EDITORIALS

Check for updates

Is It Time to Head Home for the Night? Home Sleep Testing in Young Children

8 Kristie R. Ross, M.D., M.S.¹, and Susan Redline, M.D., M.P.H.²*

¹UH Rainbow Babies and Children's Hospital, Case Western Reserve University, Cleveland, Ohio; and ²Harvard Medical School, Brigham and Women's Hospital, Boston, Massachusetts



The gold standard for the diagnosis of obstructive sleep apnea (OSA) in children is in-laboratory polysomnography (PSG), and the American Academy of Pediatrics recommends that polysomnography be performed in children with snoring and symptoms or signs of OSA (1). The field's focus on this tool for evaluating OSA (as well as evaluating other sleep disorders) reflects the ability of PSG to comprehensively collect physiological data on respiration, sleep, heart rate, and leg movements in controlled settings where issues of sensor loss can be readily addressed. However, there are key limitations to use and interpretation of such data from single-night laboratory assessments. These include the cost and

burden of testing and representativeness of data collected in the laboratory compared with what is typically experienced at home. When ordering an attended in-laboratory PSG, the response from caregivers is rarely enthusiasm. Instead, parents are concerned about the ability of the child to sleep in a strange place, the logistics of finding childcare for other children who cannot accompany the patient, and out-of-pocket expenses related to testing. In the era of coronavirus disease (COVID-19), concerns about bringing children into a sleep laboratory environment now include not just comfort but also safety. Although firstnight effects and night-to-night variability in OSA severity estimates are wellrecognized limitations to single-night in-laboratory PSG (2, 3), additional concerns are the early termination of studies by technicians working fixed shifts (that end before the child awakens), losing data for the last REM period when OSA may be most severe.

Although home-based sleep apnea testing is widely used in adults to diagnose OSA, its use in children has been much more limited, reflecting concerns about the safety and feasibility of collecting multiple respiratory signals in this population. In this issue of AnnalsATS, Vézina and colleagues (pp. 1238-1246) present data on use of home sleep cardiorespiratory monitoring in a large sample (n = 562) of very young children using a novel pilot sleep scoring algorithm (4). Notably, with technicians setting equipment up and activating the study in the children's homes and providing families with audio and video equipment to self-monitor potential safety issues (tangled and/or misplaced sensors), 91% of studies were deemed acceptable and no safety issues were reported. Importantly, the average duration of sleep monitored was 573 minutes, a period

likely to provide representative sleep data for young children, and substantially longer than total sleep time reported from attended PSGs (5, 6). Although these data support the feasibility of home sleep studies in young children, there are important questions regarding the extent to which this protocol can generalize to other samples (older children, diverse households), is broadly acceptable to families, or can be simplified such that caregivers can be trained to set up equipment.

The primary objective of their study was to provide normative data in a historically understudied population-a worthwhile endeavor that provides data critical to age-specific PSG interpretation. The data reported in this sample on distributions of such parameters as the obstructive and central apnea index, oxygen desaturation index, apnea-hypopnea index, and average oxygen saturation levels are needed for understanding the variation of these parameters in a community sample of children. Notably, the frequency of central apneas exceeded that of obstructive apneas, supporting the relatively high frequency of central events in young children. The mean oxygen desaturation index exceeded 5 (a level generally considered abnormal) and no association was seen between objective parameters and elevated scores on the Pediatric Sleep Ouestionnaire subscore, underscoring the discordance between objective and caregiver-reported information. Further research testing the ability of statistically defined thresholds for sleep-disordered breathing (SDB) metricsand their association with subjective reports-to predict clinical morbidity is needed to further inform the utility of sleep studies for clinical decision-making.

A unique aspect of the study by Vézina was the use of a scoring algorithm for

OThis article is open access and distributed under the terms of the Creative Commons Attribution Non-Commercial No Derivatives License 4.0 (http://creativecommons.org/licenses/by-nc-nd/ 4.0/). For commercial usage and reprints, please contact Diane Gern (dgern@thoracic.org).

^{*}S.R. is Deputy Editor of *AnnalsATS*. Her participation complies with American Thoracic Society requirements for recusal from review and decisions for authored works.

DOI: 10.1513/AnnalsATS.202008-970ED

Ann Am Thorac Soc Vol 17, No 10, pp 1207–1212, Oct 2020 Internet address: www.atsjournals.org

classifying sleep state from heart rate and movement data in infants. Such data are readily available not only from cardiorespiratory monitors but also from many wearable health devices used in older children and adults, which similarly use those types of data for noninvasively estimating sleep state. Nonburdensome collection of data for estimating sleep state in real-world settings, including over multiple nights, holds tremendous promise for elucidating the role of sleep in health, including how sleep changes with interventions. Further research that defines the role of non-electroencephalogramdefined sleep states in characterizing sleep physiology across the populationincluding children-could inform a myriad of clinical and research applications, including elucidating longitudinal changes in sleep and sleep disorders with growth and development. Whether such data are best extracted with manual, expert-directed approaches, as done by Vézina and colleagues (4), or can be done more efficiently and objectively using artificial intelligence is a central question as the sleep field adopts advanced technologies.

As the pediatric sleep medicine field also evaluates newer measurement approaches, it is useful to remember the often-quoted refrain "Children are not small adults" when considering multiple aspects of sleep and polysomnography in young children and the implications for adapting home sleep testing in this population. Unattended portable monitoring is recommended for use in adults with a high pretest probability of OSA when performed in conjunction with a comprehensive

evaluation by a medical provider (7, 8). In addition to addressing safety concerns about sleeping with sensors attached, and the need for parental oversight/involvement, we need to consider the indications for testing, accuracy of testing, and approach to sensor placement, all of which distinguish pediatric home-based sleep testing from standard adult protocols. The data presented by Vézina and colleagues suggest we can, and perhaps should, view these considerations not as barriers but as opportunities to innovate to study children where they and their parents are most comfortable. Innovations in technology demonstrated by Vézina and colleagues and many others (9) should do much to assuage concerns about safety and feasibility. Although further research is needed to validate these approaches in children, even the gold standard attended PSG-derived apneahypopnea index has limitations in terms of assigning severity and predicting long-term consequences.

Finally, as in many areas of health care, disparities in diagnosis, access to care, and treatment outcomes in children with SDB are a significant public health problem (10). SDB is more common in Black children than white children (11), and Black children are less likely to have spontaneous resolution of SDB (12). Higher poverty rates and percentage of children living in singlefemale-headed households are associated with higher apnea-hypopnea indices (13). What role does the reliance on in-lab PSG play in long-term health disparities related to SDB? In a study of more than 200 children referred from primary care physicians for evaluation of SDB, of whom 87% had public insurance, half of them were lost to follow-up (14). Among those referred for polysomnography, more than threequarters were lost to follow-up. These parents are voting with their feet-the barriers to bringing their children to a sleep laboratory for an attended PSG are too high for most of them to overcome. In a separate study, it took twice as long for children with public insurance to be treated with adenotonsillectomy following PSG than their peers with private insurance (15). Delay in treatment is likely to have longterm consequences. Treatment of sleep disorders in young children improves outcomes (16), but complete response is the exception rather than the rule (17, 18), suggesting there are critical windows of exposure that have lifelong consequences. Race and poverty adversely affect response to treatment (16, 19). We owe it to children and their families to leverage technology to make it easier to be diagnosed and receive treatment for SDB and other sleep disorders.

Because of the complexities of successfully studying sleep in young children, is it time to challenge ourselves that the default approach of laboratorybased attended sleep studies provides the best care? We have seen an unprecedented rate of change in healthcare delivery in 2020 and a groundswell of support to address health disparities. Using that momentum, as well as advances in technology, to improve comfort and access to care for children with sleep disorders is the right thing to do. ■

Author disclosures are available with the text of this article at www.atsjournals.org.

References

- Marcus CL, Brooks LJ, Draper KA, Gozal D, Halbower AC, Jones J, *et al.* Diagnosis and management of childhood obstructive sleep apnea syndrome. *Pediatrics* 2012;130:576–584.
- 2 Scholle S, Scholle H-C, Kemper A, Glaser S, Rieger B, Kemper G, et al. First night effect in children and adolescents undergoing polysomnography for sleep-disordered breathing. *Clin Neurophysiol* 2003;114:2138–2145.
- 3 Verhulst SL, Schrauwen N, De Backer WA, Desager KN. First night effect for polysomnographic data in children and adolescents with suspected sleep disordered breathing. *Arch Dis Child* 2006;91: 233–237.
- 4 Vézina K, Mariasine J, Young R, Reyna M, Lu Z, Subbarao P, et al. Cardiorespiratory monitoring data during sleep in healthy Canadian infants. Ann Am Thorac Soc 2020;17:1238–1246.
- 5 Katz ES, Greene MG, Carson KA, Galster P, Loughlin GM, Carroll J, et al. Night-to-night variability of polysomnography in children with suspected obstructive sleep apnea. J Pediatr 2002;140: 589–594.

- 6 Daftary AS, Jalou HE, Shively L, Slaven JE, Davis SD. Polysomnography reference values in healthy newborns. J Clin Sleep Med 2019;15: 437–443.
- 7 Collop NA, Anderson WMD, Boehlecke B, Claman D, Goldberg R, Gottlieb DJ, et al.; Portable Monitoring Task Force of the American Academy of Sleep Medicine. Clinical guidelines for the use of unattended portable monitors in the diagnosis of obstructive sleep apnea in adult patients. J Clin Sleep Med 2007;3:737–747.
- 8 Rosen IM, Kirsch DB, Carden KA, Malhotra RK, Ramar K, Aurora RN, et al.; American Academy of Sleep Medicine Board of Directors. Clinical use of a home sleep apnea test: an updated American Academy of Sleep Medicine position statement. J Clin Sleep Med 2018;14:2075–2077.
- 9 Bertoni D, Isaiah A. Towards patient-centered diagnosis of pediatric obstructive sleep apnea-a review of biomedical engineering strategies. *Expert Rev Med Devices* 2019;16:617–629.
- 10 Boss EF, Smith DF, Ishman SL. Racial/ethnic and socioeconomic disparities in the diagnosis and treatment of sleep-disordered breathing in children. *Int J Pediatr Otorhinolaryngol* 2011;75:299–307.

EDITORIALS

- 11 Redline S, Tishler PV, Schluchter M, Aylor J, Clark K, Graham G. Risk factors for sleep-disordered breathing in children: associations with obesity, race, and respiratory problems. *Am J Respir Crit Care Med* 1999;159:1527–1532.
- 12 Chervin RD, Ellenberg SS, Hou X, Marcus CL, Garetz SL, Katz ES, *et al.*; Childhood Adenotonsillectomy Trial. Prognosis for spontaneous resolution of OSA in children. *Chest* 2015;148:1204–1213.
- 13 Wang R, Dong Y, Weng J, Kontos EZ, Chervin RD, Rosen CL, et al. Associations among neighborhood, race, and sleep apnea severity in children: a six-city analysis. Ann Am Thorac Soc 2017;14:76–84.
- 14 Harris VC, Links AR, Kim JM, Walsh J, Tunkel DE, Boss EF. Follow-up and time to treatment in an urban cohort of children with sleep-disordered breathing. *Otolaryngol Head Neck Surg* 2018;159:371–378.
- 15 Boss EF, Benke JR, Tunkel DE, Ishman SL, Bridges JFP, Kim JM. Public insurance and timing of polysomnography and surgical care for children with sleep-disordered breathing. *JAMA Otolaryngol Head Neck Surg* 2015;141:106–111.

- 16 Marcus CL, Moore RH, Rosen CL, Giordani B, Garetz SL, Taylor HG, et al.; Childhood Adenotonsillectomy Trial (CHAT). A randomized trial of adenotonsillectomy for childhood sleep apnea. N Engl J Med 2013; 368:2366–2376.
- 17 Chervin RD, Ruzicka DL, Giordani BJ, Weatherly RA, Dillon JE, Hodges EK, et al. Sleep-disordered breathing, behavior, and cognition in children before and after adenotonsillectomy. *Pediatrics* 2006;117: e769–e778.
- 18 Biggs SN, Vlahandonis A, Anderson V, Bourke R, Nixon GM, Davey MJ, et al. Long-term changes in neurocognition and behavior following treatment of sleep disordered breathing in school-aged children. Sleep (Basel) 2014;37:77–84.
- 19 Amin R, Anthony L, Somers V, Fenchel M, McConnell K, Jefferies J, et al. Growth velocity predicts recurrence of sleep-disordered breathing 1 year after adenotonsillectomy. Am J Respir Crit Care Med 2008;177:654–659.

Copyright © 2020 by the American Thoracic Society

Check for updates

Riding a New Wave: Computational Fluid Dynamics Brings Clinical Trials for Tracheomalacia within Reach

3 Joseph Piccione, D.O., M.S.¹, and Alfin G. Vicencio, M.D.²

¹Division of Pulmonary Medicine and Center for Pediatric Airway Disorders, Children's Hospital of Philadelphia, University of Pennsylvania School of Medicine, Philadelphia, Pennsylvania; and ²Division of Pediatric Pulmonology, Mount Sinai Kravis Children's Hospital, Icahn School of Medicine at Mount Sinai, New York, New York

ORCID ID: 0000-0002-0691-3551 (J.P.).

Although tracheomalacia is a widely recognized cause of respiratory morbidity, diagnosis of the condition remains difficult, and prevalence is likely underestimated because of limitations in noninvasive testing (1). Flexible bronchoscopy has long been the gold standard diagnostic test, but because of its invasive nature and because it



³This article is open access and distributed under the terms of the Creative Commons Attribution Non-Commercial No Derivatives License 4.0 (http://creativecommons.org/licenses/by-nc-nd/ 4.0/). For commercial usage and reprints, please contact Diane Gern (dgern@thoracic.org).

DOI: 10.1513/AnnalsATS.202007-847ED

requires a careful anesthetic approach (i.e., balancing depth of anesthesia that maximizes patient safety and comfort while allowing spontaneous, unobstructed breathing), it has typically been employed for select patients with moderate to severe symptoms to establish initial diagnosis and less so for subsequent evaluation of disease. Importantly, visual assessment of tracheal collapse via flexible bronchoscopy is subject to significant intra- and interrater variability, even at a qualitative level (2). Confounding these limitations, there is a lack of clarity regarding the "normal" degree of tracheal collapse that is present in healthy infants during inspiration and expiration as well as a lack of data that objectively characterize age-dependent variations.

A noninvasive diagnostic modality that can reliably and quantitatively assess tracheal collapse has long been elusive, with earlier methods, including airway fluoroscopy, contrast tracheobronchography, and computed tomography, falling short in one or more of these parameters. Recently, Hysinger and colleagues validated ultrashort echo time magnetic resonance imaging (UTE MRI) as a means to noninvasively assess tracheomalacia in neonates without sedation or ionizing radiation, thereby offering an unprecedented opportunity to objectively characterize tracheal dynamics as well as assess changes over time and in response to potential therapies (3). In an editorial accompanying the Hysinger and colleagues manuscript, we noted our optimism with this technology, but we also lamented that UTE MRI "fails to measure the magnitude of force required to produce that collapse," which is necessary to objectively assess tracheal compliance and determine whether the airway collapse is due to inherent defect in the trachea or excessive forces imposed on it by obstruction in the small airways (4). Though this has yet to be accomplished, an exciting new approach adds a functional component to this structural assessment.

In this issue of *AnnalsATS*, Gunatilaka and colleagues (pp. 1247–1256) demonstrate the utility of UTE MRI to quantify tracheal resistance in infants with tracheomalacia (5). Using computational fluid dynamics (CFD), the authors extracted clinically relevant physiological data from this noninvasive imaging modality, allowing calculation of work of breathing attributed to the defect. This three-dimensional