122</td>
</tr>
<tr>
<td>Length of time in Canada &lt; 5 years</td>
<td>2 (4.8%)</td>
<td>0 (0.0%)</td>
<td>3.63</td>
<td>.123</td>
</tr>
<tr>
<td>Race/Ethnicity</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Arab</td>
<td>2 (4.8%)</td>
<td>0 (0.0%)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Black</td>
<td>2 (4.8%)</td>
<td>1 (2.3%)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>East Asian</td>
<td>6 (14.3%)</td>
<td>2 (4.7%)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>South Asian</td>
<td>5 (11%)</td>
<td>1 (2.3%)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Latin/South American</td>
<td>2 (4.8%)</td>
<td>1 (2.3%)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>White/Caucasian</td>
<td>24 (57.1%)</td>
<td>35 (81.4%)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Mixed race</td>
<td>1 (2.4%)</td>
<td>3 (7.0%)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Highest level of school completed</td>
<td></td>
<td></td>
<td>14.66</td>
<td>.001</td>
</tr>
<tr>
<td>Elementary/grade school</td>
<td>0 (0.0%)</td>
<td>2 (4.7%)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>High school</td>
<td>10 (23.8%)</td>
<td>2 (4.7%)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>College or university</td>
<td>26 (61.9%)</td>
<td>22 (51.2%)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Graduate degree</td>
<td>6 (14.3%)</td>
<td>17 (39.5%)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Paid employment</td>
<td></td>
<td></td>
<td>10.10</td>
<td>.041</td>
</tr>
<tr>
<td>Full time</td>
<td>15 (35.7%)</td>
<td>22 (51.2%)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Part time</td>
<td>7 (16.7%)</td>
<td>12 (27.9%)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Unemployed</td>
<td>16 (38.1%)</td>
<td>4 (9.3%)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Self-employed</td>
<td>3 (7.1%)</td>
<td>4 (9.3%)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Retired</td>
<td>1 (2.5%)</td>
<td>0 (0.0%)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Maternity leave</td>
<td>0 (0.0%)</td>
<td>1 (2.3%)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Annual household income</td>
<td></td>
<td></td>
<td>19.96</td>
<td>&lt; .01</td>
</tr>
<tr>
<td>&lt; $25,000</td>
<td>9 (21.4%)</td>
<td>1 (2.3%)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>$25,000–$39,000</td>
<td>8 (19.0%)</td>
<td>2 (4.7%)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>$40,000–$59,000</td>
<td>7 (16.7%)</td>
<td>5 (11.6%)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>$60,000–$99,000</td>
<td>9 (21.4%)</td>
<td>9 (20.9%)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>≥ $100,000</td>
<td>9 (21.4%)</td>
<td>26 (60.5%)</td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

Note. Continuous data are shown as mean (SD); Independent Groups t-tests were used for all continuous variables; Mann Whitney-U tests were used for those with non-normal distributions; categorical data are shown as number (percentages); Chi-Square was used for categorical variables, Fisher’s Exact test was used when more than 20% of the expected cell frequencies were < 5. BMI = Body Mass Index, CMT = children who depend on medical technology, FC = family caregivers

<sup>a</sup>n = 39. <sup>b</sup>n = 42.
No differences in proportions were found between the cohorts for the demographic variables of family caregiver age, gender, relationship to child, number in household < 18 years, number in household > 18 years, length of time in Canada (i.e., > 5 years, and birth in Canada. The majority of the sample was found to self-identify as White/Caucasian \( n = 59, 69.4\% \), with ethnic diversity (i.e., other than White/Caucasian) found among approaching half of the family caregivers of CMT \( n = 18, 42.9\% \) and fewer \( 18.6\%, n = 8 \) of the family caregiver controls.

Differences in the proportions between the groups were found in select sociodemographic characteristics associated with lower socioeconomic status. Five times as many family caregivers of CMT group were single (i.e., not in a partnered relationship) compared to family caregivers in the control group \( n = 10, 23.8\% \) FC CMT vs. \( n = 2, 4.7\% \) FC controls \( \chi^2 = 7.62, p = .008 \)). More than double the number of family caregivers of CMT \( n = 10, 23.8\% \) had high school education or less compared to family caregiver controls \( n = 4, 9.3\%; \chi^2 = 14.66, p = .001 \). On the baseline characteristic of total family income, more than half of family caregivers of CMT \( n = 23, 54.76\% \) reported an income of under $60,000 per household, while more than 80% of family caregiver controls \( n = 35, 81.4\% \) had a total household income exceeding $60,000 per year \( \chi^2 = 19.96, p = < .01 \). Importantly, four times as many family caregivers of CMT reported being unemployed \( n = 16, 38.1\% \) compared to family caregiver controls \( n = 4, 9.3\%; \chi^2 = 10.10, p = .041 \).

No differences were found in proportions between the groups for any indicators of family caregiver health status, including number of visits to a health care provider in the last 12 months and the presence of a chronic health condition. Moreover, there was no difference in self-reported body mass index between the cohorts.
Child Clinical Characteristics

All of the family caregivers of CMT ($n = 42$) had children who depended on the routine use of medical technology at home during the nighttime period from 9 p.m. to 9 a.m., per the inclusion criteria, and the majority ($n = 33, 78.6\%$) also had children who depended on technology use over the daytime period from 9 a.m. to 9 p.m (see Table 5). Half of the children ($n = 21, 50.0\%$) depended on the nightly use of a single technology, while the remaining depended on the use of multiple medical technologies. Over 70\% of the family caregivers reported that their child used respiratory technologies on a routine basis ($n = 30$) and more than a third of the participants’ children ($n = 15, 37.5\%$) depended on its use 24 hours per day. Enteral feeding pumps were used by the large majority of the participants ($n = 29, 69.2\%$) and were most often used in combination with other technologies ($n = 17, 58.6\%$). Two children depended on the use of four medical technologies, including a tracheostomy, invasive positive pressure ventilator, oxygen delivery equipment, and enteral feeding devices. There was a wide range in amount of technology use (1–12 hours of use per time period), with 37.5\% ($n = 15$) of the participants’ children dependent on the use of medical technology 24 hours per day.

Approximately half of the participants used oxygen saturation monitors on a routine basis ($n = 23, 54.8\%$) also ranging from 1–12 hours of use during the day or nighttime period. The concurrent use of other kinds of medical technologies was reported among a small number of family caregivers of CMT ($n = 8, 19.0\%$). These technologies included those seldom used and/or considered less likely to fail at night (e.g., central venous lines).

The most common primary diagnosis among the children was a genetic/metabolic condition (e.g., Down Syndrome or other chromosomal disorders; $n = 14, 33.3\%$). Parent-
reported neurocognitive delay or impairment was present in the majority of the CMT \( n = 26, 61.9\% \).

Table 5

**Clinical characteristics of Children Dependent on Medical Technology**

<table>
<thead>
<tr>
<th>Clinical characteristics</th>
<th>Mean (SD) or Count (%)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Quantity of technology use per 24-hours (hours)</td>
<td>16.38 (7.1)</td>
</tr>
<tr>
<td>Daytime use (hours)</td>
<td>6.7 (5.1)</td>
</tr>
<tr>
<td>Nighttime use (hours)</td>
<td>9.68 (3.1)</td>
</tr>
<tr>
<td>Typical period of technology use</td>
<td></td>
</tr>
<tr>
<td>Daytime</td>
<td>33 (78.6%)</td>
</tr>
<tr>
<td>Nighttime</td>
<td>42 (100%)</td>
</tr>
<tr>
<td>Typical amount of monitor use per day (hours)</td>
<td>6.42 (6.7)</td>
</tr>
<tr>
<td>Daytime use only</td>
<td>0.95 (2.7)</td>
</tr>
<tr>
<td>Nighttime use only</td>
<td>5.48 (5.6)</td>
</tr>
<tr>
<td>Typical period of monitor use</td>
<td></td>
</tr>
<tr>
<td>Daytime</td>
<td>6 (15.0%)</td>
</tr>
<tr>
<td>Nighttime</td>
<td>18 (45.0%)</td>
</tr>
<tr>
<td>Main diagnosis</td>
<td></td>
</tr>
<tr>
<td>Cardiac</td>
<td>6 (14.3%)</td>
</tr>
<tr>
<td>Genetic/Metabolic</td>
<td>14 (33.3%)</td>
</tr>
<tr>
<td>Neurological</td>
<td>9 (21.4%)</td>
</tr>
<tr>
<td>Neuromuscular</td>
<td>5 (11.9%)</td>
</tr>
<tr>
<td>Respiratory</td>
<td>7 (16.7%)</td>
</tr>
<tr>
<td>Musculoskeletal</td>
<td>1 (2.4%)</td>
</tr>
<tr>
<td>Neurocognitive delay/impairment (parent report)</td>
<td>26 (61.9%)</td>
</tr>
<tr>
<td>Type of technology</td>
<td>Frequency*</td>
</tr>
<tr>
<td>Invasive ventilator</td>
<td>5 (11.9%)</td>
</tr>
<tr>
<td>Non-invasive ventilator</td>
<td>10 (23.8%)</td>
</tr>
<tr>
<td>Continuous positive pressure</td>
<td>1 (2.4%)</td>
</tr>
<tr>
<td>Tracheostomy</td>
<td>12 (28.6%)</td>
</tr>
<tr>
<td>Oxygen delivery device</td>
<td>14 (33.3%)</td>
</tr>
<tr>
<td>Enteral/pump</td>
<td>24 (57.1%)</td>
</tr>
<tr>
<td>Enteral/drip</td>
<td>5 (4.8%)</td>
</tr>
<tr>
<td>Type of monitor use</td>
<td></td>
</tr>
<tr>
<td>Oxygen saturation</td>
<td>23 (54.8%)</td>
</tr>
<tr>
<td>Commercial*</td>
<td>6 (15.0%)</td>
</tr>
</tbody>
</table>

Note. *may use more than one technology and/or monitoring device at the same time. CMT = children who depend on medical technology, \( n = 42, * n = 41 \).
Community and Homecare

Use of the term homecare in this study refers to the presence of nursing care services, in the home and/or outside the home, providing care for the children with medical complexity, including those who depend on medical technologies. Family caregiver CMT provided care at home for their technology-dependent children, on average, for 6.0 (4.5) years, reporting a wide range of experience from 0.6–15.1 years (see Table 6). Family caregiver CMT also reported using 24.2 (22.1) hours of in-home and 10.7 (16.7) hours of out-of-home nursing services per week, all in shifts of 4 or more hours, rather than shorter visits. The range of total homecare hours funded was wide, from 0–115 hours per week. The majority (67.0%) of the homecare hours were reported as being used at nighttime. The most common source of homecare funding was from the Ministry of Health and Long Term Care funded via the Community Care Access Centre (CCAC; $n = 30, 71.4\%$), ranging from 7–46 (27.4 [22.8]) hours per week. Half of the family caregivers of CMT ($n = 21$) used other sources of government funding, ranging from $1,000–$10,000 per year (e.g., Enhanced Respite, Special Services at Home) to purchase additional homecare nursing services. Only two families used funding for homecare from a private insurance source, and one used funding from a charitable foundation. No families reported it to be typical that homecare nurses missed or cancelled their shifts.

Among those using nursing services out-of-the-home, $n = 12$ family caregivers of CMT used government funded school-based nursing care services ranging from 30–45 hours per week (36.8 [4.8]). No families used out-of-home nursing services on a weekly basis for the care of their technology-dependent children anywhere other than at school (i.e., none reported typically using of out-of-home over-night respite, although this doesn’t preclude occasional use for this purpose). Two families described that they declined the use of out-of-home nursing services.
Table 6

Use of Homecare Among Family Caregivers of Children who Depend on Medical Technology

<table>
<thead>
<tr>
<th>Homecare construct</th>
<th>Mean (SD) or Count (%)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Duration of caregiving in years</td>
<td>6.01 (4.5)</td>
</tr>
<tr>
<td>Source of homecare/payer</td>
<td></td>
</tr>
<tr>
<td>CCAC</td>
<td>30 (71.4%)</td>
</tr>
<tr>
<td>Enhanced respite</td>
<td>21 (50%)</td>
</tr>
<tr>
<td>SS@Home</td>
<td>11 (26.2%)</td>
</tr>
<tr>
<td>Philanthropic</td>
<td>1 (2.4%)</td>
</tr>
<tr>
<td>Private Insurance</td>
<td>2 (4.8%)</td>
</tr>
<tr>
<td>Typical reasons for missed homecare shifts</td>
<td></td>
</tr>
<tr>
<td>Shift unfilled by nurses</td>
<td>0 (0.0%)</td>
</tr>
<tr>
<td>Shift cancelled by family caregiver</td>
<td>8 (20.0%)</td>
</tr>
<tr>
<td>Hours nursing/week - approved</td>
<td>In-home 11.43 (16.8)</td>
</tr>
<tr>
<td></td>
<td>Out-of-home 10.68 (16.7)</td>
</tr>
<tr>
<td>Hours nursing/week typically used - total</td>
<td></td>
</tr>
<tr>
<td></td>
<td>24.17 (22.1)</td>
</tr>
<tr>
<td>Hours nursing/week typically used – nocturnal only</td>
<td></td>
</tr>
<tr>
<td></td>
<td>18.26 (22.6)</td>
</tr>
</tbody>
</table>

Note. CCAC = Community Care Access Centre (provincially funded through Ministry of Health), CMT = children who depend on medical technology, SS@Home = Special Services at Home (provincially funded through the Ministry of Community and Social Services)

Analysis of Sleep Outcomes in Family Caregivers

Over a 7-night period of actigraphy monitoring, family caregivers \( n = 80 \) in the study achieved less than 7 hours of sleep \( 6.88 \pm 1.1 \) during the nighttime period between 9 p.m. and 9 a.m.. Self-reported sleep time in the sample was just over 6 hours \( 6.33 \pm 1.1 \). Daytime sleep was common, with 70% of family caregivers found on actigraphy to nap (continuous sleep of 10 mins or more between 9 a.m. and 9 p.m.) at least once per week \( 1.65 \pm 1.6 \).

Primary Research Question: Difference in Sleep

The primary aim of this prospective cohort study was to compare sleep disturbance of family caregivers of CMT to that of healthy controls. It was hypothesized that sleep in family
caregivers of CMT would be of lesser quality and quantity than sleep in family caregivers of healthy controls.

**Question 1.** The primary research question was: Across 1 week, was there a difference between the mean: a) total sleep time—nocturnal (TST-N), b) total sleep time—daytime (TST-D), c) longest stretch of continuous sleep—nocturnal (LSS-N), and d) longest stretch of continuous sleep—daytime (LSS-D) in minutes in family caregivers of CMT compared to controls. This question was answered from data acquired from 6 days and 7 nights of actigraphy, which is an objective measure of sleep–wake patterns in healthy adults. Before analyses, data checks were completed, and unless specified, the variables were normally distributed.

For the primary outcome of interest, this study found family caregivers of CMT achieved 40 minutes less sleep per night than family caregiver controls (6.56 [1.37h] vs. 7.21 [0.56], $p = .019$; see Table 7) and less than what is typically recommended for healthy adults (7–9h) to achieve a good night’s sleep (National Sleep Foundation, 2005).

Family caregivers of CMT also achieved more sleep during the day along with longer stretches of consolidated sleep during the day, but relatively shorter stretches of consolidated sleep at night compared to controls (see Table 7). Most of the participants ($n = 30$ FC CMT [75.0%], $n = 27$ [67.5%] FC controls) took a nap during the study period, as evidenced by periods of sleep during the daytime on actigraphy. Family caregivers of CMT took twice as many naps per week compared to controls (2.07 [1.8] vs 1.20 [1.0]). Moreover, family caregivers of CMT slept more than twice as long during the day (21.57 [24.0] mins vs. 9.14 [9.9], $p = .004$).
Table 7

Between-group Differences on Objective Sleep Outcomes

<table>
<thead>
<tr>
<th>Sleep outcome</th>
<th>FC CMT(^a)</th>
<th>FC Control(^a)</th>
<th>p value</th>
</tr>
</thead>
<tbody>
<tr>
<td>TST-N (hrs)</td>
<td>6.56 (1.4)</td>
<td>7.21 (0.6)</td>
<td>*019</td>
</tr>
<tr>
<td>TST-D (hrs)</td>
<td>0.36 (0.4)</td>
<td>0.15 (0.2)</td>
<td>*004</td>
</tr>
<tr>
<td>LSS-N (hrs)</td>
<td>2.96 (1.3)</td>
<td>4.21 (1.7)</td>
<td>.001</td>
</tr>
<tr>
<td>LSS-D (hrs)</td>
<td>0.24 (0.3)</td>
<td>0.10 (0.1)</td>
<td>.004</td>
</tr>
</tbody>
</table>

Note. Independent samples t-tests were used for normally distributed data.*Mann-Whitney U was used for non-normally distributed data.
CMT = children who depend on medical technology, FC = family caregivers, LSS-D = longest stretch of continuous sleep – daytime, LSS-N = longest stretch of continuous sleep - nocturnal, TST-D = total sleep time - daytime, TST-N= total sleep time – nocturnal
\(^a\) n = 40

One marked outlier (e.g., average TST-N of 118 minutes per night) was identified in the family caregivers of CMT cohort. This case was examined, and analysis confirmed that the data represented the participant’s actual sleep and not a problem with recording or artifacts. As a sensitivity analysis, this case was removed, and the results did not change qualitatively, with family caregivers of CMT (n = 39) still getting fewer minutes of sleep per night than family caregiver controls (n = 40; FC CMT 400 [69.7] mins vs. FC controls 433 [33.1], p = .05). Thus, this atypical case has been left in the final analysis of all sleep and related outcomes.

A difference was observed in the distribution of the TST-N between the cohorts (see Figure 5). FC CMT had more variation in TST-N (118-548 mins) compared to the more homogeneous distribution found among FC Controls (282-518 mins; Polit, 2010). Thus analysis of the difference in night-to-night variability between groups was completed and identified that nearly twice as many family caregivers of CMT (n = 25, 62.5%) experienced a large difference
(i.e., >30%) in minutes of sleep between consecutive nights compared to family caregiver controls \((n = 14, 35\%)\). A three-fold difference was observed between the cohorts in the mean number of consecutive nights during the study period that greatly varied in length (FC CMT 1.33 [1.5] vs. FC controls 0.45 [0.7], \(p = .001\)). A thirty-percent difference in sleep time was chosen as the expression of variability between nights, as it approximated the difference between sleeping for a very short time (\(~ 6\) hours) and then “catching up” by sleeping for a very long time the next night (\(~ 9\) hours; Taylor, Wright & Lack, 2008) or vice versa.

*Figure 5.* Distribution of average TST-N in FC CMT versus FC controls from 9 p.m.–9 a.m. in minutes
**Question 2.** Aggregates across 7 nights of actigraphy data were used to answer the question “Was there a difference in the number of sleep deprived nights (< 6 hours per night) across 1 week in family caregivers of CMT compared to controls?” Family caregivers of CMT had a wider range of sleep deprived nights (0–7) compared to family caregiver controls who ranged from having 0–3 nights per week with a TST-N under 6 hours (2.18 [2.4], FC vs. 0.60 [.8], \( p < .001 \)). Thus, family caregivers of CMT had more than three times as many sleep deprived nights (83 nights) during the study period compared to family caregiver controls (24 nights; \( p < .001 \)).

**Question 3.** The third research question asked, “Across 1 week, was there a difference between the mean number of a) nocturnal awakenings—objectively measured (NNA-O) and b) nocturnal awakenings—self-reported (NNA-S) in family caregivers of CMT compared to family caregiver controls?” Family caregivers of CMT, on Mann-Whitney U test (for non-normally distributed data), experienced more objectively measured nocturnal awakenings (8.00 [3.9]) compared to family caregiver controls (6.01 [3.7]; \( p = 0.014 \)). Notably, family caregivers of CMT recalled more awakenings than their comparators (2.35 [2.0] vs. 1.42 [1.5], \( p = .005 \)). This represents a disparity between the objectively measured and subjectively reported number of nocturnal awakenings in both groups. Sleep onset latency (i.e., how long it took to fall asleep) differed between the groups, with twice as many family caregivers of CMT (\( n = 68, 23.1\% \)) compared to FC controls (\( n = 33, 11.0\% \)) reporting that it took longer than 30 minutes to fall asleep (\( \chi^2 = 15.36, p = .001 \); see Table 8).
Table 8

Sleep Onset Latency Subjectively Reported in Repeated Entries in Sleep Diary

<table>
<thead>
<tr>
<th>Time to sleep onset</th>
<th>FC CMT&lt;sup&gt;a&lt;/sup&gt;</th>
<th>FC Controls&lt;sup&gt;b&lt;/sup&gt;</th>
</tr>
</thead>
<tbody>
<tr>
<td>Less than 10 minutes</td>
<td>120 (40.8%)</td>
<td>146 (48.5%)</td>
</tr>
<tr>
<td>Between 10 and 30 minutes</td>
<td>96 (32.7%)</td>
<td>108 (35.9%)</td>
</tr>
<tr>
<td>More than 30 minutes</td>
<td>68 (23.1%)</td>
<td>33 (11.0%)</td>
</tr>
</tbody>
</table>

Note. Chi-square was used to test difference in proportions. Represents repeated measures across 7 nights of actigraphy; derived from 280 possible sleep diary entries per group (7x40). CMT = children who depend on medical technology, FC = family caregivers
<sup>a</sup> n missing = 10 (3.4%). <sup>b</sup> n missing = 14 (4.7%). *p = .001

**Question 4.** Question 4 asked, “Was there a difference between the proportion of family caregivers of CMT that reported poor sleep quality (> 5 as a cut-off on the PSQI) compared to family caregiver controls?” Family caregivers of CMT self-reported having poorer sleep than family caregiver controls, but both groups mean scores were above the cut-off for the total score (> 5) of poor sleep quality on the PSQI (FC CMT 7.75 [2.93] vs. FC controls 5.45 [2.8], p = .001; Mann-Whitney U test). Proportionately, almost twice as many family caregivers of CMT were classified as poor sleepers compared to family caregiver controls (n = 31, 73.8% FC CMT vs. n = 17, 39.5% FC controls).

**Secondary Research Question**

A secondary aim of this study was to describe the health and related outcomes of sleep disruption in family caregivers. Family caregivers of CMT were hypothesized to experience lessened health-related quality of life (HRQoL), heightened depression, excessive daytime sleepiness and increased fatigue compared to family caregiver controls.

**Question 5.** The study asked, ”Is there a significant difference in: a) HRQoL; b) depression; c) excessive daytime sleepiness; and d) fatigue reported in family caregivers of CMT
compared to healthy controls? A series of independent \( t\)-tests were used to answer this question (see Table 9), except where the use of a non-parametric test was indicated.

Table 9

**Group Comparisons on Measures of Quality of Life, Depression, Sleepiness, and Fatigue**

<table>
<thead>
<tr>
<th>Construct</th>
<th>FC CMT Mean (SD) or Count (%)</th>
<th>FC Control Mean (SD) or Count (%)</th>
<th>( p ) value</th>
</tr>
</thead>
<tbody>
<tr>
<td>HRQoL</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>SF-12 TM - Physical functioning component</td>
<td>41.81 (5.8)</td>
<td>42.18 (5.0)</td>
<td>.647</td>
</tr>
<tr>
<td>SF-12 TM - Mental functioning component</td>
<td>47.25 (7.5)</td>
<td>49.81 (52)</td>
<td>.072</td>
</tr>
<tr>
<td>Depression</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>CES-D</td>
<td>11.70 (8.7) ( a )</td>
<td>6.95 (6.0) ( b )</td>
<td>.008</td>
</tr>
<tr>
<td>CES-D 16-21</td>
<td>7 (16.7%) ( a )</td>
<td>3 (7.0%) ( b )</td>
<td></td>
</tr>
<tr>
<td>CES-D &gt; 21</td>
<td>5 (11.9%) ( a )</td>
<td>1 (2.3%) ( b )</td>
<td></td>
</tr>
<tr>
<td>Excessive Daytime Sleepiness</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>ESS</td>
<td>8.35 (4.2)</td>
<td>5.07 (3.3)</td>
<td>&lt;.001</td>
</tr>
<tr>
<td>ESS &gt;10</td>
<td>16 (38.1%)</td>
<td>5 (11.6%)</td>
<td></td>
</tr>
<tr>
<td>Fatigue</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>MAF</td>
<td>22.12 (9.1) ( c )</td>
<td>17.44 (9.0) ( d )</td>
<td>.021</td>
</tr>
</tbody>
</table>

*Note. SF-12® = Health Related Quality of Life—Short Form-12, 12 items, Physical Component Summary and Mental Component Summary—standardized to 50, with scores above and below 50 representing above and below average respectively; CES-D = 20-item, scores range from 0–60, 20% normal population score; > 16 suggests depression, > 21 suggests clinically meaningful depression; ESS = Epworth Sleepiness Scale, 8 items, summed with scores ranging from 0–24 (higher scores are suggestive of more sleepiness, with a > 10 cut-off for problematic daytime sleepiness); MAF = Multidimensional Assessment of Fatigue, 16 items, scores range from 1 (no fatigue) to 50 (severe fatigue). \( a \) \( n \) = 40. \( b \) \( n \) = 39. \( c \) \( n \) = 40. \( d \) \( n \) = 42.*

The cohorts differed on baseline global measures of depression, excessive daytime sleepiness, and fatigue. More than a third \( (n = 12, 33.3\%) \) of family caregivers of CMT compared to 9.8\% \( (n = 4) \) family caregiver controls scored in the range considered to be at risk for clinically meaningful depression on the CES-D. Differences in global depression scores \( (11.70 [8.7] \text{ vs. } 6.95 [6.0], \ p = .008) \) were also detected. More than a third \( (n = 16, 38.1\%) \) of the family caregivers of CMT compared to few family caregiver controls \( (n = 5, 11.6\%) \) scored above the cut-off \( (> 10) \) on the Epworth Sleepiness Scale, suggesting a risk of excessive daytime sleepiness. Family caregivers of CMT also scored higher on the measure of fatigue \( (22.12 [9.1] \text{ vs. } 17.44 [9.0], \ p = .021) \).
vs. 17.44 [9.0], \( p = .021 \)). Both groups scored below population norms for HRQoL (the SF-12\textsuperscript{TM} is a standardized to the population mean measure) but did not differ on the physical functioning component score (\( p = .647 \)) or the mental functioning component score (\( p = .072 \)) of the SF-12\textsuperscript{TM}.

**Other group differences.** Other measures were administered that targeted modifiable risk factors for family caregiver sleep. Among them, child sleep problems (CSHQ) as reported by the family caregivers, differed between groups. That is, using a non-parametric test, family caregivers of CMT appraised that their child had more sleep disturbance (50.88 [10.0] vs. 43.60 [6.2], \( p = .008 \)) compared to family caregiver controls. It is notable that both groups scored higher (with an overall total score ranging from 33–99, with higher scores suggesting more sleep disturbance) than the reported cut-off on the CSHQ for good sleep quality (> 41). Family caregivers of CMT also scored their children’s sleep problems to be higher on a number of the CSHQ subscales, including bedtime resistance (\( p = .010 \)), sleep anxiety (\( p = .003 \)), night awakenings (\( p = .043 \)), and parasomnias (\( p = .023 \)). No scores on the CSHQ signifying heightened risk on the sleep disordered breathing subscale (i.e., equal to or above seven out of nine) were identified for children who were not already being seen by a paediatric sleep or respiratory medicine specialist. Moreover, there were no differences on the subscales of sleep duration, sleep disordered breathing, or daytime sleepiness. Group scores also did not differ on the Sleep Hygiene Index, a 13-item index, in which higher scores suggested more maladaptive sleep hygiene (29.10 [5.8] vs. 28.19 [6.0], \( p = .479 \)).

**Secondary Analysis of Primary Question**

Other research questions aimed to describe associations and explore which individual, child, and environmental factors influenced sleep in family caregivers of CMT.
**Question 6.** Question six asked, “Across 1 week, was there a significant relationship between the mean (a) sleep quantity (TST-N) and (b) sleep fragmentation (NNA-O) in family caregivers of CMT and the following factors?” Simple linear regression analyses were completed and are reported in the tables below. Dependent variables (i.e., TST-N and NNA-O) were derived from actigraphy data and represent the means of each over available nights. Categorical variables were dummy coded, and referent groups are identified below (placed in parentheses), for these analyses.

**Caregiver factors.** Many caregiver factors were examined using linear regression to determine their relationships with objective sleep outcomes (see Table 10). Among family caregivers of CMT, only place of birth predicted TST-N, with Canadian born participants achieving a mean of 80 minutes more sleep (SE 22.93) than those who were born outside Canada ($p = .001$).
Table 10

Linear Regression Between Caregiver Factors and TST-N

<table>
<thead>
<tr>
<th>Caregiver Factors</th>
<th>B</th>
<th>SE</th>
<th>p value</th>
</tr>
</thead>
<tbody>
<tr>
<td>Age</td>
<td>2.38</td>
<td>1.54</td>
<td>.130</td>
</tr>
<tr>
<td>Gender (female)</td>
<td>63.62</td>
<td>35.34</td>
<td>.080</td>
</tr>
<tr>
<td>Marital status (partnered)</td>
<td>40.46</td>
<td>28.70</td>
<td>.167</td>
</tr>
<tr>
<td>Level of education (high school)</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>High school vs. elementary</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>High school vs. college/university</td>
<td>-25.33</td>
<td>31.40</td>
<td>.425</td>
</tr>
<tr>
<td>High school vs. graduate</td>
<td>-29.53</td>
<td>43.07</td>
<td>.494</td>
</tr>
<tr>
<td>Employment status</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Full-time vs. part-time</td>
<td>-14.29</td>
<td>37.42</td>
<td>.705</td>
</tr>
<tr>
<td>Full-time vs. unemployed</td>
<td>-33.13</td>
<td>30.04</td>
<td>.278</td>
</tr>
<tr>
<td>Full-time vs. self-employed</td>
<td>-40.00</td>
<td>51.43</td>
<td>.442</td>
</tr>
<tr>
<td>Full-time vs. maternity leave</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Full-time vs. retired</td>
<td>140.00</td>
<td>83.67</td>
<td>.103</td>
</tr>
<tr>
<td>Annual household income</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>&lt;$25,000 vs. $25,000–$39,000</td>
<td>-65.00</td>
<td>39.27</td>
<td>.107</td>
</tr>
<tr>
<td>&lt;$25,000 vs. $40,000–$59,000</td>
<td>21.00</td>
<td>40.65</td>
<td>.609</td>
</tr>
<tr>
<td>&lt;$25,000 vs. $60,000–$99,000</td>
<td>-52.11</td>
<td>38.16</td>
<td>.181</td>
</tr>
<tr>
<td>&lt;$25,000 vs. $100,000</td>
<td>8.13</td>
<td>39.27</td>
<td>.837</td>
</tr>
<tr>
<td>Ethnicity (Caucasian)</td>
<td>-83.24</td>
<td>58.78</td>
<td>.165</td>
</tr>
<tr>
<td>Born in Canada</td>
<td>79.69</td>
<td>22.93</td>
<td>.001</td>
</tr>
<tr>
<td>Length of time in Canada (&lt; 5 years)</td>
<td>95.29</td>
<td>65.28</td>
<td>.163</td>
</tr>
<tr>
<td># children &lt; 18 years in household</td>
<td>3.23</td>
<td>14.69</td>
<td>.827</td>
</tr>
<tr>
<td># adults &gt; 18 years in household</td>
<td>-8.25</td>
<td>14.69</td>
<td>.578</td>
</tr>
<tr>
<td>Duration of caregiving (months)</td>
<td>.43</td>
<td>.24</td>
<td>.082</td>
</tr>
<tr>
<td>Health variables</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>BMI</td>
<td>-4.83</td>
<td>2.64</td>
<td>.076</td>
</tr>
<tr>
<td>Chronic health condition</td>
<td>-32.52</td>
<td>28.96</td>
<td>.269</td>
</tr>
<tr>
<td># visits to HCPs in previous year</td>
<td>-2.50</td>
<td>5.35</td>
<td>.643</td>
</tr>
<tr>
<td>Sleep variables</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Sleep quality (PSQI)</td>
<td>-1.58</td>
<td>4.62</td>
<td>.973</td>
</tr>
<tr>
<td>Sleep hygiene (SHI)</td>
<td>-1.97</td>
<td>2.25</td>
<td>.388</td>
</tr>
<tr>
<td>Vigilance, intrusive thoughts</td>
<td>3.40</td>
<td>5.76</td>
<td>.558</td>
</tr>
</tbody>
</table>

Note. B = unstandardized coefficient, BMI = Body mass index, HCP = health care provider, PSQI = Pittsburgh Sleep Quality Index, SE = standard error, SHI = Sleep Hygiene Index

On analyses using NNA-O as the dependent variable, the only predictors for nighttime awakenings in family caregivers CMT identified were level of education and income (see Table 11). Those with college/university education as compared to those with high school, had a mean of 3 fewer (SE 1.43) nighttime awakenings (p = .052). Family caregivers of CMT with an
annualized family income of >$100,000 as compared with those with an annualized income <$25,000 had a mean of more than four fewer awakenings (SE 1.84) per night.

Table 11

*Linear Regression Between Caregiver Factors and Number of NNA-O*

<table>
<thead>
<tr>
<th>Caregiver factors</th>
<th>B</th>
<th>SE</th>
<th>p value</th>
</tr>
</thead>
<tbody>
<tr>
<td>Age</td>
<td>-.04</td>
<td>.08</td>
<td>.586</td>
</tr>
<tr>
<td>Gender (female)</td>
<td>-1.86</td>
<td>1.74</td>
<td>.292</td>
</tr>
<tr>
<td>Marital status (partnered)</td>
<td>-1.40</td>
<td>1.39</td>
<td>.320</td>
</tr>
<tr>
<td>Level of education</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>High school vs. elementary</td>
<td>-</td>
<td>-</td>
<td>-</td>
</tr>
<tr>
<td>High school vs. college/university</td>
<td>-2.88</td>
<td>1.43</td>
<td>.052</td>
</tr>
<tr>
<td>High school vs. graduate</td>
<td>-.98</td>
<td>1.97</td>
<td>.622</td>
</tr>
<tr>
<td>Employment status</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Full-time vs. part-time</td>
<td>1.01</td>
<td>1.82</td>
<td>.583</td>
</tr>
<tr>
<td>Full-time vs. unemployed</td>
<td>1.51</td>
<td>1.46</td>
<td>.310</td>
</tr>
<tr>
<td>Full-time vs. self-employed</td>
<td>4.82</td>
<td>2.50</td>
<td>.062</td>
</tr>
<tr>
<td>Full-time vs. maternity leave</td>
<td>-</td>
<td>-</td>
<td>-</td>
</tr>
<tr>
<td>Full-time vs. retired</td>
<td>.84</td>
<td>4.07</td>
<td>.839</td>
</tr>
<tr>
<td>Household income</td>
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<td></td>
<td></td>
</tr>
<tr>
<td>&lt;$25,000 vs. $25,000–$39,000</td>
<td>1.55</td>
<td>1.78</td>
<td>.391</td>
</tr>
<tr>
<td>&lt;$25,000 vs. $40,000–$59,000</td>
<td>-.65</td>
<td>1.84</td>
<td>.727</td>
</tr>
<tr>
<td>&lt;$25,000 vs. $60,000–$99,000</td>
<td>-2.11</td>
<td>1.73</td>
<td>.231</td>
</tr>
<tr>
<td>&lt;$25,000 vs. ≥ $100,000</td>
<td>-4.29</td>
<td>1.78</td>
<td>.021</td>
</tr>
<tr>
<td>Race (Caucasian)</td>
<td>3.53</td>
<td>2.83</td>
<td>.219</td>
</tr>
<tr>
<td>Born in Canada</td>
<td>-1.20</td>
<td>1.24</td>
<td>.341</td>
</tr>
<tr>
<td>Length of time in Canada &lt; 5 years</td>
<td>-1.32</td>
<td>2.70</td>
<td>.630</td>
</tr>
<tr>
<td># children &lt; 18 years in household</td>
<td>.78</td>
<td>.69</td>
<td>.267</td>
</tr>
<tr>
<td># adults &gt; 18 years in household</td>
<td>-.46</td>
<td>.70</td>
<td>.518</td>
</tr>
<tr>
<td>Duration of caregiving in months</td>
<td>.00</td>
<td>.01</td>
<td>.984</td>
</tr>
<tr>
<td>Health variables</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>BMI</td>
<td>.011</td>
<td>.132</td>
<td>.931</td>
</tr>
<tr>
<td>Chronic health condition</td>
<td>1.26</td>
<td>1.39</td>
<td>.373</td>
</tr>
<tr>
<td># visits to HCPs in previous year</td>
<td>.31</td>
<td>.25</td>
<td>.228</td>
</tr>
<tr>
<td>Sleep variables</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Sleep quality (PSQI)</td>
<td>-.004</td>
<td>.19</td>
<td>.985</td>
</tr>
<tr>
<td>Sleep hygiene (SHI)</td>
<td>.14</td>
<td>.11</td>
<td>.189</td>
</tr>
<tr>
<td>Vigilance, intrusive thoughts</td>
<td>.07</td>
<td>.28</td>
<td>.806</td>
</tr>
</tbody>
</table>

*Note.* B = unstandardized coefficient, SE = standard error, referent groups in (parentheses). BMI = Body Mass Index, HCP = health care provider, PSQI = Pittsburg Sleep Quality Index, SHI = Sleep Hygiene Index.

*Child factors.* The TST-N improved by a mean of almost six minutes of sleep per night (SE 2.4) with every year of increasing age of the CMT (*p* = .029). Family caregivers of CMT
achieved a mean of 4 minutes less (SE 1.48) sleep per night per increasing hour of reported
daytime technology use ($p = .019$; see Table 12).

Table 12

*Linear Regression Between Child Factors and TST-N*

<table>
<thead>
<tr>
<th>Child factors</th>
<th>B</th>
<th>SE</th>
<th>p value</th>
</tr>
</thead>
<tbody>
<tr>
<td>Child’s age</td>
<td>5.55</td>
<td>2.44</td>
<td>.029</td>
</tr>
<tr>
<td>Gender (female)</td>
<td>-.84</td>
<td>26.42</td>
<td>.975</td>
</tr>
<tr>
<td>Neurocognitive delay or impairment</td>
<td>83.51</td>
<td>83.10</td>
<td>.321</td>
</tr>
<tr>
<td># hrs technology use—nighttime/wk</td>
<td>.40</td>
<td>1.62</td>
<td>.804</td>
</tr>
<tr>
<td># hrs technology use—daytime/wk</td>
<td>-3.63</td>
<td>1.48</td>
<td>.019</td>
</tr>
<tr>
<td>Sleep problems per CSHQ</td>
<td>-2.23</td>
<td>1.42</td>
<td>.126</td>
</tr>
</tbody>
</table>

*Note.* B = unstandardized coefficient, SE = standard error. CSHQ = Child Sleep Habits Questionnaire (CSHQ), overall total score ranging from 33–99, with higher score suggesting more sleep disturbance.

Notably, no significant relationships were found between child variables and the number
of nocturnal awakenings (see Table 13).

Table 13

*Linear Regression Between Child Factors and NNA-O*

<table>
<thead>
<tr>
<th>Child factors</th>
<th>B</th>
<th>SE</th>
<th>p value</th>
</tr>
</thead>
<tbody>
<tr>
<td>Child’s age</td>
<td>-.012</td>
<td>.12</td>
<td>.923</td>
</tr>
<tr>
<td>Gender (female)</td>
<td>-1.39</td>
<td>1.25</td>
<td>.368</td>
</tr>
<tr>
<td>Neurocognitive delay or impairment</td>
<td>.88</td>
<td>4.03</td>
<td>.827</td>
</tr>
<tr>
<td># hours technology use—nighttime/wk</td>
<td>-.04</td>
<td>.08</td>
<td>.635</td>
</tr>
<tr>
<td># hours technology use—daytime/wk</td>
<td>.03</td>
<td>.08</td>
<td>.675</td>
</tr>
<tr>
<td>Sleep problems per CSHQ</td>
<td>.03</td>
<td>.08</td>
<td>.711</td>
</tr>
</tbody>
</table>

*Note.* B = unstandardized coefficient, SE = standard error. CSHQ = Child Sleep Habits Questionnaire (CSHQ), overall total score ranging from 33–99, with higher score suggesting more sleep disturbance.

*Environmental factors.* In family caregivers of CMT, there were no significant
relationships between any of the environmental factors and TST-N (see Table 14).
Table 14

Linear Regression Between Environmental Factors and TST-N

<table>
<thead>
<tr>
<th>Environmental factors</th>
<th>B</th>
<th>SE</th>
<th>p value</th>
</tr>
</thead>
<tbody>
<tr>
<td>Use of oxygen saturation monitor</td>
<td>10.12</td>
<td>26.27</td>
<td>.702</td>
</tr>
<tr>
<td>Sleep ecology (own bed, own room, alone or with spouse)</td>
<td>.95</td>
<td>2.05</td>
<td>.647</td>
</tr>
<tr>
<td># homecare hrs/in-home/approved/wk</td>
<td>.10</td>
<td>.53</td>
<td>.845</td>
</tr>
<tr>
<td># homecare hrs/in-home/used/wk</td>
<td>.22</td>
<td>.59</td>
<td>.715</td>
</tr>
<tr>
<td># homecare hrs/out-of-home/approved/wk</td>
<td>.70</td>
<td>.82</td>
<td>.400</td>
</tr>
<tr>
<td># homecare hrs/out-of-home/used/wk</td>
<td>.93</td>
<td>.82</td>
<td>.266</td>
</tr>
<tr>
<td># nocturnal homecare hrs used/wk</td>
<td>.55</td>
<td>.58</td>
<td>.343</td>
</tr>
</tbody>
</table>

Note. B = unstandardized coefficient, SE = standard error.

Only the number of homecare hours approved per week predicted NNA-O, such that for every additional hour approved for out-of-home homecare (e.g., for nursing shifts at school), family caregivers of CMT had a mean of less than one additional awakening per night (SE .04, p = .053; see Table 15).

Table 15

Linear Regression Between Environmental Factors and NNA-O

<table>
<thead>
<tr>
<th>Environmental factors</th>
<th>B</th>
<th>SE</th>
<th>p value</th>
</tr>
</thead>
<tbody>
<tr>
<td>Use of oxygen saturation monitor</td>
<td>-.29</td>
<td>1.26</td>
<td>.819</td>
</tr>
<tr>
<td>Sleep ecology (own bed, own room, alone or with spouse)</td>
<td>-.02</td>
<td>.10</td>
<td>.824</td>
</tr>
<tr>
<td># homecare hrs/in-home/approved/week</td>
<td>.01</td>
<td>.03</td>
<td>.788</td>
</tr>
<tr>
<td># homecare hrs/in-home/used/week</td>
<td>.01</td>
<td>.03</td>
<td>.665</td>
</tr>
<tr>
<td># homecare hrs/out-of-home/approved/week</td>
<td>.08</td>
<td>.04</td>
<td>.053</td>
</tr>
<tr>
<td># homecare hrs/out-of-home/used/week</td>
<td>.07</td>
<td>.04</td>
<td>.083</td>
</tr>
<tr>
<td># nocturnal homecare hrs used/week</td>
<td>&lt;.001</td>
<td>.03</td>
<td>.996</td>
</tr>
</tbody>
</table>

Note. B = unstandardized coefficient, SE = standard error.

**Question 7.** Question 7 asked, “Which combination of ‘modifiable’ caregiver (i.e., sleep hygiene), child (i.e., sleep problems), and environmental (i.e., nocturnal homecare) factors and sociodemographic risk factors (i.e., caregiver age, gender, and marital status) best predicted the objectively measured (a) sleep quantity (TST-N) and (b) sleep fragmentation (NNA-O) in family caregivers of CMT?”
Multiple linear regression analyses were completed to explore the relationship between modifiable (caregiver, child, environmental factors) and non-modifiable (i.e., sociodemographic) variables and objectively measured sleep outcomes (i.e., TST-N and NNA-O). As multiple regression analysis is sensitive to extreme values, the outlier (118 mins) for average TST-N per night in FC CMT was removed, but overall results were not found to change ($p = .13$), thus the extreme value was left in.

**Model 1.** In model 1, the TST-N (dependent variable) in minutes was regressed on the independent variables of: caregiver: 1) sleep hygiene, child: 2) child sleep problems and environmental: 3) nocturnal use of homecare, and sociodemographics: 4) age, 5) gender, and 6) marital status (see Table 16). Overall the combination of predictor factors (independent variables) was not found to have a significant relationship with TST-N ($R^2 = .28$, $F(6,31) = 1.98$, $p = .099$). Family caregiver gender approached significance for predicting TST-N, such that female caregivers achieved a mean total sleep time of 85.2 more minutes per night (SE 43.51) than male caregivers ($p = .059$). Family caregiver age also approached significance such that 3 minutes more sleep was achieved per night (SE 1.75) for every one year of increase in age ($p = .062$).

Table 16

*Regression: TST-N on Multiple Modifiable and Non-modifiable Factors*

<table>
<thead>
<tr>
<th>Independent variables</th>
<th>B</th>
<th>SE</th>
<th>$p$ value</th>
</tr>
</thead>
<tbody>
<tr>
<td>Sleep hygiene (caregiver)</td>
<td>-2.07</td>
<td>2.38</td>
<td>.390</td>
</tr>
<tr>
<td>Sleep problems (child)</td>
<td>-.33</td>
<td>1.56</td>
<td>.834</td>
</tr>
<tr>
<td>Environmental: Hrs nocturnal homecare/week</td>
<td>.88</td>
<td>.59</td>
<td>.143</td>
</tr>
<tr>
<td>FC gender (female)</td>
<td>85.24</td>
<td>43.51</td>
<td>.059</td>
</tr>
<tr>
<td>FC marital status (partnered)</td>
<td>-27.79</td>
<td>29.39</td>
<td>.352</td>
</tr>
<tr>
<td>FC age</td>
<td>3.38</td>
<td>1.75</td>
<td>.062</td>
</tr>
</tbody>
</table>

*Note.* Independent variable, entered in a block, * $p < .05$, ** $p < .01$. B = unstandardized coefficient, FC = family caregiver, SE = standard error.
Model 2. In model 2, NNA-O (dependent variable) in minutes was regressed on the independent variables of: caregiver: 1) sleep hygiene, child: 2) child sleep problems and environmental: 3) nocturnal use of homecare and sociodemographics: 4) age, 5) gender, and 6) marital status (see Table 17). Overall the combination of predictor factors (independent variables) was not found to have a significant relationship with NNA-O ($R^2 = .179$, Adjusted $R^2 = .172$, $F(6,31) = 1.07, p = .400$). Only family caregiver gender was found to predict objectively measured sleep quality (i.e., NNA-O), such that male caregivers had a mean of four and a half more nocturnal awakenings per night (SE 2.21) than female caregivers ($9.58±2.8$ [range 7-14] male vs. $7.72±4.1$ [range 2-20] female, $p = .052$).

Table 17

Regression: NNA-O on Multiple Modifiable and Non-modifiable Factors

<table>
<thead>
<tr>
<th>Independent variables</th>
<th>B</th>
<th>SE</th>
<th>p value</th>
</tr>
</thead>
<tbody>
<tr>
<td>Sleep hygiene (caregiver)</td>
<td>.17</td>
<td>.12</td>
<td>.166</td>
</tr>
<tr>
<td>Sleep habits (child)</td>
<td>-.07</td>
<td>.08</td>
<td>.413</td>
</tr>
<tr>
<td>Environmental: Hours nocturnal homecare/week</td>
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<td>.08</td>
<td>.525</td>
</tr>
<tr>
<td>FC gender (female)</td>
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<td>2.21</td>
<td>.052</td>
</tr>
<tr>
<td>FC marital status (partnered)</td>
<td>-1.76</td>
<td>1.49</td>
<td>.248</td>
</tr>
<tr>
<td>FC age</td>
<td>-.09</td>
<td>.09</td>
<td>.310</td>
</tr>
</tbody>
</table>

Note. Independent variable, entered in a block. B = unstandardized coefficient, FC = family caregiver, SE = standard error.

Study feasibility and acceptability.

Study recruitment was expected to occur within six months, but took 7 months owing mainly to challenges with recruitment of family caregivers of adolescents to the control cohort.

Participants in this study ($n = 12$ FC CMT, $n = 18$ FC Control) indicated in a final question on the sleep diary—”What did you like most about this study?”—that they appreciated participating in the study and that it helped them become more aware of their own sleep and healthy sleep practices. Many participants volunteered that completing the sleep diary was quick and estimated that it took less time (i.e. <15 mins) than expected to complete. However, a few family caregivers provided feedback ($n = 4$ family caregiver CMT, $n = 3$ family caregiver controls) that
completing the sleep diary was “the part of the study they liked the least.” At study conclusion, the administration of validated questionnaires during a home visit was expected to take less than 30 minutes, and this was found to be achievable in the vast majority of cases.
Chapter 5: Discussion

This chapter begins with a discussion of the study’s primary research question, which addressed the difference in sleep between family caregivers of CMT and family caregiver controls. The study was powered to identify a clinically important difference between the groups on the primary outcome of total sleep time—nocturnal (TST-N). This result, and that of other sleep outcomes, is discussed in light of existing literature, including that summarized in a recent systematic review of studies reporting on sleep outcomes in family caregivers of CMT (Keilty et al., 2014). Immediately following that, this chapter will discuss the secondary research questions, which examined the relationship between sleep and related health outcomes in family caregivers of CMT. To conclude the discussion of research questions, a discussion of more exploratory questions is provided, including which factors may have influenced sleep disturbance in family caregivers of CMT. Finally, the limitations and strengths of the study are presented.

Primary Research Question

Differences in sleep quantity.

Nighttime sleep. The primary aim of this study was to examine the difference in total sleep time—nocturnal (TST-N) between family caregivers of CMT and family caregiver controls. The family caregivers of CMT achieved a full 40 minutes less sleep per night than controls and averaged only six and a half hours of sleep per night (6.56 [1.4] vs. 7.21 [0.6], \( p = .007 \)). While the difference between the cohorts was less than what was established a priori as a minimal important difference (1 hour; Meltzer et al, 2006), this study confirms that family caregivers of CMT did not get as much sleep as their peers with healthy children and importantly less than what is recommended for healthy middle-aged adults (National Sleep Foundation, 2005). As such, findings from this study can be considered clinically meaningful and therefore
suggest a reason for intervention or change (Guyatt, et al. 2002).

Family caregivers of CMT achieved a total sleep time below the recommended cut off (7–9 hrs) for healthy middle-aged adults (National Sleep Foundation, 2005). Public health campaigns emphasize that when sleep drops below this level, it generates concerns for the health of the individual and the safety of the population. Family caregivers of CMT achieved almost a full 2 hours less sleep per night than that self-reported in the most recent nation-wide poll that examined sleep in adult Canadians (n = 9500) with one child (8.01hrs) or more (7.88hrs; Statistics Canada, 2005; http://www.statcan.gc.ca/pub/11-008-x/2008001/article/10553-eng.htm#11). More recently, 250 Canadian adults responding to an international poll (National Sleep Foundation, 2013) self-reported getting approximately one hour more sleep per night than that measured in family caregivers of CMT in this study. Thus, family caregivers of CMT appear to achieve less nighttime sleep than that consistently self-reported by the average Canadian adult, with and without children. Importantly, and by comparison, family caregiver controls in this study achieved a total sleep time within the range typically recommended for healthy middle age adults.

Notably no directly comparable data have been identified in earlier studies of family caregivers of CMT (Keilty et al., 2014). The evidence that served as the basis for this study’s sample size calculation was based on a single study using self-report measures, but considered to be the strongest study in the field (used a comparator group) at the time (Meltzer et al, 2006). To compare, a recent meta-analysis of studies in adults with insomnia, a 30-minute increase in self-reported total sleep time was appraised as a meaningful improvement (Okajima, et al., 2011). In other studies of adult family caregivers, similarly small improvements in caregiver sleep duration have been associated with positive improvements in sleep-related outcomes such
as caregiver depression (Carter, 2006; Carter et al., 2009), anxiety (Akkerman & Ostwald, 2004), and daytime sleepiness (McCurry et al., 2005).

To appreciate the magnitude of a 40 minute difference in TST-N, consider that family caregivers of CMT in this study, on average, provided homecare for their technology-dependent child for six years (6.01 [4.5] yrs); as a consequence, they each achieved over the same time period, the equivalent of approximately 208 fewer nights of sleep (i.e., calculated at, on average, 280 fewer minutes of sleep each week) than that of family caregivers of healthy same-aged children. The difference in total sleep time between the cohorts represents a massive amount of accumulated sleep debt (i.e., the difference between how much sleep one needs and how much one gets) in the successive years of extraordinary caregiving in family caregivers of CMT versus the comparator cohort.

Very few studies have reported on total sleep time, day or night, in studies of family caregivers of children with chronic illness, disability, and/or medical complexity. In the single out-patient pilot study that reported TST-N using objective measurement, parents of children with cancer were reported to have a wide range in TST-N (4.5–7.8hrs), but average sleep time was not reported (Gedaly-Duff et al., 2006). These data were derived from a very small sample size (n = 13) and only three nights of actigraphy. While some similarities between the participants existed, quantity of sleep in the Gedaly-Duff et al. (2006) study was reported during an acute phase of the child’s cancer treatment that is typically associated with crisis and uncertainty about disease trajectory. In contrast, sleep in this study was measured once chronic care in the home for the CMT was established.

Two studies have described self-reported sleep times in family caregivers of children with chronic medical conditions that either approximated or exceeded the sleep time measured
(objectively) in family caregivers of CMT in this study. These studies did not report a TST for the entire study period as was the case in this study. In both studies sleep duration was reported to vary with family caregivers getting less sleep on the weekday than on the weekend. In parents of children with cancer, family caregivers got 6.8 [1.3] hours of sleep on weekdays and 7.2 [1.0] hours on the weekends (Wright, 2011). Another investigator reported on findings of an observational study and reported a similar pattern to the self reported sleep in family caregivers of children with physical disabilities (6.5 [1.3] hrs; 6.9 [1.5]; Wright et al., 2006).

Only one study identified a TST-N in a clinical sample of family caregivers (i.e., children with chronic illness) that was shorter than that in this study (4.5 hrs/night; Cottrell and Khan, 2005). These authors described the subjective appraisal of sleep in parents (n = 50) of children with epilepsy who reported poorer sleep outcomes associated with higher frequency (r = .32, p < .05) and severity (r = .42, p < .05) of the child’s seizure activity. A marked limitation to this study design (Cottrell and Khan, 2005) included that sleep duration was measured using a single self-report item that asked about ‘usual sleep time’.

Sleep duration in family caregivers of CMT has been previously reported in three diverse studies, none of which employed objective measures of sleep (see Table 3). In the first, Feeley et al. (2014) reported an average sleep duration that was a full 48 minutes less than that measured in this study. These family caregivers had young children with a mean age of only 13.4 months who required home oxygen, which has been characterized as a demanding caregiving responsibility (McLean, Townsend, Clark, et al., 2000). It is also well known, however, that new parents of healthy infants commonly experience highly disturbed sleep (Lee & DeJoseph, 1992; Hunter, Rychnovsky, Yount, 2009; Nishihara, Horiuchi, Eto, Uchida, 2000; Insana, Williams, & Montgomery-Downes, 2013; Stremler et al., 2006). Hence, family caregivers of CMT with
infants under 12 months of age were excluded from this study, rendering it difficult to compare findings to those reported by Feeley et al. (2014).

The studies conducted by Meltzer and colleagues (Meltzer et al., 2010; Meltzer & Mindell, 2006) are the two best comparators to this study in terms of participant characteristics. In the first of the studies, TST-N was reported to be very similar (6.31 [1.2] hours; Meltzer & Mindell, 2006) to this study, and in the second, although the range was reported (5.98–6.86 h/night), the study omitted reporting the mean TST-N (Meltzer et al., 2010). Thus, while these two studies included participants with many of the same caregiver (e.g., level of education), child (e.g., age), and environmental characteristics (e.g., use of homecare) as those in this study, the comparisons on sleep outcomes are hampered owing to differences in the choice of variables and methods of measurement and reporting. Moreover, the studies by Meltzer and colleagues (2006; 2010) were highly prone to recall bias and other threats to validity, including social acceptability bias, as have most studies of sleep and health-promoting behaviours that have relied entirely on subjective self-report.

The larger number of consecutive nights with wide variability in TST-N for family caregivers of CMT suggests that they had a mean (SD) sleep duration that was often too short or too long than ideal (family caregivers of CMT 1.33 [1.5] vs. family caregiver controls 0.45 [0.7], \( p = 0.001 \)). While there is individual variation in how much sleep is required, it is recognized that good sleep hygiene practices include getting to sleep and getting up at around the same time each day (Mastin et al., 2006). Wide individual variability in sleep times between consecutive nights may signal and/or contribute to problems with disorganized circadian rhythms (Borbely, 1982; Czeisler, Weitzman, Moore-Ede, Zimmerman & Knauer, 1980). An approximate 30% difference (i.e., three hour difference) in sleep duration among healthy adults in clinical (family caregivers
of children in critical care) and laboratory (healthy adult volunteers) samples has been found to be associated with increased daytime sleepiness and fatigue (Stremler et al., 2013; Taylor et al., 2008). In future studies, it will be important to better understand the relationship between night-to-night variation in total sleep time and midpoint in sleep time (measured by actigraphy) in family caregivers of CMT to better describe the potential disorganization of their circadian rhythms, providing a potential target for sleep intervention trials (Ancoli-Israel et al., 2003).

**Daytime sleep.** In this study, the majority of family caregivers took at least one nap per week. In a review of the benefits of napping, Milner & Cote (2009) summarized the literature and described that naps of approximately 20–30 minutes in length were beneficial to shift-workers’ performance on vigilance (Purnell, Feyer, & Herbison, 2002) and psychomotor speed tests (Smith et al., 2007). Also summarized are findings from laboratory-based controlled studies, in which daytime naps of a minimum of 10 minutes among healthy adult volunteers resulted in better scores on performance-based outcomes, including accuracy (Takahashi & Arito, 2000) and reaction times (Betrus, 1986). Thus, family caregivers of CMT with nap lengths of more than 20 minutes (0.24 [0.3]) and twice as long as their comparators (0.10 [0.1], \( p = .004 \)) may find their daytime sleep to be a practical solution to enhance daytime function and performance. On the other hand, family caregivers of CMT in this study were more likely to be unemployed and therefore may simply have had more time at home to nap than their comparators. What is not known is how often the naps taken by participants in this study were too long, potentially resulting in sleep inertia (Ruggierio & Redeker, 2014), or too close to bedtime, such that they could have negatively interfered with their homeostatic sleep drive (i.e., Process S) and thus their ability to fall asleep or stay asleep at night (Dhand & Sohal, 2006). No other studies have reported on the amount of daytime sleep (measured objectively or
subjectively) in family caregivers of CMT. In future studies of sleep in family caregivers of CMT that use actigraphy, it will be important to better explore the impact of napping on overall sleep-wake patterns and daytime performance in family caregivers of CMT.

**Longest sleep session.** This study found a large difference between the two cohorts on the longest sleep session at night, a variable linked to the restorative benefits of sleep. In this study, family caregivers of CMT had much shorter continuous sleep sessions at night (30% shorter, by a full 75 minutes) than family caregiver controls \((p = .001)\) and only slightly longer than that found among post-partum women (Stremler et al., 2006). While not precisely characterized, laboratory studies suggest that when sleep is consolidated enough to enable passing through 6–8 full sleep cycles in a night, it assists in processing events of the day before and optimally preparing the mind and body for the demands of the next day (Carskadon & Dement, as cited in Kryger et al., 2010 Maquet, 2001).

Family caregivers of CMT achieved, on average, only 8 minutes more of consolidated sleep during their daytime napping periods than controls \((14.47 [15.4] \text{ vs. } 6.17 [8.0], p = .004)\). While the difference in longest sleep session in the daytime is small, the finding may be physiologically important, because, as described previously in this chapter, naps lasting longer than 10 minutes may improve feelings of restoration and daytime performance (Dhand & Sohal, 2006). In future research, it will be important to examine napping behaviours in family caregivers of CMT and its relationship to sleep wake cycles, circadian rhythms (melatonin levels), daytime performance (objective and subjective), and related outcomes, such as depression.

**Overall sleep quantity.** Collectively, the findings from this study strongly support the hypotheses put forward at the outset of this investigation and confirm that sleep in family
caregivers of CMT is of lesser quantity than sleep in family caregivers of healthy children. It has provided reliable information about total sleep time in family caregivers of CMT and their comparators, based on actigraphy rather than self-report. The study concludes that family caregivers of CMT do not achieve adequate sleep quantity and it is the first that we know of to report on sleep consolidation (longest sleep sessions, day or night) in family caregivers of children with medical complexity.

**Question 2**

**Difference in sleep deprived nights.** Importantly, this study suggests that family caregivers of CMT are at very high risk for the negative effects of acute sleep deprivation. During the study period, it found that five times as many family caregivers of CMT averaged under 6 hours of sleep per night compared to family caregiver controls (27.5%, \( n = 11 \) vs. 5.0%, \( n = 2 \)), with a 4-fold higher number of sleep-deprived nights per week (2.18 [2.4] vs. 0.60 [.8], \( p < .001 \)). Considering that sleeping under 7–8 hours per night has been found to predict obesity, diabetes, hypertension, and cardiovascular disease (Buxton & Marcelli, 2010), this study signals alarm for the long-term health, and thus capacity for caregiving, of family caregivers of CMT.

When sleep is routinely restricted to a level below what is needed, chronic sleep deprivation may occur. The vast majority of family caregivers of CMT in the study indicated that their sleep during the study period was typical, suggesting that chronic sleep deprivation may be a prevalent problem in family caregivers of CMT. Decrements in performance due to chronic sleep deprivation appear to accumulate, and the negative effects of sleep loss may not be appreciable to the individual (Van Dongen, Maislin, Mullington, & Dinges, 2003). The effects of chronic sleep restriction in family caregivers of CMT have not been systematically studied. Anecdotally, family caregivers of CMT have described being overwhelmed as a consequence of
their chronic sleep deprivation. In studies of family caregivers of children with disabilities, a similarly burdened population, sleep disturbances have been described as features of the caregiver experience that were associated with impaired family function, including strained parenting and spousal relationships (Gallagher et al., 2010). Thus, in future research, it will be important to examine sleep deprivation as it relates to the caregiver experience and their daytime performance, capacity for parenting, and the health and safety of the CMT.

Nocturnal awakenings. In population-based studies, almost 50% of family caregivers have reported that their sleep is disrupted on average twice per week because their child wakes them during the night (National Sleep Foundation, 2004). In this study, family caregivers self-reported nocturnal awakenings in over 80% of the observed nights (81.6%), and family caregivers of CMT were consistently found to wake up more often at night than family caregiver controls (actigraphy: 8.00 [3.9] vs. 6.01 [3.7], \( p = 0.02 \); diarized data: 2.35 [2.0] vs. 1.42 [1.5], \( p < .001 \)). There are no population-based studies that have reported on objectively measured awakenings in family caregivers of child-bearing age. In their clinical study, Gedaly-Duff et al. (2006) reported that community-based participants experienced very frequent awakenings (16.9 [74] and 19.7 [7.6] respectively). These data, however, are based on a very small sample size \( n = 6 \) mothers, \( n = 7 \) fathers) and only three nights of actigraphy, and like their reported TST-N, the number appears to fall well outside of expected norms for healthy adults. Because Gedaly-Duff et al. (2006) have not described all the necessary parameters in reporting on the use of actigraphy (Morgenthaler et al., 2007), it is not possible to rule out the likelihood that the observed differences reported in their study were due to differences in methods of actigraphy data collection or analysis.
In seeking an alternative comparator, it is notable that the number of objectively measured nocturnal awakenings found in this study is very similar to that recently reported among family caregivers of critically-ill hospitalized children using the same actiwatch devices on the same settings as in this study (7–8 times per night, averaged over 5 nights; Stremler et al., 2014). In future analysis of the data set from this study, it may be possible to characterize the awakenings experienced by family caregivers of CMT (e.g. what woke them), such that targeted interventions may be developed.

Family caregivers of CMT underestimated the number of arousals they experienced at night almost four-fold, and family caregiver controls over-estimated the number at a similar level, when comparing their subjective recall with objective measurement. Evidence suggests that even brief arousals, if frequent enough, can negatively influence sleep quality (Freedman & Roehrs, 2007) and optimal daytime performance (Chesire, Engleman, Deary, Shapiro & Douglas, 1992). The difference between perceived and actual numbers of arousals signals that subjective recall of awakenings in family caregivers is not reliable. This may pose a challenge in clinical practice, such that family caregivers of CMT may deny sleep problems and refuse or fail to pursue additional nighttime supports. In future development and testing of sleep-promoting interventions, this tendency towards under-appreciating nocturnal awakenings needs to be considered.

**Sleep onset latency.** The recommendation for sleep onset latency is 15 minutes (AASM, 2010). Family caregivers of CMT in this study documented that it took them more than 30 minutes to fall asleep twice as often compared to family caregiver controls ($n = 68$ nights/280 possible nights, 24% vs. $n = 33/280$ possible nights, 12%). There are no data however from
which to draw direct comparisons of sleep onset latency in family caregivers of CMT because of differences in measurement and reporting in the related studies.

When an adult thinks that it takes more than 20–30 minutes for them to fall asleep, they typically characterize the experience as distressing. Individuals diagnosed with select sleep disorders (e.g., insomnia) frequently complain of difficulty with sleep onset even when it is observed to be within normal limits (Lund & Browman, 2000). In this study among family caregivers of CMT, sleep onset latency was often reported to be longer than 30 minutes and may part have been due to both intrinsic (e.g., worry) and extrinsic (e.g., overly bright sleep environment) factors. In this study, family caregivers of CMT reported that they had “negative thoughts and feelings that kept them awake” on average about twice a week (2.02 [2.4]). In future analysis of existing sleep data from this study, it may be possible to further characterize sleep onset latency and explore in more detail what kept family caregivers of CMT awake at night.

**Sleep quality.** While the amount of sleep achieved is important in terms of overall restoration, the perceived quality of sleep is generally recognized as an important feature of an overall sense of well being and quality of life. In normal sleepers, sleep quality has been characterized as being associated with “alertness on waking and throughout the day, feeling rested and restored on waking, and the number of awakenings they experienced in the night” (Harvey et al., 2008, p. 390).

Almost twice as many family caregivers of CMT were classified as poor sleepers (73.8%, n = 31) compared to family caregiver controls (39.5%, n = 17). The mean global sleep quality score among family caregivers of CMT was also higher than that among controls (7.75 [2.9] vs. 5.45 [2.8], p = 0.001). These findings are consistent with those in the three other studies of sleep
quality in family caregivers of CMT that have all reported elevated scores on the PSQI (range 7.4–10.7; Feeley et al., 2014; Meltzer & Mindell, 2006; Read et al., 2010). In the one other controlled study, sleep quality was poorer among family caregivers of CMT versus family caregiver controls, and both groups scored above the cut-off for poor sleepers (10.7 [3.7] vs. 5.94 [3.2], p < .05; Meltzer & Mindell, 2006). Thus, the findings from this study affirm those of others that have used the PSQI in family caregivers of CMT. Importantly, poor sleep quality is a hallmark of those who suffer from chronic insomnia (Edinger, et al., 2004), and thus, the findings of this study call for future investigations that examine the role that insomnia plays in the experience of family caregivers of CMT.

In summary, this study has examined the difference in sleep outcomes between family caregivers of CMT and family caregiver controls. It has examined sleep using both objective (i.e., actigraphy) and subjective (i.e., sleep diaries, PSQI) measures. Study findings strongly indicate that while sleep in family caregivers of children is not always ideal, in keeping with the proposed hypotheses, sleep in family caregivers of CMT is worse in terms of both quality and quantity compared to family caregivers of healthy children, even when accounting for the child’s age. In the next section, the outcomes associated with sleep in this study will be discussed.

Secondary Research Questions

A secondary but important aim of this study was to examine whether there is a difference between the cohorts on select outcomes often associated with sleep quality and quantity. This section of the chapter will discuss how the outcomes of sleepiness, fatigue, depression, and quality of life differed between the cohorts. Finally, other group differences on modifiable risk factors for sleep will be discussed.

Health related quality of life. Self-reported health-related quality of life is described as
a measure of ‘what matters most’ to individuals living with and affected by chronic health conditions (Guyatt, et al., 1993). Health-related quality of life was measured to be slightly below the expected population mean (50 [10]) that was found in earlier validation studies (Ware et al., 1996). Both groups, however, scored similarly on summary scores of the SF-12™, for physical functioning (41.81 [5.8] vs. 42.18 [5.0], \( p = .647 \)) and mental functioning (47.25 [7.5] vs. 49.81 [5.2], \( p = .072 \)). Determining a direct comparator for these results is challenging, since no other study among family caregivers has used the SF-12™ to examine this construct. Generally, studies of family caregivers have described sleep as it relates to quality of life outcomes using the SF-36® (Caqueo-Urizar et al., 2009; Cottrell & Khan, 2005; Lee et al., 2007; McKeown et al., 2003; Read et al., 2010) or other disease specific measures (Feeley et al., 2013; Lewis-Jones et al., 2001). In a heterogenous sample, Hatzmann and colleagues (2008) compared self-reported health-related quality of life between parents of children with chronic health conditions (including those with similar conditions to those in this study) and healthy children, and they also found differences in the expected direction (TNO-AZL Questionnaire for Adult’s Health Related Quality of Life.; \( p = .004 \)). These authors noted that poorer sleep in family caregivers of children with chronic conditions represented the largest difference found between cohorts (\( p < .05 \); Hatzmann, Heymans, Ferrer, van Praag, & Grootenhuis, 2008).

In this study, little to no difference was found between the family caregiver cohorts in how they perceived their physical, emotional, and social well-being. It is plausible that a relationship was not identified due to the choice of measurement tool: the extent to which SF-12™ scores reflects the scores on the more widely used Short Form (36) Health Survey [SF-36™] has not been well studied (Riddle, Lee, & Stratford, 2001). What is known is that the earliest validation studies reported the standard errors to be nearly always larger for the SF-12™
when compared to the SF-36™ (Ware et al., 1996) and Canadians have scored higher than their US counterparts on all, and their UK counterparts on most, domains and sub-scores of the SF-36™ (Riddle, et al., 2001). Importantly, the more comprehensive SF-36™ includes multiple indicators of mental health (e.g., happy, blue/sad) that are not included in the SF-12™ and, when scored, these have been found in clinical studies to be useful in screening for depression and other psychiatric disorders (Berwick et al., 1991; Ware, Kosinski, & Keller, 1994b).

Notwithstanding the potential limitations of the SF-12™, and considering the relationship between sleep and health-related quality of life (Szentkirályi et al., 2009), it remains difficult to explain why differences were not found in this study between the two groups on these quality of life variables. It is plausible the one or both of the study samples was affected by participant bias (those perceiving a higher quality of life more inclined to participate). In future studies of sleep and related outcomes in family caregivers of CMT, it is suggested that that the more widely used SF-36™ be employed and the relationship between sleep and HRQoL continue to be explored.

**Depression.** Sleep disturbance and depression have been associated in earlier studies of sleep in family caregivers of CMT (Meltzer et al., 2010; Meltzer & Mindell, 2006; Read et al., 2010) and family caregivers of children with chronic illness (Yilmaz et al., 2008; Yuksel et al., 2007). Depression scores on the CES-D global depression score among family caregivers of CMT in this study were above historical norms in healthy adults (Radloff, 1977) and higher than in the family caregiver controls (11.70 [8.7] vs. 6.95 [6], p = .006). While it is suggestive of but not diagnostic for clinical depression, it is still alarming that almost three times as many family caregivers of CMT scored in the range for clinically meaningful depression (n = 11, 26.2% vs. n = 4, 9.3%). These data suggest that family caregivers of CMT may be at higher risk than the general population for the debilitating and pervasive negative effects of depression.
Depression has been measured in a small number of sleep studies in family caregivers of children with chronic illness (Cottrell & Khan, 2005; Moore et al., 2006; Yilmaz et al., 2008; Yuksel et al., 2007), but none have used the CES-D as a measure. Using the CES-D and thus allowing for more direct comparison, Meltzer et al. (2006) similarly found scores to be elevated in family caregivers of CMT compared to controls in that study (15.10 [9.5] vs. 9.28 [8.5], \( p < .05 \)), with higher overall depression scores compared to this study. Neither of the two other studies that have measured sleep and depression in family caregivers of CMT can be directly compared with this study owing to differences in measurement and choice of reported variables (Meltzer et al., 2010; Read et al., 2010).

Data from longitudinal studies have informed the conceptual framework of this study and suggest the likelihood of a bidirectional relationship between sleep and depression (Baglioni, Spiegelhalder, Lombardo, & Reimann, 2010; Buysse, 2004). Given the cross-sectional nature of all existing studies, the directionality between sleep and depression in family caregivers of CMT has not been duly explored. In future, carefully designed intervention studies will be needed. Moreover, with evidence for variable rates of depression in family caregivers over time (Carter, 2003) and a negative association between sleep disturbance and increased depression and parenting stress (Metzer & Mindell, 2007), it is suggested that future studies include longitudinal measurement of sleep in family caregivers of CMT.

**Daytime sleepiness.** Daytime sleepiness has seldom been investigated in family caregivers of CMT. In this study, nearly half of the sample and three times as many family caregivers of CMT compared to family caregiver controls scored above the cut-off for excessive daytime sleepiness (Epworth Sleepiness Scale; \( n = 16, 42.8\% \) vs. \( n = 5, 11.6\% \)). These findings indicate that family caregivers of CMT in this study had much higher rates of daytime sleepiness.
than the reported prevalence in the general population (10–25%; Carskadon & Dement, 2005). As a comparator, Meltzer et al., 2010 measured daytime sleepiness using the Stanford Sleepiness Scale, and while they did not report on global scores, they did describe a relationship between increased daytime sleepiness and fewer nocturnal homecare nursing hours ($p < .05$). The Stanford Sleepiness Scale differs from the Epworth Sleepiness Scale, with the former measuring the current state of sleepiness and the latter measuring the propensity to fall asleep at inopportune times. Excessive daytime sleepiness can be described as a major public health issue (Colten & Altevogt, 2006), is associated with decline in functioning (Durmer & Dinges, 2005), and has been linked to catastrophic outcomes on the job (Mitler, et al., 1988). Given that constant vigilance is required to safely monitor a child who is dependent on homecare technology, the evidence for the presence of excessive daytime sleepiness documented in this study signals heightened concern about impaired daytime function in family caregivers of CMT. It even leads one to ask whether health care providers have a duty to assess for daytime sleepiness and act if daytime sleepiness in a family caregiver is likely to put the safety of that caregiver or their child at risk (e.g., while driving).

In future studies in family caregivers of CMT, it may be informative to measure daytime sleepiness using objective measures. Multiple Sleep Latency Tests, carried out in a sleep laboratory, would generate data about the intensity of drive to fall asleep and provide clues to the effects of sleep deprivation on daytime sleepiness (Hirshkowitz, Sarwar, & Sharafkhaneh, in Kryger et al., 2010) and thus prove helpful in future evaluations of the effectiveness of sleep interventions. Psychomotor Vigilance Testing assesses for performance on simple psychomotor tests, can be carried out on an ambulatory basis, and is considered an indicator of manifest sleepiness when repeated over time (Hirshkowitz et al., 2011 in Kryger et al., 2010). It is
questionable, however, whether either of these tests would be deemed acceptable, and the Multiple Sleep Latency Test feasible, to carry out in a sample of family caregivers of CMT. Moreover, there are ethical considerations about collecting these kinds of data, since they may reveal and highlight deficiencies in the capacity of family caregivers of CMT to remain alert and awake under test conditions. Such findings could potentially create tensions among clinicians, policy-makers, and advocates of family caregivers if deficits were identified in the absence of immediately available and proven interventions.

**Fatigue.** Fatigue is a common complaint among those with and affected by chronic illness, but it has seldom been measured in family caregivers of children with medical complexity. Not unexpectedly, family caregivers of CMT in this study were more fatigued than family caregiver controls (22.12 [9.1] vs. 17.44 [9], \( p = .021 \)), with family caregiver controls’ scores closely approximating those reported in earlier studies of fatigue in healthy community-based adults (Belza, 1995). In previous studies, sleep disturbance and fatigue have been highly correlated among family caregivers of children with cancer (Gedaly-Duff et al., 2006) and moderately associated in family caregivers of CMT (Meltzer et al., 2010). Those studies, however, used different measures of fatigue, and therefore, direct comparisons with findings in this study are not possible.

Clinicians are aware that complaints of being tired and exhausted are common among family caregivers of CMT. In family caregivers of older adults, predictors of fatigue have included anxiety and social support, levels of perceived family support, and baseline morning fatigue in patients (Fletcher, et al., 2010). Thus in future studies it will be important to examine what interventions may help family caregivers of CMT feel less fatigued and explore its relationship to insomnia; differentiating lack of opportunity to sleep versus the ability to fall and
stay asleep. In the clinical setting, the Multidimensional Assessment of Fatigue may be applied to assess fatigue patterns over the prior week and promote understanding of its impacts on the daily caregiving responsibilities of family caregivers of CMT (Belza et al., 1993; Belza, 1995).

In sum, data have confirmed this study’s secondary hypotheses: that levels of depression, sleepiness, and fatigue are higher in family caregivers of CMT than in family caregiver controls. No difference in health-related quality of life was found, but importantly, the interpretation of results may be limited by the choice of instrument. Considering the differences found between the cohorts and the relative decrements in sleep-related health outcomes, it appears that family caregivers of CMT are at risk of negative health consequences and impaired daytime function that may interfere with their capacity to provide safe and sustained caregiving.

**Other Group Differences**

**Child’s sleep.** Many parents describe associations between their children’s sleep with that of their own (Johnson, 1991). In family caregivers of children with medical complexity (Boergers et al., 2007; Meltzer & Mindell, 2007) and developmental disabilities (Chu & Richdale, 2009), child’s sleep has been reported to predict maternal sleep quality. However, none of these studies, or any in existing published studies of CMT (except this one), have used the Children’s Sleep Habits Questionnaire to measure this construct. Providing the only known direct comparison, family caregivers of CMT scored within the range reported (47.88 [8.4] vs. 53.21 [13.6]) in a recent study among family caregivers of children with acute lymphoblastic leukemia (Zupanec et al., 2010) and higher than their comparators in this study on the global score for the Children’s Sleep Habits Questionnaire (50.88 [10] vs. 43.60 [6.2], p = .008). Global scores on the Children’s Sleep Habits Questionnaire have been above the cut-off (> 41) for behaviorally and medically based sleep problems in all of these study samples. In this study, both
cohorts had elevated scores on the Children’s Sleep Habits Questionnaire, and thus, on its own, children’s sleep does not explain the relatively negative sleep outcomes among family caregivers of CMT in this study. Given that the Children’s Sleep Habits Questionnaire is known to have higher scores when young children are included in the sample, as they were in this study (Goodlin-Jones, Sitnick, Tang, Liu, & Anders, 2008), in the future, it will be important to have a measure of children’s sleep that can be applied across the paediatric age span. Access to a population-specific measure of child’s sleep that is easy to complete and sensitive to the physical and psychological differences (e.g., higher rates of sleep disordered breathing, neurological differences limiting ability to consolidate sleep) found in the majority of CMT (61.9% parent report of child’s neurocognitive delay or impairment) would also be valuable for use in future studies.

**Sleep hygiene.** Sleep hygiene is assessed through measurement of behaviours and environmental variables thought to influence sleep outcomes (Mastin et al., 2006). Improvement in sleep quality has been measured in a small but promising number of intervention studies in adult family caregivers targeting sleep hygiene practices (McCurry, 1996, 2006). In this study, sleep hygiene was similar in both cohorts (29.10 [5.8] vs. 28.19 [6], \( p = .479 \)) and worse than that reported in a recent study of middle-aged adults with chronic pain (19.16 [6.6]; Cho, Kim, & Lee, 2013). Scores on the Sleep Hygiene Index in this study were lower (suggestive of better) than that reported during instrument development in a sample of healthy adults (i.e. university students; 34.66 [6.6]), but these results cannot be generalized to our sample, given the validation study sample had a much lower mean (SD) age (Mastin, Bryson & Corwyn, 2006); where advanced age is known to be associated with increased sleep disturbance.
In future studies, it will be important to develop interventions that target improvement in the unique features of sleep hygiene that are most likely to result in improved sleep outcomes such as longer TST-N and shorter sleep onset latency in family caregivers of CMT. Moreover, findings from this study suggest that family caregivers of CMT may benefit from being assessed for their knowledge of and adherence to recommended sleep hygiene practices (Canadian Sleep Society, 2004) so that healthy sleep habits may be generated early and sustained over the course of their caregiving experience.

Secondary Analysis of Primary Question

This section will discuss the relationship between select caregiver, child, and environmental factors and sleep in family caregivers of CMT. It will begin with a discussion of which factors are independently associated with TST-N and NNA-O. It will then explore the influence of select modifiable risk factors, controlling for select sociodemographic variables, on sleep outcomes in family caregivers of CMT. Implications of these findings for clinical practice, policy, and recommendations for next steps in clinical research will also be described.

Caregiver factors. In this study, a number of caregiver factors linked to sociodemographic status were independently associated with poorer sleep outcomes. In family caregivers of CMT, being born outside Canada was predictive of shorter TST-N, and lower levels of education and household income were associated with more sleep fragmentation. It is important to consider that almost six times more family caregivers of CMT (40.4%, n = 17), compared to family caregiver controls (7%, n = 3; p < .001), had a household income that placed them at or below the low-income cut-off for a family of four in the GTA (approximating the average size of families in this study; Statistics Canada, 2005). Thus, in future, sleep interventions for family caregivers of CMT need to be developed that will be acceptable and
feasible in diverse and marginalized populations. Moreover, more research is needed to better understand the nature of the relationships (e.g., confounding, interaction) between sleep and sociodemographic variables in family caregivers of CMT. It will be important to explore whether improvements in sleep enable improved earning potential in family caregivers of CMT and help close the gaps described between what they earn and the routine and extra expenses they often incur (Kuo et al., 2011).

**Child factors.** That child’s age was predictive of sleep quantity (TST-N) in family caregivers of CMT ($p = .029$) was an expected finding. Prior studies have found that the younger the child, the more likely they are to have behavioural sleep problems that trouble their parents, such as bedtime refusal and frequent awakenings (Mindell & Owens, 2010). Moreover, it is thought that the care needs of children with medical complexity are usually higher, or more variable, when the child is younger. The findings of one longitudinal study, however, have refuted this notion. Macdonald & Callery (2008) reported that family caregivers of children with complex care needs found the child’s care needs to be more intrusive over time and described an increased need for respite as the child aged (Macdonald & Callery, 2008). Therefore, in clinical practice, it is important to assess for child sleep problems and related family caregiver sleep disturbances, both in hospital and on an ongoing basis after discharge to homecare. Funding policies for homecare and other aspects of caregiver supports that enable sleep and respite in family caregivers of CMT need to reflect flexibility in service delivery maximums and enable adequate homecare nursing supports to be available during pivotal developmental stages in the caregiver experience.

Children who are the most dependent on medical technology (e.g., high risk for a negative health outcome if technology fails) typically use medical technology around the clock.
In this study, more technology use in the daytime predicted poorer sleep (i.e., increased objectively measured nocturnal awakenings) in family caregivers of CMT ($p = .05$). This study also found that daytime use of homecare technologies was highly variable across subjects and averaged 6.7 (5.1) hours, while nighttime use was more consistent (9.68 [3.1] hrs). One other study of sleep in family caregivers of CMT (Read et al., 2010) has reported on the daytime versus nighttime use of technology use, but the relationship between the amount and timing of technology use and sleep in family caregivers of CMT has not been reported. Thus, the findings from this study suggest that family caregivers of CMT experience highly irregular sleep–wake patterns owing to their child’s age and care needs. It is important to consider that paid shift workers receive time off to recover from negative effects, while family caregivers of CMT do not.

**Environmental factors.** As expected, owing to jurisdictional and other differences (Peter et al., 2007), there was a broad range reported in the number of homecare hours used by family caregivers of CMT in this study (0–102 hrs per week). Family caregivers of CMT routinely used much less homecare (24.17 [22.1] hrs per week for in-home and 10.68 [16.7] hrs per week for out-of-home) compared to that reported by Meltzer et al. (76 hrs/week; 2010). Direct relationships between the total amount of homecare use (none vs. > 48 hours per week) and sleep (subjective report; $p < .05$) in family caregivers of CMT have also been reported (Meltzer et al., 2010). In comparison, this study reported much lower overall use of homecare and only found that one variable (approved amount out-of-home nursing service) positively predicted sleep (NNA-O). When necessary, the use of homecare services is typically approved to enable a CMT to attend school with a registered nurse; thus, the number of hours of homecare used out-of-home
may be a proxy for medical complexity among CMT, in part explaining its influence on sleep in family caregivers of CMT.

It is suggested that the findings of this study should not be interpreted to mean that there is no relationship between homecare and improved sleep in family caregivers of CMT. There was a very high proportion of use of allocated homecare (in-home) hours (84.3%), signaling that it is a valued resource. In the study published by Meltzer et al. (2010) that found a direct relationship between more homecare and improved sleep outcomes, family caregivers of CMT used twice as much nocturnal homecare per week than family caregivers of CMT in this study (35 vs 18 hrs.). Therefore, based on findings of this study it is suggested that that the relationship between homecare use and sleep outcomes in family caregivers of CMT may not be linear. Meltzer and colleagues cited a 48-hour per week cut-off, which exceeds the amount of homecare typically used in this study sample. The limited presence of homecare nursing may have contributed to impaired communication, misaligned priorities, or other sub-optimal aspects of the relationship between homecare nurses and family caregivers of CMT (Graves & Hayes, 1996; Manhaus & Mitchell, 2012) that intruded on their sleep. Other studies have reported high rates of missed shifts due to nurses cancelling (e.g., due to illness) or shifts going unfilled (e.g., no nurse assigned); among this sample of family caregivers of CMT, no participants reported that their nursing shifts typically go unfilled or otherwise missed. Given that the only shifts cancelled were by family caregivers (n = 8, 19.0%), who cited a variety of reasons, including that the use of homecare was too stressful or that they didn’t need the help, future studies should explore the allocation, uptake (e.g., acceptance versus decline) and relationship among homecare, sleep and respite and related outcomes in family caregivers of CMT. So far, and even in the wake of a large-scale population-based pan-Canadian study that examined safety in homecare (Doran &
Blais, 2012), there are no data to either confirm or refute that homecare nursing protects family caregivers from sleep disturbance or other adverse health and safety outcomes.

**Factors that influence sleep in family caregivers of CMT.** While this study has helped to characterize sleep disturbance and related outcomes in family caregivers of CMT, it has not identified any modifiable caregiver (SHI), child (CSHQ), or environmental (# of nocturnal homecare hours) factors that predicted sleep disturbance in family caregivers of CMT. Few of the many factors examined in this study predicted sleep outcomes in the family caregivers CMT.

A factor that was found to have a significant relationship was birthplace of the caregiver. Participants born outside of Canada may have experienced a shorter sleep duration than those Canadian born for at least two reasons: 1) newcomers to Canada are more likely to be of a lower socioeconomic status, placing them at risk for poorer health outcomes, including getting less sleep, 2) the greater than 60 minute difference may also reflect cultural preferences and norms for sleep among those with origin of birth other than Canada (Adenekan, Pandey, McKenzie, Zizi, Casimir & Jean-Louis, 2013; Mindell, Sadeh, Kohyama & How, 2010). This study found that lower education and income predicted a shorter TST-N in family caregivers of CMT and fits with population based data on the influence of these sociodemographic characteristics on sleep duration in the general public (Ohayan, 1996; Stamatakis, Kaplan & Roberts, 2007).

It was expected that advancing child’s age would positively influence sleep in family caregivers CMT and therefore was taken into consideration in design of this study. Findings indicated that, for example, participants with adolescent children achieved approximately one hour more sleep per night than those with a 12 month-old infant at home (10 min improvement in TST-N per each year of child’s age). This highlights that caregiver experience related to sleep and other outcomes may change over time and with the child’s growth and development.
It is suggested that daytime medical technology use may act as a proxy for the complexity of care for the CMT. The difference on nighttime sleep (TST-N) per increasing hour of daytime technology use was small (4 mins per each hour of use), but likely meaningful for those with children approaching the upper limits of daytime medical technology use (e.g. for FC with CMT using technology for 12 hours in the daytime x 4 mins less/hr= < 45 minutes in TST-N). This finding aligns with thinking that the needs and uses of medical technology are likely to be very high on a 24-hour per day basis when a child is dependent on medical technology when awake during the day.

Results of the multiple regression analysis call out that gender played a role in this study. In studies that compare sleep between men and women, it is typical to find small differences in sleep quantity and quality. That the women in the family caregiver CMT cohort achieved almost one and a half hours (85 min) more sleep than the men far exceeded the difference expected. To compare, gender differences in sleep time are self-reported to be only 11 minutes in the general population (women achieving more than men; Statistics Canada, 2008). In a recent clinical study of sleep parents in hospital, gender differences were also found with mothers getting approximately 20 minutes more sleep than the fathers (on actigraphy; Stremler et al, 2013). In this study, over 80% of participants in this study were female, with mothers more often than fathers self-identifying as the primary caregiver for the CMT. This sample characteristic with many more females represented than males is similar to that reported in all other studies of sleep in family caregivers of children with chronic medical conditions (Meltzer et al., 2008). It is also aligned with the findings of research on the role of women, who provide the vast majority of caregiving for children in general, and especially the role of mothers of children with chronic health conditions (McKeever, 1992). Interestingly, despite the extraordinary care needs of
CMTs, no differences were found in the proportion of genders represented in each cohort \( (p = .51) \). The finding that on average, women woke up less through the night (4 fewer nocturnal awakenings per night on actigraphy) than men, is also intriguing. It raises questions about the whether male participants may have been experiencing the double burden of both primary caregiver and primary income-earner. In other studies females have typically reported more sleep disturbances than males but these differences have not typically been detected at a scale this large when using objective sleep measurement (Lauderdale, Knutson, Yan, Liu, & Rathoz, 2008; Ohayon, 1996). Thus findings from this study point to the importance of examining sleep amongst all caregivers in the family, not only the primary caregiver, in future studies of sleep in family caregivers of CMT.

It was important to explore the potential influence of environmental factors on sleep in family caregivers of CMT because many of them may be modifiable. This study raises questions about whether or not the uptake (use and impact of) of homecare services (namely in-home homecare [nursing]) is ideal. That the increased use of out-of-home homecare (typically allocated for school when a CMT is over 6 years of age and depends on medical technology in the daytime) was found to be associated with more nocturnal awakenings fits with the assumption that caregiver sleep is worse when a child has more complex care needs. These relationships between caregiver sleep, child’s care needs and homecare merit further exploration in future studies.

Based on findings from this study and those that have preceded it, questions remain about what factors most influence sleep disturbance and which among them may be modifiable in future studies and clinical practice with family caregivers of CMT. Sleep disturbance likely has multifactorial causes in this population, for which there are few validated measures (i.e., some of
the variables analyzed in the preceding section were derived from single-item, investigator-developed questions [e.g., vigilance]). In future studies it may be important to qualitatively explore factors that influence sleep in family caregivers of CMT, in order to better contextualize these data and discover what is perceived to be most influential on sleep disturbance directly from the perspectives of end-users (e.g., family caregivers of CMT and homecare providers). Finally, the identification and testing of caregiver, child, and environmental factors as they relate to sleep in family caregivers of CMT should be examined in future trials with adequate sample sizes, as limitations of study methods and statistical power may have hindered the ability of this study to find significant associations.

**Study Strengths and Limitations**

The methods used in this prospective cohort study addressed many of the limitations noted in prior studies of sleep in family caregivers of CMT. The internal validity of the study has been bolstered principally through stratified sampling of a comparator group to account for the confounder of child’s age. The use of reliable and valid measures, namely actigraphy, for objective measurement of sleep is also a major strength of this study. This section will describe these and other study strengths in detail, along with a discussion of study limitations.

**Methods.** This is only the second study of sleep in family caregivers of CMT that has used a control group and the first that reports accounting for the confounder of child’s age on sleep in family caregivers of CMT. Both of these strategies enhanced the validity of findings in this study compared to, for example, cross sectional studies reporting on sleep in family caregivers of CMT.

**Sampling.** During the study recruitment phase (7 months), all potentially eligible family caregivers of CMT attending in-hospital clinic were approached to participate in the study. By
reaching the entire population of eligible family caregivers of CMT during the study time period, the potential for referral bias in this study was highly limited. The representativeness of both samples was also considered to be high. Further determination of non-respondent bias, by comparing demographics in those who consented to those who did not, was not possible, as this study did not seek ethics approval to access the health records of non-participants.

Given the diversity of the referent population, it is notable that no participants were excluded from participation based on a lack of ability to communicate in English. Thus, this study has succeeded where others have not to enable access and participation for even those typically marginalized by language barriers.

Another strength of the sampling achieved in this study is that it did not limit enrollment, like some others, to samples of only mothers (Andrews & Neilson, 1988; Feeley et al., 2013). This study included anyone in the household that self-identified as the primary caregiver, and so, along with mothers, fathers and a grandparent were also included. While it is well known that maternal caregivers are largely responsible for the homecare of children with chronic illness and disability (McKeever, 1992; Stewart, Ritchie, McGrath, Thompson, & Bruce, 1994) in this study, a few families (n = 3) described that it was difficult to choose which family caregiver was considered to be primary in the household. The reasons for this included that both fathers and mothers got up through the night, and in select families of CMT, fathers tended to children at night even more often, as they were “stronger and better able to turn and position” (n = 2) the CMT, and/or “he handles going without sleep better [than I do]” (n = 1).

There were very low dropout rates in the two groups, limiting attrition bias. A study management technique—the use of daily reminders (email, text, or phone call)—was used to promote adherence to the protocol. Home visits at the conclusion of the study allowed for the PI
or RA to review the sleep diary and questionnaires for completeness with the participant present to help fill in missing responses, although this approach was resource-intensive. These strategies helped to ensure few losses to follow-up, as well as very little (< 10%) missing data across all variables.

Potential for bias related to time of year and season existed in this study, with these data having been collected over the spring and summer months. Summer sleep schedules are frequently characterized as unpredictable compared to other times of the year, when school and work commitments impose more routines on patterns of sleep and wake cycles. The effects of daylight on circadian rhythms and sleep have not been fully explored in this study, but it follows that TST-N is typically shorter in summer months (Duffy & Czeisler, 2009). Also, in considering the timing of data collection, scores on depression in this study may have underestimated what would have been found if study recruitment had been completed in the winter, when the effects of limited light on symptoms of depression with a seasonal pattern are at their highest (Rosenthal, et al., 1984). Interestingly, during a week of heavy recruitment (n = 2 family caregivers of CMT, n = 6 family caregiver controls) that coincided with a major storm in the region, a number of study participants commented at their home visits that their sleep was more disturbed than usual. Thus, while relevant to consider in future studies, this is the only investigation that has described potential for cohort bias related to the timing of data collection among studies of sleep in family caregivers of CMT.

Data management. The completeness and accuracy of the data was also assisted by the use of REDCap, a web-based data capture system that enabled forced entry of variables, along with checks for logic and range. Further to promote high quality data, double data entry was done for all of the data entry for primary outcomes and for a minimum of 30% of all other data.
To ensure consistency and agreement, inter-rater reliability of more than half the actigraphy files by two to three study personnel familiar with this type of analysis (PI, RA, and thesis supervisor) was also completed.

**Measurement.** This is the first study to report on the use of actigraphy in a sample of family caregivers of CMT. A major strength of this study is that the use of actigraphy limited recall as a source of measurement bias. Subjective appraisal of total sleep time (self-report), on its own, has been found to be less accurate when compared to findings on objective measurement (i.e., polysomnography) in study participants with a history of chronic sleep deprivation (Unruh, Redline, An, Buysse, Nieto, Yeh & Newman, 2008).

Octagonal Basic Motionloggers (AMI), one of the most commonly used technologies to measure sleep in ambulatory studies, were used in this study, set at standardized settings (AMI; Sadeh & Acebo, 2002). This technology has been validated against polysomnography in clinical (0.82) and healthy subjects (0.97; Jean-Louis, et al., 1997). The biggest limit to the use of actigraphy is that its sensitivity in discriminating between quiet awake time and sleep time is not ideal. Thus, results on TST-N in this study may represent an overestimate of actual sleep times, making the findings of sleep deprivation amongst CMT even more concerning. In this study, the likelihood of this occurring is the same for both groups.

Social desirability bias in completing the sleep diaries and the other subjective measures may have been higher among family caregivers of CMT. They may have been especially motivated to document deficits and problematic symptoms, thinking, although informed otherwise, that the results of the study may directly lead to improved services and their support in the future. Participant fatigue was of some concern, with evidence for this occurring in both
groups when, on occasion, participants omitted completing the entire sleep diary in advance of the final home visit.

The multiple methods of data collection used in this study have enabled strong characterization of sleep and sleep disturbances in family caregivers of CMT. Measurement of select secondary outcomes, however, may have been limited by the choice of instruments used in this study. For example, the SF-12™ was chosen for its ease of use and brevity, but in this short form, it may not have had the discriminate validity necessary to detect differences in quality of life between the cohorts. The validity of results of child’s sleep quality (CSHQ) in this study is also in question, as the measure has only been validated for use in toddlers and school-aged children (Goodlin-Jones, et al., 2008). Other studies of sleep in family caregivers of CMT have cited similar limitations of measurement.

To conclude, this study was powered to minimize the risk of Type I and Type II errors. The study was adequately powered to achieve its primary aim: to detect a clinically meaningful difference in TST-N between family caregivers of CMT and family caregiver controls. The model fit was substantially limited for the multiple regression analyses used to explore which caregiver, child, and environmental factors predicted TST-N in family caregivers of CMT (~ 7 participants: 1 predictor variable compared to the recommended 15:1 participants to predictor variable; Stevens, 1996, pp. 72). While there were small differences in select sociodemographic variables, the study samples did not differ on the majority of key demographic variables (e.g., FC age, child age), and were representative of their respective populations. Therefore, it is suggested that the findings from this study may be generalized to family caregivers in other jurisdictions in which CMT are typically cared for at home.
Chapter 6: Implications, Recommendations, and Conclusions

This study makes a substantive and novel contribution to the scholarship on sleep, sleep disturbances, and related outcomes in family caregivers of CMT. Hypothesized differences were found between the cohorts that can be interpreted to mean that family caregivers of CMT are at higher risk than their comparators for the negative outcomes associated with acute and chronic sleep deprivation. In this chapter, based on study findings and related literature, implications for policy, practice, and research are discussed and recommendations are made. The recommendations are aligned with current provincial and federal policies targeting enhanced supports for family caregivers across the age span. The suggested practice changes, if implemented, are expected to promote sleep, health, and related outcomes in family caregivers of CMT. Suggestions for the next steps in sleep research with family caregivers of CMT follow. Finally, this chapter presents conclusions about the most salient features and outcomes of this study.

Implications for Policy and Practice

Screening and education. This study tentatively recommends that all family caregivers of CMT be routinely screened for sleep problems and other health-related issues. Anticipatory guidance provided at the earliest possible time may help family caregivers of CMT to optimize sleep habits for the duration of their caregiving. Thus, assessments would ideally be completed before discharge from hospital and on an ongoing basis at regular intervals thereafter. To open the conversation about sleep in the family, clinicians are encouraged to embed screening questions in their standardized assessments. This recommendation has been made by the study investigator, and it was well received by the advisory council to a local caregiver framework that
targets enhanced health and social supports for family caregivers of children with complex care needs (C. Bruce-Barret, Personal communication, April 17, 2013).

Assessment of caregiver sleep using validated instruments is worth considering, and may be valuable if screening questions raise concerns for sleep disturbance in family caregivers of CMT. The PSQI is easily administered and ask questions that may help to discriminate what features of sleep are most problematic among ‘poor sleepers (Buysse, et al., 1988). The use of sleep diaries (with or without actigraphy) may also be considered helpful tools if the data are collected in a systematic way (Carney et al, 2012), and interpreted by a knowledgeable clinician with the capacity to provide appropriate sleep education and/or secure necessary interventions (e.g. psychological counseling, more homecare support).

It is also suggested, based on the experience gained from this study, that CES-D may be feasible for implementation in busy clinical settings. It was easily administered and highly acceptable (i.e., not too long, little missing data) in this study sample. During administration of the CES-D at home visits family caregivers often talked openly about their concerns about their mood, and later, when informed of scores suggestive of clinically important depression, were thankful for the information. Caregiver assessments of fatigue may also be incorporated in routine assessments. Heightened fatigue poses a potential barrier to participation and social isolation, which are described as important aspects in the lives of families of children with complex and special health care needs (Carnevale et al., 2006; Woodgate, Edwards, & Ripat, 2012). In the clinical setting, the Multidimensional Assessment of Fatigue questionnaire may be applied to assess fatigue patterns over the week prior and promote understanding of its impacts on the exceptional caregiving responsibilities faced daily by family caregivers of CMT (Belza et al., 1993; Belza, 1995).
High quality information about sleep and sleep hygiene, such as that made available to the public by the Canadian Sleep Society (2004) and National Sleep Foundation (http://sleepfoundation.org/ask-the-expert/sleep-hygiene), could be provided directly to family caregivers of CMT. This study suggests that this information may be ideally provided as early as possible, while the CMT are still young and before family caregivers become habituated to poor sleep routines. Education about sleep and chronobiology is also recommended for health care providers who commonly interact with this population. Current data indicate that there is a very limited amount of sleep content embedded in medical education across the globe (Mindell et al., 2011), including the majority of health practitioner curricula in Canada (Stremler, 2013). Evidence-based resources exist that would inform sleep training for nurses and could be included in professional development programming for nurses across the continuum of care (Lee et al., 2004). Community-based providers, and especially homecare nurses, may be ideally positioned to provide information and educate family caregivers of CMT about sleep, sleep disturbances, and sleep hygiene. They may also personally benefit from learning how to best manage sleep and shift work. To date, however, the work of homecare nurses may not be ideally conceptualized and structured to include this type of respite support for family caregivers and focuses its aims on the provision of direct care to the CMT.

**Use of homecare.** It is suggested that $\geq 48$ hours of nursing care per week are required to improve sleep outcomes in family caregivers of CMT (Meltzer et al., 2010). It is likely that current levels of homecare nursing use in the greater Toronto area, which this study found to be well below the cut-off suggested by Meltzer et al. (2010), may not be adequate to positively influence sleep outcomes in family caregivers of CMT. While there is no solid evidence for the “right” amount of homecare, anecdotal accounts from caregivers suggest that more homecare
nursing is needed and desired by family caregivers of CMT. This study identified that child’s age predicted poor sleep in family caregivers of CMT and reinforces that policy should enable the flexible use of homecare for family caregivers of CMT with additional supports available at select ages and developmental stages, key transitions; especially when the children are still very young.

It is suggested that the criteria for and number of homecare hours (namely in-home supports) for which family caregivers of CMT are eligible, both from the Ontario government and other funding agencies, is not consistent (Rosenbaum, 2008). The current study supports this observation. While there is a move to standardize procedures, as it stands, the amount of in-home homecare available to family caregivers of CMT from provincial sources is based on a combination of regionally established criteria for eligibility and the overall amount of funding available to the jurisdiction, as is the availability of local homecare personnel from third-party providers. This study, which found a wide range of available hours with very high utilization, supports the assertion that policy-makers should consider potential inequities in access to homecare nursing and other respite services for family caregivers of CMT.

A paradoxical relationship between the provision of homecare services and sleep outcomes in family caregivers of CMT has been described. In the first study to target sleep and homecare in family caregivers of CMT, Meltzer et al. (2010) concluded with a call for more homecare. Family caregivers of CMT in Meltzer’s study however reported that the presence of homecare nurses negatively influenced their sleep by making excessive noise at night. In contrast, a positive association between increasing number of homecare hours and caregiver sleep was reported (Meltzer et al., 2010). This study did not find a significant relationship between sleep and the use of nocturnal homecare, although a small and positive association was
found between the amount of nocturnal sleep and hours of daytime homecare support. Thus, in future, based on existing evidence, it behooves homecare administrators to seek better understanding and methods to match the allocation, uptake, and quality of this important resource. A shift from reliance on a child-centred assessment to a child and family-centred one with sleep as one of the quality indicators should be considered in future policy and practice with family caregivers of CMT. With the use of actigraphy in clinical assessments of ambulatory sleep gaining acceptance (Morgenthaler et al., 2007), it is timely to consider whether actigraphy data may used in future to judge the appropriate amount of night nursing to achieve improved sleep and quality of life for family caregivers of CMT.

In this study, low levels of homecare use may be related to preferences or choices made by family caregivers of CMT. In contrast with the findings of Meltzer et al. (2010), in which a quarter of the families reported that homecare nurses cancelled night shifts, this study found that no shifts were ‘typically’ cancelled or otherwise unfilled by the nurses. The family caregivers of CMT, however, cancelled some shifts themselves. In the future, it will be important for homecare nurses and their administrators to examine the reasons for cancelled homecare nursing shifts and seek to improve the fit between patient needs, including the need for better sleep, and service delivery.

Regional differences were observed in the use of homecare, with family caregivers of CMT in one jurisdiction describing that they were eligible for up to 70 hours per week for in-home nursing services funded by government agencies, compared to a maximum of 43 hours per week reported to be the ceiling for eligibility among remaining participants. Thus, it is suggested that policy is needed to ensure that the distribution of efficacious publicly funded homecare is equitable, easily accessed, and aligned with the unique needs of family caregivers of CMT.
**Safety and decision-making.** Little safety data exists for homecare in Canada, and among what does exist, 56% of adverse events were judged to be preventable (Doran & Blais, 2013). Data from the United States detail errors and mishaps in paediatric homecare, some resulting in severe morbidity when family caregivers have slept and homecare nurses were unavailable (Ong, Elder, Hill, & Reding, 2014). While these events are rare, and none were described in response to questions about what may have intruded on sleep from the diarized data collected during this study, it is noteworthy that in recent years, there have been two known cases in the Greater Toronto Area of the sudden death at home of a child on home ventilation occurring at night or very early morning (R. Amin, personal communication, June 18, 2012). This raises concerns among clinicians and others that appropriate supports are needed to ensure safety for the child and enable adequate amounts and quality of sleep in family caregivers of CMT.

Other studies have shown that sleep disturbances in family caregivers of those with chronic health conditions contribute to uncertainty about care decisions and increased visits to emergency departments and subsequent admissions to hospital (Rausch, Baker, & Boonmee, 2007; Yuksel et al., 2007). Recognizing that sleep disturbances alter performance and judgement (Balas, Scott, & Rogers, 2006; Scott, Hwang, & Rogers, 2006), and that family caregivers of CMT in this study did not achieve adequate quantity or quality of sleep, policy-makers are urged to identify ways to address the sleep and respite needs for family caregivers of CMT in support of safe homecare, sound decision-making, and better resource utilization for CMT.

**Social imperative.** These recommendations for policy and practice issues should be considered, as they may improve the sleep, health, and the ability of families to sustain homecare of their CMT. Importantly, they may also go a long way to improving on the social determinants
of health and the financial situation for family caregivers of CMT. Improved sleep, for example, may enable family caregivers of CMT to be more gainfully employed. In The Government of Canada’s most recent report on family caregivers, the 2012 General Social Survey on Caregiving and Care Receiving (2012), it is reported that family caregivers of children with chronic care needs were often depressed (38%) and more likely to have financial difficulties (28%) as a result of their caregiving responsibilities than family caregivers of spouses (20%) and parents (7%; Turcotte, 2013). Authors of this report and others have suggested that there is an compelling need to take better “care of the caregiver” in Canada (Mikkonen & Rafael, 2010; Spalding et al., 2012). In a population-based study in the United States 50% of family caregivers of children with medical complexity reported quitting their jobs to enable care for their child at home (Kuo et al., 2011). The results of this study support the social imperative that would limit or temper the costs incurred (e.g. emotional, financial) by family caregivers. The data comparing the groups on aspects of sociodemographics show key differences, and importantly, relative deficits in family caregivers of CMT compared to those with healthy children.

Recommendations for Research

To date, there is descriptive evidence for the presence of subjective sleep disturbance in family caregivers of CMT in 14 studies, this study included, and objective evidence for sleep disturbance in only this single study. This study has also contributed to what is known about other subjectively measured aspects of sleep and related outcomes in family caregivers of CMT. In the section that follows, recommendations for the next steps in the development of knowledge and evidence for sleep and related outcomes in family caregivers of CMT will be discussed.

End-user perspectives. Family caregivers of CMT and partners in the homecare sector (e.g., Toronto Central Community Care Access Centre) need to be engaged in all phases of
future sleep research so that the studies are deemed relevant and useful to end-users in paediatric homecare, both locally and across Canada. The high participation rate in this study demonstrated that family caregivers of CMT were eager to participate and keenly interested to learn more about their sleep. Next, it is recommended that comprehensive qualitative data be collected from family caregivers of CMT and homecare nursing providers to help answer the following questions from their perspectives: a) Which modifiable factors (e.g., caregiver, child, environment) are thought to most influence sleep (quantity and quality) among family caregivers of CMT?; b) What barriers and facilitators are there for implementing and testing sleep-promoting interventions among family caregivers of CMT?; c) What is the perceived utility of evidence-based sleep-promoting interventions among family caregivers of CMT?; and, d) How much improvement in sleep and related outcomes would be clinically meaningful among family caregivers of CMT? Data from a future study of this type would help shape an evidence-based and end-user informed sleep-promoting intervention that could be tested with family caregivers of CMT. As a component of post-doctoral training, this study investigator has secured small grant that will fund a qualitative study intended to answer the research questions described above from the end-users’ perspectives.

**Family caregiver sleep and chronobiology.** The influence of light and noise has not been systematically investigated in any community-based studies of sleep in family caregivers. In parents of hospitalized children, excessive noise (Meltzer, Davis, & Mindell, 2012) and other features of the environment (Stremler, Dhukai, Wong, & Parshuram, 2011) have been described to be associated with subjective sleep problems. With advances in technology, it will be both necessary and possible in future studies in family caregivers of CMT to examine the relationship between external cues (zeitgebers) in the environment and caregiver sleep. Ideally, objective
sleep data should be collected using actigraphs that include built-in noise and light meters, which are accessible now for purchase in the marketplace. Future studies should plan for analysis of the influence on sleep in family caregivers of CMT of the unique features of medical technology (e.g., false alarms) as compared to that of typical technology in the home (e.g., personal handheld devices, television). The results of these investigations will provide important information to be considered by developers and prescribers of homecare technology.

In future studies, it will also be important to more reliably collect data to highlight the potential presence of circadian rhythm problems in family caregivers of CMT. Similar to shift workers, as evidenced in this study, family caregivers of CMT may be experiencing greater sleep onset latency, more nocturnal arousals, and other unwanted awakenings because of disruptions to normal sleep–wake patterns. Family caregivers of CMT who depend on others to help care for their children around the clock are at risk for not having sleep onset and offset well-aligned with intrinsic cues for sleep. Arbitrarily enforced external cues (e.g., unwanted social cues from homecare nurses who arrive close to bedtime) may be strong signals and not ideally aligned with either Process S (drive for sleep) or Process C (internal clock). Sleep times may change from day to day as a consequence of a child’s unpredictable health status and care requirements. Studies are needed, therefore, that will more fully characterize this aspect of sleep and chronobiology in family caregivers of CMT, signaling if and to what extent sleep-promoting interventions need to target alterations or improvements in sleep–wake cycles in family caregivers of CMT.

**Education and behaviorally based interventions.** Family caregivers of CMT anecdotally report that they sleep with "one eye open and one eye closed." In one diary entry in this study, a family member described "staying awake most of the night because of not trusting a new night nurse." In future studies, it will be important to more fully explore the nature and
consequence of vigilance in family caregivers of CMT and determine if some of the well-accepted sleep interventions based in cognitive behavioural theory (e.g., CBT-i) can benefit this group. Vigilance in this context can be defined as anticipating what might be, calculating the risk, monitoring results and outcomes, and staying ready to act (Boroughs & Dougherty, 2010). A few promising studies of psychoeducation-based sleep interventions that have addressed caregiver thought processes in adult family caregivers exist (Carter, 2006; 2009; McCurry, 2005). These studies have described the development of brief home-based interventions that were acceptable and effected small but important improvements in overall sleep quality and quantity in adult family caregivers. In the future, it may be important to replicate these sleep intervention studies (with customization of manualized materials) for use in family caregivers of CMT.

**Measures of psychomotor performance.** Using handheld devices, performance on psychomotor performance tests could be useful as an outcome in future community-based investigations that target sleep quality and quantity in family caregivers of CMT. The results of these tests, which simulate activities requiring quick responses and decision-making (e.g., high-speed driving), may help characterize improvements or decrements in daytime function when used in future studies and policy decisions regarding sleep and related outcomes in family caregivers of CMT.

In this study, scores on excessive daytime sleepiness were high among family caregivers of CMT. In future studies, Multiple-Sleep Latency Testing, a validated clinical test of propensity to fall asleep in the daytime, could also be used to assess for excessive daytime sleepiness, although the acceptability of this in family caregivers of CMT is questionable. Family caregivers of CMT may not appreciate having their performance measured with the potential that deficits
may be identified. As described earlier in this chapter, there are implications to consider in advance of collecting these data. As it stands, the results of this study, which used validated measures, provide evidence for excessive daytime sleepiness and fatigue and thus pose questions about the health and safety of family caregivers of CMT and their children at home.

**Feasibility and acceptability.** Notably, there is little to no published evidence for interventions targeting improvements in physical, psychological, social, or other outcomes in family caregivers of CMT. Given the around-the-clock responsibilities that they carry, the consideration for participant burden and its potential influence on study recruitment, retention, and fidelity may be heightened in planning future sleep interventions with family caregivers of CMT. Before large scale clinical trials are undertaken, studies are strongly encouraged to test the feasibility and acceptability of any sleep promoting intervention in family caregivers of CMT. Specifically, these studies should, through appropriate randomization, measure sleep in family caregivers of CMT controlling for numerous characteristics and socioeconomic variables (not ideally balanced in this study). As a component of post-doctoral training, a feasibility and acceptability trial in this field is planned for by this study’s lead investigator.

**Methods and measurement.** Actigraphy was highly acceptable in this study, with data collected over a full week and very little missing data. Thus, actigraphy should continue to be used in studies of sleep in family caregivers of CMT and must continue to be accompanied by sleep diaries that meet current recommendations for their use in clinical practice and sleep research (Carney et al., 2012) but are customized, as in this study, to collect details of the unique features of sleep in this group (e.g., presence of nocturnal homecare nursing). In the future, longitudinal data should be collected about sleep and related outcomes in family caregivers of CMT to better define key transitions (e.g., before and after discharge) when sleep disturbances
may be heightened, and importantly, what factors (e.g., child’s age, as found in this study, or level of acuity) may contribute to this. Moreover, the effects of time on uptake and retention of sleep interventions will be important considerations for future researchers. It will also be important to repeatedly investigate sleep outcomes in family caregivers of CMT to identify how exposure to homecare and a sense of enhanced mastery (Simpson & Carter, 2013) affects sleep and related outcomes in this vulnerable group. In future studies, it may be possible to determine if sleep in family caregivers of CMT improves as the family caregivers become more confident in homecare regimes and establish, for example, trusting relationships with homecare nurses and other providers.

In future studies, the opportunity to further explore other associations with objectively measured sleep will be important. For example, there is a need for improved understanding about the relationship between sleep in family caregivers of CMT and child’s sleep. This study found a difference on the CSHQ between the cohorts, but not a significant relationship to TST-N in family caregivers of CMT. This finding is not consistent with what most caregivers and health care providers would have predicted. Given the many limitations to the use of the CSHQ measure in a clinical sample across the age-span in pediatrics, it is suggested that alternative measures may need to be used in assessing the unique features of child’s sleep in this population. For example, emerging measures of child’s sleep specifically designed for use in those with neurocognitive impairments, (characteristics shared by CMT) and their parents show promise as reliable and valid instruments for use in family caregivers of CMT (Tieze, et al., 2014). Another doctoral thesis, as yet unpublished, suggests that studies of caregiver sleep and fatigue include measures of caregiver burden (Tidwell, 2008).
**Conceptual issues.** To guide clinical practice, policy and research, a better understanding of the nature of the relationship between causes and consequences of sleep disturbances and other outcomes in family caregivers of CMT is needed. The conceptual model that guided this study helped to inform the research questions and selection of the measures (Fawcett & DeSanto-Madeya, 2013). It conceptualized that sleep disturbance directly influenced daytime sleepiness and fatigue, but owing to the study design, the nature of these relationships remains unclear. Also conceptualized in this study (as bi-directional) were sleep and depression in family caregivers of CMT. This relationship also merits further investigation given the clinical importance of finding higher scores on the CES-D in the family caregivers of CMT cohort. Continued development of the empirical basis for understanding sleep in family caregivers of CMT is recommended such that the most relevant research questions can be answered.

**Future investigation.** Future studies have the potential to generate new knowledge about what interventions developed for use in other at-risk populations may also benefit family caregivers of CMT. The use of daily bright light therapy is a relatively new and promising intervention for both sleep and depression in post-partum women and shift workers (Lee, Aycock, & Moloney, 2013; Crowley & Youngstedt, 2012; Huang, Tsai, Chen, & Hsu, 2014). Given that family caregivers of CMT are relatively isolated, and with data from this study confirming higher scores than controls on depression, this relatively novel and unobtrusive intervention merits investigation. Mindfulness-based stress reduction has also been found in several studies to have positive effects on sleep and related health outcomes (Winbush, Gross, & Kreitzer, 2007). Interventions of this type (i.e., mind and body, yoga) are currently being provided locally for parents of hospitalized children, and subjective reports indicate that the parents appreciate them (S. Zupanec, personal communication, October 14, 2014). This study
highlights concerns for family caregivers of CMT having difficulty falling asleep and staying asleep, for which these interventions have been found to be helpful in other population. Thus, given what little is known about what will work to promote sleep of adequate quantity and high quality in family caregivers of CMT, each of these interventions and their impacts on sleep, depression, and other outcomes in family caregivers of CMT deserve more exploration.

One aspect of sleep disturbance in family caregivers of CMT that requires careful exploration in future studies is how, and if, primary insomnia interferes with their ability to achieve adequate sleep quality and quantity. Insomnia is recognized as a prevalent problem in middle-aged women and adult family caregivers (with predominantly female samples; McCurry, Gibbons, Logson, Vitiello & Teri, 2009). This study did not screen for insomnia, but with the evidence in this sample of difficulty falling asleep (i.e., lengthened SOL), difficulty staying asleep (i.e., frequent nocturnal awakenings), poor quality of sleep, and daytime fatigue, the family caregivers of CMT as a collective appeared to meet many of the major criteria for the clinical diagnosis of insomnia (American Psychiatric Association, 2013). In the future, data that helps determine and/or confirm this risk may be required to ensure that interventions are focused on the right triggers for sleep disturbances.

In future planned analysis of this study data set, it will be possible to explore some associations in more detail and carry out further analyses that were not planned for as primary or secondary research questions in this study. For example, single items on some of the study instruments asked about features of sleep hygiene (e.g., use of caffeine) that, if found to be importantly related to sleep disturbances or other outcomes, may also be targets for knowledge translation and future interventions. Additional sleep diary data is also available, for which aggregates may be created across the total number of days and nights, creating new variables that
would allow more exploratory analyses of correlates and predictors of sleep in family caregivers of CMT. Finally, further exploration of the variables of sleep quality and depression and their correlates in family caregivers of CMT is planned.

**Conclusions**

Sleep is a complex process, sensitive to bio-behavioural influences that may affect its synchrony and regulation. It is considered essential for the promotion and maintenance of overall health, optimal performance, and daytime function. Sleep disturbance is prevalent and on the rise in the general population (Institute of Medicine [US] Committee on Sleep Medicine and Research, 2006), and it is a major public health concern (Mikkonen et al., 2013). In family caregivers of children, namely parents, it results from either sleep restriction (amount of sleep below the level of need), fragmentation (sleep that is highly broken up by frequent awakenings), or both, and it may lead to chronic sleep deprivation. If left untreated, sleep disturbances may contribute to negative physical (e.g., diabetes, cardiovascular disease) and mental (e.g., depression, anxiety) health problems that are associated with impaired performance in family caregivers. In new parents of normally developing children, chronic sleep curtailment has been associated with negative daytime consequences (i.e., less flexibility when implementing select tasks in a laboratory setting; Plessow et al., 2010).

In a systematic review of sleep outcomes in family caregivers of children who depend on the use of medical technology, the results of which are included in this dissertation and have been submitted for publication (Keilty et al., 2014), 13 studies were identified that examined sleep in family caregivers of technology-dependent children. The studies were highly varied with respect to study methods, with no intervention studies found, and all relied solely on subjective sleep measurement. Rates of self-reported sleep disturbance in family caregivers of technology-
dependent children ranged from 51–100%, sleep quality was found to be very poor, and quantity of sleep in family caregivers of ventilator dependent children was one hour less than that of family caregivers of otherwise healthy children. Thus, sleep disturbance, and its causes and consequences, was identified as a compelling aspect of the family caregiver experience for which there was little high quality evidence for its understanding.

This study examined sleep disturbance and related outcomes in family caregivers of children who depend on medical technology ($n = 42$) and compared them to family caregivers of healthy, age-matched children ($n = 43$). In this prospective study, 7 days and nights of actigraphy data were collected, and validated measures of depression, sleepiness, fatigue, quality of life, sleep hygiene, and child’s sleep quality were administered. Findings indicated that when measured objectively, family caregivers of children who depended on medical technology achieved on average less sleep per night (6.56 [1.37h] vs. 7.21 [0.56], $p = .019$), had more nocturnal awakenings (8.00 [3.93] vs. 6.01 [3.65], $p = 0.02$), and had more sleep deprived (< 6 hrs) nights per week (2.18 [2.40] vs. 0.60 [.778], $p < .001$) than family caregivers of healthy children. Scores on a subjective sleep quality measure also differed in the expected direction (poorer in family caregiver CMT; 7.8 [2.9] vs. 5.4 [2.8], $p = .001$) along with other important outcomes, including increased depression (CES-D), sleepiness (ESS), fatigue (MAF), and poorer child’s sleep quality (CSHQ; $p \leq .02$). The CARE to SLEEP Study also explored factors that contributed to sleep disturbance and identified no modifiable caregiver (e.g., sleep hygiene), child (e.g., sleep habits), and environmental (e.g., timing of homecare nursing) predictors of sleep in family caregivers CMT.

The implications for policy, practice, and future research from this study are many. That no modifiable risk factors were found to predict poor sleep outcomes in family caregivers of
CMT strongly suggests that this is not an easy problem to fix. It provides evidence that suggests having a CMT negatively affects caregiver sleep, health, and daytime functioning. Thus, based on study findings, recommendations have been made. This dissertation also outlines what is needed next in studies of sleep in family caregivers of CMT. It calls for research that will help develop and test targeted interventions in this vulnerable group to ensure better care for family caregivers of CMT, who we, as a society, and more importantly, their children, rely on so immensely 24 hours a day.

**Funding**

The conduct of this study was funded by the Canadian Respiratory Health Professionals, Canadian Lung Association, the Ontario Respiratory Care Society, Ontario Lung Association, and the Norman Saunders Complex Care Innovation Grant, SickKids Foundation.
References


*Annals of Internal Medicine, 118*(8), 622-629.


Huang, L., Tsai, M., Chen, C., & Hsu, S., (2014). The effectiveness of light/dark exposure to treat insomnia in female nurses undertaking shift work during the evening/night shift. *Journal of Clinical Sleep Medicine, 9*(7), 641-646.


Cambridge: Cambridge Press.


objective and subjective measures. *Journal of Clinical Sleep Medicine, 4,* 362-369.


Stremler, R. (2013, November). *Sleep content training in entry-to-practice health care professional programs in Canada*. Presentation at the seventh bi-annual Conference on Pediatric Sleep Medicine, Amelia Island, FL.


Research, 9(1), 49-64.


Appendix A

Summary of studies of sleep in family caregivers of older adults
## Studies of sleep in family caregivers of older adults

<table>
<thead>
<tr>
<th>Study</th>
<th>Methods</th>
<th>Participants</th>
<th>Intervention</th>
<th>Sleep Measures</th>
<th>Results/Notes</th>
</tr>
</thead>
</table>
| Akkerman, R 2004       | Type of study: Clinical trial with wait-list control  | Caregivers of recipients with AD  
N=35 Total  
N=18 Intervention  
N=18 Control  
N=5 for Actigraphy sub-sample  
Demographics: Female: 86%  
Age: 58.1y (13.8y)  | Type: Behavioral therapy, CBT  
Experimental condition:  
Weekly brief CBT over 9 sessions to address physical, cognitive, and behavioral components of anxiety.  
Control condition: Wait-list control  
Mode of delivery: small groups  
Location: out of home  | Actigraphy,  
48h x2  
Timing: At baseline and post-intervention  | Sleep: improvements from baseline in total minutes awake, number of wake episodes, and reduction in longest wake episode.  
Very small sample size (n=5) precluded statistical analysis. |
| Ancoli-Israel, S 2005  | Type of study: Randomized, double blind, clinical pilot study | Caregivers of recipients with Alzheimer’s Disease.  
N=31 galantamine  
N=32 donepezil  
Demographics:  
Age:  
galantamine 67.7y (15.9)  
donepezil 69.4y (11.4y)  
Female: 52.4%  | Type: Physiological, Drug therapy  
Experimental condition: head to head comparison of galantamine 4mg BID or donepezil 5 mg at hs for 8 weeks  
Control condition: none  
Mode of delivery: Individual tx  
Location: In the home  | Actigraphy, three x 7 day periods  
PSQI  
Timing: At baseline, week 4, week 7  | Sleep: Trend towards improvement in mean percent of sleep with galantamine; decline with donepezil  
No significant changes in subjective sleep scores with better than expected scores at baseline. |
| Carter, P 2006         | Type of study: Repeated measures clinical trial | Caregivers of recipients with cancer  
N= 15 Intervention  
N= 15 Control  
Demographics:  
Age:  
53y (17y)  
Female: 63%  | Type: Behavioral therapy, CASI  
Experimental condition: Brief intervention over 5 weeks with 2 CASI sessions (Week 2 & 4)  
Included Goal Attainment Scaling (GAS)  
Control condition: Attention control “Back Health” education and GAS  
Mode of delivery: Individual tx  
Location: In the home, or over the phone  | Actigraphy  
Sleep Logs  
PSQI  
Timing: baseline, week 3, and 2, 3 and 4 months post-baseline  | Sleep: Improvements in subjective sleep quality at week 5 (p = 0.02) and 4 months (p =0.03).  
Improvements in objective sleep were reported for the intervention group at 2 months (latency minutes 7(6), p=.05) and 4 months (duration hours 7.5 (1.2), p = .02). |
| Carter, P 2009         | Type of study: Longitudinal descriptive case series | Bereaved caregivers of recipients with cancer.  | Type: Behavioral Therapy  
Experimental condition: 2 hour CBT-I sessions  | Actigraphy, over 6 weeks  
Sleep logs  | Sleep: Significant improvement between baseline and week 5 across all subjective self-report measures (p<.02); latency |
| King, A. 2002 | Type of study: Randomized controlled trial | N=11  
Demographics:  
Age: 56y (11.77)  
Female: 64%  
and GAS weeks 2 and 4  
Control condition: none  
Mode of delivery: Individual  
Location: in the home | PSQI  
Timing: baseline, 3 and 5 weeks.  
No significant improvements on objective sleep measures. |
|---|---|---|---|
| Caregivers of recipients with dementia.  
n=51 Intervention  
n=49 Controls  
Demographics:  
Age= Intervention 62.2y (9.3)  
Control 63.3y(9.0)  
100% female | Type: Physiological, Exercise  
Experimental condition: Counseling session related to exercise plan and bi-monthly telephone f/u for 2 months, and monthly for 12 months.  
Control condition: Attention control, nutrition education.  
Mode of delivery: Individual tx  
Location: in the home, telephone supervised | PSQI  
Timing: baseline and 12 months f/u  
Sleep: subjective sleep quality better at 12 months ($p <0.045$) |
| Korn, L 2009 | Type of study: Randomized Controlled Trial | N=42  
Demographics:  
Type: Complementary Therapy  
Experimental condition:  
3h paid respite and 8 weekly standardized Polarity therapy sessions of 50 mins in native health centers  
Control condition: Enhanced Respite Condition- 3h paid respite and self-selected activity (out of the home)  
Mode of delivery: Individual tx  
Location: Activity, out of home Respite- In-home | PSQI  
Timing: baseline and post-intervention  
Sleep: PSQI showed changes in a positive direction without statistical significance  
Baseline subjective sleep scores were higher than the normal population when matched for age and gender. |
| Lee, D 2007 | Type: Prospective case series | Temperature  
Daily SD  
Actigraphy  
Timing: over 2 weeks at baseline, 2 weeks respite, and 2 weeks follow-up period.  
Caregiver sleep efficiency reduced at follow-up ($p <.05$)  
Sleep: All 4 participants reported | |
<table>
<thead>
<tr>
<th>Year</th>
<th>Authors</th>
<th>Study Type</th>
<th>Study Title</th>
<th>Sample Size</th>
<th>Condition Details</th>
<th>Timing</th>
<th>Outcome Measures</th>
<th>Results</th>
</tr>
</thead>
<tbody>
<tr>
<td>2005</td>
<td>McCurry, S</td>
<td>2005 series</td>
<td>recipients not reported</td>
<td>N=4 caregivers</td>
<td>Experimental condition: Manualized treatment for insomnia provided weekly over 4-8 weeks. Mode of delivery: Individual tx Location: ¼ in the home; ¼ in local hospital</td>
<td>Timing: baseline &amp; post-intervention (8wk) and follow-up (3mos)</td>
<td>improvements on 5 of 7 self-reported sleep variables at post-treatment and follow-up. Limited sample size precludes statistical analysis.</td>
<td></td>
</tr>
<tr>
<td>1996</td>
<td>McCurry, S</td>
<td>Type: Prospective case series</td>
<td>Caregivers of recipients with dementia</td>
<td>N=4</td>
<td>Type: Behavioral therapy Experimental condition: Brief, behavioral treatment for caregiver insomnia delivered for six 90 min sessions once a week Mode of delivery: Group tx Location: Out of home, follow-up by phone/voicemail</td>
<td>Daily SD Timing: 1 week before and up to 6th week PSQI Timing: At baseline, post-treatment and 3 months follow-up</td>
<td>Sleep: Trends towards improvements in total sleep time, sleep efficiency, sleep latency and wake-time after onset at post-treatment and 3 months follow-up. Trends toward improved caregiver subjective sleep.</td>
<td></td>
</tr>
<tr>
<td>1998</td>
<td>McCurry, S</td>
<td>Type: Clinical trial with wait-list control</td>
<td>Caregivers of recipients with dementia</td>
<td>N=36 dyads n= 7 Group Tx n=14 Individual Tx n=15 Wait list</td>
<td>Type: Behavioral therapy Experimental condition: 1) Group tx: 12 (7 active, 5 controls) caregivers, 6 week group sleep behavior/education intervention 2) Individual tx: 24 (14 active, 10 control) tx condensed to 4 weeks, and caregivers treated individually Control condition: Wait-list Mode of delivery: Group &amp; Individual tx, diaries by voicemail Location: out of home</td>
<td>Daily SD PSQI Timing: Measures at pre-post and 3 month follow-up</td>
<td>Sleep: Subjective sleep quality (PSQI scores) was better for intervention subjects at post-treatment and follow-up. (p &lt; .05)</td>
<td></td>
</tr>
<tr>
<td>2005</td>
<td>McCurry, S</td>
<td>Type: Randomized Controlled Trial</td>
<td>Caregivers of recipients with Alzheimer’s Disease.</td>
<td>N=36 dyads n=17 Intervention n= 19 Controls</td>
<td>Type: Behavioral therapy Experimental condition: NITE-AD education/training in sleep &amp; behavioral management along with telephone instruction and support in implementing sleep hygiene, daily walking and light exposure intervention. 6x1h sessions over a 2 month period and bi-weekly phone contact. Control conditions: Usual care- education and nondirective, supportive approach. Mode of delivery: Individual tx Location: in the home</td>
<td>Actigraphy Daily SD PSQI Timing: 1 week at baseline, post-intervention (2 months) and at 6 months follow-up.</td>
<td>Sleep: Intervention group experience significantly improved time awake at night at 2 month post-test and fewer awakenings per hour and were awake less time per waking than controls at 6 month follow-up (p &lt;.05) Treatment integrity addressed in detail.</td>
<td></td>
</tr>
<tr>
<td>Study</td>
<td>Type of study</td>
<td>Sample</td>
<td>Experimental condition</td>
<td>Control condition</td>
<td>Mode of delivery</td>
<td>Location</td>
<td>Sleep</td>
<td>Note</td>
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<tr>
<td>Rose, K 2009</td>
<td>Randomized, double-blind, controlled pilot study</td>
<td>Caregivers of recipients with AD n= 19 Intervention n= 19 control</td>
<td>Type: Physiological, CES</td>
<td>AlphaStim CES device 60 mins a day for 4 weeks</td>
<td>sham therapy</td>
<td>Individual tx</td>
<td>in the home</td>
<td>No significant differences in sleep disturbances. Known higher 'placebo effects' when testing medical devices.</td>
</tr>
<tr>
<td>Rowe, M 2010</td>
<td>randomized controlled clinical trial</td>
<td>Caregivers of recipients with dementia. n= 26 Intervention n= 27 control</td>
<td>Type: Physiological, nighttime monitoring system</td>
<td>Nighttime electronic monitoring system installed for use over a</td>
<td></td>
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</tbody>
</table>

**Note.** AD= Alzheimer’s disease, CBT-I= Cognitive Behavioral Therapy-Insomnia, ESS= Epworth Sleepiness Scale, GAS=Goal Attainment Scaling, GSDS: General Sleep Disturbance Scale, PSQI= Pittsburg Sleep Quality Index, SD= sleep diary.
Appendix B

Summary of studies of sleep in family caregivers of children with chronic illness
## Studies of sleep in family caregivers of children with chronic illness

<table>
<thead>
<tr>
<th>Authors</th>
<th>Study Design</th>
<th>Participants</th>
<th>Purpose of Study</th>
<th>FC Sleep and other measures</th>
<th>Sleep Results</th>
<th>Associations with sleep disturbances</th>
</tr>
</thead>
<tbody>
<tr>
<td>Andrews &amp; Nielson, 1988</td>
<td>Cross sectional</td>
<td>N= 74 Mothers of technology dependent children</td>
<td>To describe the experience of FC of TDC.</td>
<td>Single open-ended question about caregiver experience, and custom structured questionnaire.</td>
<td>Sleep loss one of the most important problems described by parents. 62% reported interruption in sleep (due to false monitor alarm).</td>
<td>Lack of confidence in night nurse False monitor alarms Noises from night nurse</td>
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<tr>
<td>Arvola, Tahvanainen &amp; Isolauri, 2000</td>
<td>Longitudinal, cohort design.</td>
<td>N= 81 parents of children with atopic dermatitis.</td>
<td>To evaluate influence of team based eczema treatment on parent expectations of care.</td>
<td>Open-ended questions and, two structured questions about sleep - sleep loss in child - parent’s fatigue on pre-post questionnaires.</td>
<td>Sleep loss was a moderate to severe concern for 54% of parents before treatment for AD and 34% after treatment. Parent fatigue reported to be 50% before treatment and 44% after treatment.</td>
<td>Child requires direct care Symptom support for pruritus Frequent awakenings</td>
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<tr>
<td>Asmussen, 1999</td>
<td>Qualitative</td>
<td>N=14 mothers and fathers of children with chronic otitis media</td>
<td>To describe family experiences with children with chronic otitis media.</td>
<td>Open ended non-specific question.</td>
<td>Sleep was one aspect of parent’s life that was disrupted.</td>
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<tr>
<td>Bowman, Lindahl &amp; Bjork, 2003</td>
<td>Cross-sectional design.</td>
<td>n=146 mothers n=118 fathers of children with cancer.</td>
<td>This study describes disease-related distress in parents.</td>
<td>Parent Psychological Distress in Childhood Cancer (includes 5 item sleep disturbances scale)</td>
<td>A total of 47.1% of parents reported sleep disturbances. A higher level of sleep disturbance was reported with a shorter time since diagnosis and/or child was active treatment.</td>
<td>Stress Time since diagnosis Active treatment or not</td>
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<td>Bowman, Viksten, Kogner &amp; Samuelsson, 2004</td>
<td>Cross-sectional design.</td>
<td>n=373 mothers n=301 fathers, of children with n=264 cancer or n=203 diabetes n=208 healthy control</td>
<td>To compare the incidence of disease-related distress symptoms in parents of children with cancer, diabetes and healthy controls.</td>
<td>Parent Psychological Distress in Childhood Cancer (PPDCC) (includes 5 item sleep disturbances scale)</td>
<td>No significant differences in sleep disturbances across the three groups. Strength of correlation of time from diagnosis and sleep disturbance strongest in diabetes.</td>
<td>Stress Time since diagnosis</td>
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<tr>
<td>Chamlin et al, 2005</td>
<td>Cross-sectional</td>
<td>N=270 parents of children with atopic dermatitis.</td>
<td>To develop a valid measure of the effects of atopic dermatitis (AD) on the quality of life of affected young children and their families.</td>
<td>Childhood Atopic Dermatitis Impact Scale (CADIS) (includes a 3-item sleep scale) –</td>
<td>14.8% of parents reported that “sleep issues” was the most bothersome thing about Atopic Dermatitis. (ranked 3/24 bothersome items)</td>
<td>Co-sleeping with child in bed with parent</td>
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<tr>
<td>Authors</td>
<td>Study Design</td>
<td>Sample Size</td>
<td>Study Aim</td>
<td>Measures</td>
<td>Key Findings</td>
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<td>Cottrell &amp; Khan, 2005</td>
<td>Cross-sectional</td>
<td>n= 50 parents of children with epilepsy</td>
<td>To describe parents sleep patterns and examine the relationship between parental sleep problems and other measures of parental adjustment to their child's chronic illness.</td>
<td>Pittsburg Sleep Quality Index (PSQI), Beck Depression Inventory-II (BDI-II), SF-36, Dyadic Adjustment Scale, RAND Health Survey</td>
<td>Mother's reported sleeping M=4.5 hours, and experienced ~3+ awakenings/night to check on child. Awakenings were positively correlated with the number of seizures (r=.32), severity of seizures (r=.42), maternal depression (r=.33) and emotional health (r=.35), (p &lt;.01) along with physical health (r=.31), pain (r=.29) and negative correlation with marital satisfaction (r=.28) (all, p &lt;.05). Sleep quality was correlated with seizure severity (r=-.28), seizure number (r=-.30), emotional health (r=.30), pain (r=.29) all p&lt;.05 in the expected direction and maternal depression (r=-.37) and marital satisfaction (r=-.37) and physical health (r=.30) with p &lt;.01. Heightened vigilance, elevated depression, negative subjective sleep quality, poor marital satisfaction, poor maternal health.</td>
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<tr>
<td>Ferrell, Rhiner, Shapiro &amp; Dierkes, 1994</td>
<td>Qualitative</td>
<td>n= 21 mothers n= 10 fathers of children with cancer</td>
<td>This study described child and parent experience and related sleep problems while living with childhood cancer</td>
<td></td>
<td>Child care needs and severity of illness: Increased number and severity of seizures. Required direct care - Child’s pain.</td>
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<tr>
<td>Study</td>
<td>Design</td>
<td>Participants</td>
<td>Methodology</td>
<td>Findings</td>
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</table>
| Gallagher, Phillips & Carroll, 2010* | Cross-sectional design with comparative cohort. | n=67 parents of children with developmental delay (DD) n=42 parents of healthy controls | This study examined the psychosocial predictors of poor sleep quality in parents caring for children with developmental disabilities. | PSQI
Single Q- does your child’s sleep patterns keep you awake?
QRSFSF SDQ
78% 'poor sleepers' (PSQI>5) in DD group versus 22% in controls (p<.001)
Poorer stress, SDQ, less social support- compared to controls
More sleep disturbance (p =.002) and poorer sleep quality, sleep latency, sleep duration, sleep efficiency, (all p <.001) reported in DD parents compared to healthy controls.
Parenting stress strongest predictor of sleep quality in DD parents. |
| Gedaldy-Duff, Lee, Nail, Nicholson & Johnson, 2006 | Descriptive, prospective study. | n=7 mothers n=6 fathers of children with cancer (ALL) | To determine the feasibility of collecting ambulatory sleep and fatigue data at home from children with acute lymphoblastic leukemia (ALL) and their parents. | 3 day sleep diary (SD), Actigraphy, Visual Analogue Scale (VAS) for fatigue.
Small sample size precludes inferential statistical analysis.
Mothers slept M= 7 hours and fathers M= 6 hours.
Mothers had 8.8% wake after sleep onset (WASO) and fathers had 14.3%.
Actigraphy recorded awakenings were 16.9 for mothers and 19.7 for fathers. |
| Heaton, Noyes, Sloper & Shah, 2006 | Cross sectional, mixed methods | n= 34 mothers n= 12 fathers of Tech Dep children | To examine sleep disruption and organisation of care routines in families of tech-dep children living at home in the United Kingdom. | Sleep measures?
Frequency of sleep disruptions
Themes related to the incidence, reasons, and effects of sleep disruptions
100% of families reported sleep disruptions, 22/36 % regularly (> 2 nights/week). |
| | | | | 'hyper' vigilance |
| | | | | Turning child. |
| | | | | Symptom mgmt pain & vomiting |
| | | | | Acuity/severity of illness |
| | | | | Required direct care |
| | | | | Frequent child awakenings |
| | | | | 'Noises from night nurse' |
| | | | | - Lack of confidence in night nurse |
| | | | | - Night support not provided |
| | | | | - Equipment problems |
| | | | | - Checking devices |
| | | | | - Vomiting due to machine |

* Higher BMI Female gender Elevated Parent stress Low mood
<table>
<thead>
<tr>
<th>Study</th>
<th>Design</th>
<th>Sample Size</th>
<th>Participants</th>
<th>Study Objective</th>
<th>Study Instrument</th>
<th>Key Findings</th>
</tr>
</thead>
<tbody>
<tr>
<td>Hopkins, Whetstone, Foster, Blaney &amp; Morrison, 2009*</td>
<td>Qualitative</td>
<td>n= 26 caregivers of children with a tracheostomy</td>
<td>To assess the impact of paediatric tracheostomy, on patients and their families.</td>
<td>Pediatric Tracheostomy Health Status Instrument (validated instrument)</td>
<td>Themes generated from structured interviews related to caregiver and child’s QOL</td>
<td>14/27 reported their sleep was disturbed often or all of the time by the child’s condition</td>
</tr>
<tr>
<td>Horner, 1997</td>
<td>Qualitative, grounded theory</td>
<td>n= 12 families of children with asthma</td>
<td>To examine the experience of mothers as they cared for their young children during illness episodes, prior to a diagnosis of asthma in the children.</td>
<td>Themes on uncertainty prior to diagnosis.</td>
<td>All of the mothers reported staying up through the night watching their child struggle to breathe. The need to be vigilant led to “wearing out” (p.661).</td>
<td>67% children woke at night, 14% choking, 31% needed help to breathe at least 1/wk, 19% needed help every night.</td>
</tr>
<tr>
<td>Lawson, Lewis-Jones, Finaly, Reid &amp; Owens, 1998</td>
<td>Cross sectional</td>
<td>n= 34 mothers n= 34 fathers of children with atopic dermatitis.</td>
<td>To identify the areas of family life most affected by AD and their perceived importance.</td>
<td>Dermatitis Family Impact Questionnaire (1 sleep and one fatigue related question)</td>
<td>64% of parents reporting frustration and exhaustion due to sleep problems.</td>
<td>‘hyper’ vigilance</td>
</tr>
<tr>
<td>Lewis-Jones, Finaly &amp; Dykes, 2001</td>
<td>Cross sectional</td>
<td>n= 89 parents of infants with AD n = 22 healthy control</td>
<td>To validate a 1) infant and 2) family measure of the impact of AD.</td>
<td>Family Dermatitis Index (FDI), Infants’ Dermatitis QOL Index (IDQOL), Infants Behavioural Checklist (BCL)</td>
<td>Sleep disturbance was the highest scoring question for parents, followed by tiredness or exhaustion.</td>
<td>Required direct care</td>
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<tr>
<td>Study</td>
<td>Design</td>
<td>Sample</td>
<td>Objectives</td>
<td>Measures</td>
<td>Findings</td>
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<td>Meltzer &amp; Mindell, 2006</td>
<td>Cross-sectional</td>
<td>Mothers of children n=47 healthy children n=42 CF n=29 ventilator dependent</td>
<td>To examine sleep patterns and causes of sleep disturbances in caregivers of children with and without chronic illnesses and to determine whether sleep mediates the relationship between a child's chronic illness and daytime functioning in caregivers.</td>
<td>Sleep: PSQI 24-Hours Sleep Patterns Inventory (SPI) Center for Epidemiological Studies-Depression Scale (CES-D) Iowa Fatigue Scale (IFS) Child Health Questionnaire (CHQ)</td>
<td>Compared to FC of healthy controls &amp; CF, FC of VENT: woke earlier (F=6.89, p &lt;.001) had a shorter TST (F=11.05, p&lt;.001), and poorer sleep quality (F=4.08, p=.02), longer sleep onset latency (F=5.75, p=.004), and poorer daytime functioning (F=6.57, p =.002) Reported sleep disturbances due to nighttime caregiving (52% VENT, 26% CF, 9% HEALTHY) and stress related to child’s illness (38% VENT and CF, 2% HEALTHY). Sleep quality found to mediate relationship between child health status and maternal depression and fatigue.</td>
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<tr>
<td>Meltzer, Boroughs &amp; Downes, 2010*</td>
<td>Cross-sectional, descriptive study</td>
<td>n=36 primary caregivers of ventilator-assisted children</td>
<td>To examine the relationship between home nursing coverage and the sleep and daytime function in parents of ventilator-assisted children.</td>
<td>Sleep: 24-Hour SPI Other: CES-D Iowa FS Stanford Sleepiness Scale Customized questionnaire. Follow-up: 2-week period following primary interview.</td>
<td>FC with 0-16h/wk night nursing had shorter sleep onset latency (SOL) than caregivers with 16-48 h/wk night nursing (p&lt;.05) Caregivers with &gt;48 h/wk night nursing had a TST approx 1 hour more than caregivers with 0-48h/wk. Caregivers with depression (F=4.13) and sleepiness (F=6.50) had fewer hours of night nursing per week than caregivers without significant symptoms of depression or sleepiness (p &lt;.05). 50% FC report awakenings due to noises from night nurse not associated with child’s care</td>
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<td>Stress r/t child’s illness Early morning - Awakening Required direct care False monitor alarms Presence of homecare</td>
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<tr>
<td>Authors</td>
<td>Study Design</td>
<td>Sample Size</td>
<td>Population/Methods</td>
<td>Outcomes</td>
<td>Sources of Sleep Disruption in FC</td>
<td>Comments</td>
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<tr>
<td>Montagnino &amp; Mauricio, 2004*</td>
<td>Cross sectional study</td>
<td>n= 50</td>
<td>50 family caregivers of a child with a tracheostomy and gastrostomy</td>
<td>To describe perceived stressors and coping strategies of the primary caregiver of a child with a tracheostomy and gastrostomy.</td>
<td>Early morning wakening</td>
<td></td>
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<tr>
<td>Moore, David, Murra, Child &amp; Arkwright, 2006</td>
<td>Cross-sectional</td>
<td>n=55 mothers n=37 fathers of children with either eczema or asthma</td>
<td>To compare the impact of caring for a child with atopic eczema vs. asthma on parents’ sleep and well-being.</td>
<td>Questionnaire about frequency and duration of parent night wakings. HADS</td>
<td>Mothers (39 min) and fathers (45 min) of children with eczema spent more time at night attending their child compared to 0 min for parents of children with asthma. Parents of children with eczema were twice as likely to have their sleep disturbed than those with asthma. Mothers of children with eczema with depression were more likely to experience nighttime sleep disruptions (OR 7.9 for &gt; 3 night awakenings in past 2 nights compared to mothers who had no night awakenings). Sleep disturbances also related to anxiety in both mothers (OR 4.6) and fathers (OR 4.9).</td>
<td>Maternal depression Parental anxiety Amount of nursing per week and presence of night nursing positively influences sleep if &gt; 16h/wk. Noises from night nurses</td>
</tr>
<tr>
<td>Nolan, Camfield &amp; Camfield, 2006*</td>
<td>Cross sectional, mixed methods.</td>
<td>n=24 parents with children with Dravet syndrome.</td>
<td>To investigate the experience of parents with a child with Dravet syndrome, a catastrophic form of epilepsy.</td>
<td>Sleep: Semi-structured interview and questionnaire. Other: Impact of Childhood Neurologic Disability Scale (ICND) questionnaire</td>
<td>Sleep issues add to the stress of parenting a child with Dravet’s syndrome.</td>
<td>Stress</td>
</tr>
<tr>
<td>Author(s), Year</td>
<td>Study Design</td>
<td>Sample Size</td>
<td>Methods</td>
<td>Findings</td>
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<tr>
<td>Read, Simonds, Kinali, Muntoni &amp; Gerrald, 2010*</td>
<td>Cross sectional, Mixed methods</td>
<td>n=10 family caregivers of children or youth with neuromuscular disorders receiving non-invasive ventilation</td>
<td>To assess the implications for sleep and well-being in patients and caregivers with neuromuscular disorders receiving non-invasive ventilation.</td>
<td>Sleep: PSQI Custom survey, qualitative interviews, and thematic analysis Other: SF-36 HADS FBS FAD FC: PSQI 7.4+/=- 4.7, 70% scored above cut-off (5) for sleep quality Sleep quality associated with: HADS total (r=.76, p=.009) SF-36, energy/vitality, (r=-.68, p =.03), emotional well-being, (r=.82, p =.007), general health, (r=-.69, p =.02) FBS total (r=.60, p=.04) FAD, disruption to family interactions, (r=.73, p=.01), mental health impact (r=.70, p =.02).</td>
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<tr>
<td>Reid &amp; Lewis-Jones, 1995</td>
<td>Cross sectional survey</td>
<td>n= 45 parents of children with AD</td>
<td>In this preliminary survey parents confirmed clinical impressions that sleep loss is a common feature when a child's atopic eczema is flaring.</td>
<td>Structured interview questions about child and parent sleep patterns. During an eczema flare, sleep disturbance reported for 86% of the nights (M = 2.7 wakings/night). Parents described losing M=2.6 hours of sleep per night. 48% of parents bought child to bed as a management strategy for eczema flares.</td>
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<td>Ricci, Bendandi, Bellini, Patrizi &amp; Masi, 2007*</td>
<td>Cross-sectional</td>
<td>n=45 family caregivers of children with AD</td>
<td>To determine the ways in which atopic dermatitis (AD) affects the lives of young Italian children and their families, in terms of quality of life, and correlate it with AD severity and the perception of severity as estimated by the family.</td>
<td>Sleep: Dermatitis Family Impact questionnaire (2 sleep items) Sleep disturbance was their greatest problem reported. Applying NPPV mask at night. Attending to child’s (other) care needs (ie. Toileting). 3/10 hospice support 4/10 homecare 5/10 nurses -variable allocation</td>
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<tr>
<td>Sullivan-Bolyai, Deatrick, Gruppuso, Tamborlane &amp; Grey, 2003</td>
<td>Qualitative</td>
<td>n= 28 mothers of children with type 1 diabetes</td>
<td>To describe the day-to-day experiences of mothers raising young children with type 1 diabetes.</td>
<td>Themes related to the meaning and mother’s management of child’s diabetes, and the meanings mothers attributed to them. Fear and constant vigilance was described in reports of chronic sleep deprivation and decision to change to co-located sleep. Fear &amp; constant vigilance. - If child required direct care. Child would come to parents bed as management strategy for eczema flares.</td>
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<tr>
<td>Sullivan-Bolyai, Knafl, Tamborlane &amp; Grey (2004)</td>
<td>Qualitative</td>
<td>n=14 mothers n=7 fathers of children with diabetes on insulin pumps</td>
<td>To describe parents experience managing their children’s diabetes using insulin pumps.</td>
<td>Semi-structured qualitative interviews In the quality of life theme, most parents reported that “sleep returned to normal” with the insulin pump (p.321).</td>
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<tr>
<td>Study</td>
<td>Methodology</td>
<td>Purpose</td>
<td>Results</td>
<td>Monitoring/Intervention</td>
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<td>Williams et al., 2000</td>
<td>Quantitative</td>
<td>To investigate if parental fears concerning seizure occurrence may be associated with behavioral changes within the home environment. One possible change involves sleeping arrangements.</td>
<td>Twenty-two percent of children with epilepsy and 8% of children with diabetes changed to a less independent sleeping arrangement following diagnosis. Twenty-four out of 25 parents of child with epilepsy reported co-sleeping due to concern about child’s epilepsy. Three out of 13 parents of child with diabetes reported co-sleeping due to concern about child’s diabetes. Sources of sleep disruption in FC: - Co-sleeping - Required direct care/observation</td>
<td>Monitoring glucose.</td>
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<td>Questionnaires concerning demographics, medical history, and sleeping arrangements were completed. 334 parents No control group. Diabetes (n =155) Epilepsy (n =179) Children ages 3 to 17 years (M = 9.4 years)</td>
<td>Questions about sleeping arrangements before and after diagnosis, question about reason for co-sleeping.</td>
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<td>Changing sleep location</td>
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<td>Yilmaz et al., 2008*</td>
<td>n= 62 mothers of children with asthma, n= 21 children with CF and n= 35 healthy children.</td>
<td>The aim of this study was to compare sleep quality and depression-anxiety parameters in mothers of children with cystic fibrosis (CF) asthma and healthy controls.</td>
<td>Sleep: PSQI Other: HADS sleep efficiency and total PSQI scores were significantly different (higher or lower?) between the groups ($p =0.02, p=0.01$ and $p =0.04$ respectively). Subjective sleep quality scores in mothers of children with asthma were higher than the ones in the control group (1.0±0.9 vs 0.6±0.7, $p =0.015$). Presence of a chronic respiratory disease in a child may be associated with disturbed sleep quality and increased depression and anxiety in mothers.</td>
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<tr>
<td>Study</td>
<td>Design</td>
<td>Participants</td>
<td>Methods</td>
<td>Findings</td>
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<td><strong>Yuksel, Sogut, Yilmaz, Demet, Ergin &amp; Kirmaz, 2007</strong>*</td>
<td>Cross-sectional design</td>
<td>n= 75 mothers of children with asthma, n= 46 mothers of healthy controls</td>
<td>To compare sleep quality in asthmatic children and their others as well as the stats of anxiety-depression in the mothers.</td>
<td>Higher subjective sleep quality on PSQI (mothers) scores were significantly with elevated asthma severity scores (child) (r=.49, p = .00). Total PSQI and depression scores (mother) were associated with elevated rates of ED use (child) (r = 0.39 and 0.31 respectively, p =.01)</td>
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<td><strong>Zupanec, Jones &amp; Stremler, 2010</strong>*</td>
<td>Descriptive, cross-sectional design</td>
<td>n=64 parents of children receiving maintenance chemotherapy for acute lymphoblastic leukemia (ALL).</td>
<td>To explore relationships between sleep habits, sleep disturbance, and fatigue for children receiving maintenance chemotherapy for acute lymphoblastic leukemia (ALL). Also, to describe sleep habits, sleep disturbance, and fatigue of parents of children and adolescents with ALL and determine if relationships exist between parent and child sleep disturbance and fatigue.</td>
<td>Parents had elevated GSDS and F-VAS, suggesting significant sleep disturbance and fatigue. Sleep: parent sleep (GSDS) and child sleep (CSHQ), r=.41, p&lt;.0001 Other: VAS-F and GSDS (r=.56, p&lt;.0001)</td>
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**Note.** * indicates studies not cited in review by Meltzer & Moore (2008). ALL= acute lymphoblastic leukemia, BDI-II = Beck Depression Inventory-II, CCF-P= Childhood Cancer Fatigue Scale-Parent, CES-D = Center for Epidemiological Studies – Depression Scale, CSS= Children’s Sleep Score, CSHQ= Children’s Sleep Habits Questionnaire, DD= developmental delay, FAD= Family Assessment Device, DASS-21= Depression Anxiety and Stress Scale, FBS= Family Burden Scale, F-VAS= Fatigue Visual Analogue Scale, GSDS: General Sleep Disturbance Scale, HADS= General Sleep Disturbance Scale, PPDCC= Parent Psychological Distress in Childhood Cancer, PHS= Parenting Hassles Scale, PSQI= Pittsburgh Sleep Quality Index, SDQ= Strengths and Difficulties Questionnaire, SPI = 24-Hour Sleep Patterns Inventor. Review updated as per November 26, 2011.
Appendix C

Search terms and strategy for systematic review
Search terms and strategy for systematic review: Sleep disturbance in family caregivers of children who depend on medical technology

The search strategy for:

**OvidSP MEDLINE <1946 to November Week 3 2013>**

<table>
<thead>
<tr>
<th>Set</th>
<th>History</th>
<th>Results</th>
<th>Comments</th>
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<td>exp Sleep Disorders/ or exp Sleep/</td>
<td>106148</td>
<td>Sleep Terms</td>
</tr>
<tr>
<td>2</td>
<td>caregivers/ or exp parents/ or (carer* or caregiver* or (care adj2 giver*).ti,ab. or maternal behavior/ or exp parent-child relations/ or parenting/ or paternal behavior/</td>
<td>158938</td>
<td>Caregiver terms</td>
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<td>3</td>
<td>1 and 2</td>
<td>2276</td>
<td>Base Clinical set 1</td>
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<td>4</td>
<td>limit 3 to &quot;all child (0 to 18 years)&quot;</td>
<td>1610</td>
<td>Age group limit</td>
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<td>exp health surveys/ or exp interviews as topic/ or narration/ or questionnaires/ or self report/ or monitoring, physiologic/ or actigraphy/ or monitoring, ambulatory/ or blood pressure monitoring, ambulatory/ or electrocardiography, ambulatory/ or esophageal ph monitoring/ or polysomnography/ or telemetry/</td>
<td>814451</td>
<td>Evaluation measurement terms</td>
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<td>6</td>
<td>4 and 5</td>
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<td>Final results</td>
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**OvidSP Embase <1980 to 2013 Week 50>**

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<th>History</th>
<th>Results</th>
<th>Comments</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>exp sleep disorder/ or exp sleep/ or sleep deprivation/</td>
<td>196683</td>
<td>Sleep Terms</td>
</tr>
<tr>
<td>2</td>
<td>(carer* or caregiver* or (care adj2 giver*).ti,ab. or exp parent/ or caregiver/ or caregiver burden/ or caregiver strain index/ or caregiver support/ or exp child parent relation/ or parental attitude/ or parental behavior/</td>
<td>203370</td>
<td>Caregiver terms</td>
</tr>
<tr>
<td>3</td>
<td>1 and 2</td>
<td>5030</td>
<td>Base Clinical set 1</td>
</tr>
<tr>
<td>4</td>
<td>limit 3 to (infant &lt;to one year&gt; or child &lt;unspecified age&gt; or preschool child &lt;1 to 6 years&gt; or school child &lt;7 to 12 years&gt; or adolescent &lt;13 to 17 years&gt;)</td>
<td>2840</td>
<td>Age group limit</td>
</tr>
<tr>
<td>5</td>
<td>exp health survey/ or questionnaire/ or open ended questionnaire/ or structured questionnaire/ or narrative/ or self report/ or (focus adj2 group*).mp. or (diary or diaries).mp. or physiologic monitoring/ or neuromuscular monitoring/ or actimetry/ or ambulatory monitoring/ or telemetry/</td>
<td>614993</td>
<td>Evaluation measurement terms</td>
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</table>

**OvidSP PsycINFO <1806 to December Week 3 2013>**

<table>
<thead>
<tr>
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<th>Comments</th>
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<tbody>
<tr>
<td>1</td>
<td>exp sleep/ or sleep deprivation/ or exp sleep disorders/ or sleep onset/ or sleep treatment/ or sleep wake cycle/ or sleepiness/</td>
<td>29551</td>
<td>Sleep Terms</td>
</tr>
<tr>
<td>2</td>
<td>caregivers/ or caregiver burden/ or exp child care/ or home care/ or home care personnel/ or respite care/ or parents/ or adoptive parents/ or exp fathers/ or foster parents/ or exp mothers/ or exp single parents/ or stepparents/ or inlaws/ or exp spouses/ or (carer* or caregiver* or (care adj2 giver*).ti,ab. or parent child relations/ or parenting/ or father child relations/ or mother child relations/ or exp parent child communication/ or parental expectations/ or parental involvement/ or parental role/</td>
<td>156468</td>
<td>Caregiver terms</td>
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<tr>
<td>3</td>
<td>1 and 2</td>
<td>870</td>
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<tr>
<td>4</td>
<td>limit 3 to (100 childhood &lt;birth to age 12 yrs&gt; or 120 neonatal &lt;birth to age 1 mo&gt; or 140 infancy &lt;age 2 to 23 mo&gt; or 160 preschool age &lt;age 2 to 5 yrs&gt; or 180 school age &lt;age 6 to 12 yrs&gt; or 200 adolescence &lt;age 13 to 17 yrs&gt;)</td>
<td>516</td>
<td>Age group limit</td>
</tr>
<tr>
<td>5</td>
<td>questionnaires/ or mail surveys/ or exp surveys/ or telephone surveys/ or interviews/ or qualitative research/ or questioning/ or narratives/ or storytelling/ or (questionnaire* or interview* or narrat*).mp. or apparatus/ or polysomnography/ or telemetry/ or monitor*.mp.</td>
<td>552673</td>
<td>Evaluation measurement terms</td>
</tr>
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<td>6</td>
<td>4 and 5</td>
<td>216</td>
<td>Final results</td>
</tr>
</tbody>
</table>

**EBSCOHost CINAHL <1983- to December 19, 2013>**

| S1   | (MH "Sleep Deprivation") OR (MH "Sleep") OR (MH "Dyssomnias") OR (MH "Sleep Disorders, Circadian Rhythm") OR (MH "Sleep Disorders, Intrinsic") | 7943    | Sleep Terms            |
| S2   | MH "Caregivers") OR (MH "Caregiver Burden") OR (MH "Role Stress") OR (MH "Parents") OR (TX carer* OR caregiver*) OR (TX care N2 giver*) OR (MH "Maternal Behavior") OR (MH "Parental Behavior") OR (MH "Parenting") OR (MH "Parent-Child Relations") | 85,630  | Caregiver terms        |
| S3   | (MH "Questionnaires") OR (MH "Open-Ended Questionnaires") OR (MH "Structured Questionnaires") OR (MH "Diaries") OR (MH "Daily Logs") OR (MH "Checklists") OR | 332,280 | Evaluation measurement terms |
(MH "Focus Groups") OR (MH "Interviews") OR (MH "Semi-Structured Interview") OR (MH "Structured Interview") OR (MH "Unstructured Interview") OR (MH "Narratives") OR (MH "Surveys") OR (MH "Self Report") OR (MH "Telemetry") OR (MH "Monitoring, Physiologic") OR "actigraphy" OR (MH "Blood Pressure Monitoring, Ambulatory") OR (MH "Electrocardiography, Ambulatory") OR (MH "Polysomnography")

<table>
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<th>S1 AND S2 AND S3</th>
<th>283</th>
<th>Base Clinical Set</th>
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<tbody>
<tr>
<td>S5</td>
<td>S1 AND S2 AND S3</td>
<td>196</td>
<td>Age group limit Final results</td>
</tr>
</tbody>
</table>

**HANDSEARCH**


Appendix D

Summary of studies in systematic review
<table>
<thead>
<tr>
<th>Authors, year</th>
<th>Participants</th>
<th>Purpose of study</th>
<th>Design</th>
<th>Measures</th>
<th>Sleep results</th>
</tr>
</thead>
<tbody>
<tr>
<td>Andrews &amp; Nielson, 1988</td>
<td>N= 74 mothers of CMT infants</td>
<td>To describe the experience of FC of CMT infants</td>
<td>Cross sectional</td>
<td>Sleep: single specific question</td>
<td>62.1% (n=46) routine sleep disturbances</td>
</tr>
<tr>
<td>Evans, Holden &amp; MacDonald, 2006</td>
<td>N=30 FC of children on home enteral tube feeds</td>
<td>To determine the practical problems that families of children on home enteral tube feeds experience in the first year post-hospital discharge.</td>
<td>Cross sectional</td>
<td>Sleep: semi-structured questionnaire with sleep specific questions</td>
<td>76% (n=22) overall sleep disturbance with 68% (n=20) reporting sleep disturbance as a nightly occurrence. Reasons for sleep disturbance: 76% (n=16) lying on tube, 10% (n=2) to change feed bag, 10% (n=2) false pump alarm, 5% (n=1) nausea and vomiting.</td>
</tr>
<tr>
<td>Evans, McDonald, Daly, Hopkins &amp; Holden, 2007</td>
<td>N=34 FC children on home enteral feeds with IMD</td>
<td>To identify common safety issues and carer pressures for patients with IMD on home enteral tube feeds.</td>
<td>Cross sectional</td>
<td>Sleep: semi-structured questionnaire with sleep specific questions</td>
<td>100% (n=34) sleep disturbance, 56% getting up once or twice, and 24% getting up five or more times nightly. Nighttime disturbances from: 68% children lying on the tubing and blocking the flow or 38% the pump alarming for unknown reasons. For 73%, pumps alarmed once or twice, and in 9% pumps alarmed three or more times nightly.</td>
</tr>
<tr>
<td>Evans, Shelton, Holden, Daly, Hopkins &amp; McDonald, 2010</td>
<td>N=40 FC children on home enteral feeds with IMD</td>
<td>To assess the safety aspects of carers’ enteral feeding technique when home enteral tube feeding children with inherited metabolic disorders.</td>
<td>Cross sectional</td>
<td>Sleep: questionnaire Other: home observations</td>
<td>58% (n=23) sleep disturbance due to room-sharing with their children and 18% (n=7) due to bed-sharing</td>
</tr>
<tr>
<td>Feeley, Turner-Henson, Christian, Avis, Heaton, Lozano &amp; Su, 2013</td>
<td>N= 61 maternal caregivers of young children with BPD on home oxygen</td>
<td>To investigate the relationship between sleep quality, stress, caregiver burden and quality of life in caregivers of young children with BPD.</td>
<td>Cross sectional</td>
<td>Sleep: PSQI Other: PSS, Montgomery-Borgatta Caregiver Burden Scale-Revised, WHO QOL-BREF, CES-D Self reported TST M= 5.8h +/- 1.7. 78.7% 'poor' sleep quality (PSQI M=7.6 +/- 3.7). Sleep quality most significant predictor of QOL (r=-.292**)</td>
<td></td>
</tr>
<tr>
<td>Hopkins, Whetstone, Foster, Blaney, &amp; Morrison, 2009</td>
<td>N= 26 FC of children with tracheostomy</td>
<td>To assess the impact of pediatric tracheostomy, on patients and their families.</td>
<td>Cross sectional</td>
<td>Sleep: structured interviews Other: PTHSI</td>
<td>53.8% (n=14) sleep disturbed ‘often or all of the time’ (by the child’s condition)</td>
</tr>
<tr>
<td>Meltzer &amp; Mindell, 2006</td>
<td>N=118 mothers n= 29 of HMV n=42 of CF n= 47 of healthy controls</td>
<td>To examine: 1) sleep patterns and causes of sleep disturbances in caregivers of children with and without chronic illnesses, and 2) whether sleep mediates the relationship between a child's chronic illness and daytime functioning in caregivers.</td>
<td>Cross sectional</td>
<td>Sleep: PSQI, 24-hour SPI Other: CES-D, IFS, CHQ Mothers of HMV compared to mothers of healthy children had: shorter TST** (6.31h [1.2] vs 7.34h [0.9]), woke earlier* (6:21 [39 min] vs 7:01 [46min]), more night awakenings** (1.39 [0.9] vs .85 [7.7]) and poorer global sleep quality** (PSQI 10.7 [3.7] vs 5.94 [3.2])</td>
<td></td>
</tr>
<tr>
<td>Meltzer, Boroughs, &amp; Downes, 2010</td>
<td>N= 36 FC of children on HMV n= 27 mothers, n= 7 fathers, n= 1 foster mother, n= 1 grandmother</td>
<td>To examine the relationship between home nursing coverage and the sleep and daytime function in parents of ventilator-assisted children.</td>
<td>Cross sectional</td>
<td>Sleep: 24-Hour SPI Other: CES-D, IFS, SSS SOL longer <strong>for FC without night nursing compared to those with (36.34 min ±18.28 vs 21.68 min ± 18.32). TST ~ 1 hour shorter</strong> among FC with ≤ 48h night nursing compared to those with &gt; 48h night nursing (5.98h vs 6.68h respectively)</td>
<td></td>
</tr>
<tr>
<td>Heaton, Noyes, Sloper &amp; Shah, 2006</td>
<td>N=36 FC of CMT n= 34 mothers n= 12 fathers</td>
<td>To examine sleep disruption in families of TDC in the UK.</td>
<td>Mixed methods</td>
<td>Sleep: Surveys, series of questions, with probes</td>
<td>61.1% (n=22) regular sleep disruptions Themes described: Incidence of sleep disruption; Reasons for sleep disruption; Help with care overnight; Effects of sleep disruption</td>
</tr>
<tr>
<td>Study</td>
<td>Sample Description</td>
<td>Methods</td>
<td>Findings/Results</td>
<td></td>
<td></td>
</tr>
<tr>
<td>----------------------------------------------------------------------</td>
<td>------------------------------------------------------------------------------------</td>
<td>------------------------------------------------------------------------</td>
<td>---------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------</td>
<td></td>
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</tr>
<tr>
<td>Mednick, Cogen &amp; Streisland (2004)</td>
<td>N=22 FC n=18 mothers and n=4 fathers of children who transitioned to insulin pump therapy.</td>
<td>Mixed methods</td>
<td>Overall greatest improvement was reported in ‘flexibility of sleep schedules’ (4.68 [0.57]) (5 point scale where higher scores indicated greater satisfaction)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Montagnino &amp; Mauricio(2004)</td>
<td>N=18 FC of children with tracheostomy and gastrostomy</td>
<td>Mixed methods</td>
<td>In the ‘fatigue’ theme, FC reported “taking care of him by ourselves…having him all day and all night without much sleep” (p. 379)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Read, Simonds, Kinali, Muntoni, &amp; Garralda, 2010</td>
<td>N=10 FC youth with NM using NIV n= 8 mothers, n=2 fathers</td>
<td>Mixed methods</td>
<td>70% 'poor’ sleep quality on PSQI 7.4 (4.7) Associated with: negative emotional symptoms, reports of illness, burden on family health and relationships.</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Sullivan-Bolyai, Knafl, Tamborlane &amp; Grey (2004)</td>
<td>N=21 mothers and fathers of children on insulin pumps.</td>
<td>Qualitative</td>
<td>In the ‘quality of life’ theme, most parents reported that ‘sleep returned to normal’ with the insulin pump (p. 321).</td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

**Note.** BPD, Bronchopulmonary Dysplasia; CES-D, Center for Epidemiological Studies - Depression Scale; CHQ, Child Health Questionnaire – General health scale; CF, Cystic fibrosis; CMT, Children who depend on medical technology; FAD, Family Assessment Device; FBS, Family Burden Interview Schedule; FC, Family caregivers; HMV, Home mechanical ventilation; HADS, Hospital Anxiety and Depression Scale; IFS, Iowa Fatigue Scale; IMD, inherited metabolic disorders, IPTSQ, Insulin Pump Therapy Satisfaction; NM, Neuromuscular; NIV, Non-invasive ventilation; PSQI, Pittsburg Sleep Quality Index; PSS, Perceived Stress Scale; PTHSI, Pediatric Tracheostomy Health Status Instrument; SOL = Sleep Onset Latency; SPI,24-Hour Sleep Patterns Inventory; SSS, Stanford Sleepiness Scale; SF-36, Health Survey Questionnaire Short Form; TST, Total Sleep Time; UK, United Kingdom; WHO QOL-BREF, World Health Organization’s Quality of Life Brief Scale

**Level of significance:** *p* < .001, **p** < .05
Appendix E

Characteristics of sample participants in systematic review
### Characteristics of sample participants in studies reporting on sleep outcomes in family caregivers of CMT

<table>
<thead>
<tr>
<th>Authors, year</th>
<th>Participants</th>
<th>FC Age</th>
<th>Child Age</th>
<th>Technology Use</th>
<th>Homecare hours</th>
<th>Level of education</th>
<th>Employment</th>
<th>Marital status</th>
<th>Additional children</th>
<th>Race/Ethnicity</th>
<th>Bias/limitations</th>
</tr>
</thead>
<tbody>
<tr>
<td>Evans, McDonald, Daly, Hopkins &amp; Holden (2007)</td>
<td>N=34 FC children on home enteral feeds with IMD</td>
<td>NR</td>
<td>4.1 years</td>
<td>button gastrostomy, nasogastric tube and gastrostomy tube. Duration: 59% 2–5 years, 24% &gt; 5 years</td>
<td>1 received respite care</td>
<td>NR</td>
<td>NR</td>
<td>15% single</td>
<td>56% two siblings; 18% between four and eight children</td>
<td>53% Pakistani origin, 38% Caucasian and 9% Indian origin.</td>
<td>Self-report bias. Subjective measures.</td>
</tr>
<tr>
<td>Feeley, Turn-Menson, Christian, Avis, Heaton, Lozano &amp; Su, 2013.</td>
<td>N= 61 maternal caregivers of young children with BPD on home oxygen</td>
<td>29.6 yrs</td>
<td>13.93 mos</td>
<td>home oxygen &amp; diuretics</td>
<td>NR</td>
<td>38/61 had some college education or above</td>
<td>NR</td>
<td>23/61 married</td>
<td>36/61 have at least one other child</td>
<td>67.2% African American, 32.8% Caucasian</td>
<td>Self-report bias, subjective measures. Social-desirability, not wanting to suggest their child is a burden. Volunteer convenience sample, possible self-selection bias. Caregiver burden scale, used in non-standardized manner.</td>
</tr>
<tr>
<td>Source</td>
<td>N</td>
<td>Age</td>
<td>Tracheostomy and/or nutritional support</td>
<td>Duration trach</td>
<td>Daily help</td>
<td>Unemployed/Part-time</td>
<td>Other measures</td>
<td></td>
<td></td>
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</tr>
<tr>
<td>Hopkins, Whetstone, Foster, Blaney, &amp; Morrison, 2009.</td>
<td>26</td>
<td>5 yrs</td>
<td>35% 1-2 y 31% 2-3 yrs 35% 3+ y</td>
<td>11/26 none, 5/26 1-2 days 2/26 3-4 days 1/26 5-6 0/26 7 days</td>
<td>NR</td>
<td>NR</td>
<td>NR</td>
<td>Self-report bias, Subjective measures Small sample size, No comparator group</td>
<td></td>
<td></td>
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</tr>
<tr>
<td>Meltzer &amp; Mindell, 2006.</td>
<td>118</td>
<td><em>37.9yrs</em></td>
<td>HMV, type not defined</td>
<td>NR</td>
<td>NR</td>
<td>NR</td>
<td>NR</td>
<td>Self-report bias, subjective measures Single-reporter, single time point Limited sociodemographic diversity Self-selection bias, lower refusal rates in healthy controls</td>
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<tr>
<td>Meltzer, Boroughs, &amp; Downes, 2010.</td>
<td>36</td>
<td>7.0 yrs</td>
<td>Home mechanical ventilation with 93% IPPV, 7% NIPPV</td>
<td>Received average 76 hours per week, average 35 hrs for nighttime 27% no nursing hours</td>
<td>NR</td>
<td>NR</td>
<td>NR</td>
<td>Self-report bias, subjective measures Small, convenience sample Number of nursing hours exceeds most jurisdictions Single-reporter, single time point</td>
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<td></td>
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</tr>
<tr>
<td>Heaton, Noyes, Sloper &amp; Shah, 2006.</td>
<td>34</td>
<td>7.8 yrs</td>
<td>Artificial nutrition, tracheostomy and/or suctioning, renal dialysis, assisted ventilation, or oxygen therapy</td>
<td>NR</td>
<td>NR</td>
<td>NR</td>
<td>NR</td>
<td>Self-report bias, Subjective measures.</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Montagnino &amp; Mauricio(2004).</td>
<td>18</td>
<td>30.8 yrs</td>
<td>Tracheostomy and gastrostomy</td>
<td>31.3%: 4-8h/day 31.1%: 9-12h/day 37.6%: 13-24h/day 88.9% reported</td>
<td>NR</td>
<td>NR</td>
<td>NR</td>
<td>Self-report bias, low response bias, primary language English Only mothers Race and SES not obtained</td>
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</table>
 receiving assistance from professional nursing services

<table>
<thead>
<tr>
<th>Study</th>
<th>Sample Size</th>
<th>Age (mean)</th>
<th>Duration (mean)</th>
<th>NIV Use</th>
<th>Education</th>
<th>Ethnicity</th>
<th>Notes</th>
</tr>
</thead>
<tbody>
<tr>
<td>Read, Simonds, Kinali, Muntoni, &amp; Garralda, 2010.</td>
<td>N=10 FC youth with NM using NIV, n=8 mothers, n=2 fathers</td>
<td>50.3 yrs</td>
<td>19.7 yrs</td>
<td>NIV used from an average of 9.13-12.11 hrs/nighttime and 3.7-5.3h/day</td>
<td>NR</td>
<td>NR</td>
<td>NR</td>
</tr>
<tr>
<td>Sullivan-Bolyai, Knaff, Tamborlane &amp; Grey (2004).</td>
<td>N=21 mothers and fathers of children on insulin pumps</td>
<td>38.0 yrs</td>
<td>7 yrs</td>
<td>Use of insulin pump: 3-36 months</td>
<td>NR</td>
<td>16+/−2 yrs education</td>
<td>NR</td>
</tr>
</tbody>
</table>

*Note: BPD, Bronchopulmonary Dysplasia; CF, Cystic fibrosis; CMT, Children who depend on medical technology; FC, Family caregivers, HMV, Home mechanical ventilation; IMD, inherited metabolic disorders; NM, Neuromuscular; NR, Not Reported; NIV, Non-invasive ventilation; Trach, Tracheostomy; UK, United Kingdom; *reported here for FC of children on HMV
Appendix F

Letter of information to study participants
Letter of Information to Family Caregivers

555 UNIVERSITY AVENUE
TORONTO, ONTARIO
CANADA M5G 1X8
Phone: 416-xxx-xxxx
www.sickkids.ca

THE HOSPITAL FOR SICK CHILDREN

The CARE to SLEEP Study

‘SLEEP disturbance in a prospective cohort of family CAREgivers of children who depend on medical technology and family CAREgivers of healthy children’

We would like to tell you about a research study that may be of interest to you. We are a group of researchers who study sleep in family caregivers, like parents and grandparents, of children. At an upcoming clinic visit to Paediatric Clinic you may learn more about our study and tell us if you would like to participate.

We think that sleep is important for your overall health and how you feel in the daytime. Parents and other family caregivers of children who use medical equipment at home like oxygen, breathing machines and feeding pumps sometimes describe having difficulty with their sleep.

The purpose of this research is to study the sleep in family caregivers of children who rely on the use of medical technology and compare it to the sleep of family caregivers with healthy children who do not rely on such technologies.

This information will help us learn about what might help family caregivers of children get better sleep and also help us plan future studies where we will test ways to improve sleep in family caregivers.

Family caregivers who agree to participate in this study will be asked to:

- complete a brief questionnaire asking information about them and their child.
- wear an actigraph, which is like a wristwatch for 7 days and nights. (This actigraph will collect information that helps us learn about the caregiver’s sleep.)
- fill out a sleep diary to tell us about what may have kept them awake or woken them up
- complete a questionnaire during a home visit when the actigraph is ready to come off

We will be available at your upcoming clinic appointment and will be happy to answer any of your questions and tell you more about this study to determine if you may be interested in participating.
Krista Keilty, MN, NP-Paediatrics, PhD-Candidate

Nurse Practitioner, Centre for Innovation & Excellence in Child and Family Centred Care
Project Investigator, Research Institute
The Hospital for Sick Children
Toronto, ON

PhD-Candidate
Lawrence S. Bloomberg Faculty of Nursing
University of Toronto
Toronto, ON

krista.keilty@sickkids.ca
pager: 416-xxxx-xxxx
phone: 416-xxx-xxxx, ext. xxxx
Appendix G

Letter of information: Parents of healthy adolescent children
The CARE to SLEEP Study

‘SLEEP disturbance in a prospective cohort of family CAREgivers of children who depend on medical technology and family CAREgivers of healthy children’

We would like to tell you about a research study that may be of interest to you. Another family or health care provider from xxx Paediatrics or the xxx Clinic has suggested that you may be interested in learning more about and participating in our study.

We are a group of researchers who study sleep in family caregivers, like parents and grandparents, of children. We think that sleep is important for overall health and how one feels in the daytime. Parents and other family caregivers of children who use medical equipment at home like oxygen, breathing machines and feeding pumps sometimes describe having difficulty with their sleep.

The purpose of this research is to study the sleep in family caregivers of children who rely on the use of medical technology (these family caregivers are being recruited from SickKids) and compare it to the sleep of family caregivers with healthy children who do not rely on such technologies (these family caregivers are being recruited from the community, including xxx Paediatrics and the xxx Clinic).

This information will help us learn about what might help family caregivers of children get better sleep and also help us plan future studies where we will test ways to improve sleep in family caregivers.

Family caregivers who agree to participate in this study will be asked to:

- complete a brief questionnaire asking information about them and their child.
- wear an actigraph, which is like a wristwatch for 7 days and nights. (This actigraph will collect information that helps us learn about the caregiver’s sleep.)
- fill out a sleep diary to tell us about what may have kept them awake or woken them up
- complete a questionnaire during a home visit when the actigraph is ready to come off
If you are interested in learning more about The CARE to SLEEP Study, please contact us by emailing or calling the numbers below. We will be available at your convenience to answer your questions, determine if you are a fit for our study, and find out if you may be interested in participating in this research.

Krista Keilty, NP-Paediatrics, PhD-Candidate

Nurse Practitioner, Centre for Innovation & Excellence in Child and Family-Centred Care
Project Investigator, Research Institute
The Hospital for Sick Children
Toronto, ON

PhD-Candidate
Lawrence S. Bloomberg Faculty of Nursing
University of Toronto
Toronto, ON

krista.keilty@sickkids.ca
pager: 416-xxx-xxxx
phone: 416-xxx-xxx, ext. xxxx
Appendix H

Study eligibility screening forms
Eligibility Questionnaire:
Family Caregivers of children who depend on medical technology.

**All answers in this section MUST BE ‘YES’ TO BE ELIGIBLE**

<table>
<thead>
<tr>
<th>YES</th>
<th>NO</th>
</tr>
</thead>
<tbody>
<tr>
<td>1. Are you the primary caregiver (provides majority of care among family members) for your child at home?</td>
<td></td>
</tr>
<tr>
<td>2. Is your child under 18 years of age?</td>
<td></td>
</tr>
<tr>
<td>3. Do you anticipate that your child will use medical technology* on a <strong>nightly basis</strong> over the coming week (ie. the full 7 nights)?</td>
<td></td>
</tr>
<tr>
<td>4. Do you live within a 45-minute driving distance from the clinic?</td>
<td></td>
</tr>
<tr>
<td>5. Do you understand and read English well?</td>
<td></td>
</tr>
</tbody>
</table>

**All answers in this section MUST BE ‘NO’ TO BE ELIGIBLE**

<table>
<thead>
<tr>
<th>YES</th>
<th>NO</th>
</tr>
</thead>
<tbody>
<tr>
<td>1. Do you have a diagnosed sleep disorder?</td>
<td></td>
</tr>
<tr>
<td>2. Do you have an infant other than ________________(name of child) at home under 12 mos of age?</td>
<td></td>
</tr>
<tr>
<td>3. Do you have more than one child living at home who depends on the use of medical technology?</td>
<td></td>
</tr>
</tbody>
</table>

*Example of types of medical technology that may be used at home:

1) home oxygen
2) tracheostomy
3) continuous positive airway pressure (i.e., CPAP)
4) non-invasive ventilation (e.g., BiPAP)
5) invasive ventilation via tracheostomy
6) feeding tube and electric pump
7) feeding tube and gravity set
8) other: ___________________________

The following will help us keep track of the study:

Date approached for consent: ☐☐☐☐ ☐☐☐☐ ☐☐☐☐ ☐☐☐☐

Reasons for non-consent:

☐ Not interested in this study
☐ Did not have time to participate
☐ Didn’t think sleep was a problem
☐ Other: ___________________________________________
Eligibility Questionnaire:
Family Caregivers of healthy children who do not depend on medical technology

**All answers in this section MUST BE ‘YES’ TO BE ELIGIBLE**

- 1. Are you the primary caregiver (provides majority of care among family members) for your child at home?
- 2. Is your child under 18 years of age?
- 3. Do you live within a 45-minute driving distance from the clinic?
- 4. Can you understand and read English well?

**All answers in this section MUST BE ‘NO’ TO BE ELIGIBLE**

- 1. Do you have a diagnosed sleep disorder?
- 2. Does ________ (name of child) have any known chronic health conditions (e.g. asthma, learning disability)?
- 3. Do you have an infant ________ (name of child) at home under 12 mos of age?

_The following will be filled out by a member of the research team to help us keep track of the study:_

<table>
<thead>
<tr>
<th>Date approached for consent:</th>
<th>☐☐☐☐ ☐☐☐☐ ☐☐☐☐ ☐☐☐☐</th>
</tr>
</thead>
</table>

**Reasons for non-consent:**

☐ Not interested in this study
☐ Did not have time to participate
☐ Didn’t think sleep was a problem
☐ Other: ____________________________________________________________
Appendix I

Study consent forms
Consent Form- Family Caregivers of Children who depend on Medical Technology

Title of Research Project:

The CARE to SLEEP Study- short title

‘SLEEP disturbance in a prospective cohort of family CAREgivers of children who depend on medical technology and family CAREgivers of healthy children’- long title

Investigator(s):

Krista Keilty, NP-Paediatrics, PhD-Candidate, Faculty of Nursing, University of Toronto, Toronto, ON, 416-xxx-xxxx and Centre for Nursing/Research Institute, The Hospital for Sick Children, Toronto, ON, 416-xxx-xxxx, ext. xxxx; pager: 416-xxx-xxxx

Robyn Stremler, RN, PhD, Faculty of Nursing, University of Toronto, Toronto, ON, 416-xxx-xxxx and Centre for Nursing/Research Institute, The Hospital for Sick Children, Toronto, ON

Eyal Cohen, MD, FRCPC, Paediatric Medicine, The Hospital for Sick Children, Toronto, ON, 416-xxx-xxxx

Karen Spalding, RN, PhD, Faculty of Nursing, Ryerson University, 416-xxxx-xxxx, Ext. xxxx

Eleanor Pullenayegum, PhD, Biostatistician, McMaster University, 905-xxx-xxxx, Ext xxxx

Research Staff:

Michelle Ho, RN, MA, Nursing Research Intern, The Hospital for Sick Children, Toronto, ON & Clinical Research Project Assistant, Faculty of Nursing, University of Toronto, Toronto, ON 416-xxx-xxxx, Ext xxxx; pager 416-xxx-xxxx

Purpose of the Research:

Sleep is important for your overall health and how you feel in the daytime while awake. Parents and other family caregivers of children who use medical equipment at home like oxygen, breathing machines and feeding pumps sometimes describe having difficulty with their sleep. The purpose of this research is to study sleep in family caregivers of children who use medical devices and compare it to the sleep of family caregivers with children who do not use these devices. We will also study how sleep problems in family caregivers are related to their health and how they feel in the daytime. Finally, we are interested in learning about what things may influence sleep in family caregivers. This information will help us learn about what might help
family caregivers of children get better sleep and also help us plan future studies where we will test ways to improve sleep in family caregivers.

**Description of the Research:**

In order to answer the study questions, one family caregiver (parent, grandparent, or other) who is most responsible for the care of a child 1 to 18 years old who is a patient in the complex care clinics in Paediatric Medicine or Respiratory Medicine at SickKids will be asked to participate in this study. To complete this study, a total of 40 participants will be recruited from SickKids Complex Care clinics and another 40 participants from community paediatric clinics.

Participation in the study involves the following:

1. If you consent the study investigator or a Research Assistant will help you complete fill a short questionnaire. The questionnaire asks you for some background information about you and your child. The questionnaire takes about 20 minutes to complete.
2. Once you have completed the questionnaire you will begin wearing an Actigraph. The Actigraph is similar in size to a watch. This device measures when you are at rest and when you are being active. This will help us know when you are sleeping. We ask that you wear the Actigraph for the remainder of the day when we give it to you and for the next 7 nights and days. You will wear the Actigraph around your wrist, just like a wristwatch.
3. During the time you are wearing the Actigraph we also ask that you fill out a ‘sleep diary’. This diary asks you to record when and where you are sleeping and what things wake you up. In the sleep diary there are also some questions for you to answer. The questions ask you about your sleep. It will take about 10 minutes each day to complete.
4. Over the seven days that you are wearing the Actigraph a member of the research team will call you at home or email you (your choice) to remind you to complete the sleep diary, answer any questions you may have, and to help you complete your sleep diary, if needed.
5. On the final day we will visit you at home to pick up the Actigraph and your sleep diary. This visit will be arranged at a time that is convenient for you. At this visit we will ask you to fill out a final questionnaire. The questions are aimed at helping us understand your sleep and your feelings during the time you are participating in this study. There are also questions that will ask you what you liked and disliked about being in the study and what things you think helped you to get sleep or stopped you from getting sleep. The final questionnaire will take about 45 minutes to complete.
6. If you agree, we may take a picture of where you and your child sleep so that we can use it to show other doctors and nurses and researchers who want to hear about our study. There is a separate consent for you to complete if you agree to having us take photos.

**Potential Harms:**

If you have an allergy to metal, wearing the actigraph may give you a rash. If this happens and you still want to participate, you can wear the actigraph over your clothing or a sweatband that we will provide. There are no other potential harms associated with participation.
in this study. There are no consequences if you should choose not to participate in this research study.

**Potential Discomforts or Inconvenience:**

As part of the study a member of the research team will call you at home or email you on each of the seven days that you wear the actigraph. We will also schedule a home visit at the end of the study. Every attempt will be made to ensure minimal disruption to your schedule. It may be inconvenient for you to wear the actigraph, take our phone calls, have us visit you at home and for you to fill out the sleep diary over a seven-day period.

**Potential Benefits:**

You and your child are not expected to benefit directly from participating in this study. If by completing the study questionnaires it becomes known that you may benefit from support, and if you agree, we will refer you to a relevant health care provider.

We hope that the information gained from the study will help us learn more about improving sleep for parents and other family caregivers of children. This information will also help us plan a bigger study in the future.

If you are interested in the outcome of the study, please let the researcher know by filling out the attached sheet and you will be sent a summary of the results after the study is completed.

**Confidentiality:**

We will respect your privacy. No information about who you are will be given to anyone or be published without your permission, unless the law makes us do this. For example, the law could make us give information about you

- If a child has been abused
- If you have an illness you could spread to others
- If you or someone else talks about suicide (killing themselves), or
- If the court orders us to give them the study papers.

When you answer questions about your feelings, your answers might tell us that you are at higher risk for depression. If the study nurse thinks that you are at higher risk for depression, she will recommend to you that you see your family doctor or another health care provider who specializes in mental health. If you request, the social worker in the Complex Care clinic will also be available to talk to you about your feelings.

Records identifying you and your child may be given to and inspected by the SickKids Clinical Research Office Monitor. By signing this consent form you agree to let these people look at you and your child’s records. We will put a copy of this research consent form in your child’s patient health record.

All information collected during the study will be stored in a secure, locked location. Only a copy of this consent will be placed on the health record. You will also be provided with a copy of the consent. The results of this study will be grouped together and your information will not be individually identified. Only members of the research team (and maybe those individuals
described above) will have access to the data. Following completion of the research study the data will be kept as long as required by the SickKids “Records Retention and Destruction” policy. The data will then be destroyed according to this same policy. Published study results will not reveal your identity.

**Reimbursement:**
We will provide you with some compensation, a $25 gift card to Shoppers Drug Mart, in recognition of your time and effort.

**Participation:**
It is your choice to take part in this study. You can stop at any time. The care you get at Sick Kids will not be affected in any way by whether you take part in this study. You may also still participate in the study if you choose not to receive the daily reminder phone calls or emails.

New information that we get while we are doing this study may affect your decision to take part in this study. If this happens, we will tell you about this new information. And we will ask you again if you still want to be in the study.”.

During this study we may create new tests, new medicines, or other things that may be worth some money. Although we may make money from these findings, we cannot give you [your child] any of this money now or in the future because you [or your child] took part in this study.”

For you to participate in this study, and if your child is able, we will also ask your child and document his/her consent or assent if he/she agrees to having you tell us about his/her health condition as it relates to the research study.

If you (your child) become ill or are harmed because of study participation, we will treat you (your child) for free. Your signing this consent form does not interfere with your legal rights in any way. The staff of the study, any people who gave money for the study, or the hospital are still responsible, legally and professionally, for what they do.

**Sponsorship:**
The funder of this research is The Lung Association. The sponsor of the research is the Centre for Nursing, Hospital for Sick Children.

**Conflict of Interest:**
Krista Keilty and the other research team members have no conflict of interest to declare.
The CARE to SLEEP Study

Investigator(s):
Krista Keilty, NP-Paediatrics, PhD candidate, Faculty of Nursing, University of Toronto, Toronto, ON, 416-xxx-xxxx and Centre for Nursing/Research Institute, The Hospital for Sick Children, Toronto, ON, 416-xxx-xxxx, ext. xxxx; pager: 416-xxx-xxxx
Robyn Stremler, RN, PhD, Faculty of Nursing, University of Toronto, Toronto, ON, 416-xxx-xxxx and Centre for Nursing/Research Institute, The Hospital for Sick Children, Toronto, ON Eyal Cohen, MD, FRCPC, Paediatric Medicine, The Hospital for Sick Children, Toronto, ON
Karen Spalding, RN, PhD, Faculty of Nursing, Ryerson University, 416-xxx-xxxx, Ext. xxxx
Eleanor Pullenayegum, PhD, Biostatistician, McMaster University, 905-xxx-xxxx, Ext xxxxx

Consent:
“By signing this form, I agree that:
1) You have explained this study to me. You have answered all my questions.
2) You have explained the possible harms and benefits (if any) of this study.
3) I know what I could do instead of taking part in this study. I understand that I have the right not to take part in the study and the right to stop at any time. My decision about taking part in the study will not affect my health care at Sick Kids.
4) I am free now, and in the future, to ask questions about the study.
5) I have been told that my medical records will be kept private except as described to me.
6) I understand that no information about who I am will be given to anyone or be published without first asking my permission.
7) I agree, or consent, to take part in this study.

_____________________________________________________
Printed Name of Subject & Age  Subject’s signature & date

_____________________________________________________
Printed Name of person who explained consent & date  Signature of Person who explained consent

_____________________________________________________
Printed Witness’ name (if the subject/legal guardian does not read English)  Witness’ signature & date

If you have any questions about this study, please call Krista Keilty at 416-xxx-xxxx, ext.xxxx.

If you have questions about your rights as a subject in a study or injuries during a study, please call the Research Ethics Manager at 416-xxx-xxxx.”
The CARE to SLEEP Study:

SLEEP disturbance in a prospective cohort of family CAREgivers of children who depend on medical technology and family CAREgivers of healthy children

If you would like to receive a summary of the study results, please print your name and address where we can send the summary to below:

Name: ________________________________

Address: ________________________________

__________________________________________________________________
Title of Research Project:

The CARE to SLEEP Study- short title

‘SLEEP disturbance in a prospective cohort of family CAREgivers of children who depend on medical technology and family CAREgivers of healthy children’- long title

Investigator(s):

Krista Keilty, NP-Paediatrics, PhD-Candidate, Faculty of Nursing, University of Toronto, Toronto, ON, 416-xxx-xxxx and Centre for Nursing/Research Institute, The Hospital for Sick Children, Toronto, ON, 416-xxx-xxxx, ext. xxxx; pager: 416-xxx-xxxx

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Karen Spalding, RN, PhD, Faculty of Nursing, Ryerson University, 416-xxxx-xxxx, Ext. xxxx

Eleanor Pullenayegum, PhD, Biostatistician, McMaster University, 905-xxx-xxxx, Ext xxxx

Research Staff:

Michelle Ho, RN, MA, Nursing Research Intern, The Hospital for Sick Children, Toronto, ON & Clinical Research Project Assistant, Faculty of Nursing, University of Toronto, Toronto, ON 416-xxx-xxxx, Ext xxxx; pager 416-xxx-xxxx

Purpose of the Research:

Sleep is important for your overall health and how you feel in the daytime while awake. Parents and other family caregivers of children who use medical equipment at home like oxygen, breathing machines and feeding pumps sometimes describe having difficulty with their sleep. The purpose of this research is to study sleep in family caregivers of children who use medical devices and compare it to the sleep of family caregivers with children who do not use these devices. We will also study how sleep problems in family caregivers are related to their health and how they feel in the daytime. Finally, we are interested in learning about what things may
influence sleep in family caregivers. This information will help us learn about what might help family caregivers of children get better sleep and also help us plan future studies where we will test ways to improve sleep in family caregivers.

**Description of the Research:**

In order to answer the study questions, one family caregiver (parent, grandparent, or other) who is most responsible for the care of a child 1 to 18 years old who is a patient in a Paediatric Community Clinic will be asked to participate in this study. To complete this study, a total of 40 participants will be recruited from SickKids Complex Care clinics and another 40 participants from community paediatric clinics.

Participation in the study involves the following:

7. If you consent the study investigator or a Research Assistant will help you complete fill a short questionnaire. The questionnaire asks you for some background information about you and your child. The questionnaire takes about 20 minutes to complete.

8. Once you have completed the questionnaire you will begin wearing an Actigraph. The Actigraph is similar in size to a watch. This device measures when you are at rest and when you are being active. This will help us know when you are sleeping. We ask that you wear the Actigraph for the remainder of the day when we give it to you and for the next 7 nights and days. You will wear the Actigraph around your wrist, just like a wristwatch.

9. During the time you are wearing the Actigraph we also ask that you fill out a ‘sleep diary’. This diary asks you to record when and where you are sleeping and what things wake you up. In the sleep diary there are also some questions for you to answer. The questions ask you about your sleep. It will take about 10 minutes each day to complete.

10. Over the seven days that you are wearing the Actigraph a member of the research team will call you at home or email you (your choice) to remind you to complete the sleep diary, answer any questions you may have, and to help you complete your sleep diary, if needed.

11. On the final day we will visit you at home to pick up the Actigraph and your sleep diary. This visit will be arranged at a time that is convenient for you. At this visit we will ask you to fill out a final questionnaire. The questions are aimed at helping us understand your sleep and your feelings during the time you are participating in this study. There are also questions that will ask you what you liked and disliked about being in the study and what things you think helped you to get sleep or stopped you from getting sleep. The final questionnaire will take about 45 minutes to complete.

12. If you agree, we may take a picture of where you and your child sleep so that we can use it to show other doctors and nurses and researchers who want to hear about our study. There is a separate consent for you to complete if you agree to having us take photos.

**Potential Harms:**
If you have an allergy to metal, wearing the actigraph may give you a rash. If this happens and you still want to participate, you can wear the actigraph over your clothing or a sweatband that we will provide. There are no other potential harms associated with participation in this study. There are no consequences if you should choose not to participate in this research study.

**Potential Discomforts or Inconvenience:**
As part of the study a member of the research team will call you at home or email you on each of the seven days that you wear the actigraph. We will also schedule a home visit at the end of the study. Every attempt will be made to ensure minimal disruption to your schedule. It may be inconvenient for you to wear the actigraph, take our phone calls, have us visit you at home and for you to fill out the sleep diary over a seven-day period.

**Potential Benefits:**
You and your child are not expected to benefit directly from participating in this study. If by completing the study questionnaires it becomes known that you may benefit from support, and if you agree, we will refer you to a relevant health care provider.

We hope that the information gained from the study will help us learn more about improving sleep for parents and other family caregivers of children. This information will also help us plan a bigger study in the future.

If you are interested in the outcome of the study, please let the researcher know by filling out the attached sheet and you will be sent a summary of the results after the study is completed.

**Confidentiality:**
We will respect your privacy. No information about who you are will be given to anyone or be published without your permission, unless the law makes us do this. For example, the law could make us give information about you
- If a child has been abused
- If you have an illness you could spread to others
- If you or someone else talks about suicide (killing themselves), or
- If the court orders us to give them the study papers.

When you answer questions about your feelings, your answers might tell us that you are at higher risk for depression. If the study nurse thinks that you are at higher risk for depression, she will recommend to you that you see your family doctor or another health care provider who specializes in mental health.

Records identifying you and your child may be given to and inspected by the SickKids Clinical Research Office Monitor. By signing this consent form you agree to let these people look at you and your child’s records. We will put a copy of this research consent form in your child’s patient health record.

All information collected during the study will be stored in a secure, locked location. Only a copy of this consent will be placed on the health record. You will also be provided with a copy of the consent. The results of this study will be grouped together and your information will
not be individually identified. Only members of the research team (and maybe those individuals described above) will have access to the data. Following completion of the research study the data will be kept as long as required by the SickKids “Records Retention and Destruction” policy. The data will then be destroyed according to this same policy. Published study results will not reveal your identity.

**Reimbursement:**
We will provide you with some compensation, a $25 gift card to Shoppers Drug Mart, in recognition of your time and effort.

**Participation:**
It is your choice to take part in this study. You can stop at any time. The care you get at Sick Kids and/or your community paediatric clinic will not be affected in any way by whether you take part in this study. You may also still participate in the study if you choose not to receive the daily reminder phone calls or emails.

New information that we get while we are doing this study may affect your decision to take part in this study. If this happens, we will tell you about this new information. And we will ask you again if you still want to be in the study.

During this study we may create new tests, new medicines, or other things that may be worth some money. Although we may make money from these findings, we cannot give you [your child] any of this money now or in the future because you [or your child] took part in this study.

For you to participate in this study, and if your child is able, we will also ask your child and document his/her consent or assent if he/she agrees to having you tell us about his/her health condition as it relates to the research study.

If you (your child) become ill or are harmed because of study participation, we will treat you (your child) for free. Your signing this consent form does not interfere with your legal rights in any way. The staff of the study, any people who gave money for the study, or the hospital are still responsible, legally and professionally, for what they do.

**Sponsorship:**
The funder of this research is The Lung Association. The sponsor of the research is the Centre for Nursing, Hospital for Sick Children.

**Conflict of Interest:**
Krista Keilty and the other research team members have no conflict of interest to declare.
The CARE to SLEEP Study

Investigator(s):
Krista Keilty, NP-Paediatrics, PhD candidate, Faculty of Nursing, University of Toronto, Toronto, ON, 416-xxx-xxxx and Centre for Nursing/Research Institute, The Hospital for Sick Children, Toronto, ON, 416-xxx-xxxx, ext. xxxx; pager: 416-xxx-xxxx
Robyn Stremler, RN, PhD, Faculty of Nursing, University of Toronto, Toronto, ON, 416-xxx-xxxx and Centre for Nursing/Research Institute, The Hospital for Sick Children, Toronto, ON
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Karen Spalding, RN, PhD, Faculty of Nursing, Ryerson University, 416-xxx-xxxx, Ext. xxxx
Eleanor Pullenayegum, PhD, Biostatistician, McMaster University, 905-xxx-xxxx, Ext xxxxx

Consent:
“By signing this form, I agree that:
1) You have explained this study to me. You have answered all my questions.
2) You have explained the possible harms and benefits (if any) of this study.
3) I know what I could do instead of taking part in this study. I understand that I have the right not to take part in the study and the right to stop at any time. My decision about taking part in the study will not affect my health care at Sick Kids.
4) I am free now, and in the future, to ask questions about the study.
5) I have been told that my medical records will be kept private except as described to me.
6) I understand that no information about who I am will be given to anyone or be published without first asking my permission.
7) I agree, or consent, to take part in this study.

_________________________ _________________________________
Printed Name of Subject & Age  Subject’s signature & date

_________________________ _________________________________
Printed Name of person who explained consent & date  Signature of Person who explained consent

_________________________ _________________________________
Printed Witness’ name (if the subject/legal guardian does not read English)  Witness’ signature & date

If you have any questions about this study, please call Krista Keilty at 416-xxx-xxxx ext. xxxx.

If you have questions about your rights as a subject in a study or injuries during a study, please call the Research Ethics Manager at 416-xxx-xxx at SickKids.
Assent Form- Children who Depend on Medical Technology

Title of Research Project:

The CARE to SLEEP Study- short title

‘SLEEP disturbance in a prospective cohort of family CAREgivers of children who depend on medical technology and family CAREgivers of healthy children’- long title

Investigator(s):

Krista Keilty, NP-Paediatrics, PhD-Candidate, Faculty of Nursing, University of Toronto, Toronto, ON, 416-xxx-xxxx and Centre for Nursing/Research Institute, The Hospital for Sick Children, Toronto, ON, 416-xxx-xxxx, ext. xxxx; pager: 416-xxx-xxxx

Robyn Stremler, RN, PhD, Faculty of Nursing, University of Toronto, Toronto, ON, 416-xxx-xxxx and Centre for Nursing/Research Institute, The Hospital for Sick Children, Toronto, ON

Eyal Cohen, MD, FRCPC, Paediatric Medicine, The Hospital for Sick Children, Toronto, ON, 416-xxx-xxxx

Karen Spalding, RN, PhD, Faculty of Nursing, Ryerson University, 416-xxxx-xxxx, Ext. xxxx

Eleanor Pullenayegum, PhD, Biostatistician, McMaster University, 905-xxx-xxxx, Ext xxxxx

Research Staff:

Michelle Ho, RN, MA, Nursing Research Intern, The Hospital for Sick Children, Toronto, ON & Clinical Research Project Assistant, Faculty of Nursing, University of Toronto, Toronto, ON 416-xxx-xxxx, Ext xxxx; pager 416-xxx-xxxx

Why are we doing this study?

Sleep is important for overall health and how you feel in the daytime. Parents and other family caregivers of children who use medical equipment at home like oxygen, breathing machines and feeding pumps often describe having difficulty with their sleep. The purpose of this research is to study the sleep in family caregivers of children who use medical devices and compare it to the sleep of parents/family caregivers with children who do not use these medical devices. We will also study how sleep problems in family caregivers are related to their health and how they feel in the daytime. Finally, we are interested in learning about what things may influence sleep problems in family caregivers. This information will help us learn about what
might help family caregivers of children get better sleep and also help us plan future studies where we will test ways to improve sleep in family caregivers.

**What will happen during the study?**

1. If you say ‘yes’ to being in the study, your parent or other family member will fill out a short questionnaire. It will ask some background information, like your age, your health and how well you usually sleep. The questionnaire takes about 15 minutes to complete.

2. Once your parent or other family member has finished the short questionnaire he/she will begin wearing an actigraph. It is the size of a watch and is worn on the wrist. The actigraph measures when someone is sleeping and when someone is awake by measuring how often they move. Your parent or other family member will wear the actigraph for the rest of the day when we give it to him/her and for the next 7 nights and days at home.

3. While they are wearing the actigraph your parent or family member will also fill out a 'Sleep Diary'. This diary asks them to record when they are sleeping, when they are awake and any things that may interrupt their sleep.

4. Over the seven days that your parent or family member is wearing the actigraph a researcher will call them at home to help them fill in the sleep diary and answer any questions.

5. The researcher will ask your parents or family member some questions about how well you slept and what, if any, care they may have provided to you through the night.

6. When we visit on the final day of the study we will pick up the actigraph and the Sleep Diary and ask your parent or family member to fill in some forms. This visit will be arranged at a time that is good for your parents or family member. You do not need to be home for this.

7. If you agree, we may take a picture of where you and your parent or family member sleep so that we can use it to show other doctors and nurses and researchers who want to hear about our study.

**Are there good things and bad things about the study?**

If your parent or family member is allergic to metal he/she might get a rash from wearing the actigraph. If this happens he/she can wear it over clothes or over an armband. Having to wear the actigraph and fill out the Sleep Diary might bother them because it is something extra that they need to do for the day. This is not expected to bother you in any way.

If in this study we learn anything about you or your parent that tells us you need support we may, if you agree, refer you to another health care provider for help or advice.

The good thing about this study is that the information we get from it might help other parents and family members with children sleep better.

**Who will know about what I did in the study?**

No one will know about your information unless you say it is okay. If we feel your health may be in danger, we may have to report your results to your doctor. Your information will be locked away safely and only the people on the research team can see your information.
**Can I decide if I want to be in the study?**

It is your decision to decide if you want to be in the study. Nobody will be angry or upset if you do not want to be in the study. We are talking to your parent/legal guardians about the study and you should talk to them about it too.

I was present when _______________________________ read this form and said that he or she agreed, or assented, to take part in this study.

_________________________________________ _________________
Printed Name of person who obtained assent  Signature & Date

If you have any questions about this study, please call **Krista Keilty at 416-xxx-xxxx, ext. xxxx.**
If you have questions about your rights as a subject in a study or injuries during a study, please call the Research Ethics Manager at 416-xxx-xxxx at SickKids.
Assent Form- Children- Control Group

Title of Research Project:

The CARE to SLEEP Study - short title

‘SLEEP disturbance in a prospective cohort of family CAREgivers of children who depend on medical technology and family CAREgivers of healthy children’ - long title

Investigators:

Krista Keilty, NP-Paediatrics, PhD-Candidate, Faculty of Nursing, University of Toronto, Toronto, ON, 416-xxx-xxxx and Centre for Nursing/Research Institute, The Hospital for Sick Children, Toronto, ON, 416-xxx-xxxx, ext. xxxx; pager: 416-xxx-xxxx

Robyn Stremler, RN, PhD, Faculty of Nursing, University of Toronto, Toronto, ON, 416-xxx-xxxx and Centre for Nursing/Research Institute, The Hospital for Sick Children, Toronto, ON

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Karen Spalding, RN, PhD, Faculty of Nursing, Ryerson University, 416-xxxx-xxxx, Ext. xxxx

Eleanor Pullenayegum, PhD, Biostatistician, McMaster University, 905-xxx-xxxx, Ext xxxxx

Research Staff:

Michelle Ho, RN, MA, Nursing Research Intern, The Hospital for Sick Children, Toronto, ON & Clinical Research Project Assistant, Faculty of Nursing, University of Toronto, Toronto, ON 416-xxx-xxxx, Ext xxxx; pager 416-xxx-xxxx

Why are we doing this study?

Sleep is important for overall health and how you feel in the daytime. Parents and other family caregivers of children who use medical equipment at home like oxygen, breathing machines and feeding pumps often describe having difficulty with their sleep. The purpose of this research is to study the sleep in family caregivers of children who use medical devices and compare it to the sleep of parents/family caregivers with children who do not use these medical devices. We will also study how sleep problems in family caregivers are related to their health and how they feel in the daytime. Finally, we are interested in learning about what things may
influence sleep problems in family caregivers. This information will help us learn about what might help family caregivers of children get better sleep and also help us plan future studies where we will test ways to improve sleep in family caregivers.

What will happen during the study?
1. If you say ‘yes’ to being in the study, your parent or other family member will fill out a short questionnaire. It will ask some background information, like your age, your health and how well you usually sleep. The questionnaire takes about 15 minutes to complete.
2. Once your parent or other family member has finished the short questionnaire he/she will begin wearing an actigraph. It is the size of a watch and is worn on the wrist. The actigraph measures when someone is sleeping and when someone is awake by measuring how often they move. Your parent or other family member will wear the actigraph for the rest of the day when we give it to him/her and for the next 7 nights and days at home.
3. While they are wearing the actigraph your parent or family member will also fill out a 'Sleep Diary’. This diary asks them to record when they are sleeping, when they are awake and any things that may interrupt their sleep.
4. Over the seven days that your parent or family member is wearing the actigraph a researcher will call them at home to help them fill in the sleep diary and answer any questions.
5. The researcher will ask your parents or family member some questions about how well you slept and what, if any, care they may have provided to you through the night.
6. When we visit on the final day of the study we will pick up the actigraph and the Sleep Diary and ask your parent or family member to fill in some forms. This visit will be arranged at a time that is good for your parents or family member. You do not need to be home for this.
7. If you agree, we may take a picture of where you and your parent or family member sleep so that we can use it to show other doctors and nurses and researchers who want to hear about our study.

Are there good things and bad things about the study?
If your parent or family member is allergic to metal he/she might get a rash from wearing the actigraph. If this happens he/she can wear it over clothes or over an armband. Having to the wear the actigraph and fill out the Sleep Diary might bother them because it is something extra that they need to do for the day. This is not expected to bother you in any way.
If in this study we learn anything about you or your parent that tells us you need support we may, if you agree, refer you to another health care provider for help or advice. The good thing about this study is that the information we get from it might help other parents and family members with children sleep better.

Who will know about what I did in the study?
No one will know about your information unless you say it is okay. If we feel your health may be in danger, we may have to report your results to your doctor. Your information will be locked away safely and only the people on the research team can see your information.

Can I decide if I want to be in the study?
It is your decision to decide if you want to be in the study. Nobody will be angry or upset if you do not want to be in the study. We are talking to your parent/legal guardians about the study and you should talk to them about it too.

I was present when ___________________________ read this form and said that he or she agreed, or assented, to take part in this study.

Printed Name of person who obtained assent  Signature & Date

If you have any questions about this study, please call Krista Keilty at 416-xxx-xxxx, ext. xxxx. If you have questions about your rights as a subject in a study or injuries during a study, please call the Research Ethics Manager at 416-xxx-xxxx at SickKids.
Title of Research Project:

‘SLEEP disturbance in a prospective cohort of family CAREgivers of children who depend on medical technology and family CAREgivers of healthy children’ (long title), the ‘CARE to SLEEP Study’ (short title).

Investigator(s):

Krista Keilty, MN, NP-Paeds, PhD Candidate, Faculty of Nursing, University of Toronto, Toronto, ON, and Centre for Nursing/Research Institute, The Hospital for Sick Children, Toronto, ON, 416-813-xxxx, ext. xxxx, pager: 416-xxxx-xxxx

Robyn Stremler, RN, PhD, Faculty of Nursing, University of Toronto, Toronto, ON, 416-xxxx-xxxx and Centre for Nursing/Research Institute, The Hospital for Sick Children, Toronto, ON

Eyal Cohen, MD, FRCPC, Paediatric Medicine, The Hospital for Sick Children, Toronto, ON, 416-813-xxxx

Karen Spalding, RN, PhD, Faculty of Nursing, Ryerson University, 416-xxx-xxxx

Eleanor Pullenayegum, PhD, Biostatistician, McMaster University, 416-xxx-xxxx

Research Staff:

Michelle Ho, RN, MA, Nursing Research Intern, The Hospital for Sick Children, Toronto, ON & Clinical Research Project Assistant, Faculty of Nursing, University of Toronto, Toronto, ON 416-813-xxxx, ext 1697; pager: 416-xxxx-xxxx

Confidentiality:

“The pictures or tapes produced from this study will be stored in a secure, locked location. Only members of the research team (and maybe the SickKids monitor, or employees of the company sponsoring the study or the regulator eg., Health Canada) will have access to them. Following completion of the study the tapes/pictures will be kept as long as required in the SickKids
“Records Retention and Destruction” policy. They will then be destroyed according to this same policy.”

Consent:

“By signing this form,
1) I also agree to be photographed during this study. These photographs will be used to help us describe to other researchers and clinicians what your sleep is like at home.
2) I understand that I have the right to refuse to take part in this study. I also have the right to withdraw from this part of the study at any time. e.g., before or even after the photographs are made. My decision will not affect my health care at SickKids.
3) I am free now, and in the future, to ask questions about the picture taking.
4) I have been told that my medical records will be kept private. You will give no one information about me, unless the law requires you to.
5) I understand that no information about me (including these tapes/pictures) will be given to anyone or be published without first asking my permission.”
6) I have read and understood pages 1 to _____ of this consent form. I agree, or consent, to having my picture taken/being taped as part of the study.

__________________________________________  ______________________________________
Printed Name of Subject     Subject’s signature & date

__________________________________________  ______________________________________
Printed Name of person who explained consent  Signature & date

__________________________________________  ______________________________________
Printed Witness’ name (subject does not read English)   Witness’ signature & date

In addition, I agree or consent for this photograph(s) to be used for:

1. Other studies on the same topic  o
2. Teaching and demonstration at SickKids.  o
3. Teaching and demonstration at meetings outside SickKids.  o
4. Not to be used for anything else.  o

In agreeing to the use of the tape(s)/photograph(s) for other purposes, I have been offered a chance to view/hear the tape(s)/photograph(s). I also have the right to withdraw my permission for other uses of the tape(s)/photograph(s) at any time.

__________________________________________  ______________________________________
Printed Name of Subject     Subject’s signature & date
If you have any questions about this study, please call Krista Keilty at 416-xxx-xxxx, ext. xxxx. If you have questions about your rights as a subject in a study or injuries during a study, please call the Research Ethics Manager at 416-813-xxxx at SickKids.
Appendix J

Baseline sociodemographic data forms
Baseline Demographics Collection Sheets:
Family Caregivers of Children who depend on Medical Technology

Today's date: ☐☐☐☐-☐☐☐☐

*Please complete the following questions by indicating responses by marking in the square ☑ or by filling in the blanks. Thank you.*

**Section A: Child Info**

*Please tell us about your child:*

1. Child DOB (year/month): ☐☐☐☐-☐☐☐☐
2. Gender: ☐ Female ☐ Male
3. Which of the following types of medical equipment does your child use at home? Please indicate approximately how many hours (daytime and/or night time) the type of equipment is used. Indicate all that may apply.

<table>
<thead>
<tr>
<th>MEDICAL DEVICES</th>
<th>NO</th>
<th>YES</th>
<th>Daytime (9am to 9pm)</th>
<th>Nighttime (9pm to 9am)</th>
<th>Other, please explain</th>
</tr>
</thead>
<tbody>
<tr>
<td>Invasive ventilator (via tracheostomy)</td>
<td>☐</td>
<td>☐</td>
<td>☐☐☐☐ hours</td>
<td>☐☐☐☐ hours</td>
<td></td>
</tr>
<tr>
<td>Non-invasive ventilator (e.g., Bi-PAP)</td>
<td>☐</td>
<td>☐</td>
<td>☐☐☐☐ hours</td>
<td>☐☐☐☐ hours</td>
<td></td>
</tr>
<tr>
<td>Continuous Positive Pressure Ventilation (CPAP)</td>
<td>☐</td>
<td>☐</td>
<td>☐☐☐☐ hours</td>
<td>☐☐☐☐ hours</td>
<td></td>
</tr>
<tr>
<td>Tracheostomy</td>
<td>☐</td>
<td>☐</td>
<td>☐☐☐☐ hours</td>
<td>☐☐☐☐ hours</td>
<td></td>
</tr>
<tr>
<td>Oxygen delivery device</td>
<td>☐</td>
<td>☐</td>
<td>☐☐☐☐ hours</td>
<td>☐☐☐☐ hours</td>
<td></td>
</tr>
<tr>
<td>Enteral feeding tube and electric pump</td>
<td>☐</td>
<td>☐</td>
<td>☐☐☐☐ hours</td>
<td>☐☐☐☐ hours</td>
<td></td>
</tr>
<tr>
<td>Enteral feeding tube and drip set</td>
<td>☐</td>
<td>☐</td>
<td>☐☐☐☐ hours</td>
<td>☐☐☐☐ hours</td>
<td></td>
</tr>
<tr>
<td>Other (please describe):</td>
<td>☐</td>
<td>☐</td>
<td>☐☐☐☐ hours</td>
<td>☐☐☐☐ hours</td>
<td></td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>MONITORING DEVICES</th>
<th>NO</th>
<th>YES</th>
<th>Daytime (9am to 9pm)</th>
<th>Nighttime (9pm to 9am)</th>
<th>Other, please explain</th>
</tr>
</thead>
<tbody>
<tr>
<td>Oxygen saturation monitor</td>
<td>☐</td>
<td>☐</td>
<td>☐☐☐☐ hours</td>
<td>☐☐☐☐ hours</td>
<td></td>
</tr>
<tr>
<td>Cardiorespiratory monitor</td>
<td>☐</td>
<td>☐</td>
<td>☐☐☐☐ hours</td>
<td>☐☐☐☐ hours</td>
<td></td>
</tr>
<tr>
<td>Commercial (e.g. baby) monitor</td>
<td>☐</td>
<td>☐</td>
<td>☐☐☐☐ hours</td>
<td>☐☐☐☐ hours</td>
<td></td>
</tr>
<tr>
<td>Other (please describe):</td>
<td>☐</td>
<td>☐</td>
<td>☐☐☐☐ hours</td>
<td>☐☐☐☐ hours</td>
<td></td>
</tr>
</tbody>
</table>

4. Your child’s main medical diagnosis is:
☐ Cardiac (e.g. Congenital Heart Disease, CHARGE Syndrome):
☐ Craniofacial (e.g. Pierre Robin Syndrome, Apert’s Syndrome):

☐ Gastrointestinal (e.g. Gastroesophageal Reflux Disease), please describe:

☐ Genetic/Metabolic (e.g. Down’s Syndrome), please describe:

☐ Neurological (e.g. Cerebral Palsy, Acquired Brain Injury), please describe:

☐ Neuromuscular (e.g. SMA, Muscular Dystrophy), please describe:

☐ Respiratory (Chronic Lung Disease, Hypoventilation), please describe:

☐ Other, please describe ___________________________________________

Revised Section B: Homecare info

*We are interested in your use of homecare/respite services.\
* may be from combined sources (e.g. CCAC, private insurance)*

5. When did ___________ (child’s name) begin to depend on technology support at home?\
(year/month): ☐☐☐☐

6. How many hours of in-home homecare/respite* are you approved for per week? ☐☐\
Please describe the allocations of these hours:

From CCAC ☐ No ☐ Yes If yes, # hr/wk ☐☐☐
Enhanced respite: ☐ No ☐ Yes If yes, # hr/wk ☐☐☐
SS@Home: ☐ No ☐ Yes If yes, # hr/wk ☐☐☐
Private insurance: ☐ No ☐ Yes If yes, # hr/wk ☐☐☐
Philanthropic sources: ☐ No ☐ Yes If yes, # hr/wk ☐☐☐
Other: ☐ No ☐ Yes If yes, # hr/wk ☐☐☐
Please describe:____________________________________________________

7. How many hours of out-of-home homecare/respite* are you approved for per week (e.g. school, respite facility)? ☐☐☐

Please describe the allocations of these hours:

From CCAC ☐ No ☐ Yes If yes, # hr/wk ☐☐☐
Enhanced respite: ☐ No ☐ Yes If yes, # hr/wk ☐☐☐
SS@Home: ☐ No ☐ Yes If yes, # hr/wk ☐☐☐
Private insurance: ☐ No ☐ Yes If yes, # hr/wk ☐ ☐
Philanthropic sources: ☐ No ☐ Yes If yes, # hr/wk ☐ ☐
Other: ☐ No ☐ Yes If yes, # hr/wk ☐ ☐
Please describe:____________________________________________________

8. How many hours of in-home homecare/respite * do you typically use per week? ☐ ☐ ☐ hours/wk

9. How many hours of out-of-home homecare/respite * do you typically use per week? ☐ ☐ ☐ hours/wk

10. How many of the total in-home homecare hours* per week are typically used at night? ☐ ☐ ☐ hours/wk

11. If applicable, What are the reasons for not using all of your homecare hours/respite in a week?
☐ Non applicable, I typically use all of my homecare/respite hours.
☐ Shifts go unfilled by nurse/agency (e.g. sick calls, staff shortage)
☐ I cancel the shifts, please explain why: ________________
☐ Other, please explain: ________________________________

Section C: Family Caregiver Info

Please tell us about you.

12. What year and month were you born in? ☐ ☐ ☐ ☐ year ☐ ☐ month

13. Gender: ☐ Female ☐ Male

14. Are you your child's? ☐ Mother ☐ Father ☐ Other: ________________

15. Including ____________ (child's name), how many children < 18 years of age live at home with you? ☐ ☐ total

16. Including yourself, how many adults >18 years of age live at home with you? ☐ ☐ total

17. Do you have any diagnosed chronic health conditions? ☐ No ☐ Yes

If yes, check all that apply:
☒ ☐ Neurological, please describe ________________________________
18. How many times did you visit a health care provider for medical needs in the last 12 months (e.g. to see primary care provider, medical specialists, visit urgent care or emergency department)? ☐☐ times/yr

What is your current (may weigh self in clinic or recall, if known):

19. Height? ☐ ft, ☐ inches or ☐ cm

20. Weight? ☐ pounds or ☐ kg

21. What is your current marital status?
☐ Married or living with partner
☐ Single
☐ Other (please specify): ________________________________

22. What is your highest level of schooling completed?
☐ Elementary/grade school
☐ High School
☐ College or University Diploma
☐ Graduate Degree

23. What is your current paid employment status?
☐ Full time
☐ Part time
☐ Unemployed
☐ Other (please explain): ________________________________

24. What is your total family income?
☐ under $25,000
☐ $25,000-$39,000
☐ $40,000-$59,000
☐ $60,000-$99,000
☐ $100,000 or more

25. Were you born in Canada?
☐ Yes  ☐ No ➔ If no, length of time living in Canada?
  ☐ < 5 years
  ☐ 5 or more years

26. What is your race?
☐ African American/Canadian, Caribbean, Black
☐ Arab (e.g. Afgani, Iranian)
☐ East Asian (e.g. Chinese, Vietnamese, Fillipino, Japanese, etc.)
☐ First Nations/Aboriginal
☐ South Asian (e.g. East Indian, Pakistani, Sri Lankan, etc)
☐ Latin, Central or South American
☐ Caucasian, White
☐ Mixed race: please describe: ____________________________
☐ Other, please describe: ____________________________
Baseline Demographics Collection Sheets:
Family Caregivers of Children who do not use Medical Technology

We are interested in knowing more about you and your child. Please complete the following by either filling in the blanks or indicating your response by marking in the square ☑

**Please tell us about your child:**
1. Child DOB (year/month): ☐☐☐☐-☐☐
2. Gender: ☐ Female ☐ Male

**Please tell us about you.**
3. What year and month were you born in? ☐☐☐☐ year ☐☐ month
4. Gender: ☐ Female ☐ Male
5. Are you your child’s?
☐ Mother  
☐ Father  
☐ Other: ______________________
6. Including ____________ (child’s name), how many children < 18 years of age live at home with you? ☐☐ total
7. Including yourself, how many adults > 18 years of age live at home with you? ☐☐ total
8. Do you have any diagnosed chronic health conditions? ☐ No ☐ Yes
   If yes, check all that apply
   ➔ ☐ Neurological, please describe ________________________________
   ➔ ☐ Cardiovascular, please describe ________________________________
   ➔ ☐ Respiratory, please describe ________________________________
   ➔ ☐ Psychiatric, please describe ________________________________
   ➔ ☐ Endocrine, please describe ________________________________
   ➔ ☐ Autoimmune, please describe ________________________________
   ➔ ☐ Chronic pain, please describe ________________________________
   ➔ ☐ Other, please describe ________________________________
9. How many times did you visit a health care provider for medical needs in the last 12 months (e.g. to see primary care provider, medical specialists, visit urgent care or emergency department)? ☐☐ times
What is your current (may weigh self in clinic or recall, if known):

10. Height? □ ft, □ inches or □ □ □ cm

11. Weight? □ □ □ pounds or □ □ □ kg

12. What is your current marital status?
   □ Married or living with partner
   □ Single
   □ Other (please specify): ___________________________________

13. What is your highest level of schooling completed?
   □ Elementary/grade school
   □ High School
   □ College or University Diploma
   □ Graduate Degree

14. What is your current paid employment status?
   □ Full time
   □ Part time
   □ Unemployed
   □ Other (please explain): ___________________________________

15. What is your total family income?
   □ under $25,000
   □ $25,000-$39,000
   □ $40,000-$59,000
   □ $60,000-$99,000
   □ $100,000 or more

16. Were you born in Canada?
   □ Yes □ No ➔ If no, length of time living in Canada?
     □ < 5 years
     □ 5 or more years

17. What is your race?
   □ African American/Canadian, Caribbean, Black
   □ Arab (e.g. Afgani, Iranian)
☐ East Asian (e.g. Chinese, Vietnamese, Fillipino, Japanese, etc.)
☐ First Nations/Aboriginal
☐ South Asian (e.g. East Indian, Pakistani, Sri Lankan, etc)
☐ Latin, Central or South American
☐ Caucasian, White
☐ Mixed race: please describe: __________________________
☐ Other, please describe: __________________________
Appendix K

Daily sleep diaries
DAILY SLEEP DIARY

Family Caregivers of Children who depend on Medical Technology

The CARE to SLEEP Study

Principle Investigator: Krista Keilty, NP-Paediatrics, PhD-Candidate
Faculty of Nursing
University of Toronto
155 College Street, Room 290
Toronto, ON M2T 1P8

416-813-xxxx, Ext xxxx
pager 416-xxx xxxx
**EXAMPLE: Family Caregiver of Children**

**INSTRUCTIONS:** Fill in the times you are asleep with shaded boxes. Leave blank the times you are awake. Below the last line, please mark: X = awake, stayed in bed O = awake, got up out of bed

**For example:** You went to sleep at 10 pm in your child’s room but he/she was restless and you were both up for an hour, then you fell asleep, and woke at 12 am to let the dog out. By 12:15 am you went back to sleep in your own room and woke again at 2 am when you heard a noise from your child’s room and listened to see if he/she would settle again. You stayed in bed and fell back asleep, and woke again at 6 am when you got up to use the washroom. After this you laid in bed awake until 7 am when your alarm went off and you got up.

| Night-time: | 9P | 9:30 | 10P | 10:30 | 11P | 11:30 | 12A | 12:30 | 1A | 1:30 | 2A | 2:30 | 3A | 3:30 | 4A | 4:30 | 5A | 5:30 | 6A | 6:30 | 7A | 7:30 | 8A | 8:30 | 9A |
|-------------|----|------|-----|-------|-----|-------|-----|-------|---|------|---|------|---|------|---|------|---|------|---|------|---|------|---|
| My sleep:   | X  | X    | O   |     X  | O    | O     |   X | O     |   | X    |   | X    |   | X    |   | X    |   | X    |   | X    |   | X    |   |

| Night-time: | 9P | 9:30 | 10P | 10:30 | 11P | 11:30 | 12A | 12:30 | 1A | 1:30 | 2A | 2:30 | 3A | 3:30 | 4A | 4:30 | 5A | 5:30 | 6A | 6:30 | 7A | 7:30 | 8A | 8:30 | 9A |
|-------------|----|------|-----|-------|-----|-------|-----|-------|---|------|---|------|---|------|---|------|---|------|---|------|---|------|---|
| My sleep:   | X  | X    | O   |     X  | O    | O     |   X | O     |   | X    |   | X    |   | X    |   | X    |   | X    |   | X    |   | X    |   |
DAY 1 – DAYTIME

INSTRUCTIONS: Fill in the times you are asleep with shaded boxes. Leave blank the times you are awake.

Below the last line, please mark:  X= awake, stayed in bed  O= awake, got up out of bed

Use from 9:00 am to 9:00 pm

| 9A | 9:30 | 10A | 10:30 | 11A | 11:30 | 12P | 12:30 | 1P | 1:30 | 2P | 2:30 | 3P | 3:30 | 4P | 4:30 | 5P | 5:30 | 6P | 6:30 | 7P | 7:30 | 8P | 8:30 | 9P |
|----|------|-----|-------|-----|-------|-----|-------|---|------|---|------|---|------|---|------|---|------|---|------|---|------|---|------|---|------|
| My sleep: |     |     |       |     |       |     |       |   |      |   |      |   |      |   |      |   |      |   |      |   |      |   |      |   |      |
DAY 1 - EVENING

COMPLETE THIS PAGE BEFORE YOU GO TO SLEEP

1. Time you are filling this out: AM / PM (circle one)

2. Did anything happen today that might affect your sleep tonight?

   No  Yes ➔ If yes, what happened? ______________________________
INSTRUCTIONS: Fill in the times you are asleep with shaded boxes. Leave blank the times you are awake.

Below the last line, please mark: X= awake, stayed in bed

Below the last line, please mark: O= awake, got up out of bed

**Night-time:**
Use from 9:00 pm to 9:00 am

<table>
<thead>
<tr>
<th>9P</th>
<th>9:30</th>
<th>10P</th>
<th>10:30</th>
<th>11P</th>
<th>11:30</th>
<th>12A</th>
<th>12:30</th>
<th>1A</th>
<th>1:30</th>
<th>2A</th>
<th>2:30</th>
<th>3A</th>
<th>3:30</th>
<th>4A</th>
<th>4:30</th>
<th>5A</th>
<th>5:30</th>
<th>6A</th>
<th>6:30</th>
<th>7A</th>
<th>7:30</th>
<th>8A</th>
<th>8:30</th>
<th>9A</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
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<td></td>
</tr>
</tbody>
</table>

My sleep:
1. Time you are filling this out: □□ : □□ AM / PM (circle one)

2. What time did you turn out the lights to go to sleep last night? □□ : □□ AM / PM

3. How long did it take you to fall asleep last night?
   □ less than 10 minutes
   □ between 10 and 30 minutes
   □ more than 30 minutes

4. What time did you get up out of bed for the day this morning?
   □□ : □□ AM / PM

5. How long were you awake before you got out of bed this morning?
   □ less than 10 minutes
   □ between 10 and 30 minutes
   □ more than 30 minutes

NIGHTTIME HELP

6. Did anyone help you to care for ________________(child’s name) last night?
   □ No, no one helped me
   □ Yes, I got help from
      If yes, who helped you last night?
      ➤□ a Registered Nurse (RN)?
      ➤□ a Registered Practical Nurse (RPN)?
      ➤□ a Personal Support Worker (PSW)?
      ➤□ paid help (e.g. nanny)
      ➤□ unpaid help (e.g. family, spouse, grandparent)
      ➤□ other , please explain ________________________________

TYPICAL NIGHT

7. Compared to most nights how was your overall sleep last night?
   □ Much worse than usual
   □ Somewhat worse than usual
   □ About the same as usual
☐ A bit better than usual
☐ A lot better than usual

8. Compared to most nights how demanding were your child’s ______________(child’s name) care needs last night?
☐ Much more demanding than usual
☐ Somewhat more demanding than usual
☐ About the same as usual
☐ A bit better than usual
☐ A lot better than usual

SLEEPING ARRANGEMENTS
9. Please describe what surface(s) you slept on last night (check all that apply)
☐ Own bed
☐ My child’s bed: ______________ (child’s name)
☐ A bed separate from my child but in his/her room
☐ My other children’s bed: ______________
☐ A chair
☐ A sofa/couch
☐ Other: Please explain: ______________

10. Please describe what room(s) you slept in last night (check all that apply)
☐ My own room
☐ My child’s room: ______________ (child’s name)
☐ My other children’s room: ______________
☐ Family/living room
☐ Other: Please explain: ______________

11. Please describe if you shared bed(s) or sleep surface(s) last night (check all that apply)
☐ No, I slept alone
☐ Yes, with my spouse/partner
☐ Yes, with my child: ______________ (child’s name)
☐ Yes, with my other child(ren)
☐ Other: please explain____________________

NIGHT AWAKENINGS
12. Did you wake up at any point throughout the night before you got up for the day?
☐ No ☐ Yes ➔ If yes, how many times? ☐ ☐
13. Were you kept awake at any point last night by negative thoughts & feelings, when you wanted to be asleep?

☐ No  ☐ Yes ➔ If yes, please select from the following reasons all that apply.
☐ felt ‘too mentally alert or active’
☐ had repetitive negative thoughts
☐ had thoughts keep running through my mind
☐ had a jittery, nervous feeling in your body
☐ worried about my child’s overall health or health care
☐ worried that something will go wrong with my child at night
☐ worried about something else: please explain_______________________

14. Were you kept awake at any point last night by noises or other things in the home or your environment when you wanted to be asleep?

☐ No  ☐ Yes ➔ If yes, please select from the following reasons all that apply.
☐ by your child: ____________ (child’s name)?
☐ by other child(ren)
☐ by family members for reasons not related to my child’s care
☐ by homecare personnel (e.g. RN, RPN, PSW) for reasons not related to my child’s care
☐ by family members for reasons that were related to my child’s care
☐ by homecare personnel (e.g. RN, RPN, PSW) for reasons that were related to my child’s care
☐ by your paid help for reasons not related to your child’s care (e.g., nanny)
☐ by your paid (e.g., nanny) help for reasons that were related to your child’s care
☐ noises made by technology/monitor (alarms)
☐ lights from homecare technology/monitors
☐ Other, please explain:__________________________________

15. After falling asleep last night, did you get out of bed before your planned rise time?

☐ No  ☐ Yes ➔ If yes, was it to:
➔take care of ______________(child’s name) by yourself?
☐ No  ☐ Yes
➔help someone else take care of ______________(child’s name)?
☐ No  ☐ Yes
If yes, what care needed to be provided to ________________(child’s name)? Check all that apply

☐ comfort and reassurance  
☐ routine activities of living (e.g., toileting, turning)  
☐ scheduled health care needs (e.g. start feeding pump, give medication)  
☐ unscheduled health care needs (e.g. suction, turn up oxygen, give fever medicine)  
☐ to cancel or respond to equipment alarms  
☐ To take care of my other child(ren)  
☐ To take care of others who live with me  
☐ To use the washroom or other personal needs  
☐ Other: please explain: _____________________________________________________________________  

Please comment on anything not listed prior that you think may have disturbed your sleep last night: ____________________________________________________________________________________

TECHNOLOGY USE

16. Which of the following did ______________(child’s name) use last night between 9pm-9am? Please select all that apply.

<table>
<thead>
<tr>
<th>MEDICAL DEVICES</th>
<th>NO</th>
<th>YES</th>
<th># of hours of use, please describe schedule</th>
</tr>
</thead>
<tbody>
<tr>
<td>Invasive ventilator (via tracheostomy)</td>
<td>☐</td>
<td>☐</td>
<td>☐ ☐ hours</td>
</tr>
<tr>
<td>Non-invasive ventilator (e.g., Bi-PAP)</td>
<td>☐</td>
<td>☐</td>
<td>☐ ☐ hours</td>
</tr>
<tr>
<td>Continuous Positive Pressure Ventilation (CPAP)</td>
<td>☐</td>
<td>☐</td>
<td>☐ ☐ hours</td>
</tr>
<tr>
<td>Tracheostomy</td>
<td>☐</td>
<td>☐</td>
<td>☐ ☐ hours</td>
</tr>
<tr>
<td>Oxygen delivery device</td>
<td>☐</td>
<td>☐</td>
<td>☐ ☐ hours</td>
</tr>
<tr>
<td>Enteral feeding tube and electric pump</td>
<td>☐</td>
<td>☐</td>
<td>☐ ☐ hours</td>
</tr>
<tr>
<td>Enteral feeding tube and drip set</td>
<td>☐</td>
<td>☐</td>
<td>☐ ☐ hours</td>
</tr>
<tr>
<td>Other (please describe):</td>
<td>☐</td>
<td>☐</td>
<td>☐ ☐ hours</td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>MONITORING DEVICES</th>
<th>NO</th>
<th>YES</th>
<th># of hours of use, please describe schedule</th>
</tr>
</thead>
<tbody>
<tr>
<td>Oxygen saturation monitor</td>
<td>☐</td>
<td>☐</td>
<td>☐ ☐ hours</td>
</tr>
<tr>
<td>Cardiorespiratory monitor</td>
<td>☐</td>
<td>☐</td>
<td>☐ ☐ hours</td>
</tr>
<tr>
<td>Commercial (e.g. baby) monitor</td>
<td>☐</td>
<td>☐</td>
<td>☐ ☐ hours</td>
</tr>
<tr>
<td>Other (please describe):</td>
<td>☐</td>
<td>☐</td>
<td>☐ ☐ hours</td>
</tr>
</tbody>
</table>
Thank you for completing today’s sleep diary.

The information that you have provided will help us in our research to better understand what your sleep is like and what things may affect it.
DAILY SLEEP DIARY

Family Caregivers of Healthy Children

The CARE to SLEEP Study

Principle Investigator: Krista Keilty, NP-Paediatrics, PhD-Candidate
Faculty of Nursing
University of Toronto
155 College Street, Room 290
Toronto, ON M2T 1P8

416-xxx-xxxx, Ext xxxx
krista.keilty@sickkids.ca
**EXAMPLE: Family Caregiver of Children**

**INSTRUCTIONS:** Fill in the times you are asleep with shaded boxes. Leave blank the times you are awake. Below the last line, please mark:  
- **X** = awake, stayed in bed
- **O** = awake, got up out of bed

**For example:** You went to sleep at 10 pm in your child’s room but he/she was restless and you were both up for an hour, then you fell asleep, and woke at 12 am to let the dog out. By 12:15 am you went back to sleep in your own room and woke again at 2 am when you heard a noise from your child’s room and listened to see if he/she would settle again. You stayed in bed and fell back asleep, and woke again at 6 am when you got up to use the washroom. After this you laid in bed awake until 7 am when your alarm went off and you got up.

**Night-time:**

| 9P | 9:30 | 10P | 10:30 | 11P | 11:30 | 12A | 12:30 | 1A | 1:30 | 2A | 2:30 | 3A | 3:30 | 4A | 4:30 | 5A | 5:30 | 6A | 6:30 | 7A | 7:30 | 8A | 8:30 | 9A |
|----|------|-----|-------|-----|-------|-----|-------|---|-----|---|-----|---|-----|---|-----|---|-----|---|-----|---|-----|---|
|    |      |     |       |     |       |     |       |  X |     |   |     |   |     |   |     |   |     |   |     |   |     |   |     |   |
|    |      |     |       |     |       |     |       |   |     |   |     |   |     |   |     |   |     |   |     |   |     |   |     |   |

**Practice example:** You went to sleep at 1130 pm in your own room and slept until 4 am when your child woke crying for you. You woke up and went to his/her room and tried to settle him/her but after 30 minutes decided to sleep in your child’s room. You slept through till 7am in the morning when you had to get up to get the children ready for school.

**Night-time:**

<table>
<thead>
<tr>
<th>9P</th>
<th>9:30</th>
<th>10P</th>
<th>10:30</th>
<th>11P</th>
<th>11:30</th>
<th>12A</th>
<th>12:30</th>
<th>1A</th>
<th>1:30</th>
<th>2A</th>
<th>2:30</th>
<th>3A</th>
<th>3:30</th>
<th>4A</th>
<th>4:30</th>
<th>5A</th>
<th>5:30</th>
<th>6A</th>
<th>6:30</th>
<th>7A</th>
<th>7:30</th>
<th>8A</th>
<th>8:30</th>
<th>9A</th>
</tr>
</thead>
<tbody>
<tr>
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</tr>
</tbody>
</table>

My sleep:  

| X | O | X | OX | O |
**INSTRUCTIONS:** Fill in the times you are asleep with shaded boxes. Leave blank the times you are awake.

Below the last line, please mark:  

- **X=** awake, stayed in bed  
- **O=** awake, got up out of bed

Use from 9:00 am to 9:00 pm

<table>
<thead>
<tr>
<th>9A</th>
<th>9:30</th>
<th>10A</th>
<th>10:30</th>
<th>11A</th>
<th>11:30</th>
<th>12P</th>
<th>12:30</th>
<th>1P</th>
<th>1:30</th>
<th>2P</th>
<th>2:30</th>
<th>3P</th>
<th>3:30</th>
<th>4P</th>
<th>4:30</th>
<th>5P</th>
<th>5:30</th>
<th>6P</th>
<th>6:30</th>
<th>7P</th>
<th>7:30</th>
<th>8P</th>
<th>8:30</th>
<th>9P</th>
</tr>
</thead>
<tbody>
<tr>
<td>My sleep:</td>
<td></td>
<td></td>
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</tbody>
</table>

**DAY 1 – DAYTIME**

**DAY OF THE WEEK______________**
1. Time you are filling this out: __ : __ AM / PM (circle one)

2. Did anything happen today that might affect your sleep tonight?
   □ No  □ Yes ➔ If yes, what happened? ________________________________
INSTRUCTIONS: Fill in the times you are asleep with shaded boxes. Leave blank the times you are awake.

Below the last line, please mark:  

- X= awake, stayed in bed  
- O= awake, got up out of bed

Night-time:  
Use from 9:00 pm to 9:00 am

| 9P | 9:30 | 10P | 10:30 | 11P | 11:30 | 12A | 12:30 | 1A | 1:30 | 2A | 2:30 | 3A | 3:30 | 4A | 4:30 | 5A | 5:30 | 6A | 6:30 | 7A | 7:30 | 8A | 8:30 | 9A |
|-----|------|-----|-------|-----|-------|-----|-------|---|------|---|------|---|------|---|------|---|------|---|------|---|------|---|

My sleep: | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | | |
DAY 2 – MORNING

DAY OF THE WEEK __________

COMPLETE THESE PAGES WHEN YOU WAKE UP

1. Time you are filling this out: □□ : □□ AM / PM (circle one)

2. What time did you turn out the lights to go to sleep last night? □□ : □□ AM / PM

3. How long did it take you to fall asleep last night?
   ☐☐ ☐☐ less than 10 minutes
   ☐☐ ☐☐ between 10 and 30 minutes
   ☐☐ ☐☐ more than 30 minutes

4. What time did you get up out of bed for the day this morning? □□ : □□ AM / PM

5. How long were you awake before you got out of bed this morning?
   ☐☐ ☐☐ less than 10 minutes
   ☐☐ ☐☐ between 10 and 30 minutes
   ☐☐ ☐☐ more than 30 minutes

NIGHTTIME HELP

6. Did anyone help you to care for _______________ (child’s name) last night?
   ☐☐ No, no one helped me
   ☐☐ Yes, I got help from
If yes, who helped you last night?

☐☐ ☐☐ paid help (e.g. nanny), please explain ___________________________

☐☐ ☐☐ unpaid help (e.g. family, spouse, grandparent)

☐☐ ☐☐ other, please explain _________________________________________

TYPICAL NIGHT

7. Compared to most nights how was your overall sleep last night?

☐ ☐ Much worse than usual
☐ ☐ Somewhat worse than usual
☐ ☐ About the same as usual
☐ ☐ A bit better than usual
☐ ☐ A lot better than usual

8. Compared to most nights how demanding were your child’s ____________(child’s name) care needs last night?

☐ ☐ Much more demanding than usual
☐ ☐ Somewhat more demanding than usual
☐ ☐ About the same as usual
☐ ☐ A bit better than usual
☐ ☐ A lot better than usual

SLEEPING ARRANGEMENTS

9. Please describe what surface(s) you slept on last night (check all that apply)

☐ ☐ Own bed
☐ ☐ My child’s bed: ____________ (child’s name)
☐ ☐ A bed separate from my child but in his/her room
☐ ☐ My other children’s bed: __________________
☐ A chair
☐ A sofa/couch
☐ Other: Please explain: _______________________

10. **Please describe what room(s) you slept in last night (check all that apply)**
    ☐ My own room
    ☐ My child’s room: ___________ (child’s name)
    ☐ My other children’s room: _____________________
    ☐ Family/living room
    ☐ Other: Please explain: _________________________

11. **Please describe if you shared bed(s) or sleep surface(s) last night (check all that apply)**
    ☐ No, I slept alone
    ☐ Yes, with my spouse/partner
    ☐ Yes, with my child: ________________ (child’s name)
    ☐ Yes, with my other child(ren)
    ☐ Other: please explain_____________________

**NIGHT AWAKENINGS**

12. **Did you wake up at any point throughout the night before you got up for the day?**
    ☐ No ☐ Yes ➔ If yes, how many times? ☐ ☐

13. **Were you kept awake at any point last night by negative thoughts & feelings, when you wanted to be asleep?**
    ☐ No ☐ Yes ➔ If yes, please select from the following reasons all that apply.
☐ felt ‘too mentally alert or active’
☐ had repetitive negative thoughts
☐ had thoughts keep running through my mind
☐ had a jittery, nervous feeling in your body
☐ worried about my child’s overall health or health care
☐ worried that something will go wrong with my child at night
☐ worried about something else: please explain_______________________

14. Were you kept awake at any point last night by noises or other things in the home or your environment when you wanted to be asleep?

☐ No  ☐ Yes ➔ If yes, please select from the following reasons all that apply.

☐ by your child: _______________ (child’s name)?
☐ by other child(ren)
☐ by family members for reasons not related to my child’s care
☐ by family members for reasons that were related to my child’s care
☐ by your paid help for reasons not related to your child’s care (e.g., nanny)
☐ by your paid (e.g., nanny) help for reasons that were related to your child’s care
☐ noises made by technology/monitor (alarms)
☐ lights from homecare technology/baby monitors
☐ Other, please explain:______________________________

15. After falling asleep last night, did you get out of bed before your planned rise time?

☐ No  ☐ Yes

➔ If yes, was it to:


到底是否要親自照顧______________（孩子的名字）？

☑ No ☐ Yes

是否要幫其他人照顧______________（孩子的名字）？

☑ No ☐ Yes

如果要，照顧______________（孩子的名字）所需的護理是哪些？（請選中適用的）

☐ ☐ comfort and reassurance
☐ ☐ routine activities of living (e.g., toileting, turning)
☐ ☐ unscheduled health care needs (e.g. give fever medicine)
☐ ☐ to cancel or respond to monitor alarms
☐ ☐ To take care of my other child(ren)
☐ ☐ To take care of others who live with me
☐ ☐ To use the washroom or other personal needs
☐ ☐ Other: please explain: ____________________________

請評論任何未列項但你認為可能打擾到你昨晚睡眠的事情：

________________________________________________________________________________

TECHNOLOGY USE
16. Which of the following did ________________ (child’s name) use last night between 9pm-9am? Please select all that apply.

<table>
<thead>
<tr>
<th>MONITORING DEVICES</th>
<th>NO</th>
<th>YES</th>
<th># of hours of use, please describe schedule</th>
</tr>
</thead>
<tbody>
<tr>
<td>Oxygen saturation monitor</td>
<td>☐</td>
<td>☐</td>
<td>☐☐☐ hours</td>
</tr>
<tr>
<td>Cardiorespiratory monitor</td>
<td>☐</td>
<td>☐</td>
<td>☐☐☐ hours</td>
</tr>
<tr>
<td>Commercial (e.g. baby) monitor</td>
<td>☐</td>
<td>☐</td>
<td>☐☐☐ hours</td>
</tr>
<tr>
<td>Other (please describe):</td>
<td>☐</td>
<td>☐</td>
<td>☐☐☐ hours</td>
</tr>
</tbody>
</table>
Appendix L

Home visit questionnaires
In this package we have selected questionnaires that will help us learn more about your sleep, your child’s sleep and things related to sleep like how you feel and function in the daytime.

All together the questionnaires are expected to take 30-45 minutes to complete.

We will help you to complete these forms and will be happy to answer any questions you may have about them and our research study.

Many thanks for your participation in our research study.
Questionnaire A: CAREGIVER SLEEP QUALITY

These questions will help us understand more about your sleep quality over this past week. Thinking about this week:

1. When have you usually gone to bed?  
   TIME: \[\square:\square:\square\]

2. How long (in minutes) has it taken you to fall asleep each night?  
   TIME: \[\square:\square:\square\]

3. When have you usually gotten up in the morning?  
   TIME: \[\square:\square:\square\]

4. How many hours of actual sleep did you get at night?  
   TIME: \[\square:\square:\square\]
   (This may be different than the number of hours you spent in bed)

5. During the past week, how would you rate your sleep quality overall?  
   - \[\square\] very good  
   - \[\square\] fairly good  
   - \[\square\] fairly bad  
   - \[\square\] very bad

Over the past week:

<table>
<thead>
<tr>
<th></th>
<th>Not during the past week</th>
<th>Less than once a week</th>
<th>Once or twice a week</th>
<th>Three or more times a week</th>
</tr>
</thead>
<tbody>
<tr>
<td>6. How often have you had trouble sleeping because you...</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>a. Cannot get to sleep within 30 minutes</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>b. Wake up in the middle of the night or early morning</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>c. Have to get up to use the bathroom</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>d. Cannot breathe comfortably</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>e. Cough or snore loudly</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>f. Feel too cold</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>g. Feel too hot</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>h. Had bad dreams</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>i. Have pain</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>j. Other reason(s), please describe:</td>
<td></td>
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</tr>
</tbody>
</table>

7. How often have you taken medicine to help you sleep (prescribed or "over the counter")? 

8. How often have you had trouble staying awake while driving, eating meals, or engaging in social activity? 

9. How much of a problem is it for you to keep up enthusiasm to get things done?
Questionnaire B: CHILD’s SLEEP HABITS

The following statements are about your child’s sleep habits and possible difficulties with sleep. Think about the most recent typical week in your child’s life when answering these questions. Answer USUALLY if something occurs 5 or more times in a week; answer SOMETIMES if it occurs 2 – 4 times in a week; answer RARELY if something occurs never or 1 time during a week. Note: Some of the questions may not apply to your child.

1. **Bedtime**

Write in child’s USUAL bedtime: Weeknights: □□:□□□□ Weekends: □□:□□□□

<table>
<thead>
<tr>
<th></th>
<th>Usually (5-7)</th>
<th>Sometimes (2-4)</th>
<th>Rarely/ N/A (0-1)</th>
</tr>
</thead>
<tbody>
<tr>
<td>a. Child goes to sleep at the same time at night</td>
<td></td>
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</tr>
<tr>
<td>b. Child falls asleep within 20 minutes after going to bed</td>
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<td></td>
</tr>
<tr>
<td>c. Child falls asleep alone in own bed</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>d. Child falls asleep in parent’s or sibling’s bed</td>
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</tr>
<tr>
<td>e. Child needs parent in the room to fall asleep</td>
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</tr>
<tr>
<td>f. Child struggles at bedtime (cries, refuses to stay in bed, etc.)</td>
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<td></td>
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</tr>
<tr>
<td>g. Child is afraid of sleeping in the dark</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>h. Child is afraid of sleeping alone</td>
<td></td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

2. **Sleep Behavior**

Child’s usual amount of sleep each day (combining nighttime sleep and naps): Weekdays: □□:□□□□

<table>
<thead>
<tr>
<th></th>
<th>Usually (5-7)</th>
<th>Sometimes (2-4)</th>
<th>Rarely/ N/A (0-1)</th>
</tr>
</thead>
<tbody>
<tr>
<td>a. Child sleeps too little</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>b. Child sleeps the right amount</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>c. Child sleeps about the same amount each day</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>d. Child wets the bed at night</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>e. Child talks during sleep</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>f. Child is restless and moves a lot during sleep</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>g. Child sleepwalks during the night</td>
<td></td>
<td></td>
<td></td>
</tr>
</tbody>
</table>
h. Child moves to someone else’s bed during the night (parent, brother, sister, etc)  
   □  □  □  0-1

i. Child grinds teeth during sleep (your dentist may have told you this)  
   □  □  □  0-1

j. Child snores loudly  
   □  □  □  0-1

k. Child seems to stop breathing during sleep  
   □  □  □  0-1

l. Child snorts and/or gasps during sleep  
   □  □  □  0-1

m. Child has trouble sleeping away from home (visiting relatives, vacation)  
   □  □  □  0-1

<table>
<thead>
<tr>
<th></th>
<th>Usually (5-7)</th>
<th>Sometimes (2-4)</th>
<th>Rarely/ N/A (0-1)</th>
</tr>
</thead>
<tbody>
<tr>
<td>n. Child awakens during night screaming, sweating, and inconsolable</td>
<td>□  □  □  0-1</td>
<td></td>
<td></td>
</tr>
<tr>
<td>o. Child awakens alarmed by a frightening dream</td>
<td>□  □  □  0-1</td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

3. **Waking During the Night**

<table>
<thead>
<tr>
<th></th>
<th>Usually (5-7)</th>
<th>Sometimes (2-4)</th>
<th>Rarely/ N/A (0-1)</th>
</tr>
</thead>
<tbody>
<tr>
<td>a. Child awakes once during the night</td>
<td>□  □  □  0-1</td>
<td></td>
<td></td>
</tr>
<tr>
<td>b. Child awakes more than once during the night</td>
<td>□  □  □  0-1</td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

Write in number of minutes a night that waking usually lasts: _______________________

4. **Morning Waking**

Write in the time of day child usually wakes in the morning:

<table>
<thead>
<tr>
<th></th>
<th>Hours</th>
<th>Minutes</th>
</tr>
</thead>
<tbody>
<tr>
<td>Weekdays:</td>
<td>□□ : □□</td>
<td></td>
</tr>
<tr>
<td>Weekends:</td>
<td>□□ : □□</td>
<td></td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th></th>
<th>Usually (5-7)</th>
<th>Sometimes (2-4)</th>
<th>Rarely/ N/A (0-1)</th>
</tr>
</thead>
<tbody>
<tr>
<td>a. Child wakes up by him/herself</td>
<td>□  □  □  0-1</td>
<td></td>
<td></td>
</tr>
<tr>
<td>b. Child wakes up in negative mood</td>
<td>□  □  □  0-1</td>
<td></td>
<td></td>
</tr>
<tr>
<td>c. Adults or siblings wake up child</td>
<td>□  □  □  0-1</td>
<td></td>
<td></td>
</tr>
<tr>
<td>d. Child has difficulty getting out of bed in the morning</td>
<td>□  □  □  0-1</td>
<td></td>
<td></td>
</tr>
<tr>
<td>e. Child takes a long time to become alert in the morning</td>
<td>□  □  □  0-1</td>
<td></td>
<td></td>
</tr>
</tbody>
</table>
5. **Daytime Sleepiness**

<table>
<thead>
<tr>
<th></th>
<th>Usually (5-7)</th>
<th>Sometimes (2-4)</th>
<th>Rarely/ N/A (0-1)</th>
</tr>
</thead>
<tbody>
<tr>
<td>a. Child seems tired</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

During a usual week has your child appeared sleepy or fallen asleep during the following (check all that apply):

<table>
<thead>
<tr>
<th></th>
<th>Very Sleepy</th>
<th>Falls Asleep</th>
<th>Not Applicable</th>
</tr>
</thead>
<tbody>
<tr>
<td>b. Watching TV</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>c. Riding in a car</td>
<td></td>
<td></td>
<td></td>
</tr>
</tbody>
</table>
Questionnaire C: CAREGIVER FEELINGS AND MOOD

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

<table>
<thead>
<tr>
<th></th>
<th>Rarely or none of the time (less than 1 day)</th>
<th>Some or a little of the time (1-2 days)</th>
<th>Occasionally (3-4 days)</th>
<th>Most or all of the time (5-7 days)</th>
</tr>
</thead>
<tbody>
<tr>
<td>1.</td>
<td>I was bothered by things that usually don’t bother me.</td>
<td>0</td>
<td>1</td>
<td>2</td>
</tr>
<tr>
<td>2.</td>
<td>I did not feel like eating; my appetite was poor.</td>
<td>0</td>
<td>1</td>
<td>2</td>
</tr>
<tr>
<td>3.</td>
<td>I felt that I could not shake off the blues even with help from my family or friends.</td>
<td>0</td>
<td>1</td>
<td>2</td>
</tr>
<tr>
<td>4.</td>
<td>I felt I was just as good as other people.</td>
<td>0</td>
<td>1</td>
<td>2</td>
</tr>
<tr>
<td>5.</td>
<td>I had trouble keeping my mind on what I was doing.</td>
<td>0</td>
<td>1</td>
<td>2</td>
</tr>
<tr>
<td>6.</td>
<td>I felt depressed.</td>
<td>0</td>
<td>1</td>
<td>2</td>
</tr>
<tr>
<td>7.</td>
<td>I felt that everything I did was an effort.</td>
<td>0</td>
<td>1</td>
<td>2</td>
</tr>
<tr>
<td>8.</td>
<td>I felt hopeful about the future.</td>
<td>0</td>
<td>1</td>
<td>2</td>
</tr>
<tr>
<td>9.</td>
<td>I thought my life had been a failure.</td>
<td>0</td>
<td>1</td>
<td>2</td>
</tr>
<tr>
<td>10.</td>
<td>I felt fearful.</td>
<td>0</td>
<td>1</td>
<td>2</td>
</tr>
<tr>
<td>11.</td>
<td>My sleep was restless.</td>
<td>0</td>
<td>1</td>
<td>2</td>
</tr>
<tr>
<td>12.</td>
<td>I was happy.</td>
<td>0</td>
<td>1</td>
<td>2</td>
</tr>
<tr>
<td>13.</td>
<td>I talked less than usual.</td>
<td>0</td>
<td>1</td>
<td>2</td>
</tr>
<tr>
<td>14.</td>
<td>I felt lonely.</td>
<td>0</td>
<td>1</td>
<td>2</td>
</tr>
<tr>
<td>15.</td>
<td>People were unfriendly.</td>
<td>0</td>
<td>1</td>
<td>2</td>
</tr>
<tr>
<td>16.</td>
<td>I enjoyed life.</td>
<td>0</td>
<td>1</td>
<td>2</td>
</tr>
<tr>
<td>17.</td>
<td>I had crying spells.</td>
<td>0</td>
<td>1</td>
<td>2</td>
</tr>
<tr>
<td>18.</td>
<td>I felt sad.</td>
<td>0</td>
<td>1</td>
<td>2</td>
</tr>
<tr>
<td>19.</td>
<td>I felt that people dislike me.</td>
<td>0</td>
<td>1</td>
<td>2</td>
</tr>
<tr>
<td>20.</td>
<td>I could not get “going.”</td>
<td>0</td>
<td>1</td>
<td>2</td>
</tr>
</tbody>
</table>
Questionnaire D: CAREGIVER DAYTIME SLEEPINESS

The following questions will help us understand how sleepy you may feel during the day.

**Over the past week, we are interested in knowing:**

How likely are you to doze off or fall asleep in the following situations, in contrast to feeling just tired? This refers to your usual way of life in recent times. Even if you have not done some of these things recently try to work out how they would have affected you. Use the following scale to choose the most appropriate choice for each situation:

<table>
<thead>
<tr>
<th>Situation</th>
<th>no chance of dozing</th>
<th>slight chance of dozing</th>
<th>moderate chance of dozing</th>
<th>high chance of dozing</th>
</tr>
</thead>
<tbody>
<tr>
<td>C1. Sitting and reading</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>C2. Watching TV</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>C3. Sitting inactive in a public place (e.g. a theater or a meeting)</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>C4. As a passenger in a car for an hour without a break</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>C5. Lying down to rest in the afternoon when circumstances permit</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>C6. Sitting and talking to someone</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>C7. Sitting quietly after a lunch without alcohol</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>C8. In a car, while stopped for a few minutes in traffic</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
</tbody>
</table>
Questionnaire E: CAREGIVER FATIGUE

These questions will help us understand about your fatigue and the effect of fatigue on your activities.

For each of the following questions, indicate the number that most closely indicates how you have been feeling during the past week.

D1. To what degree have you experienced fatigue?

<table>
<thead>
<tr>
<th>Not at all</th>
<th>1</th>
<th>2</th>
<th>3</th>
<th>4</th>
<th>5</th>
<th>6</th>
<th>7</th>
<th>8</th>
<th>9</th>
<th>10</th>
<th>A great deal</th>
</tr>
</thead>
</table>

If no fatigue (i.e. answered “1” “not at all”, stop here and go to next section. Otherwise continue.

D2. How severe is the fatigue which you have been experiencing?

<table>
<thead>
<tr>
<th>Mild</th>
<th>1</th>
<th>2</th>
<th>3</th>
<th>4</th>
<th>5</th>
<th>6</th>
<th>7</th>
<th>8</th>
<th>9</th>
<th>10</th>
<th>Severe</th>
</tr>
</thead>
</table>

D3. To what degree has fatigue caused you distress?

<table>
<thead>
<tr>
<th>No distress</th>
<th>1</th>
<th>2</th>
<th>3</th>
<th>4</th>
<th>5</th>
<th>6</th>
<th>7</th>
<th>8</th>
<th>9</th>
<th>10</th>
<th>A great deal of distress</th>
</tr>
</thead>
</table>

To what degree HAS FATIGUE interfered with your ability to do the following activities in the past week.

For activities you don’t do, for reasons other than fatigue (e.g. you don’t work because you are retired), indicate you don’t do this activity.

D4. Household chores

<table>
<thead>
<tr>
<th>Don’t do this activity</th>
</tr>
</thead>
</table>

<table>
<thead>
<tr>
<th>Not at all</th>
<th>1</th>
<th>2</th>
<th>3</th>
<th>4</th>
<th>5</th>
<th>6</th>
<th>7</th>
<th>8</th>
<th>9</th>
<th>10</th>
<th>A great deal</th>
</tr>
</thead>
</table>

D5. Cook

<table>
<thead>
<tr>
<th>Don’t do this activity</th>
</tr>
</thead>
</table>

<table>
<thead>
<tr>
<th>Not at all</th>
<th>1</th>
<th>2</th>
<th>3</th>
<th>4</th>
<th>5</th>
<th>6</th>
<th>7</th>
<th>8</th>
<th>9</th>
<th>10</th>
<th>A great deal</th>
</tr>
</thead>
</table>

D6. Bathe or wash

<table>
<thead>
<tr>
<th>Don’t do this activity</th>
</tr>
</thead>
</table>

<table>
<thead>
<tr>
<th>Not at all</th>
<th>1</th>
<th>2</th>
<th>3</th>
<th>4</th>
<th>5</th>
<th>6</th>
<th>7</th>
<th>8</th>
<th>9</th>
<th>10</th>
<th>A great deal</th>
</tr>
</thead>
</table>

D7. Dress

<table>
<thead>
<tr>
<th>Don’t do this activity</th>
</tr>
</thead>
</table>

<table>
<thead>
<tr>
<th>Not at all</th>
<th>1</th>
<th>2</th>
<th>3</th>
<th>4</th>
<th>5</th>
<th>6</th>
<th>7</th>
<th>8</th>
<th>9</th>
<th>10</th>
<th>A great deal</th>
</tr>
</thead>
</table>

D8. Work

<table>
<thead>
<tr>
<th>Don’t do this activity</th>
</tr>
</thead>
</table>

<table>
<thead>
<tr>
<th>Not at all</th>
<th>1</th>
<th>2</th>
<th>3</th>
<th>4</th>
<th>5</th>
<th>6</th>
<th>7</th>
<th>8</th>
<th>9</th>
<th>10</th>
<th>A great deal</th>
</tr>
</thead>
</table>
D9. Visit or socialize with friends  ○ Don’t do this activity

D10. Engage in sexual activity  ○ Don’t do this activity

D11. Engage in leisure and recreational activities  ○ Don’t do this activity

D12. Shop and do errands  ○ Don’t do this activity

D13. Walk  ○ Don’t do this activity

D14. Exercise, other than walking  ○ Don’t do this activity

D15. Over the past week, how often have you been fatigued?
○ Every day
○ Most, but not all days
○ Occasionally, but not most days
○ Hardly any days

D16. To what degree has your fatigue changed during the past week?
○ Increased
○ Fatigue has gone up and down
○ Stayed the same
○ Decreased
### Questionnaire F: SLEEP & ACTIVITY

These questions will help us understand more about your activities and sleep behaviors over the past week.

<table>
<thead>
<tr>
<th></th>
<th>Always</th>
<th>Frequently</th>
<th>Sometimes</th>
<th>Rarely</th>
<th>Never</th>
</tr>
</thead>
<tbody>
<tr>
<td>E1. I take daytime naps lasting two or more hours</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>E2. I go to bed at different times from day to day</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>E3. I get out of bed at different times from day to day</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>E4. I exercise to the point of sweating within 1 hour of going to bed</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>E5. I stay in bed longer than I should two or three times a week</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>E6. I use alcohol, tobacco, or caffeine within 4 hours of going to bed or after going to bed</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>E7. I play video games, use the internet, or clean the house before going to bed (or other things that may stimulate me before bedtime)</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>E8. I go to bed feeling stressed, angry, upset, or nervous</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>E9. I watch television, read, eat, or study in bed (i.e. use my bed for things other than sleeping or sex)</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>E10. I sleep on an uncomfortable bed (for example: poor mattress or pillow, too many or not enough blankets)</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>E11. I sleep in a bedroom that is too bright, too stuffy, too hot, too cold, or too noisy (i.e. uncomfortable)</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>E12. I do work tasks before bedtime (for example: pay bills, schedule, or study)</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>E13. I think, plan, or worry when I am in bed</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
</tbody>
</table>
Questionnaire G: CAREGIVER HEALTH & WELLBEING

This survey asks for your views about your health. This information will help keep track of how you feel and how well you are able to do your usual activities.

For each of the following questions, please mark an ✗ in the one box that best describes your answer.

1. In general, would you say your health is:

<table>
<thead>
<tr>
<th>Excellent</th>
<th>Very good</th>
<th>Good</th>
<th>Fair</th>
<th>Poor</th>
</tr>
</thead>
<tbody>
<tr>
<td>▼ ▼ ▼ ▼</td>
<td>▼ ▼ ▼ ▼</td>
<td>▼ ▼</td>
<td>▼ ▼</td>
<td>▼ ▼</td>
</tr>
<tr>
<td>1 2 3 4</td>
<td>5 6 7 8</td>
<td>9 10</td>
<td>11 12</td>
<td>13 14</td>
</tr>
</tbody>
</table>

2. The following questions are about activities you might do during a typical day. Does your health now limit you in these activities? If so, how much?

a. Moderate activities, such as moving a table, pushing a vacuum cleaner, bowling, or playing golf

   | Yes, limited a lot | Yes, limited a little | No, not limited at all |
   | ▼ ▼ ▼ | ▼ ▼ ▼ | ▼ ▼ ▼ |
   | 1 2 3 | 4 5 6 | 7 8 9 |

b. Climbing several flights of stairs

   | 1 2 3 |
   | ▼ ▼ ▼ |
3. During the past week, how much of the time have you had any of the following problems with your work or other regular daily activities as a result of your physical health?

<table>
<thead>
<tr>
<th>All of the time</th>
<th>Most of the time</th>
<th>Some of the time</th>
<th>A little of the time</th>
<th>None of the time</th>
</tr>
</thead>
<tbody>
<tr>
<td>▼</td>
<td>▼</td>
<td>▼</td>
<td>▼</td>
<td>▼</td>
</tr>
</tbody>
</table>

a. Accomplished less than you would like ................................... □ 1 □ 2 □ 3 □ 4 □ 5

b. Were limited in the kind of work or other activities ................ □ 1 □ 2 □ 3 □ 4 □ 5

4. During the past week, how much of the time have you had any of the following problems with your work or other regular daily activities as a result of any emotional problems (such as feeling depressed or anxious)?

<table>
<thead>
<tr>
<th>All of the time</th>
<th>Most of the time</th>
<th>Some of the time</th>
<th>A little of the time</th>
<th>None of the time</th>
</tr>
</thead>
<tbody>
<tr>
<td>▼</td>
<td>▼</td>
<td>▼</td>
<td>▼</td>
<td>▼</td>
</tr>
</tbody>
</table>

a. Accomplished less than you would like ................................... □ 1 □ 2 □ 3 □ 4 □ 5

b. Did work or other activities less carefully than usual ............... □ 1 □ 2 □ 3 □ 4 □ 5

5. During the past week, how much did pain interfere with your normal work (including both work outside the home and housework)?

<table>
<thead>
<tr>
<th>Not at all</th>
<th>A little bit</th>
<th>Moderately</th>
<th>Quite a bit</th>
<th>Extremely</th>
</tr>
</thead>
<tbody>
<tr>
<td>▼</td>
<td>▼</td>
<td>▼</td>
<td>▼</td>
<td>▼</td>
</tr>
<tr>
<td>□ 1</td>
<td>□ 2</td>
<td>□ 3</td>
<td>□ 4</td>
<td>□ 5</td>
</tr>
</tbody>
</table>

6. These questions are about how you feel and how things have been with you during the past week. For each question, please give the one answer that comes closest to the way you have been feeling. How much of the time during the past week...

<table>
<thead>
<tr>
<th>All of the time</th>
<th>Most of the time</th>
<th>Some of the time</th>
<th>A little of the time</th>
<th>None of the time</th>
</tr>
</thead>
<tbody>
<tr>
<td>▼</td>
<td>▼</td>
<td>▼</td>
<td>▼</td>
<td>▼</td>
</tr>
</tbody>
</table>

a. Have you felt calm and peaceful? ........................................ □ 1 □ 2 □ 3 □ 4 □ 5

b. Did you have a lot of energy? ........................................... □ 1 □ 2 □ 3 □ 4 □ 5
7. During the past week, how much of the time has your physical health or emotional problems interfered with your social activities (like visiting with friends, relatives, etc.)?

<table>
<thead>
<tr>
<th>Have you felt downhearted and depressed?</th>
<th>1</th>
<th>2</th>
<th>3</th>
<th>4</th>
<th>5</th>
</tr>
</thead>
<tbody>
<tr>
<td>All of the time</td>
<td>▼</td>
<td></td>
<td></td>
<td></td>
<td>▼</td>
</tr>
<tr>
<td>Most of the time</td>
<td>▼</td>
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<td></td>
<td></td>
<td>▼</td>
</tr>
<tr>
<td>Some of the time</td>
<td>▼</td>
<td>▼</td>
<td>▼</td>
<td></td>
<td>▼</td>
</tr>
<tr>
<td>A little of the time</td>
<td>▼</td>
<td>▼</td>
<td>▼</td>
<td>▼</td>
<td>▼</td>
</tr>
<tr>
<td>None of the time</td>
<td>▼</td>
<td>▼</td>
<td>▼</td>
<td>▼</td>
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</tr>
</tbody>
</table>

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(IQOLA SF-12v2 Acute, English [Canada])
Thank you for completing this questionnaire.

Your responses are valuable and will help us to learn more about sleep and related health outcomes in family caregivers.

Please take a moment to tell us what it was like for you to participate in this study.

What I most liked about the study was:
__________________________________________________________________
__________________________________________________________________
__________________________________________________________________

What I least liked about the study was:
__________________________________________________________________
__________________________________________________________________
__________________________________________________________________