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Editorial

Laryngomalacia, GER, and sleep apnea are BRUEing – Sometimes!

David Gozal

Department of Child Health, the Child Health Research Institute and MU Women and Children's Hospital, University of Missouri School of Medicine, Columbia, Missouri, MO, USA

"Breathing dreams like air"
F. Scott Fitzgerald, *The Great Gatsby*

Keywords

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Corresponding author

David Gozal, MD, MBA, Department of Child Health, University of Missouri, 400 N. Keene Street, Suite 010, Columbia, MO 65201, USA; email: gozald@health.missouri.edu.

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The field of unexplained events in infancy that are perceived by their caretakers as potentially life threatening has evolved, and prompted assignment of different criteria along with a novel nomenclature aimed at better refining the phenotypic presentation of such cases [1]. Indeed, separation of low-risk and high-risk brief resolved unexplained events (BRUE) has helped reduce the extent of the diagnostic work-up that traditionally was implemented in all previously designated apparent life-threatening events (ALTE) [2], even if considerable divergence and heterogeneity exists among pediatricians [3, 4]. Furthermore, few if any testing appears to be positive among higher risk BRUE cases being evaluated in the emergency room [5].

Since few if any of the BRUE events take place during cardiorespiratory monitoring, particularly among the low-risk BRUE category, we are only to assume that such events occur during infant sleep. In an earlier study, Nosetti and colleagues explored potential polysomnographic characteristics of different infants being evaluated for BRUE, obstructive sleep apnea (OSA) or OSA and BRUE [6]. In this study, laryngomalacia was identified relatively frequently in the young cohort (< 12 months of age) suggesting that it might play a role in BRUE, an impression that was also shared by the parents [7]. In a subsequent study published in this issue of the Journal [8], Nosetti and collaborators conducted a retrospective review of 448 children (age < 12 months) evaluated for BRUE, in whom endoscopic evaluation of the upper airways as well as a nap polysomnography and 24-h cardiorespiratory monitoring were conducted as part of the initial work-up follow-up. Interestingly, 1 of every 9-10 infants presented evidence compatible with laryngomalacia and of these, two thirds of the infants also demonstrated the presence of obstructive respiratory events when asleep [8]. Overall outcomes were extremely favorable with resolution of the upper airway structural deficits along with normalization of sleep respiratory patterns in the vast majority of the cases.

As is always the case, we need to put these findings in a contextual setting and explore their potential relevance when considering whether and when flexible endoscopic evaluation is needed in BRUE. First, the selection of the cases who were evaluated with flexible endoscopy was not random and was prompted by the underlying symptoms or by high level of suspicion in a tertiary medical center. As such, we are unclear what the true prevalence of laryngomalacia would be if all cases of BRUE being evaluated in the emergency room would be included. Second, the association of laryngomalacia with sleep-disordered breathing and gastroesophageal reflux (GER) is not surprising since both of these conditions have been associated with laryngomalacia [9-11]. Furthermore, these issues will progressively improve and resolve over time even without supraglottoplasty [12]. As such, it is unclear whether laryngoscopic evaluation is really needed, considering that few if any of the infants will need to be surgically treated. Finally, an unanswered question revolves around the positioning of the infant with laryngomalacia for sleep. On the one hand, it is assumed that the supine position (as recommended for most healthy infants) might exacerbate the frequency and severity of the upper airway obstructive events in infants with BRUE who already manifest laryngomalacia [13]. However, this may not be the case. Although no specific studies have been conducted in BRUE, and the authors of the current paper do not specifically address this issue, studies in infants with micrognathia supported adopting the prone position [14], while studies in infants with cleft palate failed to reveal differences in the severity of sleep-disordered breathing in the supine vs. prone position [15].

In summary, the triad of GER, obstructed sleep-disordered breathing and laryngomalacia emerges as a relatively frequent clinical cluster of problems potentially facilitating the emergence of a BRUE. Awareness to this association may permit a more detailed scrutiny of its presence, and promote a more structured approach to cardiorespiratory monitoring and follow-up of those infants with high-risk BRUE.

Declaration of interest

The Author declares that there is no conflict of interest.

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