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Feasibility of a Research Protocol to Investigate the Effect of the TherapressureTM Program Using Salivary Cortisol

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Abstract

Background: There is an absence of high quality research to support the use of the Therapressure Program^{TM}. This pilot study aimed at developing appropriate research protocols to investigate the effectiveness of the Therapressure Program^{TM} on the stress response in children with sensory overresponsivity.

Method: A one-group pre-test/post-test repeated measures design was conducted using a convenience sample. Six children (6-8 years of age) with sensory overresponsivity received 14 consecutive days of the Therapressure Program^{TM} by their parents at home. Parents concurrently collected salivary cortisol samples from their children.

Results: Children with sensory overresponsivity displayed both hyper- and hypo-cortisolism at baseline. All of the children's cortisol levels shifted toward a normative range after intervention. Aspects have been identified related to the data collection protocol.

Conclusion: When testing children with sensory overresponsivity who are constantly activating their stress response system, we raise awareness of the need to check for both hyper- and hypo-cortisolism during statistical analysis. Preliminary pilot data may also show modulation of sympathetic arousal following the intervention. Further research is warranted and recommendations are made related to data collection protocols.

Keywords
Feasibility study, Wilbarger protocol, Therapressure ProgramTM, sensory overresponsivity, cortisol, HPA-axis, stress, pediatrics

Credentials Display
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Background and Literature Review

Parents of children who are hyper-responsive to sensation often seek occupational therapy for their children. Wilbarger and Wilbarger (2014) termed this hyper-responsiveness to sensation as sensory defensiveness, described as "a constellation of behaviors related to aversive or defensive reactions to non-noxious stimuli across one or more sensory systems" (p. 2). Sensory defensiveness is classified by Miller, Anzalone, Lane, Cermak, and Osten (2007) as sensory overresponsivity (SOR), described as the individual's responses to sensation as faster, more intense, or more prolonged when compared to individuals with typical responses. Under Miller's proposed nosology for the classification of sensory processing disorder, SOR is displayed as a sub-type of the disorder. Examples of clinical presentations for SOR include overly defensive responses to everyday stimuli, such as perceived overly dramatic responses to having one's hair washed or avoiding certain stimuli, such as lights that seem too bright or noises that seem too loud. At present, SOR is not recognized in the International Statistical Classification of Diseases and Related Health Problems, tenth revision (ICD-10, World Health Organization, 2015) or the Diagnostic and Statistical Manual of Mental Disorders, fifth edition (DSM-5, American Psychiatric Association, 2013). It is, however, used clinically to characterize individuals who display patterns of overresponsivity to sensory stimuli as measured by clinical observation and caregiver report.

The Wilbarger protocol provides a prescribed set of guidelines for the treatment of SOR in children (Wilbarger & Wilbarger, 2014). The protocol includes three components that are individualized to a child's treatment needs: (a) client assessment and education of SOR, (b) a set of sensorimotor activities known as the Sensory Diet, and (c) professionally guided intervention. Professionally guided intervention can include either the Therapressure Program™ and/or the Oral Tactile Technique (Wilbarger & Wilbarger, 2014). The data collection protocols for this study were administered around the Therapressure Program™.

The Therapressure Program™ administers deep touch pressure and proprioception as two consecutive components. First, the child's hands, arms, back, legs, and feet are brushed with a non-scratching Therapressure Brush™ manufactured by Clipper Mills. According to Wilbarger and Wilbarger (2014), application of the correct brushing technique is essential to avoid noxious stimuli. Subsequently, joint compressions are applied to the hands, elbows, shoulders, trunk, hips, knees, and ankles to stimulate proprioception. Parents or caregivers are trained by a certified therapist to administer the Therapressure Program™, which should be applied consistently every 90 to 120 min during the child's waking hours (or eight to 10 times per day) for a number of weeks. Intervention duration is individually determined to meet the child's specific needs (Wilbarger & Wilbarger, 2014).

Researchers theorize that the effectiveness of the Wilbarger protocol is due to its ability to assist the central nervous system (CNS) to

modulate its responses to sensory input (Kimball et al., 2007). Sensory modulation is the ability to respond "to sensory input with behavior that is graded relative to the degree, nature, or intensity of the sensory information" (Miller, Anzalone et al., 2007, p. 136). This is achieved through harmonious interaction between the CNS's ability to decrease its responsivity to familiar stimuli ("habituation") and increase its responsivity to significant or potentially dangerous stimuli ("sensitization") (Kimball et al., 2007). It is proposed that failure to modulate sensory stimuli can lead to a physiological stress response, in turn causing anxiety and stress-related behaviors (Reynolds & Lane, 2009). Thus, researchers suggest that the Wilbarger protocol influences the CNS by modulating the physiological stress response (Kimball et al., 2007).

The stress response in humans involves an interaction between two major physiological systems: the sympathetic-adrenal-medullary (SAM) system and the hypothalamic-pituitary-adrenal (HPA) axis (McCarthy et al., 2009). The SAM system is controlled by the sympathetic branch of the autonomic nervous system, providing rapid, phasic responses to stressful events (e.g., sympathetic arousal, the fight-flight reaction) through the release of epinephrine and norepinephrine. In contrast, the HPA-axis, through the release of cortisol and other "stress" hormones, provides a slower and steadier response to stress (Hanrahan, McCarthy, Kleiber, Lutgendorf, & Tsalikian, 2006).

In recent studies, physiological characteristics in the autonomic nervous system were found to support SOR as an independent clinical condition (Ben-Sasson, Carter, & Briggs-Gowan, 2009), and sympathetic arousal was shown to be one of its diagnostic markers (Miller, Anzalone et al., 2007). Studies have also shown that children with SOR have higher levels of electrodermal reactivity, a stress-response measure of sympathetic arousal, as well as slower habituation when compared to controls (Ahn, Miller, Milberger, & McIntosh, 2004). It is suggested that children with SOR are in a constant state of sympathetic arousal due to an inability to self-regulate and restore homeostasis (Schaaf et al., 2010).

The HPA-axis can be measured through salivary cortisol levels, which is advantageous for children's research because it is non-invasive, can be detected in small concentrations, and can be collected by non-health professionals (McCarthy et al., 2009). Salivary cortisol is produced in a typical diurnal fashion, with a peak just before waking and an evening nadir. This pattern is fixed in children by 4 to 6 years of age (Sumner, Bernard, & Dozier, 2010). Typical values for 4 to 10 year olds range from a mean of 0.301 μg/dL at 8 am to 0.197 μg/dL at midday to 0.119 μg/dL at 3 pm (McCarthy et al., 2009). Day to day variability of cortisol measurements in children is not well established, as most studies of cortisol levels in children employ only a small number of collections (Gunnar, Kryzer, Van Ryzin, & Phillips, 2010; McCarthy et al., 2009; Sumner et al., 2010).

Kimball et al. (2007) conducted a pilot study to investigate the effectiveness of a Wilbarger-based procedure on HPA-axis reactivity in four boys with sensory defensiveness.

The procedure consisted of single applications of brushing and joint compressions, once per week for 4 weeks, at the commencement of scheduled therapy sessions. Salivary cortisol samples were collected immediately before and 15 min after each procedure. The findings indicated that the boys modulated their salivary cortisol levels after each procedure, which, in theory, modulated their arousal. The analysis was based on what appeared to be normal cortisol levels for each child, because normative age-matched data was unavailable at the time of the study. The authors recommended that future research administer the Wilbarger protocol as it was designed and collect cortisol in natural environments for children.

The results of a recent systematic review of the Wilbarger protocol indicated that there is weak and inconsistent evidence for its effectiveness with children (Weeks, Boshoff, & Stewart, 2012). This conclusion was drawn from the low number and poor methodological quality of available studies on this topic, rather than the presence of high quality evidence to refute its effectiveness (Weeks et al., 2012). In the absence of high quality research, it is acknowledged that subjective anecdotal evidence, from occupational therapists and parents, support the efficacy of this approach for children in reducing their behavioral responses to environmental stimuli (Kimball et al., 2007). Although there is evidence to suggest that children with SOR experience higher states of sympathetic arousal than typical individuals, we are not aware of any study that has investigated the effectiveness of the Wilbarger protocol, as prescribed, in modulating children's arousal levels.

This pilot study is the first phase of a larger program of mixed methods research on the Wilbarger protocol. The primary aim of this study was to assess the feasibility of a data collection protocol implemented by parents in the home environment, while concurrently administering the Therapressure Program™. The secondary aim was to determine if the preliminary pilot data from this study warranted a full-sized trial to investigate the effectiveness of the Therapressure Program™ on the arousal levels of children with SOR. The results of this pilot study will provide recommendations for conducting a larger scale study.

Method

Research Design

A quasi-experimental, one-group pretest/posttest repeated measures design (Shadish, Cook, & Campbell, 2002) was used to investigate the effectiveness of the Therapressure Program™ on salivary cortisol levels in children with SOR. The participants served as their own controls due to the inter-individual variability of salivary cortisol (Kelly, Young, Sweeting, Fischer, & West, 2008) and the small sample size advised for pilot studies (Connelly, 2008). This pragmatic approach measured the effects of the participants engaging in their usual treatment protocol in their natural environment to replicate real-life conditions and allow the results to be generalizable to everyday practice. Ethical approval was gained from the University of South Australia's Human Research Ethics Committee. Informed consent for child and parent participation was obtained.

Participants and Recruitment

Child participants were recruited from a private practice specializing in pediatric occupational therapy in metropolitan Adelaide, South Australia. The treating occupational therapist was experienced in the assessment of SOR, certified in the Therapressure Program[TM], and was the sole therapist involved in this study. The parents were trained in the administration of the Therapressure Program[TM] during group sessions delivered by the occupational therapist through demonstration, verbal instruction, and the opportunity to practice the technique on the occupational therapist. The first author (SW) provided study information and instruction on data collection protocols at the conclusion of these group sessions. Seven children were assessed for eligibility to participate in this study. Inclusion criteria for the children included: between 4 to 8 years of age, a primary diagnosis of SOR, appropriate for the Therapressure Program[TM] as identified by the occupational therapist, and first time receiving this intervention. The participants were intially screened by the occupational therapist for inclusion based on clinical testing through the use of the somatosensory component of the Southern California Sensory Integration Tests (SCSIT) (Ayres, 1972), tactile sensitivity reported in a parent questionnaire, information gathered during a parent interview, and clinical observations of the child's response to touch during play. Exclusion criteria for the children included: a diagnosis of Autism Spectrum Disorder (ASD), because they have complex, specific, and significantly different sensory profiles to children without ASD (Tomcheck &

Dunn, 2007), and loose teeth or medical conditions related to bleeding in the mouth that would affected cortisol analysis (Schwartz & Granger, 2004).

Instruments

Salivary cortisol. Salivary cortisol was used to measure HPA reactivity, as it is a frequently used measure for stress research in children (Gunnar, Talge, & Herrera, 2009). Assessment of cortisol via saliva is a well-established, valid, and reliable method to study stress and appears within five min of a stimulus (Kelly et al., 2008; Kimball et al., 2007). Timing during the day is crucial because cortisol levels rise after waking for 30 to 45 min, then fall across the remainder of the day. Approximately 1 mL of saliva was collected at each point in sterile containers with a larger (25 ml) diameter to make collection easy.

Sensory Profile Questionnaire. The Sensory Profile (Dunn, 1999) was used to correlate changes in stress response to changes in response to sensory stimuli. The Sensory Profile is a standardized questionnaire that the parent or caregiver completes to assess their child's sensory processing abilities and the effects of sensory processing on their child's daily functional performance. The Sensory Profile is a reliable measure and has obtained content, convergent, and discriminant validity. The full version was used in this study to provide a more comprehensive assessment (Dunn, 1999).

Calendar and record sheets. A calendar was provided that contained an overview of intervention and data collection time points to allow the parents to plan their daily activites for

the duration of the study and to cue the sample collection number to the correct day. Record sheets were provided to cue the parents on the correct timing and sequence of intervention administration and data collection per day. The record sheets required the parent to note detailed information, including the date and container number of the sample, the child's estimated time of waking, and the time of day each cortisol sample was collected, to inform cortisol analysis.

Intervention

The researchers devised the data collection protocol around the Therapressure Program™. Each child had already received assessment and education for SOR by the occupational therapist and had an individualized Sensory Diet incorporated into his or her daily routine prior to the commencement of this study. Children were included in this study at the point that they began the Therapressure Program™. The intervention included the parents brushing their child's fingers, hands, arms, back, legs, and soles of feet, followed by 10 joint compressions to the fingers, wrists, shoulders, hips, trunk, knees, and ankles. Intervention was administered every 90 to 120 min during the child's waking hours, for 14 consecutive days, during the South Australian school holiday periods for Terms 1 and 2 (April and July). The occupational therapist scheduled the Therapressure Program™ to coincide with school holidays to allow the parents the best opportunity to achieve treatment fidelity. Treatment fidelity was measured in two ways. First, the occupational therapist confirmed that the parents were correctly administering the technique during the group training session when they demonstrated the technique on the occupational therapist. Second, the parents were asked to note any missed applications of brushing and joint compressions during intervention and report this when interviewed in person by the first author (SW) immediately upon completion of the intervention. The parents were also asked to provide details of any potential co-intervention.

Data Collection

The first author (SW) instructed the parents on the data collection protocol and maintained contact with them throughout the study to provide support on data collection procedures and completion of record sheets; however, the first author (SW) remained independent of the therapeutic input (e.g., advice on the Therapressure Program™) that was provided by the occupational therapist. The occupational therapist and the participants were blinded to the study outcomes.

Baseline. A Sensory Profile questionnaire was completed by the parents at home prior to collecting physiological samples. Baseline cortisol samples were collected on the first Saturday and Sunday of the school holiday period. The parents were instructed to note their child's morning waking time for cortisol analysis. The parents were asked to collect a single salivary cortisol sample (1 mL) from their child at three time points per day, including a morning sample (30 to 45 min after the child participant woke), a before lunch sample, and a bedtime sample, to establish a baseline cortisol circadian rhythm pattern (Kelly et al., 2008). The parents recorded the time that the samples were collected and whether the morning sample was collected in 30

to 45 min of the child's waking time. The child participant was not to eat or drink anything (except for water to assist with saliva production) for 30 min prior to providing a sample and to stay seated with minimal activity to minimize assay error (Kryski, Smith, Sheikh, Singh, & Hayden, 2011; McCarthy et al., 2009). The parents stored saliva samples in the refrigerator, and the first author (SW) collected the samples at the conclusion of the baseline, intervention, and follow-up periods, immediately returning them on ice to the university laboratory. The parents collected salivary cortisol samples in the home environment for two reasons. First, it has been shown that true baseline increases and recoveries in cortisol are better measured for children at home, compared to laboratory settings where baselines are higher (Kryski et al., 2011). Second, it was not feasible for the parents to take their child to a laboratory setting during such an onerous intervention schedule.

During intervention. The parents were asked to collect a single salivary cortisol sample (1 mL) between 30 to 45 min after the child woke every second day of the intervention period. Data collection was scheduled in the morning due to the onerous nature of the Therapressure Program[TM], thus increasing the potential for compliance. Moreover, the highest production of cortisol occurs in the morning, thereby allowing for a greater potential to detect change.

Follow up. The follow-up salivary cortisol measurements were an exact repeat of baseline. The 14-day Therapressure Program[TM] finished on a Sunday with the children returning to school on Monday. The follow-up

measurements occurred on a Saturday and Sunday, which was 12 days after the completion of intervention, to allow time for treatment effect. The parents completed a Sensory Profile questionnaire immediately after collecting the final follow-up salivary cortisol sample.

Data Analysis

Cortisol. Prior to analysis, all frozen saliva samples were thawed, mixed, and centrifuged at 3000 x g for 10 min at room temperature. The supernatant was collected and used for analysis. Samples were analyzed for cortisol in duplicate with a competitive immunoassay specifically validated for the quantitative measurement of salivary cortisol (Salimetrics LLC, State College, Pennsylvania) according to the manufacturer's instructions, except that internal quality control material was used rather than that provided by the manufacturer. The absorbance of each sample was read at 450 nm with correction at 630 nm using a microplate reader (LabSystems Multiskan[®] Ascent 354 Microplate Reader; Thermo Fisher Scientific, Massachusetts, USA). The analytical sensitivity was 0.003 µg/dL and the calibration range was 0.012 – 3 µg/dL. Inter-assay coefficient of variation was 16% (0.052 µg/dL) and 12% (0.52 µg/dL). The analytical sensitivity was determined by the manufacturer by interpolating the mean minus two standard deviations for 10 sets of duplicates at 0 nmol/L standard. Determination of inter-assay coefficient of variation was performed in our laboratory by analyzing two levels of internal quality control material in 10 runs.

Descriptive anlayses were used to evaluate the feasibility of the parents and children completing the data collection protocol. Two methods were then used to determine if the preliminary cortisol data would warrant a full-size trial. First, diurnal cortisol profiles were produced for each participant to compare baseline to follow-up time points to evaluate the effectiveness of the intervention. Diurnal cortisol profiles with children can provide a useful and stable measurement of HPA-axis activity over the course of a typical day (Rotenberg, McGrath, Roy-Gagnon, & Tu, 2012). Diurnal cortisol profiles were produced by calculating the means of the Saturday and Sunday samples for each of the three time points (morning, lunchtime, and bedtime) and charting a line to connect the three mean values. Each participant's diurnal profile was compared to normative salivary cortisol levels, which are based on recently published values from a large cohort of children aged 4 to 10 years (McCarthy et al., 2009). Second, cortisol levels from morning samples were charted from baseline, intervention, and follow-up periods for trend lines.

Sensory Profile. Tests for normality were run on each of the Sensory Profile subscores and quadrant items. Paired samples t-tests using a one-tail test were used to analyze complete sets of baseline and follow-up data on variables that were normally distributed. Wilcoxon signed-rank tests were used for non-normally distributed variables (DePoy & Gitlin, 2011). Power was set at 80% to avoid a Type II error with the significance level set at $p = < 0.05$.

Results

Participants

The families of all seven children who met the inclusion criteria and were referred to this study agreed to participate. Because of an ear infection, Participant 2 (boy) dropped out of the study on completion of baseline and did not receive intervention. All six of the participants who received intervention were Australian residents, with their families ranging from moderate to high levels of access to economic resources when assessing their postcodes against Socio-Economic Indexes for Areas (SEIFA) scores (Australian Bureau of Statistics, 2008). Participant 4, an 8-year-old boy, was unable to provide saliva samples after baseline due to distress caused by spitting. The parents were advised not to pressure their children, as this could confound cortisol concentration values. The results from the five participants who participated in the whole cortisol data collection protocol are reported.

At the time of baseline assessment, Participant 1 (girl) was aged 6.9 years, with a co-diagnosis of Fragile X syndrome but not ASD. Participant 3 (boy) was aged 6.4 years, Participant 5 (boy) was aged 8 years, Participant 6 (girl) was aged 6.2 years, and Participant 7 (boy) was aged 6.3 years. Participants 3, 5, 6, and 7 did not have co-morbidities. All of the participants were prescribed the Therapressure Program™ by their therapist primarily for tactile over-responsivity.

Intervention

All of the parents reported achieving treatment fidelity for the Therapressure Program™ when interviewed by the first author

(SW). Participants 4 and 7 received a single 60-min occupational therapy clinic session prior to day 3 of the Therapressure Program[TM], which the occupational therapist reported included sensory integration and motor techniques that were not intended to target sensory modulation difficulties. No other participant was receiving any additional intervention for the duration of this study.
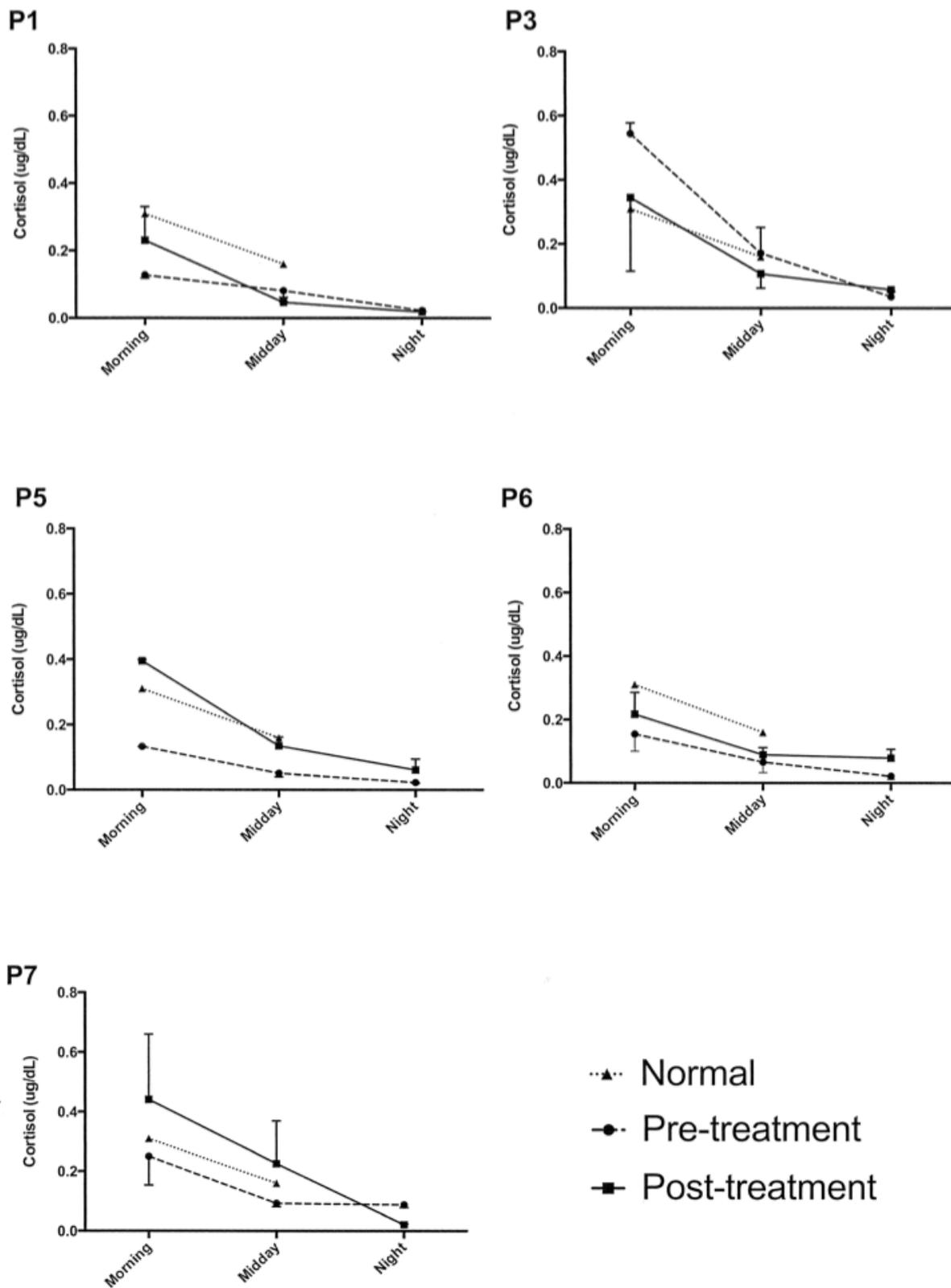
Cortisol

Completeness of saliva sample collection and adherence to data collection protocol. All of the participants collected complete sets of morning saliva samples at baseline and follow up, while four participants collected complete sets of lunchtime and bedtime samples. Only three participants could provide a complete set of morning samples during intervention; however, Participant 7 only missed one sample during this period. All saliva samples that were collected were of sufficient volume for analysis.

Feedback from the parents indicated that it was more difficult to collect the required volume for the morning samples than it was for the lunchtime and bedtime samples; however, lunchtime and bedtime samples were more difficult to collect due to busy daytime schedules. The data collection procedure was more difficult to adhere to in the second week of intervention due to the onerous nature of both intervention and data collection protocols. Two parents misinterpreted the data collection protocol. One parent missed collecting data during intervention, while the other collected too many samples and,

therefore, did not affect the results. All of the parents noted collecting morning saliva samples at the peak of the waking curve (30-45 min after their child woke), except for Participant 3 (on day 1 of intervention and day 1 of follow up) and Participant 6 (on day 1 of intervention).

Baseline diurnal cortisol profile. Prior to the intervention, Participants 1, 5, 6, and 7 all showed lower levels of salivary cortisol secretion at 30 to 45 min after waking than reported normative values for children aged 4 to 10 at the same time (McCarthy et al., 2009). These lower waking cortisol levels resulted in a flattened diurnal cortisol pattern as compared to similarly aged children. In contrast, Participant 3 showed higher than normal salivary cortisol levels prior to the intervention, resulting in an exaggerated diurnal cortisol pattern as compared to similarly aged children (see Figure 1).

Follow-up diurnal cortisol profile. After the administration of the 14-day Therapressure Program[TM], Participants 1, 5, 6, and 7, who showed abnormally low morning cortisol levels at baseline, showed a shift in waking cortisol levels toward a normative range. Participant 3, who showed abnormally high morning cortisol levels at baseline, showed a decrease in waking cortisol levels toward a normative range following intervention. Less change was observed for lunchtime and bedtime levels, with the exception of Participant 7, who experienced a significant decrease in bedtime cortisol concentration levels between baseline and follow-up (see Figure 1).

Legend. Mean waking, midday, and bedtime salivary cortisol levels (μg/dL + or – SEM) prior to and after administration of the 14-day Therapressure Program[TM] for participants 1 (P1), 3 (P3), 5 (P5), 6 (P6), and 7 (P7). Compared to normative values for children aged 4 – 10 at the sames times of day (based on time since waking).

Figure 1. Pre and postintervention salivary cortisol secretion as compared to published normative ranges.

During intervention. No consistent or reliable trend lines could be established when charting morning salivary cortisol levels through the intervention period.

Sensory Profile. All variables were normally distributed, except for the quadrant item Sensation Seeking. The results of the paired t-tests showed statistically significant differences for the subscores of Auditory ($p = 0.04$), Touch ($p = 0.05$), and Modulation of Movement Affecting Activity Level ($p = 0.03$) and the quadrant item Sensory Sensitivity ($p = 0.02$). The results of the Wilcoxon signed-rank test for the quadrant item Sensation Seeking found no significant difference ($p = 0.20$). Power could not be reached because of the small sample size, indicating a larger sample size is necessary. Effect sizes were therefore not calculated.

Discussion

The aims of this study were to use the quantitative data to evaluate whether it was feasible for parents to concurrently implement a salivary cortisol data collection protocol and the Therapressure Program[TM] in the home environment. Moreover, we aimed to determine if the preliminary pilot data from this study warranted a full-sized trial to investigate the effectiveness of the Therapressure Program[TM] on the arousal levels of children with SOR.

Decades ago, Hans Selye demonstrated that chronic activation of the stress response system resulted initially in hyperactivity of the physiological stress response. However, sustained exposure to a stressor resulted in a "collapse" of the system which manifests as a failure to elicit much, if any, HPA-axis response to any challenge

(Selye termed this the "triphasic course of the stress response" in his General Adaptation Syndrome [Selye, 1984, p. 111]). In recent years, this progression of the stress response system through over- to under-response has been largely ignored, and researchers had assumed that the response can only increase (Sapolsky, 1992). However, recent research has identified circumstances in which the cortisol response is curtailed, and it is now recognised that HPA-axis dysfunction can occur in two forms: (a) a hyper-reactive overly sensitive system that over reacts or maintains a sustained response to many routine facets of daily life (Sapolsky, 1992), or (b) a hypo-reactive system that fails to elicit a significant stress response, even to a sustained challenge (Fries, Hesse, Hellhammer, & Hellhammer, 2005). Failures of past research to find differences in the HPA-axis may have arisen because the chronically stressed individuals were composed of a mixture of hyper- and hypo-responsive individuals, leading to mean levels of a similar magnitude to the unstressed comparison group. Investigation of how the HPA-axis changes with exposure to chronic stress is further complicated by the huge number of factors which interact with the stress response system (Kelly et al., 2008).

Nonetheless, a number of studies have now reported hypercortisolism associated with a variety of medical conditions, including fibromyalgia (Riva, Mork, Westgaard, Rø, & Lundberg, 2010), post-traumatic stress disorder (Bicanic et al., 2013), and chronic fatigue syndrome (Papadopoulos & Cleare, 2012). However, whether the altered stress response is a

causal factor or a result of the condition is unclear (Tak & Rosmalen, 2010). By contrast, hypocortisolism has also been identified in groups that are exposed to chronic stress, including children raised in Romanian orphanages (Gunnar & Vazquez, 2001) and other chronically stressful environments, such as being an ethnic minority (DeSantis et al., 2007). Based on the findings of this study, we hypothesize that children with SOR are constantly activating their stress response system, and given the information above, we raise awareness for the need to check for both hyper- and hypo-cortisolism when analyzing cortisol results in this population. We are unsure, however, whether this altered stress response at baseline is a causal factor of SOR or a result of SOR.

Moreover, the preliminary findings of this study may indicate that children with SOR modulated their cortisol levels toward a normative range following 14-days of the Therapressure Program[TM]. This, in turn, may suggest that the effect of the intervention resulted in a reduction of sympathetic arousal. However, this cannot be determined from the results of this study because of the small sample size. Nonetheless, these findings warrant the need for large scale testing, with a control group, to establish if the Therapressure Program[TM] does in fact reduce sympathetic arousal as theorized. Based on our findings, we recommend future research collect cortisol samples at baseline and follow-up time periods only, without sampling during intervention, as these time points achieved the highest compliance of data collection and were the results that showed the greatest potential to detect

a change in sympathetic arousal. We recommend scheduling 2 days of baseline and 2 days of follow up on Saturday and Sunday because weekends are easier for parents to comply with data collection across the day. Researchers should also take into account the differences between collecting cortisol in home versus in the school environments. Children with SOR can feel as if they have less control over their social and physical environments at school, as it can often be more stimulating and threatening than at home (Ben-Sasson et al., 2009). We are not advocating against collecting samples at school, but suggest that researchers may consider separating weekday (school) and weekend (home) samples in statistical analyses.

The MacArthur Foundation (2008) recommends collecting six salivary cortisol samples per day in adult studies, with four or five samples the minimum needed to obtain an accurate area-under-the-curve (AUC) measurement for that day. Moreover, it is recommended that data collection should be repeated for 3 to 4 days for reliable "trait" estimates of daily concentration levels (MacArthur Foundation, 2008). Taking into account the pragmatics of researching children and the associated costs of salivary cortisol analysis, we recommend four saliva samples be collected per day: at waking, 30 to 45 min after waking, 2.5 hours post waking, and at bedtime. The first three time points should provide sufficient information of the postawakening increase (or lack thereof), and when the final time point is included, a sound understanding of the AUC measurement. While there are no

recommendations for children, other child studies have used two consecutive sample days to obtain trait estimates of daily concentration levels in children (Gunnar et al., 2010; Sumner et al., 2010).

Although not part of the primary aims for this study, we administered the Sensory Profile as an outcome measure to see if it could detect any changes in the children's responses to sensory stimuli following intervention and if these changes correlated with changes in salivary cortisol. The Sensory Profile was not validated as an outcome measure (Schaaf et al., 2014) or subjected to test-retest reliability examination in the user manual (Ohl et al., 2012). While there has been preliminary evidence of test-retest reliability in some subscores of the Sensory Profile (Ohl et al., 2012), further larger scale testing is required before it could be recommended as a reliable outcome measure (Schaaf et al., 2014). At the time this study was designed and conducted, there were limited effectiveness studies available for interventions addressing sensory modulation disorders. Several of these pilot studies did, however, administer the Sensory Profile (or its short version) as an outcome measure (e.g., Hall & Case-Smith, 2007; Kim, Bo, & Yoo, 2012; Miller, Coll, & Schoen, 2007; Miller, Schoen, James, & Schaaf, 2007) showing some degree of positive improvement following intervention. Our study may not have detected changes in the Sensory Profile due to the lack of power from the small sample size, as reported in the results. Moreover, it may not be sensitive enough to detect meaningful changes at such a short interval period (4-week frequency), where the other studies used

8 to 10 weeks frequency between pre- and posttesting. This may be due to reporter bias at a short frequency because the Sensory Profile is a subjective caregiver report (e.g., parent familiarity with the rating scale, heightened awareness of the child's sensory behaviors from completing it the first time, or parents wish that they had scored differently the first time). More recent intervention studies have administered the Sensory Profile as part of a comprehensive evaluation and inclusion criteria for sensory processing disorder, but have avoided using it to measure outcomes until validated (Schaaf et al., 2014; Schaaf, Hunt, & Benevides, 2012).

Limitations

The results of this study related to the stress response of children need to be viewed in light of the original objective of the study: to develop appropriate research protocols for investigating the stress response of children. The results, therefore, need to be interpreted with caution because of the trial nature of the study, the small sample size, the single geographical location, and the single demographic group. Moreover, this study was conducted during an academic year. As ethical approval was gained at the end of the summer holidays, this vital recruitment period (3 months) was missed. Fewer families participate during the regular school year, as holidays are only two weeks in duration. Additionally, although the morning saliva samples were recorded as being collected between 30 to 45 min after their child woke, it is possible that there may have been occasions where parents were unaware of the exact time their child woke. This may alter the position of the cortisol sample on the

waking curve. In future studies, the parent could be asked if they woke up their child. Finally, the Therapressure Program™ was administered with a concomitant Sensory Diet. This study did not obtain details of the types of activities recommended for the Sensory Diet because it was not in the aims of the research; however, this should be included in a large scale study and the Sensory Diet should not commence prior to administration of the pretest measures.

Implications for Practice and Research

This study shows promising results to further pursue the link between cortisol as a stress response marker for the effectiveness of the Therapressure Program™. A full-size trial that requires a greater sample size should consider recruitment and data collection occurring during the longer holiday period and across multiple sites. It is also possible that recruitment may have to span multiple regional areas or states to obtain a sample size that reaches statistical power. Results from a well-designed study may lead to therapists having more effective sensory-based therapies and, therefore, improving allied health service delivery for children with sensory overresponsivity.

Conclusion

The article has reported on preliminary results related to the stress response of children. We raise the awareness of the need to check for hyper- and hypo-cortisolism in similar studies. Preliminary pilot data may show modulation of sympathetic arousal following the intervention; however, these results need to be interpreted with caution due to the small sample size. The evidence warrants further robust studies. This pilot study has identified a practical protocol for evaluating Therapressure Program™ effectiveness in reducing the stress response in children with SOR. Further refinement of the research protocols is recommended.

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