Received: 22 June 2023 | Accepted: 7 January 2024

DOI: 10.1111/1440-1630.12935

FEATURE ARTICLE

Australian Occupational Therapy Journal | WILEY

Sensory processing in children with Paediatric Acute-onset Neuropsychiatric Syndrome

Michelle J. Newby[1,2] | **Shelly J. Lane**[2,3] | **Kirsti Haracz**[2] | **Janice Tona**[4] | **Kerrin Palazzi**[5] | **David Lambkin**[5]

[1]Stepping Stones Therapy for Children, Charlestown, New South Wales, Australia

[2]Occupational Therapy, College of Health, Medicine and Wellbeing, School of Health Sciences, University of Newcastle, Callaghan, New South Wales, Australia

[3]Department of Occupational Therapy, Colorado State University, Fort Collins, Colorado, USA

[4]Rehabilitation Science Department, University at Buffalo, Buffalo, New York, USA

[5]Hunter Medical Research Institute, New Lambton Heights, New South Wales, Australia

Correspondence
Michelle J. Newby, PO Box 455, The Junction, NSW 2291, Australia.
Email: michelle@steppingstonesforchildren.com.au

Funding information
This study received no specific grant from any funding agency in the public, commercial, or not-for-profit sectors.

Abstract

Introduction: Previous research indicates that children with Paediatric Acute-onset Neuropsychiatric Syndrome (PANS) experience sensory reactivity differences that impact occupational performance. The purpose of this study was to determine whether there are differences in sensory reactivity in these children across two different time points; during exacerbation and during remission, using the Sensory Processing Measure (SPM) Home-Form. The study also sought to investigate whether children with PANS experience sensory differences during remission periods, when compared with SPM Home-Form norms.

Methods: A two-period bidirectional case-crossover design was used, and an online assessment was conducted to measure sensory reactivity. Parents of children aged 4.6 to 13.1 years with a diagnosis of PANS were recruited from various sites across Australia, USA, England, Ireland, Scotland, Canada, and New Zealand. The SPM Home-Form was used to measure sensory reactivity at two time points, when PANS was in remission (T-R) and in exacerbation (T-E). Study entry was permitted at either T-E or T-R. Participant exacerbation status was monitored over a maximum 12-month period, and a follow-up SPM Home-Form was sent when a change in exacerbation status was indicated. A linear mixed model was used to assess the difference between SPM Home-Form norm-referenced scores during exacerbation and remission.

Results: The study included 82 participants, with 80 providing data at study entry, and 27 providing data at follow-up. Results showed a statistically significant decline in performance across the SPM Home-Form domains of Hearing, Social Participation, Planning and Ideas, and Total Sensory Systems T-scores during exacerbation when compared with remission data. Results also demonstrated atypical sensory reactivity across Vision, Hearing, Touch, Balance and Motion, and Total Sensory Systems domains during periods of remission compared with SPM Home-Form norms.

Conclusion: This study found that children with PANS experience significant sensory reactivity differences during exacerbation and remission across multiple sensory domains, with a decline in performance during exacerbation. Where there are occupational performance challenges, occupational therapists should consider administering sensory assessments so that effective intervention plans addressing the unique sensory reactivity needs of children with PANS can be developed.

KEYWORDS
Neuropsychiatric Disorder Associated with Streptococcal infections (PANDAS), occupational performance, Paediatric Acute-onset Neuropsychiatric Syndrome (PANS), paediatric mental health, paediatric occupational therapy, sensory modulation, sensory processing, Sensory Processing Measure (SPM), sensory reactivity

1 | INTRODUCTION

Paediatric Acute-onset Neuropsychiatric Syndrome (PANS) and Paediatric Auto-immune Neuropsychiatric Disorder Associated with Streptococcal infections (PANDAS) are related conditions that follow a similar course (Rea et al., 2021; Sigra et al., 2018; Swedo et al., 2012). For the sake of brevity, these conditions will be collectively referred to as PANS. PANS is characterised by an abrupt onset (i.e., no symptoms to full symptom onset and intensity within 24–48 hours) of Obsessive Compulsive Disorder (OCD) or eating restrictions, or both concurrently (Chang et al., 2015; Swedo et al., 2012). These symptoms must be accompanied by at least two or more comorbid neuropsychiatric or somatic symptoms that may include sensory or motor abnormalities; anxiety; emotional lability or depression; irritability, aggression, or severely oppositional behaviours; developmental/behavioural regression; deterioration in school performance; and somatic signs and symptoms such as sleep disturbances, enuresis, or urinary frequency. PANS typically follows a relapsing remitting course, with the child experiencing a sudden exacerbation of symptoms, leading to a loss of function following exposure to a trigger (Frankovich et al., 2017). For some children, PANS follows a chronic progressive course, whereby they continue to experience ongoing symptoms and do not return to their pre-exacerbation level of function (Frankovich et al., 2017). Triggers are varied and can include infections such as Influenza or Group A Streptococcus. Triggers can also be non-infectious in nature such as environmental (e.g., mould) or metabolic changes. Although there are no studies to date that describe the prevalence of PANS within the general population, prevalence has been reported across a number of different paediatric cohorts, including children with tic disorders ($n = 80$, 11% meeting abrupt symptom onset criteria for PANS) (Singer et al., 2000), children attending an outpatient OCD clinic ($n = 136$, 5% meeting PANS criteria) (Jaspers-Fayer et al., 2017), and children attending an eating disorder clinic ($n = 100$, 52% meeting PANS criteria) (Aman et al., 2022). PANS exacerbations have a sudden and pervasive impact on the child, potentially limiting participation and performance across the range of childhood occupations, including activities of daily living, education, play, and leisure (Tona et al., 2017). Performance across these occupations is limited by the impact of the condition on a range of person factors, including emotional coping, cognition, muscle strength and endurance, and sensory processing (Tona et al., 2017).

Sensory processing also impacts children's participation and performance in all occupations, including school occupations, play, sleep, activities of daily living, and social participation (Bar-Shalita et al., 2008; Foitzik & Brown, 2018; Roberts et al., 2018; Sleeman & Brown,

Key Points for Occupational Therapy

- Children with PANS present with significant differences across multiple sensory domains, with a decline in performance during exacerbation.
- Sensory reactivity may vary depending upon whether a child with PANS is in an exacerbation or remission state.
- Occupational therapists should include a comprehensive sensory assessment where children with PANS are presenting with occupational performance challenges.

2022). Sensory processing is a neurological process that involves accurately detecting or registering sensory information from the environment or within one's own body, interpreting or discriminating that sensation, and finally, organising an appropriate response (Bundy & Lane, 2020). Vision, hearing, taste, touch, and smell are the most commonly recognised senses; however, proprioception (awareness of body position in space) and vestibular (awareness of balance and movement) are also considered key sensory systems supporting occupational performance (Bundy & Lane, 2020). Sensory systems work together in an integrated manner to enable the individual to adapt and respond appropriately to environmental demands (Bundy & Lane, 2020). When this process runs smoothly, children can participate in occupations that are purposeful and meaningful to them; however, when there are sensory processing differences, difficulties with independence and participation can occur (Anzalone & Lane, 2012). The two primary indicators of meaningful differences in sensory processing are sensory reactivity differences and dyspraxia (Bundy & Lane, 2020). Sensory reactivity differences are reflected in over-responsivity or under-responsivity to sensory stimuli (Bundy & Lane, 2020). Dyspraxia occurs when there is a disruption in sensory perception within the somatosensory (i.e., touch and proprioception) and/or vestibular senses, and impacts the child's ability to accurately initiate, plan, organise, and execute movement patterns (Bundy & Lane, 2020; Cermak & May-Benson, 2020).

Sudden onset changes in sensory reactivity have been reported in children with PANS, with indications of over-reactivity to touch, including hypersensitivity to items of clothing and textures of foods; smell; taste; sound; light; and visual stimuli (Demchick et al., 2020; Frankovich et al., 2015; Gromark et al., 2019). The most detailed study to date describing sensory reactivity differences experienced by children with PANS was conducted by Tona et al. (2017). In a non-standardised online survey of parents of 111 children with PANS, the investigators found that most participants reported some degree of sensory over-reactivity, including increased sensitivity to touch, and to a lesser degree with taste, smell, proprioception, hearing, vision, and vestibular senses (Tona et al., 2017). Demchick et al.'s (2019) qualitative, phenomenological study of six families, examining family quality of life in the context of raising children with PANS aged from 7 to 13 years, also reported challenges with sensory over-reactivity including challenges with dressing due to tactile hypersensitivity. Gromark et al. (2019) conducted a systematic evaluation of 45 Swedish children aged between 3 and 13 years who met the PANS diagnostic criteria and found that half of their study participants reported sensory over-reactivity based on their developmental history, including hypersensitivity to touch (including clothing), sounds, and light. Sensory-seeking behaviours have also been described, such as a pervasive need to touch particular textures or objects (Swedo et al., 2017). Although there is an emerging body of evidence indicating that children with PANS experience sensory processing changes that create challenges, there is currently no published evidence using a norm-referenced data collection tool. Therefore, our understanding of sensory reactivity in children with PANS, and how this impacts functional performance during and between PANS exacerbations, is yet to be firmly established. Additionally, it is noteworthy that the signature characteristic of PANS (i.e., an acute onset of neuropsychiatric and somatic symptoms, including sensory changes) distinguishes this group from other populations treated by occupational therapists, who generally present with more consistent sensory difficulties (Swedo et al., 2012).

Addressing the functional impact of sensory reactivity challenges is a core component of paediatric occupational therapy practice; therefore, it is imperative to current and future occupational therapy practice to clearly define whether PANS exacerbations impact sensory reactivity. This will provide important information to guide intervention planning for children with PANS, with consideration of potential fluctuations in performance during remission and exacerbations. Although there is little research from the field of occupational therapy in this area, Tona et al. (2017) have suggested that a flexible approach to intervention be considered when working with these children. They propose that remediation strategies are most appropriate during periods of remission, whereas interventions that emphasise accommodations and adaptations are most appropriate during periods of exacerbation. Although these recommendations appear to be an appropriate model of care for children with PANS, they are based on non-standardised parent-report. Therefore, additional research is necessary to gain a more comprehensive understanding of performance during periods of remission and exacerbation, which will be fundamental in guiding occupational therapy treatment.

This study sought to answer two key questions:

1. Do children with PANS experience differences in sensory reactivity, as measured by the SPM Home-Form scores, during periods of remission and exacerbation?
2. Do children with PANS who are in remission experience differences in sensory reactivity, based on SPM Home-Form scores, when compared with the norm-referenced sample of the SPM Home-Form?

2 | METHODS

2.1 | Ethics approval

Prior to data collection, ethical approval was obtained from the University of Newcastle, Human Research Ethics Committee (approval number H-2019-0284).

2.2 | Study design

This study was a part of a broader study using a two-period bidirectional case-crossover design. An online assessment was used, whereby sensory reactivity was measured using the Sensory Processing Measure (SPM) Home-Form (Parham & Ecker, 2007) at two separate time points:

- Time-point Exacerbation (T-E): When the child had experienced a PANS exacerbation during the past month;
- Time-point Remission (T-R): When the child had not experienced a PANS exacerbation during the last month.

Participants were able to enter the study at either time point.

2.3 | Participants and recruitment

Participants were parents of children aged 4.6 to 13.1 years with a medical diagnosis of PANS. Age was originally limited to 5 to 12 years as this is the specified age range for the SPM Home-Form. It is noteworthy that our study included one participant who was 12 years old at the commencement of the research, but who turned 13 during the data collection period. There was an additional participant who was 4.6 years at study entry and subsequently turned 5 during the data collection period. Although these 2 participants fell outside the anticipated age range for some time points, their scores were not outliers and were therefore included. Participants were recruited via networks of professionals working with children with PANS at various sites across Australia, the United States of America (USA), England, Ireland, Scotland, Canada, and New Zealand. Participants were limited to these countries as they were English speaking and there were multidisciplinary PANS networks who were known to the researchers in each of these countries. Medical and allied health professionals employed in private practices, who were known to assess and treat children with PANS, were informed about the study via email and social media posts. Those indicating an interest in the study were provided with flyers including a website link that could be given to parents/carers of children with PANS. The link provided information about the study and how to indicate intention to participate. Parents/carers of children with PANS were also recruited via informative social media posts that included the link for parents to indicate intention to participate. In addition, snowballing sampling methodology was utilised, whereby parents and carers of children with PANS were invited to inform other parents and carers of children with PANS about the study.

As sensory sensitivity forms part of the restricted and repetitive section of the diagnostic criteria for Autism Spectrum Disorder (ASD), children with an additional ASD diagnosis were excluded from this study (American Psychiatric Association, 2013). This allowed for observations to be made between sensory reactivity differences that are specific to the child's PANS diagnosis. Children with neuromotor conditions were excluded as they were not a population of interest for this study. We included children who had experienced a PANS exacerbation in the past 6 months, hypothesising that these children would be most likely to experience an exacerbation and remission state during the course of the study.

2.4 | Data collection tools

2.4.1 | The Sensory Processing Measure Home-Form (SPM Home-Form) (Parham & Ecker, 2007)

The SPM Home-Form was used to collect information regarding the child's sensory processing abilities and identify sensory differences that create challenges for the child. It is a norm-referenced measure used to identify sensory processing patterns in children aged 5–12 years within the home environment. Widely used by paediatric occupational therapists, the SPM Home-Form measures sensory processing across vision, hearing, touch, body awareness (i.e., proprioception), and balance (vestibular) senses. In addition, the assessment includes Social Participation and Planning and Ideas scales. Although not specific senses, these domains each represent higher level cognitive functions that are strongly impacted by sensory input and processing. The Social Participation domain is a measure of the child's participation in social activities in the home and community, including interactions and conversational skills with friends, parents, and other significant adults. The Planning and Ideas scale is a measure of praxis, which is the ability to conceptualise, plan, and organise motor responses to complete novel tasks (Parham & Ecker, 2007).

The SPM Home-Form consists of 75 items and is completed by the child's primary home-based caregiver. Items

are rated using a 4-point Likert Scale, based on the frequency of the observed behaviour (i.e., *Never*, *Occasionally*, *Frequently*, and *Always*). The assessment provides norm-referenced T-scores across the following scales: Social Participation, Vision, Hearing, Touch, Body Awareness, Balance, and Planning and Ideas. Items within each of the scales are designed to determine vulnerabilities or differences that create challenges within that specific sensory system. A Total Sensory Systems score can also be obtained and is the composite score of the five individual sensory scales (Vision, Hearing, Touch, Body Awareness, and Balance), plus additional items that represent the child's taste and smell processing. The Total Sensory Systems score can be used to determine whether there are overall differences that create challenges in a child's sensory processing ability (Parham & Ecker, 2007).

The SPM Home-Form T-scores make it possible for a child's behavioural and sensory functioning to be accurately compared with a normative sample of typically developing children (Parham & Ecker, 2007). Three interpretative ranges are provided, including *typical range*, *some problems*, and *definite dysfunction*. T-scores that fall within the *typical range* (T-score range of 40–59) indicate that the child's behavioural and sensory functioning is similar to that expected from typically developing peers. Scores within the *some problems* range (T-score range of 60–69) indicate mild to moderate difficulties in behavioural or sensory functioning when compared with peers. Scores within the *definite dysfunction* range (T-score 70–80) indicate that the child has significant sensory processing challenges that may have a noticeable impact on the child's daily function. SPM Home-Form scores within the *some problems and definite dysfunction* ranges are considered elevated and occupational therapy intervention may be indicated (Parham & Ecker, 2007).

Parham and Ecker (2007) report strong reliability for the SPM Home-Form. Internal consistency reliability estimates ranged from $\alpha = 0.77$ to 0.90 across all subscales and $\alpha = 0.95$ for the Total Sensory Systems. Test–retest reliability estimates ranged from $r = 0.94$ to 0.98 for each subscale and $r = 0.98$ for the Total Sensory Systems. SPM items were taken from previously peer reviewed assessments including the Evaluation of Sensory Processing and the School Assessment of Sensory Integration to enhance content validity (Parham & Ecker, 2007).

2.4.2 | Researcher developed surveys

A researcher-developed survey entitled 'About you and your child demographic survey' was used to collect demographic and clinical information, including the child's age, sex, and country of residence; parents' level of education; details regarding their child's initial PANS diagnosis; comorbid diagnoses; medical interventions; and experience with occupational therapy intervention. This survey was completed by participants upon entry to the study. The participant then indicated whether their child had experienced a PANS exacerbation in the previous month (T-E) or had not, indicating remission (T-R). Additional data regarding clinical characteristics, in-depth demographic information, and experiences with occupational therapy are outside the scope of this paper and will be included in later publications.

A further researcher-developed survey entitled 'Clinical information' was also used. This was a brief survey used to collect monthly clinical information regarding exacerbation status (i.e., T-E or T-R). If an indication was given that the child was experiencing an exacerbation, further details were collected regarding exacerbation symptoms, including behavioural and somatic symptoms. A rating of how different their child's behaviour and participation was when compared with their typical presentation was also included. This information was used to confirm that the child was in an exacerbation.

2.5 | Study procedures

Upon consenting to participate in the study, participants were emailed the 'About you and your child demographic survey' survey, the 'Clinical information' survey, and the SPM Home-Form. The questions were completed based on their child's performance over the previous month. The 'Clinical information' survey was then sent each month, to determine whether there had been a change in the child's exacerbation state. Once a change in exacerbation state was indicated, participants were asked to complete a second SPM Home-Form, again based on their child's performance over the previous month. Therefore, the SPM Home-Form was completed at two different time points (i.e., T-E and T-R). Once SPM Home-Form data were collected at T-E and T-R time points, participation in the study was complete.

2.6 | Data analysis

Data were analysed in SAS v9.4. An alpha level of 0.05 was specified for all tests and confidence intervals, and two-tailed analysis was conducted. Descriptive analysis was undertaken with results presented as count (%), median (Q1, Q3), and means (standard deviation). Descriptive statistics for SPM Home-Form scores were grouped by whether the child was at the T-E or T-R time points.

A linear mixed model was chosen to assess the difference between SPM Home-Form norm-referenced scores during (T-E) and between (T-R) PANS exacerbations, as this allows for analysis of data that are non-independent, as participants could provide data for both T-E and T-R timepoints. To ensure reliability of the linear mixed model, fixed effects for exacerbation status (i.e., during or between PANS exacerbations), order (i.e., exacerbated at study entry or at follow-up; continuous), and time (i.e., number of months from study entry to follow-up) and a random effect for participant were included. Model assumptions were checked and found to be acceptable. Intraclass correlation coefficients (ICCs) were calculated using a null model to quantify within and between person correlation (presented in the Supporting Information).

3 | RESULTS

3.1 | Participants

An initial 153 participants consented to participate in the study; 62 did not meet inclusion criteria, an additional eight did not provide data, and one withdrew consent, resulting in data from 82 participants at the start of the study. Two participants provided consent at study entry but did not provide initial demographic data. They both later went on to complete the SPM Home-Form, so have been included in the analysis, but excluded from study entry descriptive statistics. Twenty-one of the participants provided data at both T-E and T-R time points, and the remaining participants provided data at either T-E or T-R, for a total of 87 surveys.

As displayed in Table 1, the mean age of children included at study entry was 9.5 years, with most being male (63%). The mean age at the first onset of PANS symptoms was 5.1 years, and the mean age of PANS diagnosis was 6.8 years. Most participants were from the USA (54%); however, there were participants from all countries included in the study (i.e., Australia, USA, England, Ireland, Scotland, Canada, and New Zealand). PANS was the only diagnosis for 44% of children, with others reporting additional diagnoses of OCD (25%), Attention Deficit Hyperactivity Disorder (ADHD) (24%), Oppositional Defiance Disorder (ODD) (13%), Tourette Syndrome (15%), and others (including a small number with Generalised Anxiety Disorder, Mood Disorder, Psychosis, Type 1 Diabetes, Dysautonomia, Eosinophilic Esophagitis, and/or seizures). Of the 80 children at study entry, 59% were reported to be in an exacerbation (see Table 2 for further details). The median time to study follow-up from study entry was 3 months.

TABLE 1 Demographics.

Characteristic	Class/Statistic	Total ($n = 80$)
Age at study entry	Mean (SD)	9.5 (2.4)
Age at first onset of PANS	Mean (SD)	5.1 (2.4)
Age at diagnosis	Mean (SD)	6.8 (2.5)
Gender	Male	50 (63%)
	Female	29 (36%)
	Other	1 (1.3%)
Country	Australia	11 (14%)
	USA	43 (54%)
	England	8 (10%)
	Ireland	1 (1.3%)
	Scotland	1 (1.3%)
	Canada	12 (15%)
	New Zealand	4 (5.0%)
Additional diagnoses	OCD	20 (25%)
	ADHD	19 (24%)
	ODD	10 (13%)
	Tourette Syndrome	12 (15%)
	Other diagnosis	19 (24%)
	PANS only	35 (44%)

Abbreviations: ADHD, attention deficit hyperactivity disorder; OCD, obsessive compulsive disorder; ODD: oppositional defiant disorder.

3.2 | SPM Home-Form PANS exacerbation analysis

As shown in Table 3, the mixed models analysis revealed that there was a statistically significant increase in the Social Participation, Hearing, and Planning and Ideas domain scores and the T-scores at T-E, indicating a significant deterioration in sensory reactivity across these domains during exacerbation. The largest impact at T-E across the individual domains was observed on the Social Participation scale, with almost one full standard deviation difference (9.8 T-score difference; $p \leq .001$). For all models, effects for order and time were statistically non-significant (estimates presented in the Supporting Information).

3.3 | SPM Home-Form PANS T-R and normative data analysis

As shown in Table 3, study participants at T-R demonstrated LSMeans T-scores within the *typical range* (T-

TABLE 2 Participant exacerbation characteristics.

Flare status	Study entry ($n = 80$)	Follow-up ($n = 27$)	Total ($n = 107$)
Exacerbation	59 (83%)	11 (41%)	70 (71%)
Remission	12 (17%)	16 (59%)	28 (29%)
Missing	9	0	9

TABLE 3 Least squares means (95%CI) for SPM standardised scores ($n = 91$ SPM Home-Forms).

SPM HOME scale	LSMean score T-R (95%CI) $n = 27$	LSMean score T-E (95%CI) $n = 60$	Difference estimate (95%CI)	*p*-value
TOT	62.8 (60.1, 65.5)	65.4 (62.9, 67.8)	2.6 (0.5, 4.6)	0.016*
SOC	57.4 (53.7, 61.1)	67.2 (64.2, 70.1)	9.8 (5.5, 14.1)	<.001*
VIS	60.8 (57.6, 64.1)	63.1 (60.1, 66.1)	2.2 (−0.2, 4.7)	0.067
HEA	60.6 (57.0, 64.2)	64.6 (61.5, 67.8)	4.1 (0.8, 7.3)	0.018*
TOU	63.6 (60.7, 66.6)	65.9 (63.3, 68.6)	2.3 (−0.1, 4.7)	0.055
BOD	57.9 (54.5, 61.4)	60.2 (57.1, 63.3)	2.3 (−0.5, 5.1)	0.099
BAL	60.0 (56.5, 63.5)	61.4 (58.3, 64.5)	1.4 (−1.4, 4.3)	0.307
PLA	59.4 (55.5, 63.3)	63.7 (60.5, 66.9)	4.3 (0.1, 8.5)	0.046*

Abbreviations: BAL, balance and motion scale; BOD, body awareness scale; CI, confidence interval; HEA, hearing scale; PLA, planning and ideas scale; SPM, sensory processing measure; SOC, social participation scale; TOT, total sensory systems scale; TOU, touch scale; VIS, vision scale.
*Results that achieved statistical significance.

score 40–59) across Social Participation (57.4), Body Awareness (57.9), and Planning and Ideas (59.4) scales but fell within the *Some Problems* range (T-score 60–69) across the domains of Vision (T-score 60.8), Hearing (60.6), Touch (63.6), Balance and Motion (60.0), and the Total Sensory Systems (62.8).

4 | DISCUSSION

We aimed to address two key questions in this study: (1) Do children with PANS experience differences in sensory reactivity, as measured by the SPM Home-Form scores, during periods of remission and exacerbation; (2) Do children with PANS who are in remission experience differences in sensory reactivity, based on SPM Home-Form scores, when compared with the norm-refenced sample of the SPM Home-Form. Our findings indicate differences in sensory reactivity between periods of PANS exacerbations and times of remission. Additionally, we observed atypical sensory reactivity within our study cohort even during remission phases. Differences were noted across multiple domains of the SPM Home-Form. Our study revealed that during periods of PANS exacerbation, children experienced a statistically and clinically significant decline in performance across the SPM Home-Form domains of Hearing, Social Participation, Planning and Ideas, and the Total Sensory Systems T-scores, when compared with their performance during remission periods. Previous studies have also indicated that children with PANS experience issues related to sensory reactivity. Tona et al. (2017) investigated the impact that PANS exacerbations had on occupational performance in a sample of 111 children with a mean age of 10 years at study entry and with a diagnosis PANS or PANDAS. These investigators found that over 71% of the children in their study reported 'sensory defensiveness', now more commonly termed sensory over-reactivity, during periods of exacerbation, including extreme tactile defensiveness impacting the ability to wear clothes. Our findings are also consistent with those of Calaprice et al. (2017) in their study of 698 children with PANS and PANDAS with a mean age of 11.8 years at study entry. These investigators reported sensory defensiveness to light, sound, and/or clothing in 79% of participants. However, our study is unique as it is the first to measure sensory reactivity in children with PANS using a more objective assessment tool, thus providing a more reliable measure of sensory reactivity in this group. Considering the well-documented association between sensory reactivity and limitations in various occupational performance areas such as academics, play, self-care, and social interactions (Bar-Shalita et al., 2008; Foitzik & Brown, 2018; Roberts et al., 2018; Sleeman & Brown, 2022), it is advisable to include a thorough evaluation of sensory reactivity for children diagnosed with

PANS. This information can be used to develop occupational therapy treatment programs that are tailored to the child's individual sensory needs.

Our study also revealed that the children on average experienced atypical sensory reactivity, not just during exacerbations, but also during remission periods. Atypical sensory reactivity was observed specifically within the SPM Home-Form domains of Total Sensory Systems, Vision, Hearing, Touch, and Balance and Motion. Bhattacharjya's (2013) thesis involving retrospective parental recall of children's functioning before and after a PANS exacerbation revealed that children with PANS in their sample experienced an observable change in sensory reactivity following exacerbations, with reactivity becoming more atypical. Bhattacharjya (2013) described a relationship between this change in sensory reactivity and an associated deterioration in occupational performance across various domains, including academic activities, leisure pursuits, and activities of daily living for many children with PANS. Importantly, their study revealed that these participants did not return to their baseline levels of occupational performance after completing a PANS exacerbation (Bhattacharjya, 2013). As all the participants in our study reported previous PANS exacerbations, it is possible that with each previous PANS exacerbation, sensory reactivity differences may have become increasingly more significant, resulting in atypical sensory reactivity even during periods of remission. Consequently, our study findings may not accurately capture this group's true baseline sensory functioning, as by the time the remission data were collected, the sensory capabilities of the children involved could have already been impacted. Further research is necessary to confirm this hypothesis. This would ideally consist of a longitudinal study objectively measuring sensory reactivity and functioning at each remission period to determine if functioning returns to baseline after an exacerbation or if functioning deteriorates after each exacerbation. Noted sensory processing difficulties during remission may mean that occupational therapy intervention at times of remission may still be warranted.

It was not possible to determine whether taste and/or smell reactivity challenges or differences were present in our study population due to the lack of individual T-scores for taste and smell processing in the SPM Home-Form. Taste and smell sensitivities have been linked to atypical feeding behaviours and social participation challenges in children (Lane et al., 2010; Smith et al., 2005). For example, Smith et al. (2005) found that almost half of the children without a previous diagnosis of autism, cerebral palsy, or Down's syndrome in their study who were identified as picky eaters refused food due to its smell. Similarly, Lane et al. (2010) reported a correlation between extreme taste and smell sensitivity and social communication challenges. As food restriction and social participation challenges are common symptoms in children with PANS, it is important for occupational therapists to understand how potential taste and smell sensitivity may be contributing to these areas of difficulty. Therefore, we recommend that future research adopt norm-referenced assessment tools that provide specific results relating to taste and smell processing. This will help us to gain a more thorough understanding of the effect of taste and smell reactivity on children's occupational performance during PANS exacerbations.

It is noteworthy that challenges in sensory reactivity reported in our study fell on average within the *Some Problems* category of the SPM Home-Form. This indicates only mild to moderate impairments in sensory functioning relative to peers (Parham & Ecker, 2007). This finding is in contrast with those of Tona et al. (2017) and Demchick et al. (2019) who reported cases of severe disturbances in sensory reactivity, specifically related to tactile sensitivities that rendered clothing intolerable. For example, a participant in Demchick et al.'s (2019, p. 191) study on family quality of life in the context of raising a child with PANS stated that it took a parent '...three hours to get out of the house due to her daughter's severe sensory sensitivity with clothing ...'. Similarly, a participant from Tona et al.'s study (2017, pp. 6) noted that their child with PANS was '...unable to wear any clothes for days because of extreme tactile defensiveness ...'. The difference between our findings and those reported by Tona et al. (2017) and Demchick et al. (2019) suggests that there is an inherent variability in the sensory experiences of children during PANS exacerbations. Some children may experience only mild sensory symptoms during periods of exacerbation, whereas others may experience sensory symptoms that are severe and incapacitating. Similarly, some periods of exacerbation may result in more severe symptoms than others. Parents may be more likely to recall and report more extreme exacerbation experiences in retrospective studies such as those conducted by Demchick et al. (2019) and Tona et al. (2017). As our current study indicates, children with definite changes in sensory processing that are not extreme should not be overlooked in evaluation and intervention.

Our study provides evidence that children with PANS experience atypical sensory reactivity, with a significant deterioration in function during PANS exacerbations, thus impacting occupational performance in a fluctuating pattern. Although efficacy of occupational therapy intervention for children with PANS is yet to be studied, our study findings offer a degree of validation for Tona et al.'s (2017) recommended occupational therapy intervention model for children with PANS, which advocated for

tailoring treatment to the child's exacerbation status. They suggest that during exacerbation periods, occupational therapy treatment should prioritise accommodations and adaptations, whereas remediation strategies should be reserved for periods of convalescence where tolerated, and remission (Tona et al., 2017). It will be important for occupational therapists to assess the effectiveness of this treatment approach for children with PANS. Nevertheless, in the interim, this approach appears to be supportive of the fluctuating pattern of occupational performance in this population of children.

This study's notable strengths include its prospective design, allowing for the capture of a child's symptoms as they manifest over time. Furthermore, this study is the first of its kind to employ a norm-referenced assessment tool to measure sensory reactivity in children both during and between PANS exacerbations. However, an important limitation of this study stems from recruitment and data collection occurring in 2020, a time when the COVID-19 pandemic had a global impact, leading to constraints on participant numbers and retention. A further limitation of this study was the disproportionate number of participants who provided data at T-E ($n = 60$) compared with T-R ($n = 27$). The underlying cause for the limited data available during T-R remains unclear; however, it is plausible that caregivers might have been less inclined to engage in the study when their child was in remission. The mean duration of 3 months between exacerbation and remission, as identified in our study, raises important considerations, particularly as families may find it easier to recognise the onset of exacerbations compared with remissions. This discrepancy suggests that what is perceived as remission may actually be an incomplete return to pre-exacerbation functioning and therefore may be a potential limitation in our study. It would therefore be beneficial for future research to consider an additional assessment period, 3 months following the perceived remission. This approach could offer deeper insights into the trajectory of sensory symptoms post-exacerbation.

5 | CONCLUSION

The results of this study confirm that children with PANS experienced significant differences during exacerbation and remission in Total Sensory System, Hearing, Planning and Ideas, and Social Participation domains of the SPM Home-Form, with a deterioration in performance observed during PANS exacerbations. A thorough sensory assessment to identify sensory reactivity differences in children with PANS should be included in occupational therapy evaluations, and consideration should be given to the potential fluctuations based on whether a child is in exacerbation or remission. It may be appropriate to repeat sensory evaluations where a child's exacerbation state changes. This will enable occupational therapists to more accurately understand how sensory reactivity differences may be impacting on functional performance and to develop more effective intervention plans.

AUTHOR CONTRIBUTIONS

Michelle J. Newby developed the study protocol and methodology, recruited and followed up with participants, interpreted data, and wrote the manuscript. Shelly J. Lane, Kirsti Haracz, and Janice Tona supervised the development of the work, helped in study design and interpretation, and edited the manuscript. Kerrin Palazzi assisted with data analysis and edited the manuscript. David Lambkin assisted with data analysis.

ACKNOWLEDGEMENTS

We would like to thank all the participants and health care providers who supported recruitment to this study. Open access publishing facilitated by The University of Newcastle, as part of the Wiley - The University of Newcastle agreement via the Council of Australian University Librarians.

CONFLICT OF INTEREST STATEMENT

The authors have no conflicts of interest to declare.

DATA AVAILABILITY STATEMENT

We appreciate the journal's commitment to data sharing. We are fully supportive of this initiative and are committed to making our data available to the scientific community. All relevant data associated with this research will be made openly available as requested.

ORCID

Michelle J. Newby https://orcid.org/0000-0002-3072-8663

Kirsti Haracz https://orcid.org/0000-0002-1321-8899

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SUPPORTING INFORMATION

Additional supporting information can be found online in the Supporting Information section at the end of this article.

How to cite this article: Newby, M. J., Lane, S. J., Haracz, K., Tona, J., Palazzi, K., & Lambkin, D. (2024). Sensory processing in children with Paediatric Acute-onset Neuropsychiatric Syndrome. *Australian Occupational Therapy Journal*, *71*(4), 475–485. https://doi.org/10.1111/1440-1630.12935