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SCIENTIFIC INVESTIGATIONS

Current Practice Patterns in the Diagnosis and Management of Sleep-Disordered Breathing in Infants

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Study Objectives: Currently, there are no universally accepted guidelines for diagnosis and management of sleep-disordered breathing (SDB) in infants. The purpose of this study was to survey pediatric sleep medicine providers regarding their current practice patterns for diagnosis and management of SDB in infants.

Methods: An anonymous, web-based survey with 71 questions was distributed via the PEDSLEEP and Ped-Lung listserv, which serve as a hub of communication for pediatric sleep and pulmonary medicine providers worldwide.

Results: Fifty-four providers from eight countries completed the survey. Ninety-six percent of providers reported performing sleep studies in infants with 53% performing more than 30 studies per year. There was no consensus on the definition of obstructive sleep apnea (OSA) in infants when using an obstructive apnea-hypopnea index (AHI) cutoff: AHI > 1 (30%), AHI > 2 (35%), AHI > 5 (24%), AHI > 10 (2%) and other (9.3%). Thirty-six percent did not use pediatric criteria to define severity of OSA in infants. Opinions regarding management of five typical SDB cases were solicited and the results varied among respondents. Most of the providers (89%) thought that more research is needed to gather normative sleep data in infants and that their practice would benefit from evidence-based guidelines for diagnosis and management of SDB in infants (98%).

Conclusions: These results demonstrate substantial variability in practice patterns for diagnosis and management of SDB in infants. Further research and consensus guidelines are needed to ensure optimal care for infants with SDB.

Keywords: infants, sleep apnea, sleep-disordered breathing, survey

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BRIEF SUMMARY

Current Knowledge/Study Rationale: Sleep-disordered breathing in infants has a unique pathophysiology, and universally acceptable diagnostic and management algorithms are lacking. This study assessed current practice patterns for diagnosing and managing sleep-disordered breathing in infants.

Study Impact: Pediatric sleep providers exhibit substantial variability in their approach to diagnosis and management of sleep-disordered breathing in infants. Our results highlight the importance of and need for developing practice parameters and guidelines to provide better care for these infants.

INTRODUCTION

The prevalence of obstructive sleep apnea (OSA) in infants is unknown because of scarcity of research in this population. In children, untreated OSA may result in suboptimal neurodevelopmental outcomes and cognitive dysfunction,^{1–3} deleterious cardiovascular sequelae,⁴ behavioral and academic challenges,⁵ and growth impairment.⁶ Although timely diagnosis and treatment of OSA is imperative to prevent neurocognitive impairment, lack of research has significantly hindered care for these infants. Guidelines regarding the diagnosis and management of sleep apnea in children exist,⁷ but are not applicable to children younger than 12 months.

Currently, there are no universally accepted criteria used for diagnosing sleep apnea in infants, no consensus guidance on what the management options are, or what the common practice patterns are among sleep providers. Therefore, the purpose of the current study was to survey sleep medicine providers regarding their current practice patterns for evaluating infants

with sleep-disordered breathing (SDB), diagnosing sleep apnea in these infants, and treating their disease.

METHODS

Participants

An invitation to participate in this research study was distributed with the PEDSLEEP and Ped-Lung listserv, which are listservs dedicated specifically to pediatric pulmonary and sleep medicine providers. The survey was housed in a REDCap database. There were no questions regarding individual patients, and the survey itself was anonymous, so individual providers would not be identified. This study was approved by the Institutional Review Board at Children's Mercy-Kansas City Hospital.

Survey

The questionnaire included questions regarding respondent background and clinical practice including specialty board certification,

years in practice, practice setting and location (state if United States, or country), percentage of practice dedicated to infants, and number of infant sleep studies performed per year. Physicians were asked to rate the order of importance of various factors in the evaluation and management of infants with SDB. Specific questions were asked regarding physician's thresholds for polysomnography (PSG) diagnosis of OSA and central sleep apnea (CSA), use of supplemental oxygen for management of OSA in neonates and infants, and the use of continuous positive airway pressure (CPAP) in this population. Respondents were asked to answer five case-based questions to determine the influence and use of various diagnostic and clinical parameters in clinical decisions pertaining to infant obstructive and CSA (supplemental material).

Data Analysis

Results for each question were separately compiled for all respondents, and summary statistics were generated using Microsoft Excel. The following definitions were determined *a priori* to compile the results: consensus ($\geq 70\%$ agreement among respondents), unanimous (100% agreement), majority ($\geq 50\%$ agreement), and no consensus ($< 50\%$ agreement).

RESULTS

Physician Background

Fifty-four pediatric sleep providers completed the survey. Beyond sleep medicine, providers were board certified/eligible in pediatrics (81%), neurology (5%), internal medicine (2%), pulmonology (65%), and family practice (3%). Most providers were in a primarily academic practice setting (85%), although 13% were in a private group and 2% in a solo practice. Providers had been practicing sleep medicine for less than 5 years (26%), 5 to 9 years (26%), 10 to 15 years (22%), or more than 15 years (26%). The average number of infants with suspected SDB seen each month by providers were 0 to 5 (54%), 5 to 9 (28%), 10 to 15 (13%), or more than 15 (5%). Infants, including neonates, comprised zero to 5% of the patient population seen by most of the providers (67%). Eighty percent of providers stated that they performed PSGs on infants younger than 2 months. Ninety-six percent of providers affirmed that PSG was performed on infants 2 to 12 months of age, whereas 80% performed PSG on neonates. Fifty-four percent of providers stated that PSG was performed more than 30 times per year in infants. Respondents were from 24 different states within the US (45 providers) and 7 other countries [(United Kingdom (2 providers), Australia (1 provider), Ireland (1 provider), South Africa (1 provider), Portugal (1 provider), Canada (2 providers), and Israel (1 provider)]. Patterns correlating with country of practice were not observed.

Practice Patterns

Diagnosis of Suspected Sleep Apnea in an Otherwise Healthy Neonate

Case A: A 3-week-old boy, born at term, is hospitalized with pauses in breathing at night since birth. He is otherwise healthy. There is no noisy breathing reported. Pulse oximetry at the bedside

reveals normal oxygen saturations awake and asleep. What is the next best step?

There was no agreement or consensus among respondents regarding the next best step toward diagnosis. Interestingly, the two most commonly endorsed responses were in direct contrast with each other (35% opting for a sleep study versus 42% opting against a sleep study). "Other" diagnostic choices (open response) chosen by 11% of respondents included pneumography, overnight oxygen desaturation study, analysis of capillary blood gas, evaluation for gastroesophageal reflux, and laryngoscopy.

Diagnosis of Suspected Sleep Apnea in a Neonate With a Craniofacial Syndrome

Case B: A 3-week-old boy, born at term, is hospitalized with pauses in breathing at night since birth. He has Pierre Robin sequence. There is no noisy breathing reported; he is growing well. Pulse oximetry at the bedside reveals normal oxygen saturations awake and asleep. What is the next best step?

There was consensus regarding obtaining a full polysomnography for assessment of SDB. Fourteen percent of respondents chose the next most common step "no sleep study is needed." "Other" diagnostic choices (open response) selected by 5% of participants included "depending on what is considered normal," pneumography, and nocturnal desaturation study.

Diagnosis of Suspected Sleep Apnea in an Infant With a Laryngomalacia

Case C: A 6-month-old, previously healthy girl presents with noisy breathing while awake and asleep. Her otolaryngologist has made the diagnosis of laryngomalacia. She is growing well. What is the next best step?

There was no agreement or consensus among respondents regarding the next best step toward diagnosis. Respondents were tied regarding the need for a sleep study: 43% responded that a sleep study is needed to assess for sleep disordered breathing versus 44% responded that no sleep study is needed. Seven percent opted for a cardiorespiratory study, 2% for an overnight oximetry, and 4% chose "other" (laryngoscopy, bronchoscopy, chest computed tomography).

Management of CSA in an Otherwise Healthy Infant

Case D: A 3-month-old, previously healthy boy undergoes overnight PSG for apneic episodes with sleep. The central apnea index is 6 events/h, total AHI = 6.3 events/h, AHIo = 0.3 events/h, No periodic breathing, oxygen saturation nadir 92%, total sleep time $< 90\% = 0$. Average oxygen saturations remain normal overnight and there is no hypoventilation. What is your next step?

There was no agreement or consensus among respondents regarding the next best step in management. Sixty-three percent chose "watchful waiting" while 22% chose "further workup for CSA." "Other" management choices (open response) chosen by 14% respondents included normal study, infant monitor, and need more information.

Management of OSA in an Otherwise Healthy Infant

Case E: A 2-month-old previously healthy girl undergoes overnight PSG for symptoms of SDB. The only significant finding reported is a total AHIo of 46 events per hour with saturation nadir at

92%. Average oxygen saturations remain normal overnight and there is no hypoventilation. What is the next best step?

There was near-unanimous agreement (92%) regarding need for further work-up for OSA in this infant.

Follow-up question: How would you manage this patient?

There was no agreement or consensus among respondents regarding the next best step in management. Responses were as follows: treatment with supplemental oxygen (18%), treatment with CPAP (16%), treatment with tracheostomy (none), treatment with other surgery (16%), no treatment because oxygenation and ventilation are normal (15%), and “other” (33%, open response) which included upper airway evaluation, ear, nose, and throat surgery, and home monitor.

Diagnosis of OSA in Infants

There was no agreement or consensus among respondents regarding optimal cutoff value for AHIO to diagnose OSA in older infants (2 to 12 months): AHIO > 1 (30%), AHIO > 2 (35%), AHIO > 5 (24%), AHIO > 10 (2%), AHIO > 15 (0%), AHIO > 20 (0%), and other (9%, open response) which included cutoffs of 1.5, 3, and 100. Likewise, there was no agreement or consensus among respondents regarding optimal cutoff value for AHIO to diagnose OSA in young infants (0 to 2 months) and neonates (Figure 1). Although there was consensus among providers regarding the use of pediatric criteria (mild: AHIO >1 to < 5, moderate: AHIO > 5 to < 10 and severe AHIO > 10) to determine the severity of OSA in older infants, 36% of providers stated they did not use these criteria in young infants (0 to 2 months).

When asked regarding indications for PSG in infants, responses were as follows: regardless of symptoms (6%), if there are symptoms of SDB (snoring, noisy breathing, increased work of breathing) awake and asleep (37%), if there are symptoms of SDB (snoring, noisy breathing, increased work of breathing) asleep only (57%), if there are symptoms of SDB (snoring, noisy breathing, increased work of breathing) awake only (0%).

Regarding the most important PSG variable(s) in determining clinical management, 50% of providers stated that abnormal oxygenation or ventilation during a PSG are more important factors in

determining management than the absolute AHI value, whereas 37% thought that an abnormal AHI warrants further management regardless of severity of hypoxemia or hypoventilation. Eleven percent stated that the severity of hypoxemia is more important than the absolute AHI in determining management options and only 2% stated that an abnormal AHI without significant hypoxemia (oxygen saturation nadir of ≥ 90%) does not need to be treated.

Management of OSA in Infants

Providers were asked whether they have prescribed supplemental oxygen or CPAP therapy for management of OSA in infants. There was consensus among providers regarding the use of low-flow supplemental oxygen via nasal cannula for the treatment of OSA in infants of all ages. Although 90% of providers stated they had prescribed CPAP for OSA management in infants between 2 to 12 months of age, the group was divided regarding their use of CPAP for younger infants (0 to 2 months, 53% said “yes”, 47% said “no”).

Diagnosis of Central Sleep Apnea in Infants

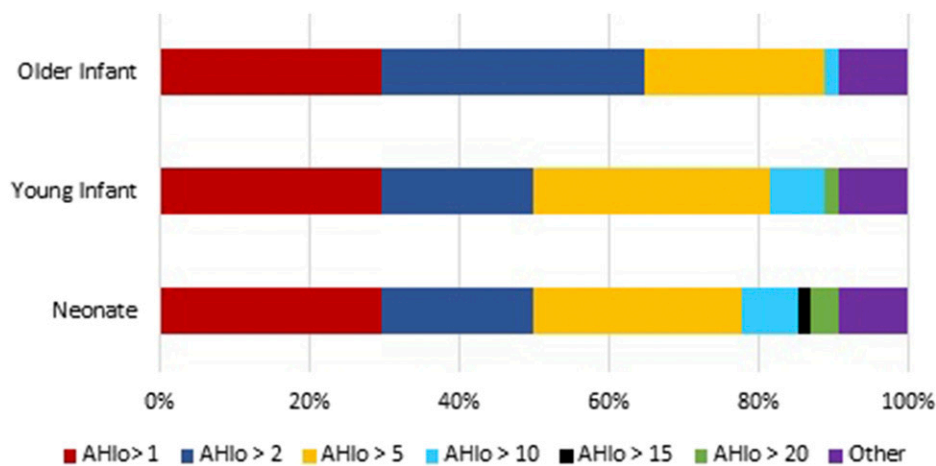
There was consensus among providers regarding using a central apnea index cutoff greater than 5 to diagnose CSA in older infants (74% of responders). The cutoff for young infants (0 to 2 months of age), was less clear: 50% choosing a cutoff of central apnea index > 5 and 25% using a cutoff of central apnea index > 10 (Figure 2). Sixty-two percent stated they use a different cutoff for diagnosing CSA in a neonate compared to older infants.

Management of CSA in Infants

When asked to rank in order of importance the factors considered when determining management of CSA in infants of all ages, there was no consensus among providers for the following: degree of oxygen desaturation associated with events, severity of CSA based on the central apnea index, length of central apneic events, and percentage of total sleep time with oxygen saturations below 90%.

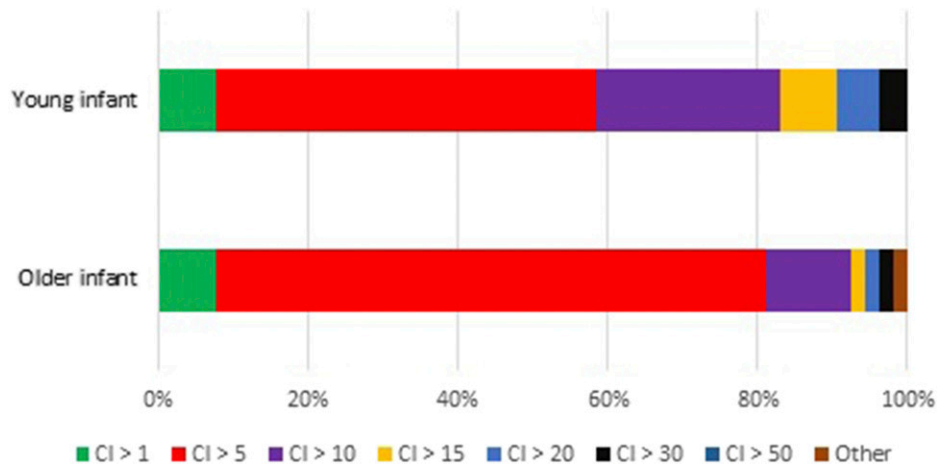
There was consensus among providers regarding the need for normative sleep data in infants (89% providers) and the group was near unanimous regarding the need for evidence-based

Figure 1—AHIO cutoff for OSA diagnosis.



AHIO = obstructive apnea-hypopnea index, OSA = obstructive sleep apnea.

Figure 2—CI cutoff for CSA diagnosis.



CI = central apnea index, CSA = central sleep apnea.

guidelines on diagnosis and management of infant OSA and CSA (98% providers).

DISCUSSION

Herein we have shown that practice patterns for diagnosis and management of SDB in infants differ among sleep providers both within the United States and abroad. There is little agreement among surveyed providers regarding diagnostic criteria and management for both OSA and CSA, especially in young infants. Key questions and results have been provided in **Table 1**.

The etiology of OSA in infants is multifactorial and compared to older children, the diagnosis and treatment is often more complex.⁸ In-laboratory overnight PSG is the gold standard for diagnosis of OSA. Although guidance exists for PSG criteria for OSA and CSA diagnosis in older children, there are no guidelines for infants. Barriers to development of guidelines are that normative sleep data are limited and the definition of an obstructive respiratory event using PSG (particularly hypopneas) has not been established in infants. Reflecting this, there was no consensus among providers in our survey regarding the optimum AHIo cutoff for diagnosis of OSA in infants even though standard pediatric criteria exist, suggesting that providers are practicing using different cutoffs. Moreover, for infants younger than 2 months, there was no consensus regarding cutoffs used for differentiating mild, moderate, or severe OSA. For infants with CSA, providers agreed that pediatric criteria (central index > 5) can be applied to infants, but the majority stated that they would not use this criteria in neonates. Therefore, despite caring for a large proportion of infants in their clinical practice, there appears to be significant variability in both diagnostic and management strategies among surveyed sleep providers within the United States and abroad. Naturally, this can have implications for clinical care and affect management of these infants.

Pediatric guidelines recommend a PSG whenever there are symptoms suggestive of SDB, particularly in those with risk factors

Table 1—Summary of key questions and responses highlighting gaps in currently available evidence.

Key Questions	Agreement Among Respondents
Is a PSG indicated to screen otherwise healthy neonate with pauses in breathing at night and normal bedside pulse oximetry?	No agreement, no consensus.
Is a PSG indicated to screen a neonate with Pierre Robin sequence with pauses in breathing at night and normal bedside pulse oximetry?	Consensus, full PSG is needed.
Is a PSG indicated to screen an infant with laryngomalacia and noisy breathing when asleep?	No agreement, no consensus.
Is further evaluation needed in an infant with severe OSA without significant hypoxemia (normal oxygen saturations overnight)?	Near unanimous, further work-up is needed.
Can pediatric criteria be used to determine severity of OSA in infants?	Consensus, yes it can be used.
Is there a need for normative sleep data in infants?	Consensus, yes
Is there a need for evidence based guidelines on diagnosis and management of infant OSA?	Near unanimous, yes

Consensus was defined as ≥ 70% agreement among respondents. No agreement indicates < 50% agreement. OSA = obstructive sleep apnea, PSG = polysomnography.

for OSA (craniofacial anomalies, prematurity, obesity, neurologic disorders, and genetic conditions).⁹ Although providers agreed upon performing PSG in an infant with SDB and a craniofacial syndrome, there was no consensus on whether PSG is indicated for otherwise healthy infants with SDB or those with laryngomalacia. Given that these infants comprise a major percentage of SDB in infants seen at pediatric sleep centers, formal guidance is needed to determine management in this cohort.

Five clinical cases, representative of common scenarios encountered in clinical practice, were selected to determine agreements in clinical care. There was no agreement on management for either CSA or OSA in infants. This suggests a gap in currently available literature, reflects the need for evidence or consensus-based guidelines, and has implications for future research in the field.

Limitations

First, a major limitation of this study is the relatively small sample size of participants. That said, 54 providers likely represents a significant proportion of pediatric sleep providers in the U.S and abroad. The survey was sent via the PEDSLEEP and Ped-Lung mailing lists, which include sleep physicians from across the country and internationally. There are about 459 members of the pediatric sleep listserv group and over 500 members of the Ped-Lung email group. Both of these email groups include a large cohort of physicians of multiple subspecialties, nurses, allied health professionals, and researchers with an interest in pulmonary and/or sleep medicine. Identifiable information (such as degree or specialty of practice) is not stored and therefore it is difficult to ascertain the true response rate without knowing the exact number of pediatric sleep providers in the group (because this survey was addressed to physicians only). Based on the information provided, if we extrapolate that there are 500 physicians currently practicing sleep medicine worldwide, the response rate is about 10%. We believe the true response rate may be higher. Moreover, given the small number of participants from countries other than the United States, we were unable to determine whether there are regional variations of practice.

CONCLUSIONS

In conclusion, these results demonstrate substantial variability in current practice patterns for diagnosing and managing SDB in infants. Not only is there a critical need for more research in this area, but a need to develop consensus-based recommendations to better guide sleep physicians in providing standardized care to their infant patients. Further research and practice parameters are needed.

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DISCLOSURE STATEMENT

All authors have seen and approved the final manuscript. The authors report no conflicts of interest.