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Sensory Processing Disorder in Preterm Infants during Early Childhood and Relationships to Early Neurobehavior

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Abstract

Background—Preterm infants are exposed to a variety of sensory stimuli that they are not developmentally prepared to handle, which puts them at risk for developing a sensory processing disorder. However, the patterns and predictors of sensory processing disorder and their relationship to early behavior at term equivalent age are poorly understood.

Objectives—The aims of the study are to: 1) describe the incidence of sensory processing disorder in preterm infants at four to six years of age, 2) define medical and sociodemographic factors that relate to sensory processing disorder, and 3) explore relationships between early neurobehavior at term equivalent age and sensory processing disorder at age four to six years.

Methods—This study was a prospective longitudinal design. Thirty-two preterm infants born ≤30 weeks gestation were enrolled. Infants had standardized neurobehavioral testing at term equivalent age with the NICU Network Neurobehavioral Scale. At four to six years of age, participants were assessed with the Sensory Processing Assessment for Young Children (SPA).

Results—Sixteen children (50%) had at least one abnormal score on the SPA, indicating a sensory processing disorder. There were no identified relationships between medical and sociodemographic factors and sensory processing disorder. Having more sub-optimal reflexes (p=.04) and more signs of stress (p=.02) at term equivalent age was related to having a sensory processing disorder in early childhood.

Conclusion—Preterm infants are at an increased risk for developing a sensory processing disorder. Medical and sociodemographic factors related to sensory processing disorder could not

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be isolated in this study, however relationships between sensory processing disorder and early neurobehavior were identified.

Keywords

NICU; Sensory Processing Disorder; Neurobehavior; Prematurity

Background

One in ten infants in the United States is born preterm[1] [1]. Infants born prematurely have an increased risk for immediate medical complications, as well as social-emotional, cognitive, language, and sensory processing problems later in life [2–5]. Studies have examined the effects of prematurity on developmental outcomes, such as cognition [3, 4, 6]; however, there is a need for more attention on sensory processing disorder in children born preterm.

A sensory processing disorder involves difficulties with interpreting and using sensory information from the environment for behavioral regulation and motor performance [7]. Sensory processing disorder manifests through atypical behaviors, referred to as patterns of sensory responsiveness, such as hypo- or hyper-responsiveness to sensory stimuli [8]. This can result in a child who does not respond to stimuli or a child with heightened responses to stimuli. Atypical behavioral manifestations of sensory processing disorder can negatively impact a child's enjoyment and frequency of participation in everyday activities [9, 10]. Poor participation in everyday activities negatively impacts developmental skill acquisition [11]. Furthermore, a child's sensory processing abilities impact the entire family unit as a child may be unable to fully participate in certain activities. This places additional stress related to planning and preparation on parents and may even lead to strained family dynamics if the child is unable to participate in activities that are meaningful, such as family gatherings [12].

Sensory processing disorder affects 39% to 52% of infants born preterm, with some evidence suggesting that infants born earlier than 32 weeks carry the greatest risk [5, 13, 14]. Patterns of low registration have been described as one of the most common types of sensory processing disorder in those born preterm [5, 13]. Many children who are born preterm also exhibit tactile defensiveness and have a hyperactive temperament [15], impacting their ability to interact and respond to the environment in appropriate ways. Literature has shown that sensory processing disorder in children born preterm persists until at least eight years of age [5, 13, 14, 16–18].

It remains unclear if sensory processing disorder is evident early in the lifespan and if it may be due to specific exposures or injuries. Although sensory development begins early in utero and continues over time, the early sensory development of the preterm infant occurs primarily in an external environment with sensory stimuli the infant is not yet prepared to integrate. The early neonatal intensive care unit (NICU) environment could play a role in the

[1]Neonatal Intensive Care Unit (NICU), NICU Network Neurobehavioral Scale (NNNS), the Sensory Processing Assessment for Young Children (SPA), Nectrotizing Enterocolitis (NEC), Patent Ductus Arteriosus (PDA), Cranial ultrasound (CUS), Magnetic resonance imaging (MRI), Continuous positive airway pressure (CPAP), Total parenteral nutrition (TPN)

development of sensory processing disorder in preterm infants. The NICU environment has external stimuli that an infant would not typically experience in utero [19]. Without the protective environment of the womb, preterm infants are exposed to intense auditory, tactile, visual, and nociceptive stimuli in the NICU [20]. These sensory exposures occur during a critical period of brain development [6], which can interfere with motor, neurological, and sensory development [19, 21–24].

Sensory processing disorder is not well understood, and the etiology is unclear. It is unclear if early medical complications or interventions may contribute to sensory processing disorder. It is also unclear if there are early signs of sensory processing disorder in infant behavioral manifestations. The aims of this study are to: 1) describe the incidence of sensory processing disorder in preterm infants at four to six years of age, 2) define predictors of sensory processing disorder, and 3) relate early neurobehavior at term equivalent age to sensory processing disorder at four to six years of age. It is hypothesized that: 1) sensory processing disorder will be common, 2) those with significant or prolonged medical interventions and conditions will have the most impairment, and 3) there will be early neurobehavioral signs of sensory processing disorder.

Methods

This study was approved by the Human Research Protection Office of Washington University School of Medicine in St. Louis. The study included a subset of infants from an overarching study aimed at understanding brain development of preterm infants. Consecutive admissions were recruited from 2007–2010. At term equivalent age, prior to NICU discharge, each infant's neurobehavior was assessed using the NICU Network Neurobehavioral Scale (NNNS) [25]. Sociodemographic factors and medical factors were collected from the medical record. At four to six years chronological age, participants returned for assessment of sensory processing disorder using the Sensory Processing Assessment for Young Children (SPA) [26].

Participants and Setting

Preterm infants born ≤30 weeks gestation were enrolled within the first 72 hours of life, and parents signed informed consent. Infants were excluded if they had a congenital anomaly or were not expected to live, as per the opinion of the attending physician. Gestational age at birth was verified using the Ballard exam [27]. The study setting was the 75-bed level III–IV neonatal intensive care unit (NICU) at St. Louis Children's Hospital. At the time of the study, it was designated as a level III unit, but with changes in NICU classification, it is now considered a level IV unit. During NICU hospitalization, study participants received standard care. At the study site, half of the bed spaces are in an open ward style (with 8–12 beds in 4 large rooms) and half are in private NICU rooms. The study site uses the EMPOWER program to educate and engage parents [28]. In addition, all infants who meet this study's inclusion criteria receive occupational therapy at the study site NICU, with the occupational therapist addressing each infant's sensory needs [29].

Independent Variables

Medical Conditions—Medical conditions that were used for this study were documented in the electronic medical record. Estimated gestational age at birth and the presence of necrotizing enterocolitis (NEC, all stages), patent ductus arteriosus (PDA, treated with indomethacin or surgical ligation), or cerebral injury were recorded. Cerebral injury was defined as the presence of either a grade III–IV intraventricular hemorrhage, cystic periventricular leukomalacia, and/or cerebellar hemorrhage, identified through routine cranial ultrasound (CUS) during the first week and month of life and/or with magnetic resonance imaging (MRI) at term equivalent age. A trained neurologist analyzed the CUS and MRI images to identify the presence of cerebral injury.

Medical Interventions—The number of days on a mechanical ventilator, days on continuous positive airway pressure (CPAP), total number of oxygen hours (including any form of ventilation, CPAP, and hours on oxygen delivered via a nasal cannula), days on total parenteral nutrition (TPN), and number of surgeries during the NICU hospitalization were obtained from the electronic medical record.

Socio-demographics—Socio-demographic information was collected from the electronic medical record and included infant sex, insurance type (public or private) as a proxy for socioeconomic status, race (African-American or non African-American), maternal age at birth, and maternal marital status at birth (single or married).

Neurobehavioral Assessment—The NICU Network Neurobehavioral Scale (NNNS) was used to assess neurobehavior at term equivalent age, prior to NICU discharge [25, 30]. The NNNS is a 115-item test with 13 summary scores: habituation, orientation, hypertonicity, hypotonicity, arousal, lethargy, asymmetry, sub-optimal reflexes, excitability, tolerance of handling, stress, quality of movement, and self regulation. A single trained and certified examiner (author, RP) completed NNNS assessments. Internal consistency, as well as predictive validity, have been established for the NNNS [30, 31].

Outcome Measures

Sensory Processing Assessment for Young Children (SPA)—At four to six years chronological age, participants returned for sensory testing. The SPA was used to determine the presence of sensory processing disorder. The SPA is a 20 minute, play-based behavioral observation assessment performed in a semi-structured format [32]. The SPA involves observation of the child's reaction to tactile, auditory, and visual stimuli while interacting with toys and unexpected sensory stimuli (puffs of air, a phone ringing, calling the child's name, etc.). The SPA examines the child's approach or avoidance to novel sensory toys, determines how the child orients and habituates to social and non-social stimuli in the environment, and identifies how the child generates novel action strategies with the toys. The SPA also examines self-stimulatory behaviors [26], including: covering ears with hands or arms, hand flapping, finger mannerisms, mouthing non-food objects, smelling non-food objects, other repetitive motor movements, and repetitive object manipulation. The SPA was completed after the child had undergone approximately four hours of testing by another examiner. The additional testing consisted of standardized assessments of motor, cognitive

and language function. Interrater and intrarater reliability for the SPA are high [32, 33]. A trained member of the research team performed the assessment. Confidence in SPA scoring in the current cohort was achieved by having a second scoring of each assessment, which was videotaped, to ensure intra-rater agreement. There was 100% agreement in scoring from the first to the second scoring of the SPA, which was done with careful review of scoring criteria outlined in the SPA manual.

The SPA has 3 subscales: avoidance, orienting, and defensive. Each has a subscore for reactions to social and non-social stimuli within each subscale score. The orienting subscale relates to hyporesponsiveness to stimuli, while the avoidance subscale relates to hyperresponsiveness [32, 33]. Defensiveness describes the child's avoidance or fear of sensory stimuli [26]. Raw scores were calculated for each subscale and subscore within each subscale. Higher scores indicate poorer performance. No normative data is currently available for the SPA subscale and subscores. For the purposes of this study, sensory processing disorder was defined as having any raw subscale or subscore above the 75th percentile in the sample of high-risk infants. However the use of a 90th percentile cut off score resulted in the number of children identified as having a sensory processing disorder to remain largely unchanged (n=15 vs. n=16).

Statistical Analysis

The IBM Statistical Package for the Social Sciences (SPSS 22) was used for statistical analyses. Descriptive statistics were used to describe the incidence of sensory processing disorder. Chi-square analyses, independent samples t-tests, and nonparametric statistics were used to determine relationships between medical conditions, medical interventions, sociodemographic factors, and sensory processing disorder. Independent samples t-tests were used to explore NNNS summary scores among those with and without sensory processing disorder. Associations with $p < 0.05$ were considered statistically significant.

Results

One hundred and thirty six preterm infants were enrolled in the overarching study. Of those enrolled, seven withdrew, one was excluded due to congenital anomaly, one transferred to another hospital, and twenty expired while in the NICU, leaving 107 in the cohort. Two infants expired and one withdrew after discharge, leaving 104 in the cohort. Of those, 84 (81%) returned for developmental follow up testing at four to six years of age, and 32 (38%) completed sensory testing. Reasons for not doing sensory testing at four to six years was due to scheduling conflicts and lack of an available tester. There were 26 infants who received both sensory testing at age four to six years and neurobehavioral testing at term equivalent age. There were no significant differences in sex, race, birthweight, age at testing, gestational age at birth, and insurance type among those who did and did not receive sensory testing at age four to six years. The average age of testing at term equivalent age was 37.7 (±1.5) weeks postmenstrual age. The average age of testing at four to six years was 66.3 (±4.3) months. See Table 1 for the descriptives of the sample.

Patterns and Predictors of Sensory Processing Scores

See Table 2 for sensory processing scores observed in the cohort. There were 16 (50%) infants with a sensory processing disorder, of which 12 (38%) had an abnormal subscale or subscore in more than one category.

Predictors of Sensory Processing Disorder

Refer to Table 1 for a breakdown of medical and sociodemographic factors in those who did and did not have sensory processing disorder. No medical conditions, medical interventions, or sociodemographic factors were related to sensory processing irregularities. Sex was not related to sensory processing disorder. Predictors of sensory processing were also explored using raw SPA subscale scores, however the findings remained unchanged.

Relationships Between Early Neuorobehavior and Sensory Processing Disorder

Of the 107 infants discharged as part of the overarching study, 81 (76%) had NNNS testing. Neurobehavioral testing at term equivalent age was not conducted in some infants due to the tester being unavailable, the infant being discharged early, or the infant being too sick to undergo testing. There were no significant differences in sex, race, birthweight, gestational age at birth, age at testing, and insurance type among those who did and did not receive neurobehavioral testing. Twenty-six infants were included in analyses investigating sensory processing disorder and neurobehavior. The relationships between early neurobehavior and sensory processing disorder can be found in Table 3. Relationships between early neurobehavior and sensory processing disorder were also explored using raw SPA subscale scores, however the findings remained largely unchanged.

Discussion

The key findings of this study were that 1) half of preterm infants born ≤30 weeks demonstrated sensory processing disorder, 2) the impact of medical and sociodemographic factors on later sensory processing disorder could not be isolated in this study, and 3) there were early behavioral markers, specifically more signs of stress and more sub-optimal reflexes, that were related to later sensory processing disorder. We were able to accept our hypothesis that sensory processing disorder would be common in this very preterm sample and that there would be early neurobehavioral signs of sensory processing disorder.

Our finding that 50% of the sample had sensory processing disorder is similar to previously reported incidences ranging between 39% and 52% [5, 13, 14]. This could relate to how preterm infants fail to receive the protective and natural sensory experiences during the second and third trimesters of pregnancy. Instead, their fragile and underdeveloped sensory system must mature outside the womb [22, 34, 35]. While developing in the NICU, preterm infants are exposed to adverse stimuli such as heel lance procedures, long periods of intubation, along with intense light and sound exposures. These altered sensory experiences, during a period of neurodevelopmental vulnerability and fragility, can result in sensory processing disorder, which can include heightened responses or being less responsive to stimuli. Altered sensory experiences and responses early in life can affect development and

impact how children will respond to their environments and participate in occupations later in life [10, 16].

Although we hypothesized that more medical interventions would be related to sensory processing disorder, this could not be isolated in this study, possibly due to the limited sample size. Studies with larger sample sizes have identified medical conditions and interventions; such as longer periods of intubation, longer NICU stay, and lower gestational age as predictors for developing sensory processing disorder between three to five years of age [14, 36]. Other studies have also defined male sex and white matter brain injury to be predictors of sensory processing disorder, but no relationships with sex or cerebral injury were observed in the current study [37].

Infants who were identified as having sensory processing disorder at four to six years of age had more stress and sub-optimal reflexes at term equivalent age. It is unclear whether these early markers are indicative of the impairment that followed, or if early impairment identified on the neurobehavioral exam resulted in altered sensory experiences leading to the subsequent sensory processing disorder. This is the first study to investigate relationships of early neurobehavior to later sensory processing disorder. Early neurobehavioral exams can be useful in identifying early alterations in development so that targeted therapeutic interventions in early infancy can be implemented. This study indicates that infants who demonstrate more stress and sub-optimal reflexes are at risk and, therefore, may benefit from innovative therapies that address early sensory skills. Early occupational therapy interventions that address the sensory needs of the young infant have been documented [29].

In contrast to previous studies of sensory processing disorder among preterm infants, this study utilized an observational tool of sensory processing instead of a parent report measure. Due to the known limitation of parent report measures [38], use of an observation-based measure likely provided a less biased, and more standardized assessment of the child's sensory related behaviors. However, the SPA, which was used in this study, continues to undergo development and does not yet have established normative data. Therefore, interpretation of the sensory processing results for each child was limited. Future studies utilizing the SPA would benefit from supplementing the assessment with a parent questionnaire on sensory processing, to ensure that findings in the clinical evaluation are consistent with the child's typical daily behaviors. Further research and development of the SPA, including better differentiating sensory versus behavioral responses, could improve our understanding of sensory processing disorder and enable future research. Limitations include having a small sample size, which can result in a Type I error. However, this study sets the stage for future work that can explore these questions in an appropriately powered sample. Another limitation is the amount of time between NICU discharge and the sensory assessment. Participants likely had many sensory exposures, from NICU discharge through early childhood, prior to returning to receive sensory testing. All the exposures and experiences during the first four to six years were not measured, but may have contributed to their sensory processing performance. Future studies could benefit from measuring sensory exposures more frequently following discharge from the NICU. This study was also limited by a large repertoire of testing that was conducted prior to the sensory processing testing at

age four to six years. It remains unclear how this impacted the child and if the SPA outcome would be different if it were the sole test administered on the day of follow-up testing.

The findings of this study support the continued need for research of preterm infant's sensory processing. Preterm infants are at an increased risk for experiencing developmental challenges, including developing a sensory processing disorder. More research in this area could aid early identification and determine the need for targeted interventions to optimize outcomes and participation in childhood activities.

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Highlights

- Half of preterm infants born ≤30 weeks demonstrated sensory processing irregularity
- Impact of medical and sociodemographic factors could not be isolated
- Early behavioral markers, specifically stress and sub-optimal reflexes, were related to later sensory processing irregularities

Table 1

Descriptives of the Sample.

	Total Sample N (%) or Mean (SD) or Median (IQ range)	*Among those without Sensory Processing Disorder N (%) or Mean (SD) or Median (IQ range)*	*Among those with Sensory Processing Disorder N (%) or Mean (SD) or Median (IQ range)*	** P value
EGA	26.4 ±1.9	26.8 ±1.4	26.1 ±2.4	.32
NEC	3 (9%)	1 (33%)	2 (67%)	.54
PDA	21 (66%)	10 (48%)	11 (52%)	.71
Sepsis	11 (34%)	4 (36%)	7 (64%)	.26
*Brain Injury	7 (22%)	4 (57%)	3 (43%)	.60
Hours on Oxygen	1,518 (678–2,318)	1,091 (678–2,168)	1,716 (510–2,322)	.49
Days on CPAP	3 (1–9)	2.5 (1–12.3)	5 (0.3–9)	.87
Days on Vent (n=26)	3 (1.0–26.3)	2 (1.0–12.3)	6 (0.3–40.5)	.52
Days on TPN	20.5 (11–37.5)	26 (13.3–36.5)	15 (9.0–54)	.40
Surgery	15 (47%)	6 (40%)	9 (60%)	.29
Insurance (Public)	15 (47%)	9 (60%)	6 (40%)	.29
Marital Status at Birth (Single)	18 (56%)	11 (61%)	7 (39%)	.15
Maternal Age at Birth	29.4 ±6.9	27.4 ±5.3	31.4 ±7.8	.10
Sex (Male)	14 (44%)	6 (43%)	8 (57%)	.48
Race (African American)	12 (38%)	5 (42%)	7 (58%)	.47

* Brain injury was defined as the presence of either a grade III–IV intraventricular hemorrhage, cystic periventricular leukomalacia, and/or cerebellar hemorrhage.

** P-value is from investigating factors related to sensory processing disorder using independent samples t-tests, nonparametrics, and chi-square analyses.

Abbreviations:Estimated gestational age (EGA), nectrotizing enterocolitis (NEC), patent ductus arteriosus (PDA), continuous positive airway pressure (CPAP), total parenteral nutrition (TPN)

Table 2

SPA subscales and subscores and incidence of sensory processing disorder.

	** Median (IQ)	*** N (%) with Abnormal Scores
Hyperresponsiveness	.11 (0–0.2)	6 (19%)
Social	.5 (0–1.0)	5 (16%)
Non-social	0 (0–0)	2 (6%)
Hyporesponsiveness	1.5 (1.0–2.0)	7 (22%)
Social	1.7 (1.0–2.0)	4 (13%)
Non-social	1.3 (1.0–2.0)	6 (19%)
Defensive	0 (0–0.1)	8 (25%)
Social	0 (0–0)	1 (3%)
Non-social	0 (0–0.2)	0 (0%)
*Sensory Processing Disorder**		16 (50%)

* Sensory processing disorder defined as having any one score above the 75th percentile in any SPA subscale or subscore.

** Higher scores indicate poorer performance or more abnormality.

*** Abnormal scores were determined from frequencies and distributions of raw scores within each SPA subscale and subscore, with abnormal scores identified among those scoring >75th percentile on each subscale or subscore.

Table 3

Relationships between early neurobehavior and sensory processing disorder.

NNNS Subscore *n=26*	Total Sample Mean (SD)	Among those without Sensory Processing Disorder Mean (SD)	Among those with a Sensory Processing Disorder Mean (SD)	* P-value
Habituation	7.7 ±1.0	8.1 ±0.3	6.5 ±2.1	.29
Orientation	2.5 ±0.8	2.2 ±0.7	3.5 ±0.1	.43
Handling	0.6 ±0.2	0.6 ±0.2	0.7 ±0.1	.24
Quality of Movement	3.7 ±0.8	3.8 ±0.9	3.3 ±0.2	.29
Self Regulation	4.4 ±0.9	4.2 ±1.0	4.8 ±0.5	.81
Sub-Optimal Reflexes	7 ±2.4	6.3 ±2.1	9.5 ±2.1	.04*
Stress	0.4 ±0.1	0.3 ±0.1	0.4 ±0.1	.02*
Arousal	3.7 ±0.8	3.8 ±0.9	3.2 ±0.1	.81
Hypertonia	1.9 ±1.3	1.8 ±1.4	1.9 ±1.1	.73
Hypotonia	0.7 ±0.6	0.6 ±0.5	0.7 ±0.7	.61
Asymmetry	1.8 ±1.6	1.3 ±1.4	3.5 ±0.7	.16
Hyperexcitability	4.1 ±2.6	4.4 ±2.9	3.0 ±1.4	.41
Lethargy	8.0 ±2.9	8.1 ±3.0	8.0 ±3.0	.94

* p value is from exploring the relationships between early neurobehavior and sensory processing disorder using independent samples t-test using $p<.05$.