



Sleep In Children with Medical Complexity

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Presenter Disclosure

- **Presenter:** Caroline Okorie, MD, MPH
- **Relationships with commercial interests:**
 - Grants/Research Support: none
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Objectives

- Review the importance of adequate sleep and the consequences of poor sleep, especially in children with medical complexity (CMC).
- Recall some common sleep-disordered breathing (SDB) issues that arise in CMC.
- Review some evidence-based treatments for SDB and insomnia including the benefits of a multidisciplinary, comprehensive and individualized approach when considering sleep concerns
- Discuss insomnia often seen in children with medical complexities, including various pharmacologic and non-pharmacologic treatment options

What is Sleep?

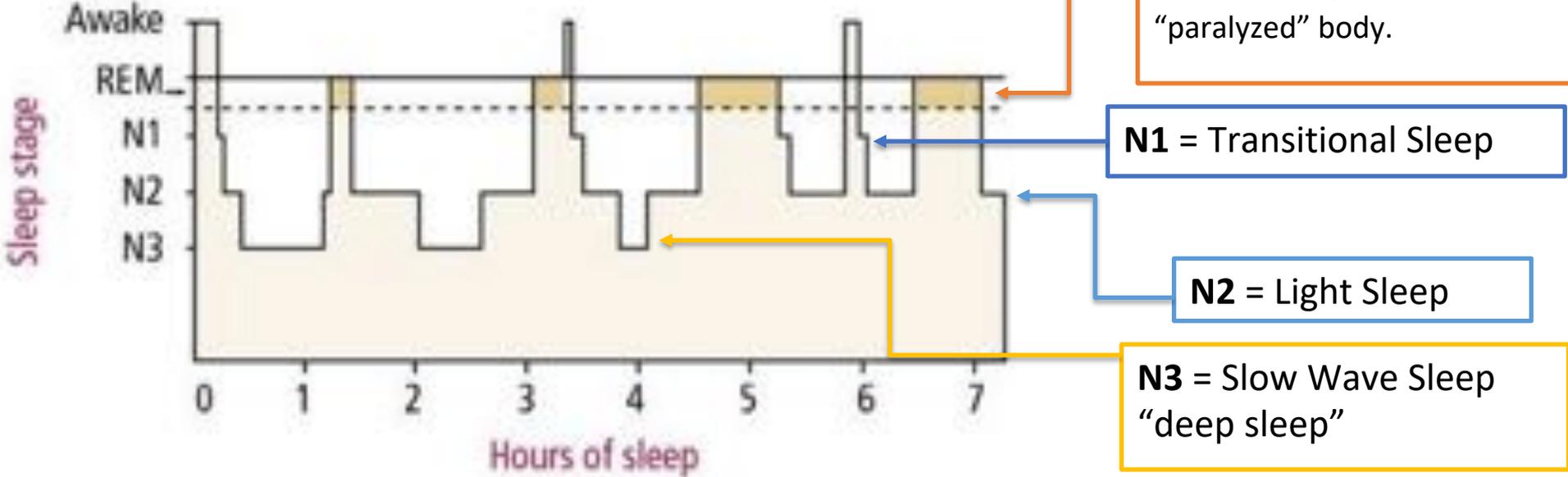


Image credit: <https://ericroth.org/portfolio/sleep-tight/>

More N3 sleep in the first part of the night
More REM sleep in the later part of the night.



Why Do We Sleep?

- **Sleep is thought to be restorative:**
Building muscle and bone, activating the immune system, and consolidating learning and memories.
- **Poor sleep causes problems:**
Poor learning, decreased immune function, low mood, anxiety, depression, overall poor health and lower quality of life.
- Studies also associate poor sleep with increase depression/suicide risk

What are Some examples of Pediatric Sleep Disordered Breathing?



Obstructive sleep apnea – upper airway obstruction with breathing effort



Central sleep apnea – pauses in breathing



Hypoventilation – retention of carbon dioxide



Hypoxemia – low oxygen level

Sleep Disordered Breathing Signs and Consequences

Symptoms:

- Snoring
- Restless sleep
- Night sweats
- Daytime sleepiness
- Teeth grinding
- Sleep walking
- Daytime irritability
- Attention issues
- Decreased growth

Consequences

- Impairments of neurocognitive development
- Low mood/anxiety/depression
- Poor school performance
- Cardiovascular morbidity
- Metabolic derangements (insulin resistance)
- Increased healthcare utilization



CMC Risk for SDB

- Children with Medical Complexity can be at increased risk for sleep disordered breathing due to:
 - Orofacial structure
 - Elevated BMI
 - Neuromuscular weakness or hypotonia
 - Spasticity
 - Decreased control of breathing (by brainstem)

Why do a Sleep Study?

Respiratory Indications Include:

- Concern for SDB
- Screening patients at risk for SDB
 - e.g. craniofacial, lung disease
- Peri-operative risk stratification before T&A
- Candidacy for tracheostomy decannulation

Non-Respiratory Indications Include:

- Concern for intrinsic disruptor of sleep causing parasomnias
- Concern for nocturnal seizure (though history may be more effective)
- Sleep related movement disorder
- Hypersomnia if narcolepsy suspected



Contents lists available at ScienceDirect

Paediatric Respiratory Reviews



Mini-Symposium: Sleep disordered breathing in children: indications, interpretation and implications

Pediatric polysomnography—A review of indications, technical aspects, and interpretation

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How to Prepare for the Sleep Study

- Preparation, anticipation, and shared decision making are important
- Consider sensory issues, developmental issues, or insomnia
- Showing parents/patients video to introduce Sleep study set up
- Make the process fun for the child
- Encourage transition items if used
- Consider desensitization interventions at home
 - *Practice with stickers, nasal cannula*



What are treatments for SBD

- Adenotonsillectomy
- Intranasal steroids/montelukast
- Orthodontia (e.g. palatal expanders)
- PAP therapy
- Weight loss (if appropriate)
- High Flow Nasal Cannula
- Hypoglossal nerve stimulator
- Medication?



Adenotonsillectomy

- Effective when indicated
- Watchful waiting is reasonable approach in patients with mild OSA, or high risk for surgery

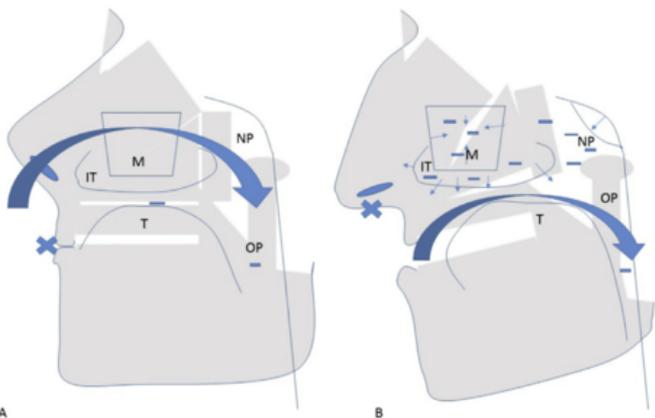


Review Article

Gravitational forces, negative pressure and facial structure in the genesis of airway dysfunction during sleep: a review of the paradigm

Howard D. Stupak*, Steven Y. Park

Albert Einstein College of Medicine, Department of Otorhinolaryngology, Bronx, NY, USA



Mouth-breathing can contribute to Rotational displacement of the maxillary palate and mandibular body relative to the mandibular ramus (gonial angle).

Additionally, nasopharyngeal negative pressure and subsequent intra-luminal tissue expansion can lead to worsened obstruction

Oral breathing can affect orofacial growth and increase risk of OSA

Sleep Breath
DOI 10.1007/s11325-015-1154-6

ORIGINAL ARTICLE

Mouth breathing, “nasal disuse,” and pediatric sleep-disordered breathing

Seo-Young Lee · Christian Guilleminault ·
Hsiao-Yean Chiu · Shannon S. Sullivan

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- Oral breathing, even after adenotonsillectomy is associated with continue OSA symptoms and findings on PSG.
- A retrospective study on non-overweight and non-syndromic prepubertal children with SDB treated by T&A with pre- and post-surgery clinical and polysomnographic (PSG) evaluations including systematic monitoring of mouth breathing
- Patients who had oral breathing treated with myofunctional therapy showed improved PSG findings.

Medication Management of OSA

Systematic Review/Meta-analysis

Montelukast and Nasal Corticosteroids to Treat Pediatric Obstructive Sleep Apnea: A Systematic Review and Meta-analysis

- Intranasal steroids and montelukast are modestly effective short-term treatment of mild OSA (reduction in AHI and increase in oxygenation)
- Further prospective research is needed for moderate and severe OSA

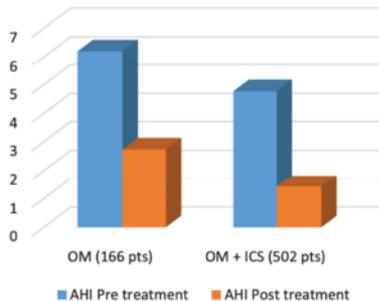


Figure 2. Effect of treatment on AHI. AHI, apnea-hypopnea index; ICS, intranasal corticosteroids; OM, oral montelukast.

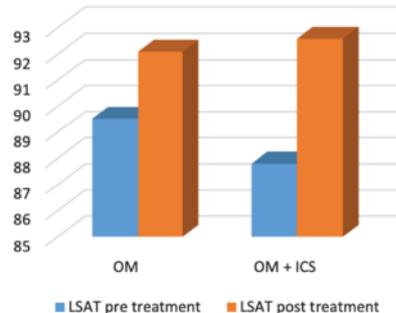


Figure 3. Effect of treatment on LSAT. ICS, intranasal corticosteroids; LSAT, lowest oxygen saturation; OM, oral montelukast; SaO₂, saturation of oxygen.

For Montelukast – The FDA requires Boxed Warning about serious mental health side effects (including suicidal thoughts/actions) and advises restricting use for allergic rhinitis

Systematic Review

Rapid Maxillary Expansion for Pediatric Obstructive Sleep Apnea: A Systematic Review and Meta-Analysis

Macario Camacho, MD; Edward T. Chang, MD, MS; Sungjin A. Song, MD; Jose Abdullatif, MD; Soroush Zaghi, MD; Paola Pirelli, DDS; Victor Certal, MD, PhD; Christian Guilleminault, MD



Image: Indian Journal of Orthodontics and Dentofacial Research, April-June 2016;2(2):50-55

- Rapid Maxillary Expansion an orthodontic procedure to expand the hard palate which is intended to reduce teeth crowding, expand nasal base to improve nasal breathing.

Review Conclusions:

- Improvement in AHI and lowest oxygen saturation (short term <3 years follow up)
- Need more data for >3 years follow up

MYOFUNCTIONAL THERAPY TO TREAT OSA: REVIEW AND META-ANALYSIS

Myofunctional Therapy to Treat Obstructive Sleep Apnea: A Systematic Review and Meta-analysis

Macario Camacho, MD¹; Victor Certal, MD²; Jose Abdullatif, MD³; Soroush Zaghi, MD⁴; Chad M. Ruoff, MD, RPSGT⁵; Robson Capasso, MD⁶; Clete A. Kushida, MD, PhD¹

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- Myofunctional therapy is a program of specific exercises that target the facial muscles used to speak, chew, swallow. Specifically, they often focus on improving tongue tone, strength and position.
- Studies suggests that it is effective
- ↓ AHI by 50% in adults, 62% in children
- ↓ sleepiness, ↓ snoring, improved oxygen saturations



AOMT

ACADEMY of OROFACIAL
MYOFUNCTIONAL THERAPY

What about PAP/CPAP?

Consider PAP if:

- Obesity
- Craniofacial abnormalities
- Residual OSA after surgery
- Family prefers non-surgical option
- No additional surgery recommended
- Primary pulmonary condition

Issues to Consider:

- Acclimation may take time
- **Be careful about mid face retrusion!!**
(hindered face growth associated with pressure from the mask on midface structures)

CPAP limitations...

- *Adherence, adherence, adherence...*
- Challenging for adults and children alike to tolerate.
- Increase adherence is associated with
 - non-Latinx white race
 - younger age
 - high baseline AHI
 - presence of neurodevelopmental disorders

Risk for poor adherence is increased:

- Children <6 yo, >15 yo
- Poor mask fit and inadequate pressure all contribute to treatment failure.

Deliberate age-appropriate desensitization protocols and close follow-up can improve adherence rates

Auto-titrating CPAP for the treatment of obstructive sleep apnea in children

- In-lab PSG with PAP titration is standard
- AutoCPAP considered to avoid delay in care, and to allow more time for desensitization at home.
- AutoCPAP has been studied to be an effective treatment in older pediatric patients, with careful/close follow up afterwards.

High flow nasal cannula treatment for obstructive sleep apnea in infants and young children

- Retrospective review of treatment with heated humidified high flow nasal cannula (HFNC) as treatment for OSA in young children intolerant of PAP, or at high risk for midface hypoplasia
- 22 children with OSA (AHI 4.8-89.2 events/hr)
- HFNC can serve as a bridge to surgery or spontaneous resolution of OSA

Update on Hypoglossal Nerve Stimulation in Children With Down Syndrome and Obstructive Sleep Apnea

- Case series of 20 children with Down Syndrome, ages 10-21 yo
- All participants had severe OSA (AHI >10, <50/hr) who failed CPAP therapy.
- Reduction of AHI by 85%
- Mean use 9.21hr/night. Improved quality of life
- **HGN still appears to be a reasonable option for patients with severe OSA, at risk for severe sequelae from untreated OSA, who are intolerant of PAP therapy**
- Further study continues



Novel pharmacotherapeutics for OSA

- Combination medication Atomoxetine (a norepinephrine reuptake inhibitor) and Oxybutynin (an antimuscarinic) *Taranto-Montemurro, et al 2020*
 - Increase genioglossus muscle responsiveness (upper airway dilator muscle)
 - Reduced OSA severity by 63%
- New Studies suggest Long term use (>1 year) is effective *Chen, et al 2021*
- Clinical Trial of Medication *ClinicalTrials.gov NCT04115878*
 - Children with Down Syndrome, ages 6-17 yo
 - Comparing high dose and low dose
 - Primary outcome: oAHI
 - Secondary outcomes: OSA QoL measures, arousal index, caregiver impression, sleep architecture

Taranto-Montemurro L, et al . Effects of the Combination of Atomoxetine and Oxybutynin on OSA Endotypic Traits. Chest. 2020 Jun;157(6):1626-1636. doi: 10.1016/j.chest.2020.01.012. Epub 2020 Jan 30.

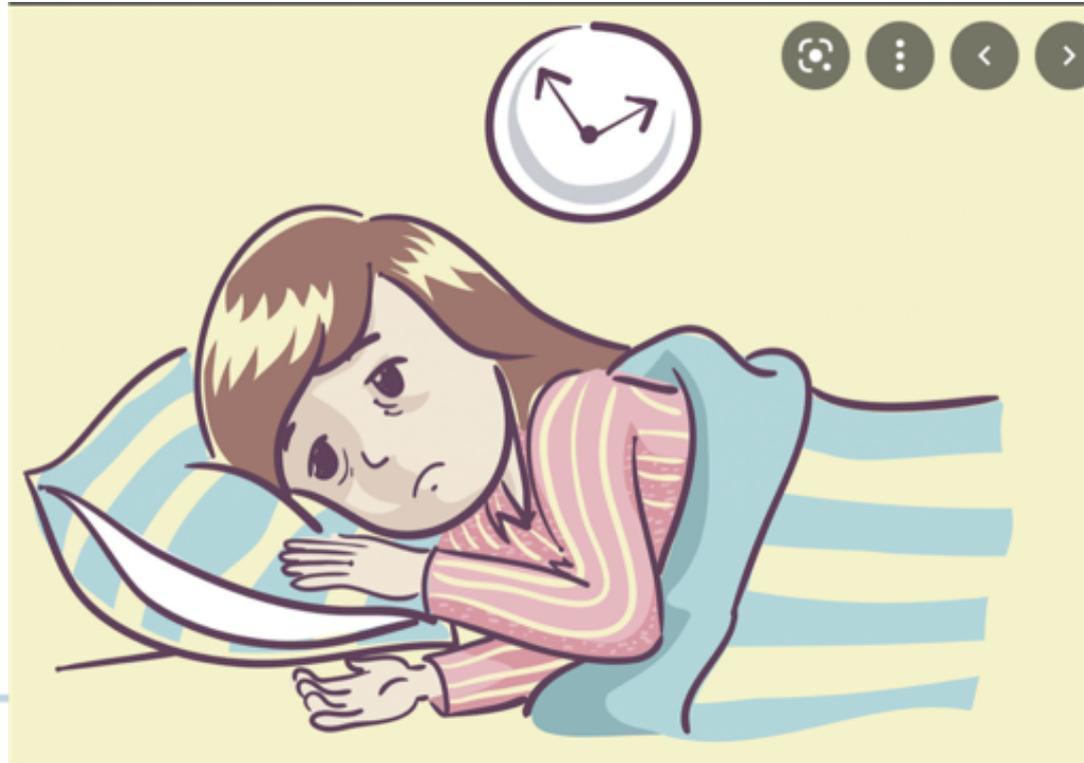
Chen, TY., et al. Long-term atomoxetine-oxybutynin combination use may be beneficial for the prevention of obstructive sleep apnea. Sci Rep 11, 12526 (2021). <https://doi.org/10.1038/s41598-021-91988-5>



Objectives for part 2

- Discuss insomnia often seen in children with medical complexity
- Summarize various pharmacologic and nonpharmacologic treatments for insomnia
- Convey the benefits of a multidisciplinary, comprehensive and individualized approach when considering sleep concerns

What is insomnia?



Insomnia



Chronic Insomnia Disorder

- Reports of difficulties falling asleep, staying asleep, and/or early waking
- Daytime consequences of sleep problem
- Adequate opportunity for sleeping
- Frequent ($\geq 3x/wk$) and chronic (≥ 3 mos)
- Not explained by another sleep-wake disorder, medical condition or mental health disorder



How common is pediatric insomnia?

- 30% of typically developing children meet criteria for a sleep disorder, with most meeting criteria for insomnia
- 85% or more of children with neurodevelopmental disabilities (NDD) meet criteria for a sleep disorder, mostly insomnia
- Probably similar prevalence in children with medical complexities
- Commonly last into adolescence and adulthood

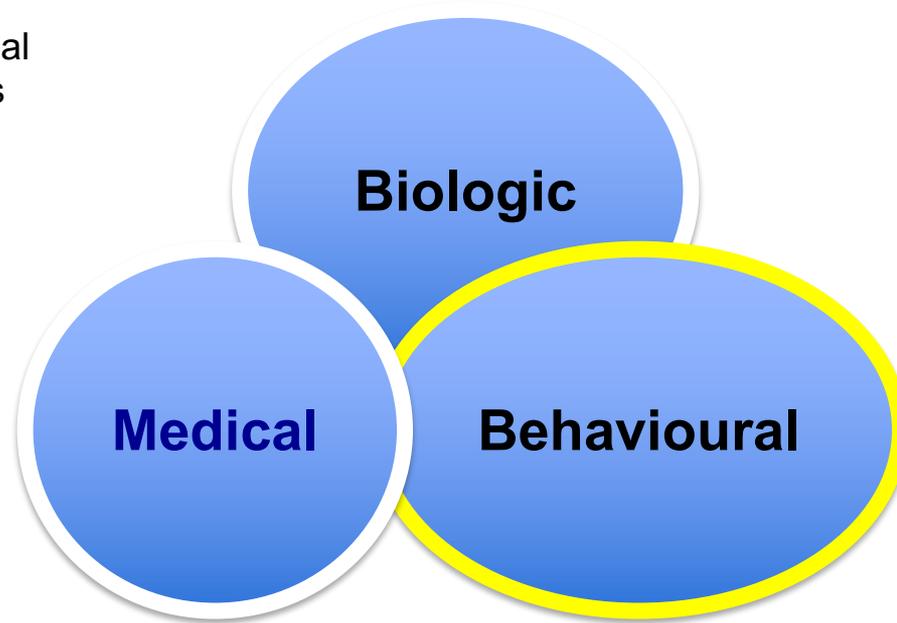
Robinson-Shelton A, Malow BA. Curr Psych Reports June (1) 2016

Tietze A et al. Sleep Medicine Reviews April 16(2) 2012

Insomnia in children with medical complexity

Neurochemical/Hormonal
Circadian Clock Factors
Sensory Differences
Mental Health issues
(e.g. anxiety)

Sleep Apnea
Vision/Hearing
Seizures
GERD
Spasticity
Medication



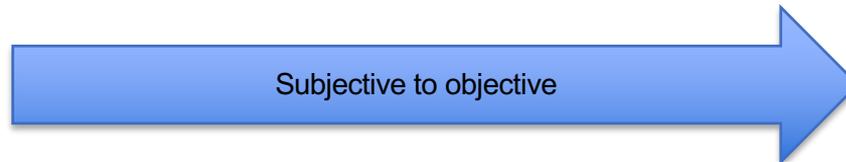
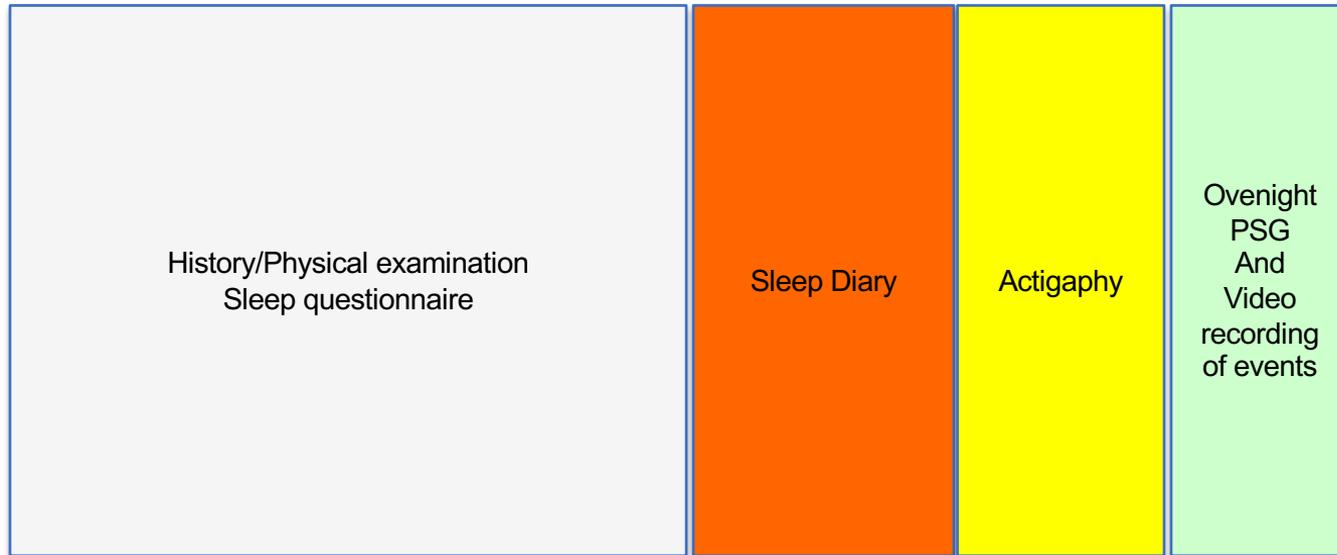
Inconsistent sleep practices
Lack of Exercise
Screen time
Parental concerns or stress

MULTIDISCIPLINARY APPROACH NEEDED

Examples of Behavioural Insomnia

Disorder	Age of occurrence	Features
Sleep-onset association	6 months to school age	Infants/children who require parental presence to initiate sleep and at times of night waking
Limit setting	Toddler to school age	Difficulty establishing limits, including (but not always limited to) bedtime routines
Sleep timing	School age to adolescent	Children/adolescents who prefer an early or later sleep onset and offset; in teenagers, this is typically the later type, called a delayed sleep phase preference

Evaluation of sleep



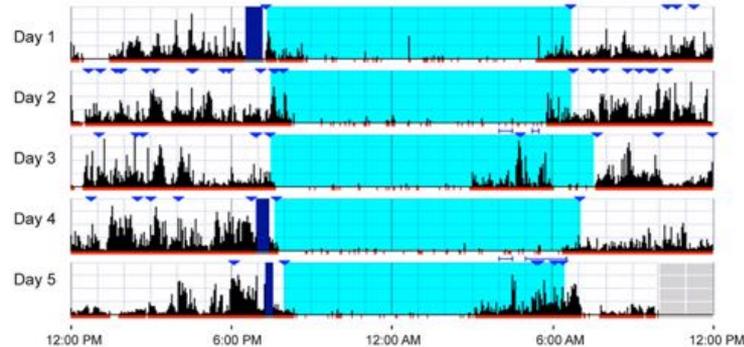
BEARS Sleep Screening Questionnaire

- “BEARS” instrument is divided into five major sleep domains
 - Each sleep domain has a set of age-appropriate “trigger questions” for use in the clinical interview.
-
- **B** = bedtime problems
 - **E** = excessive daytime sleepiness
 - **A** = awakenings during the night
 - **R** = regularity and duration of sleep
 - **S** = snoring



Actigraphy

- Measures sleep patterns and responses to treatment
- Requires parents to keep accurate sleep diaries as actigraph needs to be interpreted in context of when child went to bed



Polysomnography

- Useful to rule out obstructive or central sleep apnea, periodic limb movements
- May not get representative night of sleep
- Access differs in different countries and within same country to pediatric sleep labs
- Adult labs – may not use pediatric scoring rules





Treatment of Insomnia

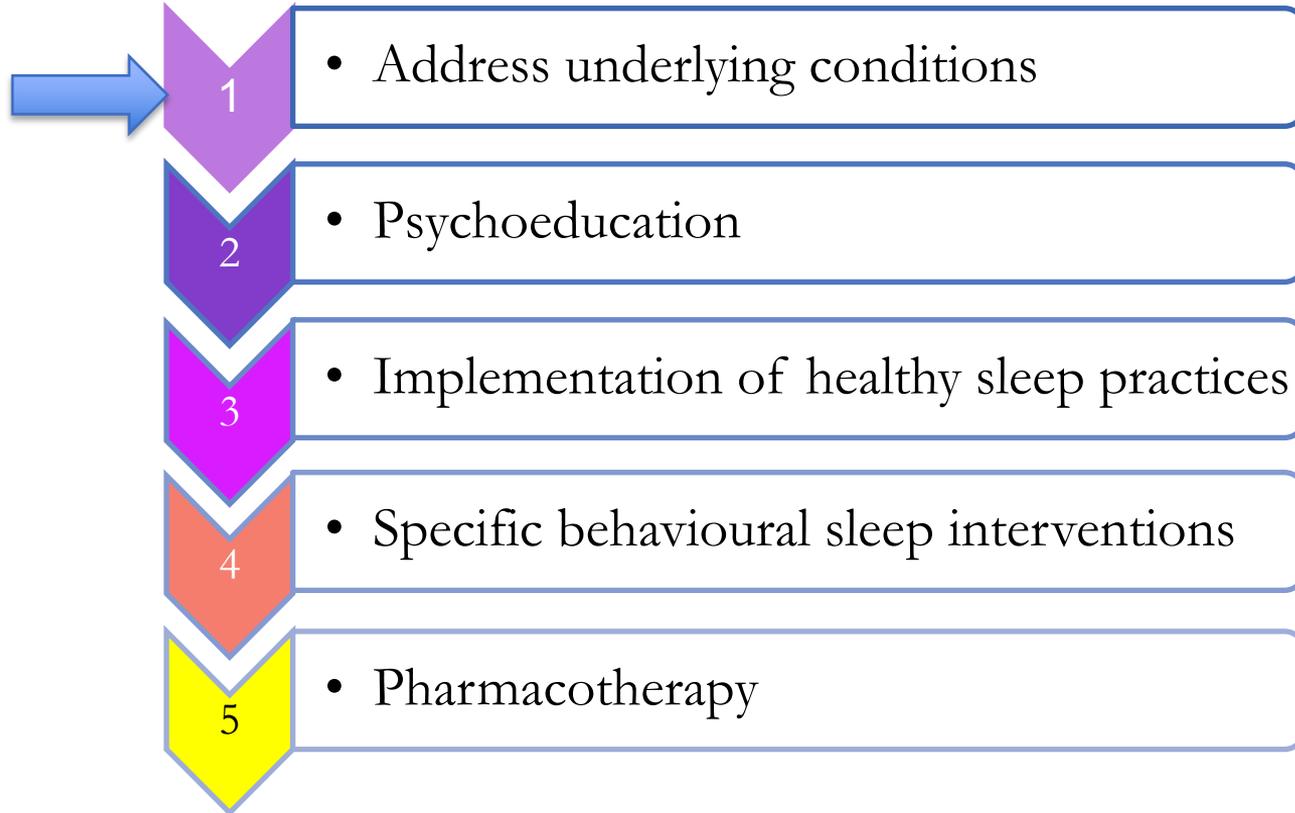
Good news

- We know what works to treat insomnia
- Robust evidence for behaviourally-based treatment for insomnia

Bad news

- Less than 1% of children receive evidence-based treatment
- Rather, most treated with medication, especially children with NDD
- Probably similar for children with medical complexity

Treatment- Best Practices



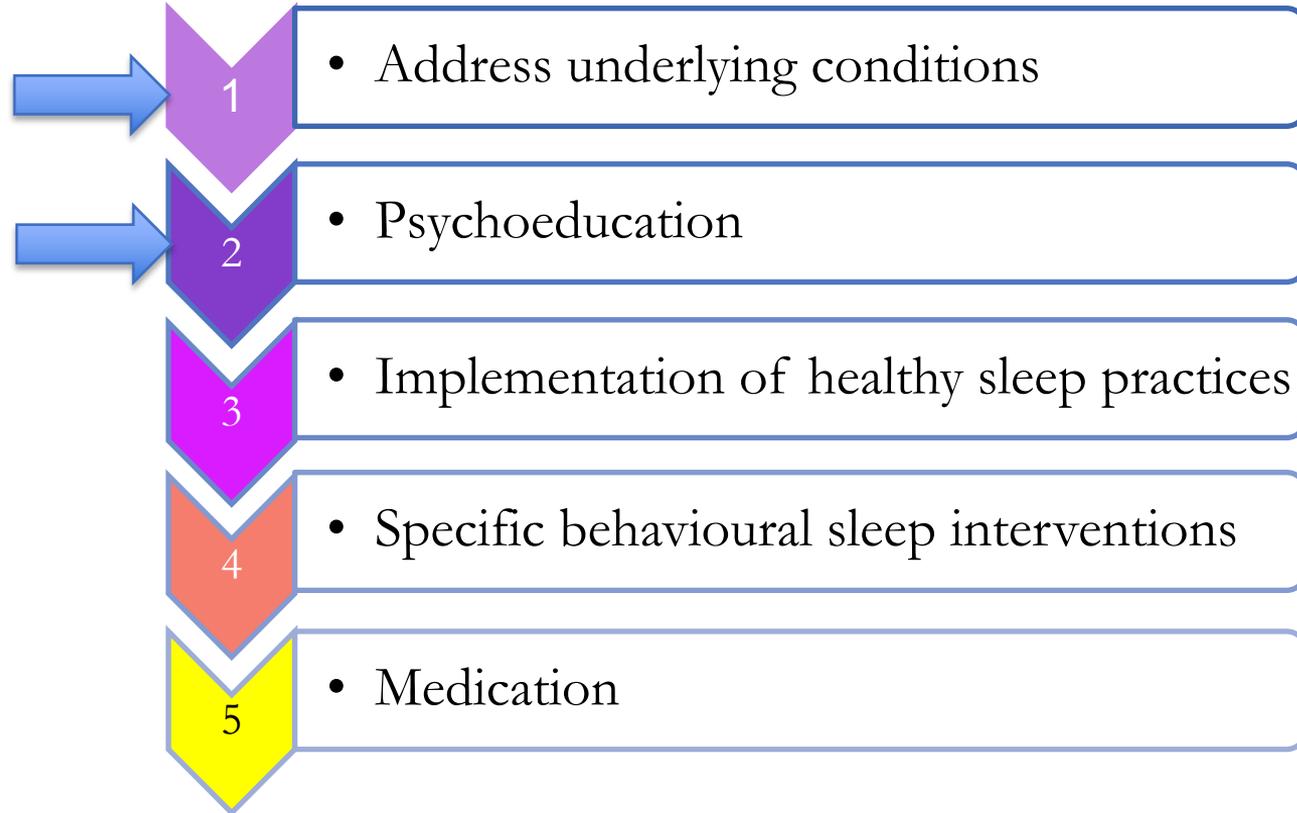


Addressing Other Concerns

Sleep issues will continue unless other concerns are addressed. Examples of other concerns include:

- ❖ Reflux
- ❖ Allergies
- ❖ Eczema
- ❖ Seizures
- ❖ Snoring/Apnea
- ❖ Enteral feedings (G-Tube at night)
- ❖ Pain
- ❖ Spasticity
- ❖ Anxiety

Best Practices: Treatment

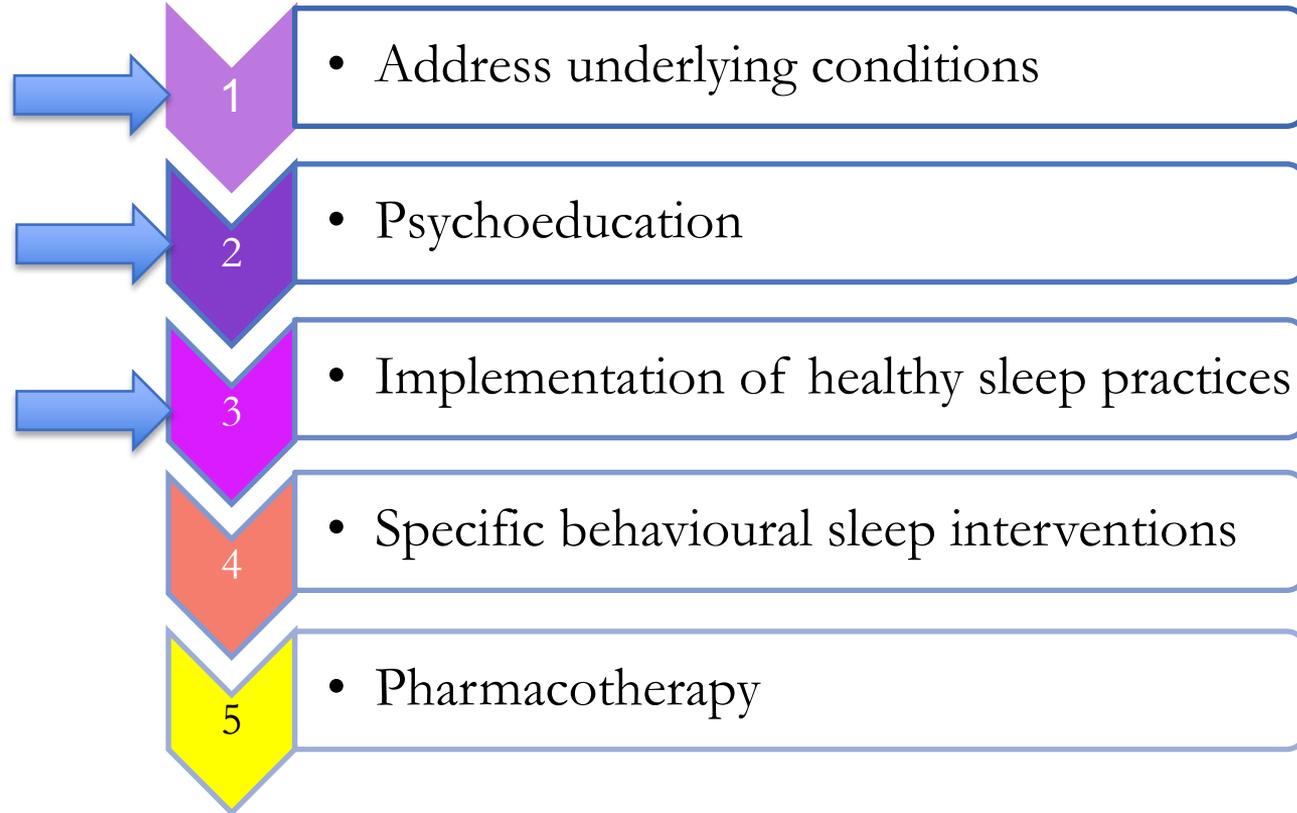


Psycho-education

- Sometimes sleep problems are a result of lack of knowledge and due to faulty beliefs, for example...
 1. Expecting child to have same chronotype as parents (e.g., owl/lark)
 2. Thinking that keeping the child up later will result in the child sleeping in later in the morning
 3. Thinking that arousals at night are not typical (rather than seeing this as self-soothing problem)
 4. Feeling that sleep problems in children with medical complexity are inevitable

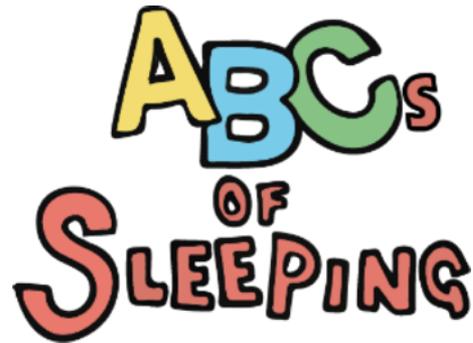


Best Practices: Treatment





Implementation of health sleep practices



Healthy Sleep Practices

Age-appropriate

Bedtimes, wake-times and naps, with

Consistency

Schedule and routines

Location

no Electronics in the bedroom or before bed

Exercise and diet

Positivity and relaxation

Independence when falling asleep

Needs met during the day

....all of the above equals Great sleep!

ABCs
OF
SLEEPING



Take electronics out of the bedroom

- Mobile devices pose unique threats to sleep
- Portability and versatility- hard to keep out of bedtime routines



Healthy sleep practices for children with Autism

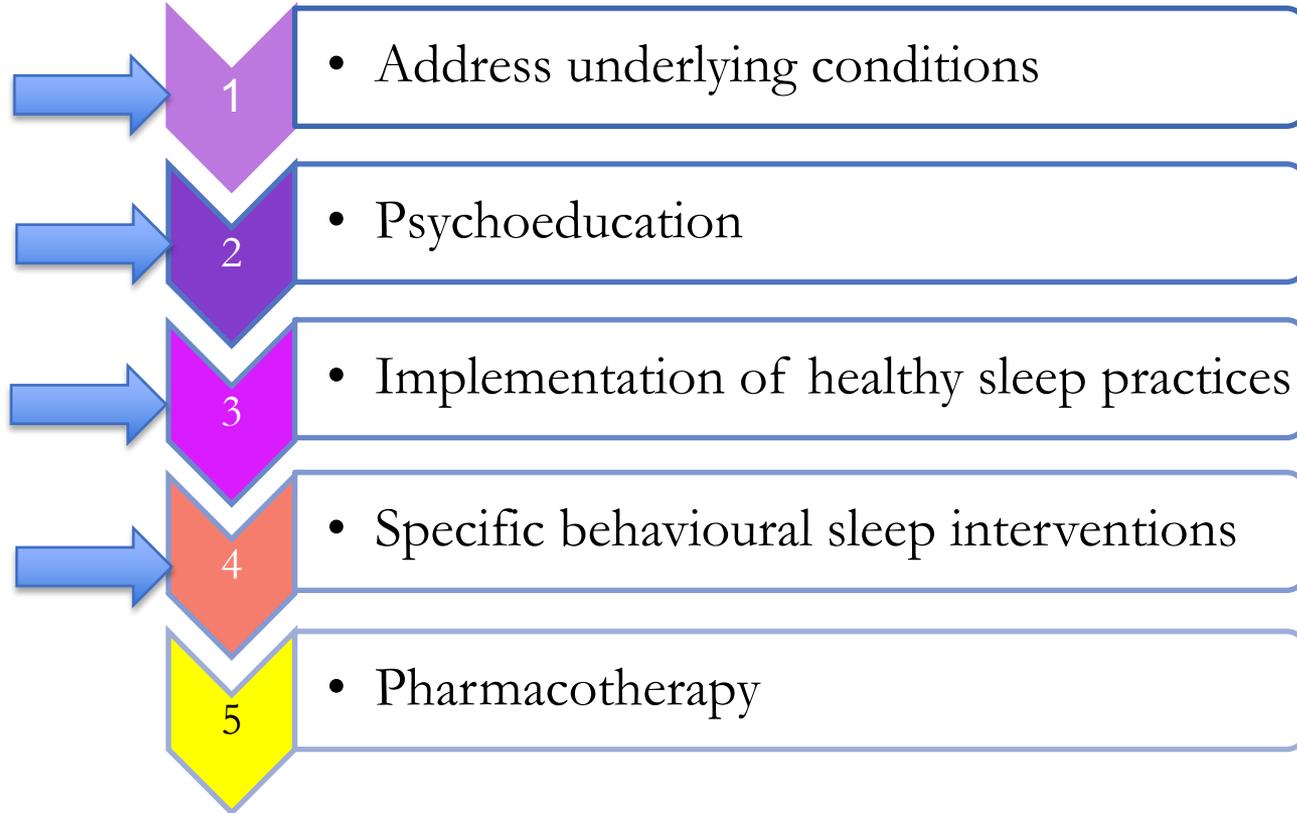
Sleep Strategies for Children with Autism



A Parent's Guide



Best Practices



Specific Sleep Strategies

- Goal typically involves some combination of developing positive sleep-related associations, establishing routines, and implementing relaxation/self-soothing skills
- Strong evidence for sleep intervention programs
 - 94% of studies found behavioural interventions to be effective
 - 80% of children had clinically significant improvements
 - Improvements in sleep onset latency, frequency and duration of night wakings and sleep efficiency (not sleep duration)
 - Improvements lasted 3 to 6 months

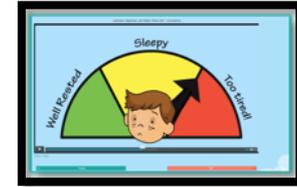
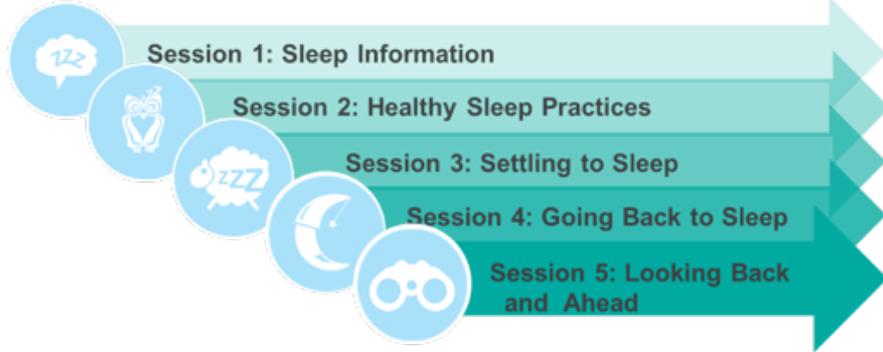
Behavioural interventions



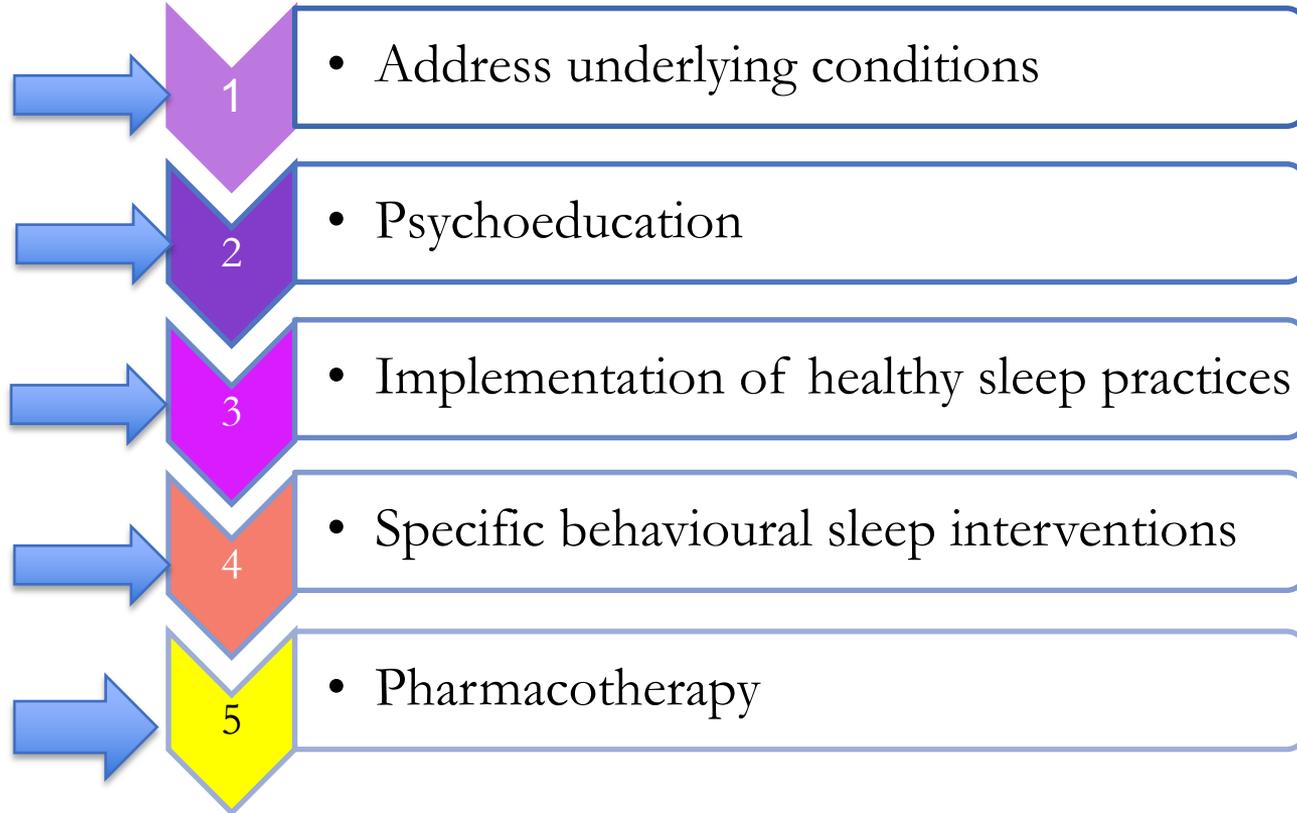
Vriend, J. & Corkum, P. Clinical management of behavioral insomnia of childhood. *Psychology Research and Behavior Management*. 4, 2011.

Corkum, P., Davidson, F., Tan-MacNeill, K., & Weiss, S. Sleep in Children with Neurodevelopmental Disorders: A focus on insomnia in children with ADHD and ASD. *Sleep Medicine Clinics*. 9(2) 2014

Better Nights, Better Days- example of an eHEALTH intervention for insomnia



Best Practices



Role of pharmacotherapy





General principles

- Best used after behavioral treatments have been tried unsuccessfully, and in combination with behavioral therapies
- Potential to use at beginning of treatment when a family is not able to implement behavioral strategies
- Use medications sparingly– to facilitate behavioral strategies rather than substitute for them
- Whenever possible, choose a medication that will treat a co-occurring condition such as epilepsy, anxiety, or a mood disorder
- Start at low doses, to avoid excess sedation and adverse effects

Which treatment?

- Gabapentin
- Alpha agonists (clonidine, guanfacine)
- Clonazepam
- Antidepressants
- Ramelteon
- Antihistamines
- Chloral Hydrate
- Z drugs





Melatonin

- Typically developing children with insomnia should respond to behavioral strategies and do not need supplemental melatonin
- For children with medical complexity, melatonin may be used as part of the stepped care approach once:
 - Underlying physical/mental health issues are addressed
 - Education, healthy sleep practices and behavioral strategies are implemented



Future directions

- Research is needed comparing medications in combination with behavioural strategies, including eHealth – e.g. Better Nights Better Days for treating insomnia
- eHealth plus any of following (melatonin, Slenyto -Pediatric prolonged release melatonin, CBD)
- Strategies to prevent/improve airway resistance
- Improved exploration into desensitization protocols for PAP
- More alternative therapies to PAP for the sensory sensitive

Prolonged release preparations improve sleep duration, sleep latency, and night wakings
(Gringras, *Am Acad Child Adol Psych* 2017; Maras, *J Child Ad. Psychopharm*, 2018)



Thank you for your attention!

Questions?