Introduction

Sleep difficulties are estimated to occur at a prevalence of 50% to 80% in children with autism spectrum disorders (ASD) [1-3]. Sleep disturbances in these children are also of a great concern for their parents due to the detrimental effects of poor sleep on daytime behaviors and overall level of functioning [4,5]. Children with ASD and sleep difficulties may exhibit behavioral dysregulation such as hyperactivity, sensory alteration with self-stimulatory behaviors, anxiety, aggression and ultimately poor general health with diminished quality of life and wellbeing [6-10]. Sleep disorders are commonly reported by parents of children with ASD and are present in 53-78% of individuals with ASD [11]. In addition, sleep disorders in children with ASD can occur with other concomitant psychiatric and, medical conditions [11]. If not addressed, sleep disorders can exacerbate other ASD behaviors such as

- Aggression
- Depression
- Hyperactivity
- Increased behavioral problems
- Irritability
- Poor learning and cognitive performance
- Self-injury
Causes of sleep disorders in children with ASD

Although the exact etiology of sleep disturbances in children with ASD is yet to be determined, several theories have been suggested including:

- **Misinterpretation of social cues:** From an early age, children with ASD spend less time engaged in social interaction compared to their age group peers and as such, potential social exchanges are avoided because other environmental stimuli are more highly valued by default [12]. In general children usually recognize that night time is associated with going to sleep and utilize their social cues of seeing their siblings getting ready to go to bed to sleep at bed time. Children with ASD, may not be tuned to these social cues and could misinterpret them.

- **Melatonin dysregulation:** Melatonin, which normally regulate sleep-wake cycles and is produced internally from the amino acid tryptophan, was found to be lower than normal in some children with ASD [13]. Typically, melatonin levels rise in response to darkness at night and decrease during the daylight hours. Studies have shown that some children with ASD have melatonin dysregulation that could contribute to disturbed sleep cycles [14].

- **Sensory sensitivity:** Children with ASD may have trouble falling asleep or awaken in the middle of the night due to an increased sensitivity to touch or sound stimuli. Although sensitivities in all sensory domains are similarly associated with sleep problems, hypersensitivity towards touch exhibit the strongest relationship with sleep disturbances in children with ASD. This may indicate the existence of a specific sleep disturbance mechanism that is associated with sensitivity to touch, which has been considered in several clinical studies [15].

- **Concomitant psychiatric disorders:** Children with ASD have higher rates of concomitant psychiatric disorders which could adversely affect sleep and may include:

  - **Anxiety disorders:** These disorders are always characterized by excessive fear and/or anxiety, which leads to impairment and/or distress and, include separation anxiety, specific phobias, social anxiety, panic disorder, agoraphobia, and generalized anxiety disorder [16]. Although anxiety disorders affect millions of people and has been shown to co-occur in combination with sleep disturbances. Children with ASD tend to have a higher level of anxiety than other children which would adversely affect their sleep [17].

  - **Depressive disorders:** Children with ASD may be more vulnerable to depression than the general population. Depressive disorders in ASD are associated with poor quality-of-life, including sleep disturbances, medical illness and hospitalization [18].

  - **Bipolar disorder:** When bipolar disorder is concomitant with ASD it involves a higher level of manic symptoms. Some of the cardinal symptoms of mania include difficulties with initiating sleep and a decreased need for sleep [18].

  - **Obsessive compulsive disorder:** Obsessive compulsive disorder is highly prevalent in individuals with ASD. The overlap between these two conditions is still unclear. Children with either condition may have unusual responses to sensory experiences. The response to sensory experiences could be manifested throughout the day, however when it occurs toward bedtime, it could lead to difficulty with initiating and maintaining sleep [18].

  - **Oppositional defiant disorder:** This condition is characterized by a pattern of angry, irritable mood, argumentative and defiant behavior, or vindictiveness [16]. In contrast children with ASD who tend to show irritability and defiance not for its own sake, but as either an instinctive response to a frustrating situation or according to some reason that may simply not be obvious to a bystander. When the two conditions exist together, there is an increased level of irritability, anger, and defiance to follow rules in the context regular bedtime and sleep schedules.

  - **Conduct disorder:** This disorder can be defined as a persistent and pervasive problem with social interaction and the perceptions
in others, that usually arises from abnormal social learning, rather than the intrinsic nature of ASD. Conduct disorder is also characterized by a repetitive and persistent pattern of behaviors where the basic rights of others and major age-appropriate social norms or rules are violated. The co-occurrence of ASD with conduct disorder also alter the sleep pattern of these children.

- **Schizophrenia and psychotic disorders:** These disorders are characterized by abnormal behaviors, including delusions, hallucinations, disorganized speech and or negative symptoms such as flat emotional expression and avolition [16]. The association between ASD, schizophrenia and psychosis are manifested by several cognitive features common to both conditions such as weak central coherence, difficulties in set-shifting, deficiency of communication, deficiency of attention, and social impairment. These features when present in both conditions could also affect the quality and quantity of sleep.

- **Gilles de la Tourette syndrome:** This neuropsychiatric disorder is characterized by multiple motor and one or more phonic tics, lasting at least 1 year [16]. The prevalence of ASD in Tourette’s Syndrome patients can vary from 2.9 to 20% [19]. Common tics include repetitive throat clearing, blinking or grimacing. The presence of Gilles de la Tourette syndrome and ASD could exacerbate difficulty with falling asleep and staying asleep through the night. These sleep issues could tend to worsen behavioral challenges, interfere with learning and decrease the overall quality of life in these children.

- **Attention deficit hyperactivity disorder:** Sleep disturbance is often a problem for children with either ASD or attention deficit hyperactivity disorder. Psychostimulant medications which are commonly used to treat attention deficit hyperactivity disorder symptoms could further worsen sleep difficulties in children with ASD.

**Concomitant medical conditions**

Some of the common co-occurring medical conditions include gastrointestinal disorders and seizure disorders:

- **Gastrointestinal disorders:** These are frequently associated with chronic constipation, abdominal pain, gastroesophageal reflux and bowel inflammation [20], which could all lead to ongoing sleep disturbances.

- **Seizure disorders:** Children with ASD have an increased frequency of seizure disorders [21], which can be presenting as red flags and may include unexplained staring spells, involuntary movements, unexplained confusion, severe headaches, and disrupted sleep.

**General management of sleep difficulties**

Timely and appropriate treatment of concomitant psychiatric and medical conditions could eventually lead to an overall improvement in the sleep pattern of children with ASD [18]. The various therapies [22,23], that could improve sleep in children with ASD may include [23] the following interventions:

- Behavioral intervention programs that focus on teaching certain skills that would help in reducing the social, language and behavioral difficulties associated with ASD.

- Programs that focus on teaching children how to act in social situations and to improve communication with others.

- Applied behavior analysis could also help children learn new skills and generalize these skills to multiple situations through a reward-based motivation system.

- **Educational therapies:** Highly structured educational programs which typically include a team of specialists and a variety of activities to improve social skills, communication and behavior especially in preschool children.

- **Family therapies:** Parents and other family members can learn how to play and interact with their children in ways that promote social interaction skills, manage problem behaviors, and teach daily living skills and communication.

- **Other therapies:** Depending on each child needs. For instance, speech therapy could improve communication skills, occupational...
therapy could be used to improve activities of daily living, and physical therapy to improve movement and gait balance. Practicing various sleep hygiene principles [24], such as:

- Avoiding all intake of caffeine, sugary drinks or sweet before bedtime.
- Establishing a nighttime routine and empathize going to bed at the same time every night.
- Helping to relax before bed by reading a book, giving a gentle back massage, or turning on soft music.
- Avoiding stimulating activities such as watching television, playing video games, or using electronics at least an hour before bedtime.
- Preventing sensory distractions during the night, such as blocking outside light with dark curtains, installing thick carpeting, fixing creaky doors, regulating room temperature and adjusting bedding sheets to fit with the sensory needs.

**Medications:** Although it is not recommended to use medications for the treatment of sleep difficulties in children with ASD. Medications can be used to treat the concomitant psychiatric and medical conditions. Some children with ASD who were prescribed medications for sleep had worse daytime behaviors and overall quality of life than the children who were not taking medications.

Psychiatric medications could be used to stabilize some of the psychological and behavioral symptoms in ASD [11,18,25]. Risperidone and aripiprazole have been approved by the US Food and Drug Administration (FDA) for treatment of irritability associated with ASD. However, associated metabolic side-effects are concerning and the adverse effects of long-term use can be burdensome. The stimulant methylphenidate and the non-stimulant atomoxetine are FDA approved for the treatment of attention deficit hyperactivity disorder symptoms. Clonidine and guanfacine appear to be also somehow helpful for the management of attention deficit.

The serotonin reuptake inhibitor antidepressants seem to be poorly tolerated in ASD and have not shown any beneficial effects in reducing restricted repetitive behaviors, anxiety, or depression. Buspirone have shown some benefits in the reduction of restricted repetitive behaviors. Antiseizure medications have been inconsistent in the overall management of ASD except for their effects on stabilizing concomitant seizure disorders.

Glutamatergic agents, Gamma-aminobutyric acid, cholinergic agents, oxytocin have been anecdotal and inconsistent in their clinical outcomes. Other antidepressants, antihistamines, antipsychotics, benzodiazepines, α-agonists, β-blockers, sedatives, iron supplements, and vitamins/dietary supplements B6 and magnesium, multivitamin/mineral folic acid, omega-3, L-Carnosine, chelation, N-Acetylcysteine have not shown efficacy in the treatment of ASD core symptoms or in the management of sleep disturbances. Probiotics and other gastrointestinal medications could be used only for ASD patients with gastrointestinal symptoms and iron supplementation can be used in those with low serum ferritin level.

Melatonin: This is an endogenous neurohormone released by the pineal gland in response to decreasing levels of light, its levels begin to rise shortly after nightfall, and plays an important role in promoting sleep by regulating the sleep-wake cycle and setting the body's sleep clock. Although melatonin itself is generally safe, it is not clear if all its over the counter products contain the hormone, it is also considered a supplement rather than an FDA approved sleep medication. Melatonin has been found to help initiate sleep in some children with ASD [26].

**Complementary and alternative interventions**

Massage Therapy, Acupuncture, Exercise, Music Therapy, Animal-Assisted Therapy, Neurofeedback, although found acceptable for the management of some of ASD core symptoms, they have not been proven to be beneficial for all children with ASD and sleep disturbances and need more replications in controlled research studies before being generalized as alternative effective treatment interventions [27].

**Case Study: The Benefits of Storytelling in the Management of Sleep Disturbances**

Several camps offer specialized support and programs for children with ASD and the following case study describes the effect of storytelling on inducing sleep in some children with ASD who participated in a Summer camp in the Northeastern region of Toronto, Canada.

This particular Summer camp enrolled male children with ASD between the age of 7 and 10 years old. The camp had 48 children enrolled for a 7 days period who participated in various sport and outdoor activities that were specially designed to meet the cognitive and behavioral needs of children with ASD. Children were also divided into groups and each group had 6 children. Each group had their own coach, two assistant coaches and they had their own schedules of activities, meals time, periods of rest, bedtime and waking up time. Many of the children had ongoing sleep difficulties that included:

- Difficulty falling asleep
- Inconsistent sleep routines
- Restlessness or poor sleep quality
- Waking early and waking frequently

The following story was told consistently and repeatedly at bedtime to one group of children and resulted in the remission of the difficulty in falling asleep in five of the six children in that group.

**The tallest tree in the woods**

Once upon a time there was an old tall tree which did not grow any leaves although it was alive and big and was taller than all the other trees in the woods. No birds or squirrels would build their nests on its branches because they thought it was not a living tree. The tree was sad and lonely and kept wishing that one day some birds or squirrels will use its branches to build their nest and keep her company specially in the long cold winter months. One day a squirrel who was lost in the woods and was starting to feel the cold breeze of air and realized that he needed to build his own nest before Winter arrives. He looked all around and saw that all the trees were already occupied by nests of birds and squirrels and was not welcomed by their inhabitants. He then noticed the tall tree and thought that it would be the perfect one for building his nest. The tree welcomed him and showed him many spots on one of its twisted branches to build a secure and sheltered nest. Other squirrels made fun of this plan because they still believed that this tall tree was dead, and it will not survive the Winter storms and that when the snow falls, his nest will be destroyed and crashed to the cold frozen grounds. Birds flew over and laughed at the squirrel and the squirrel was very happy that he finally finished building his nest and the tree was so glad to have his company. He would climb on its tallest branches and could see as far as he is able some fields that were abundant with acorns. The tree also pointed areas of fallen pecans and walnuts that were hidden from sight of other squirrels and birds. The squirrel worked hard and gathered so many acorns, walnuts and pecans and stored them in his large nest for the long cold Winter. That Winter came with unusual extremely cold air, frigid winds and lot of snow. The tree with its long strong branches offered an amazing protection for the squirrel nest in addition to providing extra warmth against the blustery cold winds. As the frigid cold days continued with snowflakes turning to ice, the other squirrels and birds’ nests that were built on other trees began to fall from the weight of the ice. Their stored nuts were scattered and buried under the ice and they found themselves cold frozen, homeless and hungry.

The tall tree was very, very sad to see what was happening and had love and compassion toward these destitute birds and squirrels and asked her only inhabitant her squirrel if he could invite all the birds and squirrels who lost their nests to come and share his very spacious and large nest. The squirrel came down from the tallest tree and welcomed all the homeless birds and squirrels with their babies to his large nest. He was so happy to shelter them from the harsh Winter weather and to share his abundance of stored food. The tall tree was so joyful to have the company of so many birds, squirrels and their babies. This amazing and unexpected company took away the so many years of lonely and sad living. The cold Winter gradually left to make room for the Spring, the ice melted, and the sun began to shine on...
other trees with their branches and leaves. The tall tree then started to think that sooner than later she will lose her abundant company of friends who will abandon her to live on other trees with leaves and she will be left once more all alone with so many branches and no leaves. The squirrels and the birds felt the tall tree’s sadness and began to wonder on how to help that loving tree growing leaves on her so many branches. They noticed that in nearby fields, the farmers place a kind of a dark soil on the ground before they water to grow their green crops. They whispered among themselves and found that this soil is full of top soil. So, they carried in their little mouths that dark soil and spread it all around the tall tree. When the rain came, the dark soil disappeared and seemed to be absorbed by the tall tree who then grew bright green leaves all over its many countless branches. The squirrels and birds invited so many other squirrels and birds to come build their nests in this tall tree who was filled with joy and gratitude and her first squirrel friend became her ambassador and envoy to let all the other trees know that his tall tree is not just the tallest but the most amazing and awesome tree of the woods. And so, for many years that came and gone, the tallest tree of the woods became the only tree where all the squirrels and birds that lived in these woods built their nests and stored their Winter food. And everybody lived happily after.

The children asked their coaches to repeat that story every night at their bedtime until they fell asleep. By the end of the 7th day of camping all seven children had memorized the story of the tallest tree in the woods and told the same story to their parents, when they returned home from their Summer camp. The parents repeated that same story every night at bedtime and it helped them fall asleep and remain asleep throughout the night.

Discussion

There is increasing evidence of severe sleep problems in children with ASD. Sleep disorders are often untreated and ignored as other behavioral difficulties tend to take precedence. Some research supports the efficacy of melatonin in decreasing sleep-onset latency and increasing total sleep time when administered close to bedtime [26]. In contrast, other studies have suggested that melatonin is only may not be effective for many children with ASD [2]. After the initiation of treatment for concomitant psychiatric and medical conditions, parent-based education and behavioral interventions are the first line of treatment for sleep disruption in ASD [11]. Behavioral interventions such as sleep hygiene approaches which focus on changing the environment to promote regular sleep-wake cycle have been shown to be effective interventions in improving sleep onset and maintenance in ASD [11]. The basic principles of sleep hygiene include selecting an appropriate bedtime and set routine, minimizing television watching, and reducing emotional and behavioral stimulation at night [18]. Given the associations between inadequate sleep, intensified daytime problem behaviors, and parental stress in ASD, there is a strong need to develop other effective sleep interventions.

Storytelling can be used as an additional intervention to help minimize and resolve sleep difficulties in children with ASD who are willing to listen to story telling at bedtime. Since most of our lives are spent telling tales. Storytelling is an exciting way to engage children in listening and speaking. The gift of storytelling may be one of the most powerful tools one can use to help children with ASD not just to fall asleep but to continue to develop their language and cognitive skills as they master and repeat the stories that they have listened to and later memorized [28]. Repetition of the same story becomes a key factor. As the children become more familiar with the story, they can focus on the subtler aspects of its content and its plot. Repetition allows the children to visualize the various characters and identify their emotions and correlate them with their own emotions and as such minimize distraction and outward sensory stimulation, especially in folktales. The distinctive features of folktales make them ideal for beginning storytellers in children with ASD. The folktale usually begins with an opening, "Once upon a time...." The setting is generalized or universal, such as a road or a forest. The characters are flat and stereotypical, such as givers or selfish. These features can be presented in a few sentences, thus allowing the children to readily become familiar with the story while allowing the storyteller to concentrate on the development of the plot [29]. Generally, there are two plot structures: linear and cumulative [14]. In the linear plot, a problem is introduced and followed by a series of events as the character attempts to solve the problem. In cumulative plots, the character’s, problem, and actions build on each other. It often includes a repetition of phrases. Folktales usually have abrupt endings with a short, sweet resolution: "...and they lived" [30].
Storytelling Effects on Sleep Difficulties in Children with Autism Spectrum Disorders: Case Study

Conclusion

Storytelling can be an intervention to consider for the management of sleep disruption manifested by difficulty falling asleep in some children with ASD. This benign intervention could not be generalized and used as an effective management of sleep difficulties in children with ASD until such outcome is replicated in double-blind, placebo randomized controlled trials with long-term follow-up and reliably diagnosed samples of participants with ASD, using standard treatment outcome measures, while continually monitoring potential adverse effects and changes in concomitant treatment. In the meantime, clinician need to be aware of the importance of addressing sleep disturbances as an important component of the overall management of the emotional and behavioral disorders in children with ASD and in providing guidance to the parents and caregivers. Treating sleep difficulties in children with ASD has great potential to improve daytime behavior and family functioning in this special and unique yet vulnerable population.

Conflicts of Interests

No conflicts of interests. The materials described in this review are those of the author and do not reflect the view of the Department of Veterans Affairs or the Sacramento VA Medical Center or UC Davis, School of Medicine, Sacramento, California.

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