

# Losing Sleep: Considering Caregivers of Children with Rare Epilepsy Syndromes



The burden of care for children with rare epilepsies is very high. These patients often have treatment-resistant seizures, complex medical comorbidities, and challenges with learning and behavior. Moreover, these children are at heightened risk for sudden unexpected death in epilepsy (SUDEP) and for death due to other causes related to their underlying conditions.<sup>1</sup> In this volume of *The Journal: X*, investigators from the Rare Epilepsy Network provide important and novel insight into the impact on caregivers of caring for children with rare epilepsies.<sup>2</sup> Sleep disturbances in these children are associated with fatigue, sleep-related disturbance, and sleep impairment in their caregivers, often leading to parental anxiety and depression.

We expect parents or guardians to care for their children at all hours of the day and night; hours worked by parents of children with medical complexity can easily surpass the safety limits imposed on professional medical providers.<sup>3</sup> Analyses of data from the National Survey of Children with Special Health Care Needs, a nationally representative sample of 40 242 parents and guardians, estimated that 31.5% of caregivers of children with epilepsy spend >21 hours per week providing health care at home.<sup>4</sup>

Across cultures, disturbances in the sleep of young children influence the patterns of their parents' sleep and daytime functioning.<sup>5</sup> This relationship may be heightened for caregivers of children with epilepsy. There is increased room sharing, co-sleeping, sleep disturbance, daytime sleepiness, and fatigue among caregivers of children with epilepsy compared with caregivers for a general pediatrics population.<sup>6</sup> Concern about nocturnal seizures was noted by 69% of epilepsy caregivers studied, and 44% of caregivers reported never or rarely feeling rested. Despite the association of co-sleeping with decreased sleep quality, caregivers of children with epilepsy were more likely to initiate co-sleeping compared with their general pediatric counterparts (for whom co-sleeping, when it occurred, was usually initiated by the child). This phenomenon emphasizes caregivers' worries about the potentially grave sequelae of nocturnal seizures, confirmed by the data reported by the Rare Epilepsy Network.<sup>2</sup>

A population-based case-control study reported that caregivers of children with epilepsy had similar levels of parenting stress as caregivers of children with non-epilepsy neurologic conditions. However, unlike the caregivers of children with other neurologic conditions, parents of children with epilepsy reported that much of their stress was attributed to sleep disruption.<sup>7</sup> An assessment of 50 parents of children aged ≤5 years with epilepsy found that these parents' total

nocturnal sleep time averaged just 4 hours and they had 3 or more awakenings per night. The number of nighttime awakenings was inversely correlated with parental sleep quality, marital satisfaction, and parental health.<sup>8</sup> Importantly, the American Academy of Sleep Medicine recommends >7 hours of sleep per night for healthy adults to avoid sequelae of sleep deprivation.<sup>9</sup>

Taking care of medically complex children in a sleep-deprived state is not an easy task. Studies of sleep deprivation among interns and residents have consistently reported important impacts on performance. A study of sleep-deprived anesthesiology residents found increased errors in drug administration and dosage recall, delay in the recognition of hypotension, and missing communication compared with their non-sleep-deprived counterparts during a simulation experiment.<sup>10</sup> A case-crossover study in which 2737 medical interns completed a survey during rotations with ≥5 extended shifts compared with ≤4 extended shifts found that residents who worked more extended shifts were more likely to report making at least 1 fatigue-related medical error and 1 fatigue-related preventable adverse event, and to have trouble paying attention during clinical activities.<sup>11</sup> Caregivers of children with epilepsy, who have frequently disrupted sleep and limited total sleep time, likewise may find it increasingly difficult to take care of a child with long-term medical needs and to perform their daytime duties well. Indeed, increased fatigue and decreased productivity are reported for caregivers of children with epilepsy compared with caregivers of non-epilepsy neurologic conditions.<sup>12</sup>

Chronic caregiver sleep deprivation can jeopardize the overall health and well-being of the very people we rely on to care for vulnerable children with epilepsy. A study from the UK Biobank demonstrated that among 461 347 participants free of cardiovascular disease, getting <6 hours of sleep per night can increase the risk of myocardial infarction, after adjusting for genetic sleep traits and genetic risk for coronary artery disease.<sup>13</sup> A study of community-based adolescents found that sleep deprivation and depression are reciprocal risk factors,<sup>14</sup> and a study of medical interns found that increased sleep deprivation and sleep disturbance are associated with an increased probability of depression over time.<sup>15</sup> Experiments have clearly demonstrated that sleep deprivation and sleep restriction can induce anxiety.<sup>16</sup>

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SUDEP sudden unexpected death in epilepsy

R.S. is supported by the National Institutes of Health (Grant R01 HL147261), the Patient-Centered Outcomes Research Institute (2015C2-1507-31187), the Pediatric Epilepsy Research Foundation, and the University of Michigan; receives royalties from UpToDate for authorship of topics related to neonatal seizures; is a consultant for the Epilepsy Study Consortium; and serves as an Editorial Board member for *The Journal of Pediatrics*. G.G. is supported by National Institutes of Health Grant T32 HL110952 and R01 NS111166.

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<https://doi.org/10.1016/j.jmpdx.2020.100022>

Sleep and circadian rhythm play a role in immune function and mediate the movement of B and T cells between the blood and lymphoid tissues.<sup>17</sup> Among healthy men, reduction of sleep time by 2 hours resulted in increased levels of ghrelin (promoting appetite) and decreased leptin levels (decreasing satiety).<sup>18</sup> This finding is thought to explain, at least in part, the association between obesity and sleep deprivation. Thus, sleep-deprived caregivers of our patients with rare epilepsies are at risk not only for obesity, but also for its downstream consequences, such as diabetes and hypertension, along with anxiety and depression.

In a population with an increased risk of sleep-related morbidity, such as children with rare epilepsies, it is important to evaluate and optimize resources for caregivers. A sense of mastery over the treatment of epilepsy and a sense of optimism are associated with an improved quality of life among mothers of children with epilepsy,<sup>19</sup> as echoed in the Rare Epilepsy Network study.<sup>2</sup> Targeted education about pediatric sleep disorders and provision of tools, such as behavioral therapy interventions,<sup>20</sup> could provide caregivers with an increased sense of mastery over their child's disordered sleep. Recognition and resource provision for parental sleep disorders is a clear gap in our current child-focused care model.

The use of sleep monitoring devices can provide a subjective sense of relief at times; however, frustration due to alarm fatigue from false-positives and heightened anxiety due to potential false-negative results may be of concern.<sup>21</sup> None of the available monitoring devices has been clearly demonstrated to reduce the risk for SUDEP in children. In the Rare Epilepsy Network study, bed-sharing and the use of sleep monitoring devices were associated with an elevated risk for caregiver sleep impairment and sleep-related disturbance.<sup>2</sup> Further refinement of seizure monitoring devices could greatly improve the well-being of both caregivers and children with epilepsy. Until then, clinicians must pay careful attention to the data that support (or refute) the use of these devices when they counsel families.

Physicians are compelled "above all" to "do no harm." By advising or inadvertently encouraging caregivers to be vigilant at all times while their children with epilepsy are sleeping, we expose parents to risks of sleep deprivation (eg, accidents from drowsy driving, risk for cardiovascular disease and worsening anxiety and depression, among many others) and cannot guarantee that we are protecting their children. The Rare Epilepsy Network study<sup>2</sup> should serve as a call to action to optimize comprehensive care for everyone who cares for families affected by these disorders. ■

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