ARTICLE

Validity and reliability of the behavioral signs of respiratory instability (BSRI) © scale during activity for infants with bronchopulmonary dysplasia

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OBJECTIVE: There is no reliable evidence on how best to evaluate the overall status of infants with severe forms of bronchopulmonary dysplasia (BPD). The Behavioral Signs of Respiratory Instability (BSRI) scale was developed as an objective measure of developmental capacity during occupational and physical therapy sessions. The purpose of this study was to determine the psychometric properties of the BSRI Scale.

STUDY DESIGN: The BSRI and Respiratory Severity Score (RSS) were compared for 25 infants with BPD and 15 infants without BPD. A cross-sectional design was used to test inter-rater reliability among 10 NICU occupational and physical therapists. A prospective cohort design was used to evaluate validity.

RESULTS: The BSRI demonstrated good to excellent inter-rater reliability ($\rho = 0.47–0.91$) and was strongly correlated with RSS ($\rho = -0.77$, $p < 0.001$; concurrent validity).

CONCLUSION: The BSRI Scale has preliminary psychometric support. Standardized measures like the BSRI may provide accurate, objective data that can improve care planning within interdisciplinary teams that supports brain growth and potentially improves neurodevelopment.

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INTRODUCTION

Preterm birth is the most common cause of abnormal lung development in infancy, and Bronchopulmonary Dysplasia (BPD) is the most common pulmonary complication of prematurity which can lead to lifelong sequelae [1]. Levels of severity of BPD are recognized by the need for supplemental oxygen, and mode of delivery. Severe BPD (sBPD) is defined as the need for positive pressure at 36 weeks post menstrual age (PMA) [2, 3]. An estimated 16% of infants born less than 32 weeks PMA have sBPD, translating to roughly 13,000 babies diagnosed with sBPD annually in the US alone [3]. Infants with sBPD typically require long hospitalizations for medical management [2, 4]. The clinical presentation of sBPD is diverse and complex. Management strategies optimizing survival and long-term outcomes are controversial and uncertain [3, 5]. For example, the association between sedatives and poor developmental outcomes suggests a need for prudent use of these drugs for infants with sBPD [6]. Interventions vary by institution and may include differing combinations of steroids, diuretics, oxygen concentration, and ventilation management [2, 3, 5, 7, 8].

Medical interventions are typically prescribed based on physiologic evidence of respiratory status, such as oxygen saturation, respiratory rate, carbon dioxide levels, and chest x-rays [4, 9]. However, physiologic evidence alone may not provide an accurate assessment of the steady-state respiratory status of infants with BPD. It is possible that behavioral signs during typical daily activity and physical manifestations of respiratory status provide additional insight regarding an infant's response to medical interventions. Functional status informs clinical decision-making when treating both adults and children. For example, functional classification scales have been developed for children and adults with pulmonary hypertension to objectively describe the impact of this chronic health condition on individuals [10]. There is some evidence that the Silverman Andersen Respiratory Severity Score (RSS) can guide medical management for infants with respiratory compromise. For example, RSS scores may be useful for assessing the need to escalate oxygen therapy for term infants in low economically resourced settings [11]. The RSS does not address the impact of respiratory instability on capacity for participating in developmental challenges, which is pertinent when infants with BPD are receiving rehabilitation services. Accurate representation of a patient's functional status, makes it possible to review progress with time, and the response to interventions, ultimately driving treatment plans [10].

The Behavioral Signs of Respiratory Instability (BSRI) Scale was developed to provide therapists and the interdisciplinary medical team objective data about current capacity for participation in

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Table 1. BSRI© Administration Guidelines and Clinical Considerations.

Domain	Description
Frequency	The BSRI can be completed and documented during all therapy sessions. Ideally, 2–4 therapy sessions/ week can occur, as staffing allows
Timing	Coordinate the assessment with typical wakeful periods for the infant (typically with nursing care times)
Interaction	Provide just right support to encourage alertness. This may require calming, or gentle stimulation, based on each infant.
Midline	If the infant has a head preference, passive stretches are completed prior to assessing midline orientation, to give the infant the best chance of success.
Extension patterns, Tachypnea, WOB	Typically assessed during and after 1–5 min in a supported sitting position.
	If the baby becomes physiologically unstable, supported upright is discontinued

WOB Work of Breathing.

developmentally appropriate activities. The BSRI Scale can be used serially to determine response over time. These data can inform collaborative decisions related to medical management that supports best outcomes for infants with sBPD [12, 13]. The psychometric properties of the BSRI Scale have not yet been investigated. The purposes of this study were to determine the inter-rater reliability, concurrent validity, and construct (known-groups) validity of the BSRI when used to assess hospitalized infants with BPD. We hypothesized that (1) clinicians would have good inter-rater reliability following standardized training; (2) BSRI Scale scores would have a direct relationship with other measures of respiratory stability in infants with BPD (concurrent validity); and (3) BSRI scores would distinguish between infants with BPD and those without BPD (construct validity).

METHODS

This study was conducted in two phases. Phase 1 was a prospective cross-sectional design to investigate inter-rater reliability. Nationwide Children's Hospital (NCH) Institutional Review Board (IRB) approval was granted (IRB14-00892) for Phase I. It was exempt from attaining informed consent because procedures occurred in the context of standard care, with no changes made in the infants' plan of care. Data collection for Phase 1 occurred from 2015 to 2016. Phase 2 was a prospective cohort design to evaluate concurrent and construct validity. The study was approved by the NCH IRB (STUDY00000807). Informed consent was provided by parents. Data collection for Phase 2 occurred from 2020 to 2021, with a pause in data collection due to Covid-19. Eligibility criteria for Phase 1 and for testing concurrent validity were admission to the hospital neonatal intensive care unit (NICU), age between 37 weeks and 1 year corrected gestational age, eligibility to receive occupational therapy and/or physical therapy services, BPD diagnosis, and supplemental oxygen via nasal continuous positive airway pressure (NCPAP) or nasal cannula. For testing construct validity in Phase 2, infants without BPD were included.

Exclusion criteria were oral intubation, tracheostomy, and complex chronic illness, such as periventricular leukomalacia, cerebellar infarct, chromosomal abnormalities, hypoxic ischemic encephalopathy, and neonatal abstinence syndrome that may negatively bias BSRI Scale scores. The electronic medical record was reviewed to determine eligibility for recruitment to the study.

Measures

Behavioral signs of respiratory instability (BSRI) scale. The BSRI Scale captures infant behaviors during handling, from 37 weeks corrected age through 1 year [14]. The 5 domains of the BSRI Scale include Interaction, Midline, Extension Patterns of Movement, Tachypnea, and Work of Breathing. Each domain is scored 0, 1, or 2. A sum score of all domains yields a maximum possible score of 10. Lower scores indicate greater respiratory impact, or instability. The BSRI Scale is scored in the context of the baby performing developmentally appropriate motor activity during physical and/or occupational therapy sessions. For example, at our institution, infants with sBPD receive therapy services 2–4 times per week, for neuro-regulatory support and/or neuro-motor skill acquisition, based on their current level of physiologic stability. The BSRI Scale is scored after each of these sessions. Scoring criteria for each domain are found in Supplemental Material 1. Administration guidelines can be found in Table 1.

Silverman andersen respiratory severity score (RSS). The RSS quantifies the severity of respiratory distress in infants. Five items are scored based on observations during routine care of the infant. The score for each item is 0, 1 or 2, with higher scores indicating more severe respiratory distress [4, 11, 15]. There is evidence that RSS scores are associated with respiratory distress [11] and strong reliability has been reported for trained nurses [15]. Studies on the psychometric properties of the RSS are lacking.

Phase 1 procedures: inter-rater reliability

A convenience sample of twenty ($n = 20$) infants with BPD was included for testing of inter-rater reliability of the BSRI Scale. Ten raters were occupational and physical therapists in the NICU Therapy Department at NCH who voluntarily participated in the study. Raters completed standardized training. The BSRI Scale was scored within the context of routine developmental treatment. Scores were based on the first 15 min of each treatment session. Independent scoring by two of the 10 raters was completed concurrently, and each rater was blinded to the scoring of the other. Each of the 10 raters scored four different sessions, for a total of 40 scored sessions. Several of the infants in the study sample were scored more than once over the course of their hospitalization.

Phase 2 procedures: concurrent and construct (known-groups) validity

The results of the Phase 1, inter-rater reliability, were known prior to the initiation of Phase 2. Lower reliability for the WOB domain appeared to result from inadequate scoring criteria descriptors. Based on this, the scoring criteria in this single domain, was revised to include the words "expiratory grunting and wheezing" as additional descriptors for a WOB score of "0". From 2016 to the present, the BSRI has been used clinically by the physical and occupational therapists at NCH. This revision to the BRSI Scale, based on reliability results, was important to report the most accurate information to the interdisciplinary medical team. The additional descriptors were included for Phase 2 of the study, which was initiated in 2020.

Forty ($N = 40$) hospitalized infants were included for testing the validity of the BSRI. Twenty-five ($n = 25$) infants with BPD were included for testing concurrent validity of the BSRI Scale. During routine daily assessment, a nurse practitioner (NP) scored the RSS. A physical therapist or occupational therapist independently and simultaneously scored the BSRI Scale. The NP and therapist were not privy to one another's scores. BSRI Scale scores from the same 25 ($n = 25$) infants with BPD and a sample of 15 ($n = 15$) infants without BPD were used for testing construct (known-groups) validity. For infants without BPD, a physical or occupational therapist scored the BSRI Scale during a routine treatment session.

Data analysis

Descriptive statistics were calculated for demographic characteristics of the participants. Due to non-normal distribution of the data, appropriate nonparametric tests were used. Inter-rater reliability for the BSRI was calculated using Spearman's rank correlations. The relationship between BSRI and RSS scores was also measured using Spearman correlations. Wilcoxon Rank Sum Tests were used to determine differences in BSRI scores for infants with and without BPD to investigate differences in known

Table 2. Descriptive Statistics of Participant Characteristics in Phase 1 and Phase 2.

	Phase 1		Phase 2	
	Infants at onset of scoring ($n = 20$)	**Infants with BPD ($n = 25$)**	**Infants without BPD ($n = 15$)**	**Total ($n = 40$)**
Gestational age at birth (weeks)				
Mean (SD)	26 (2.35)	26 (3.10)	34 (4.86)	29 (5.25)
Gestational age when scored (weeks)				
Mean (SD)	48 (6.60)	44 (6.58)	42 (4.59)	43 (5.93)
Sex[a]				
Male	9 (45%)	13 (52%)	8 (53%)	21 (53%)
Female	11 (55%)	12 (48%)	7 (47%)	19 (47%)
BSRI				
Median (IQR)		7 (2)	9 (1)	8 (1.50)
RSS				
Median (IQR)		2 (2)		

BPD Bronchopulmonary Dysplasia, *BSRI* Behavioral Signs of Respiratory Instability (Total Score), *RSS* Respiratory Severity Score.
[a]Frequency (proportion).

clinical groups. All analyses and figures were completed using R (version 4.1.2).

RESULTS

Participant characteristics

For Phase 1, 9 infants (45%) were male, and 11 (55%) were female. Average gestational age at birth was 26 weeks. Average gestational age when scoring occurred was 51 weeks. Phase 2 included 13 (52%) males and 12 (48%) females with BPD, and 8 (53%) males and seven (47%) females without BPD. Mean gestational age at birth was 26 weeks for infants with BPD and 34 weeks for those without. Scoring on the BSRI occurred at an average age of 44 weeks for the infants with BPD and 42 weeks for the infants without BPD. These characteristics are summarized in Table 2.

Phase 1: Inter-rater reliability

Inter-rater reliability of the BSRI Scale total score was good ($\rho = 0.70$, $p < 0.001$). Inter-rater reliability of the individual domains of the BSRI ranged between 0.47 and 0.91. Work of Breathing was determined to be moderately reliable, whereas the other categories had good to excellent inter-rater reliability (Table 3).

Phase 2: concurrent validity

The overall sample had a median BSRI score of 8 (IQR = 1.50). The BSRI Scale showed a significant negative correlation with RSS scores ($\rho = -0.77$ [95% CI: −0.77, −0.48], $p < 0.001$). Higher BSRI scores correlated with lower RSS scores, both of which indicate minimal respiratory distress. There were no significant differences between the NCPAP and nasal cannula groups on the BSRI [$W = 76$, $p = 0.74$, $r = 0.07$ (small effect)] or RSS [$W = 52$, $p = 0.29$, $r = 0.22$ (small effect)].

Phase 2: construct (known-groups) validity

Infants with BPD had significantly lower BSRI total scores compared to the infants without BPD [$W = 51$, $p < 0.001$, $r = 0.62$ (large effect)]. The BPD group had a median BSRI score of 7 (IQR = 2) and the non-BPD group had a median BSRI score of 9 (IQR = 1). These results are in Fig. 1.

Table 3. Spearman's Correlation Coefficient Rho for BSRI Scale Domains between Raters.

	ρ	*p*-value
Interaction	0.91	<0.001
Midline	0.75	<0.001
Extension Patterns of Movement	0.85	<0.001
Tachypnea	0.83	<0.001
Work of Breathing	0.47	0.003
Total Score	0.70	<0.001

BSRI Behavioral Signs of Respiratory Instability.

DISCUSSION

Infants with sBPD are at greater risk for lifelong impairments in cognitive, language, academic achievement, executive, emotional, social and physical functioning when compared with infants without BPD [16]. Not only do infants with sBPD show delayed acquisition of many critical motor skills compared to age-specific norms, but these skills often develop atypically [6]. Occupational and physical therapy are recommended early for infants with BPD to address limitations in functional capacity, muscle strength, and motor function [17]. Negative trajectories of motor skill acquisition have also been shown to be more strongly and consistently associated with routine exposure to sedating medications than with the level of respiratory support [6]. There is undoubtedly a connection between respiratory function, neurodevelopment, medication exposure, and nutrition and growth that warrants further research.

Infancy and early childhood represent a period of life with both exquisite opportunity and vulnerability for neurodevelopment [18]. Physical and occupational therapy services promote the neurodevelopmental skills of hospitalized infants with sBPD but can be limited by medical instability. Optimal pulmonary function, brain growth, and development can only occur when these are properly balanced [12]. The BSRI Scale was developed to provide an objective measure of respiratory instability observed during developmentally appropriate activity. The Interaction domain is based on the knowledge that infants are able to visually engage with objects in their environment only when they have not exceeded the capacity of their respiratory system, as described previously by The Newborn Individualized Developmental Care and Assessment Plan (NIDCAP) [19]. The remaining four domains of the BSRI Scale incorporate knowledge of developmental movement patterns and breathing mechanics. The trunk maintains a dual role to support respiration and postural control [20]. Midline orientation of the head in the transverse plane is essential for postural control and motor skill development, including coordination of visual and upper extremities skills [19, 21]. Cervical extension may be seen in respiratory conditions, including BPD, because accessory muscles are recruited to support respiration [22]. Extension of the head reduces resistance by splinting large airways open to allow greater air flow, lower airway resistance, and decreased WOB [20]. Tachypnea, the single most sensitive sign of respiratory distress, is a compensatory mechanism for hypercarbia, hypoxemia, or acidosis, especially in neonates [23]. The final domain, WOB, includes excessive recruitment of accessory muscles in the neck, chest wall, or abdomen for respiration [23]. Grunting, stridor, wheezing and nasal flaring are subconscious attempts at decreasing airway resistance [4, 11, 24].

A published case study describes the conceptual framework and clinical application of the BSRI Scale for infants with BPD [25]. Since that time, an iterative process of repeat and review resulted in expert consensus on BSRI Scale domains and scoring criteria. Domains were adjusted slightly for continuity in skills through 1 year of corrected age. Scoring criteria were updated so that BSRI scores are ordinal in nature, with higher

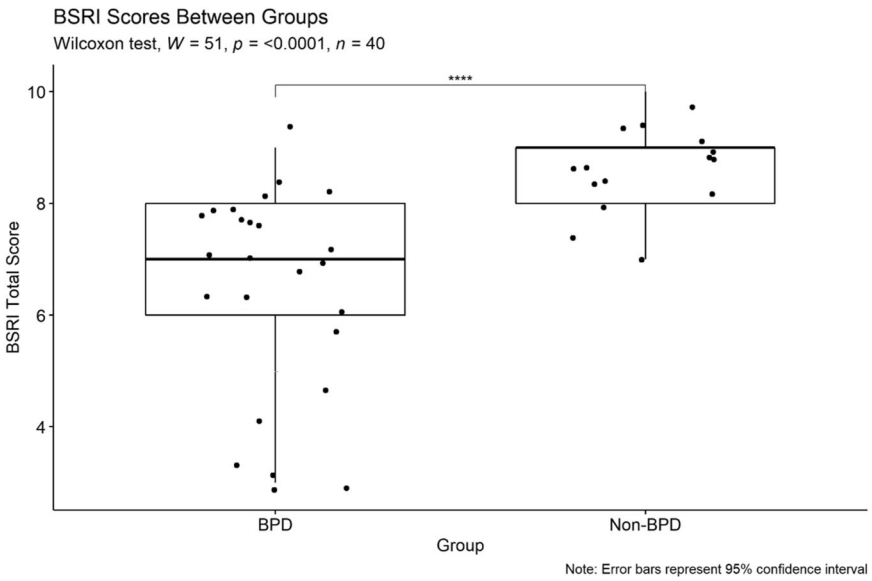

Fig. 1 BSRI Scale Scores between infants with BPD and those without BPD. BSRI Behavioral Signs of Respiratory Instability, BPD bronchopulmonary dysplasia. ****$p < 0.0001$.

scores reflecting better performance and less respiratory instability. Additional descriptors were added to the criteria for scoring WOB. The length of time indicated to observe the testing items was clarified. Finally, updated criteria were added to score the Interaction and Midline domains as "observed" versus "not observed". The remaining domains were scored based on the most persistent behavior observed during the assessment. Training using didactic content and case studies was developed to optimize the consistency of scoring among clinicians using the BSRI Scale. A copyright was obtained for the final version in 2016 (Supplemental Material 1).

Physical and occupational therapists have historically provided subjective information about infant capacity to participate in developmental activity. Standardized tools, such as the BSRI Scale, limit the subjective nature of assessment, enhancing the rigor of data provided by NICU therapists and allow longitudinal comparison. With standardized training, the therapists in this study achieved good to excellent inter-rater reliability for all domains of the revised BSRI Scale, including the total score. Optimally, information from the BSRI scale is shared with the interdisciplinary team to address the complex needs of this population.

No differences were found in the validity of the BSRI Scale between infants supported by NCPAP or NC. This was surprising as we had assumed that the need for NCPAP would reflect a greater underlying disease burden that would lead to poor tolerance of developmental activity. Given the small group sizes for NCPAP ($n = 10$) and NC ($n = 14$), these results are considered exploratory. Nonetheless, we speculate that the similarity in scores suggests that the infants were benefiting from the use of NCPAP, which provided them the respiratory stability to engage in developmentally appropriate activities. The BSRI scale could, therefore, be a useful tool to quantify developmental capacity by teams evaluating the impact of factors that affect developmental outcomes.

Our results indicate psychometric support for concurrent and construct validity of the BSRI Scale. Scores were inversely related to RSS scores, where lower BSRI scores and greater RSS scores both indicate greater respiratory instability. This may indicate that the BSRI Scale captures the respiratory stability of infants. Additionally, BSRI scores for infants with BPD were significantly lower than scores for infants without BPD, confirming our hypothesis. Unlike the RSS, the BSRI Scale is scored while the infant is performing functional activities. This adds meaningfully to NICU physical and occupational therapy practice because BSRI scores are a means of objective communication about capacity for participation in developmental activities among hospitalized infants with sBPD.

With standardized training, clinicians can reliably apply the BSRI Scale, and scores appear to reflect respiratory instability that may impact neurodevelopmental progress. For training and support for implementation of the BSRI, please contact the authors for available resources. The total score on the BSRI Scale can be tracked over time, reflecting the infant's capacity to participate in developmental activity, during their NICU admission. The BSRI has the potential to be utilized in collaboration with an interdisciplinary medical team to provide clear and objective measure of developmental competence. This supports the ultimate goal of NICU therapy services: optimizing long term neurodevelopmental outcomes for hospitalized infants.

This study had a variety of limitations. The small sample size at a single institution limits generalizability. The BPD group had highly variables scores, which may have reduced our ability to detect a true difference between infants supported by NCPAP versus NC. Given the lack of statistical power, it is possible that we were unable to detect true differences between infants with BPD and those without. Including a large sample in a randomized, multicenter study is recommended for future research. Babies supported by mechanical ventilation were omitted from the study. Occupational and physical therapy services in the NICU can be initiated even when infants require high levels of respiratory support, including intubation. Use of the BSRI Scale among this population warrants further study. Finally, we hypothesize that supporting inpatient neurodevelopment will improve long term outcomes, yet we did not include that data as part of this study.

DATA AVAILABILITY

All phases of this study were performed in accordance with the Declaration of Helsinki. Datasets for each phase of this study are available upon request.

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AUTHOR CONTRIBUTIONS

K Susey--scale development, IRB submission, experimental design, and manuscript author. MH--scale development, experimental design, and manuscript author. AW--experimental design, IRB submission, data analysis, and manuscript author. M Mandich--experimental design, data analysis, and manuscript editor. K Scott—manuscript editor. K Stephenson—experimental design, data analysis, and manuscript author. ES—scale development, manuscript editor. M Mehling—IRB submission, manuscript editor.

COMPETING INTERESTS

The authors declare no competing interests.

ADDITIONAL INFORMATION

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