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Evaluating a sham-controlled sensory testing protocol for non-verbal adults with neurodevelopmental disorders: Self-injury and gender effects

Frank J. Symons, Ph.D.,
Department of Educational Psychology, University of Minnesota

Vicki Harper, M.A.,
Department of Psychology, J. Iverson Riddle Developmental Center

Satomi K. Shinde, Ph.D.,
Department of Communicative Disorders, University of Wisconsin-River Falls

Jamie Clary, M.A., and
Department of Psychology, J. Iverson Riddle Developmental Center

James W. Bodfish, Ph.D.
Center for Development & Learning, University of North Carolina – Chapel Hill

Abstract

Ambiguous or blunted responses to sensory and painful stimuli among individuals with severe intellectual disabilities and co-morbid communicative impairments put them at risk for having their experience of pain discounted and their expression of pain misinterpreted. Valid measurement procedures of behavioral expression are critical for this vulnerable group of individuals. We investigated a sham-controlled sensory testing protocol as an approach to guard against observer bias during non-verbal behavioral recording for individuals with intellectual disabilities. Participants were 44 (52% male) adults (mean age = 46, sd = 10) with moderate (14%) and severe to profound (86%) intellectual impairment. The facial behavior of the participants before, during, and after five sensory stimulation modalities (pin prick, light touch, deep pressure, cool, warm) was coded by three raters using the Facial Action Coding System (FACS). For each participant, their 5 active sensory trials were randomized with sham trials during which no stimulation was applied. Observers were blinded to active vs. sham stimulation status. FACS scores increased significantly during active sensory trials ($p < 0.05$) compared with sham trials. There were significant effects for gender with females more expressive than males ($p < 0.05$). There were also significant effects for the presence of self-injurious behavior (SIB) with individuals with SIB more expressive than individuals without SIB ($p < 0.05$). The results suggest that the procedure was valid (i.e., distinguished between active vs. sham sensory stimulation) and provides additional evidence that individuals with significant intellectual impairments are sensitive to tactile stimulation consistent with quantitative sensory testing protocols.

Corresponding Author: Frank Symons, Department of Educational Psychology, Education Science Building 56 River Road, University of Minnesota, Minneapolis, MN 55455, (t) 612.626.8697, (e) symon007@umn.edu.

1. Introduction

Measuring and evaluating pain in the presence of communicative and cognitive impairments is one of the most pressing but difficult requirements in the delivery of appropriate health care for individuals with intellectual and associated developmental disabilities (e.g., cerebral palsy) and related disorders (e.g., autism). As a group, individuals with intellectual impairments are vulnerable to a set of assumptions regarding their ability to experience[4] and express[1] pain leading, at times, to disastrous consequences.[3]

Promising results are emerging from a number of groups working independently on the problem of assessment and measurement of pain among pre- and non-verbal populations.[18, 23, 22] One strategy has been to rely on nonverbal indicators (e.g., gross motor behavior, facial expression, mood/affect, posture, heart rate, etc.) as candidate signs for pain. A routinely used procedure is to record individual responses before, during, and after a specific noxious event (e.g., needle stick associated with a vaccination or blood draw) and measure whether there are observable behavioral (e.g., facial musculature) or physiological (e.g., heart rate variability) changes time locked to the onset and offset of a noxious stimulus and not before or after.[20, 25, 26, 27] Based on this approach, it has been shown that individuals for reasons of age (neonate) or cognitive ability (dementia, intellectual disability) who cannot reliably self-report verbally are capable of displaying a pain response that corresponds to the timing of a noxious or painful event.[9, 13, 17, 18, 24] Extending this line of work, Defrin et al.,[10] used a quantitative sensory testing procedure in which a reaction-time free measure was used to provide the first evidence that heat-pain sensory thresholds were lower (i.e., sensitivity to noxious stimuli was greater) among a sample of individuals with mild levels of intellectual disability compared with no-disability control individuals (although cf. Hennequin et al.[15]).

Most approaches for measuring sensory behavior in general and acute pain expression in particular among nonverbal individuals rely on measurement procedures in which observers are not ‘blind’ to the application of the stimulus.[17, 24] Thus, observers may be aware of the timing of the stimulus (e.g., needle stick) and most likely the intensity of the experience which runs the risk of introducing bias into the procedure. The primary purpose of this preliminary study was to evaluate an approach for reducing observer bias by introducing a sham control procedure into a standardized sensory evaluation protocol. Facial activity was measured using facial action coding across five sensory modalities (pin prick, light touch, deep pressure, cool, warm) among a sample of adults with intellectual disability and associated communicative impairments. The specific aims were to test whether changes in facial activity were time locked to stimulus application (not before or after) and specific to active but not sham stimulus trials.

2. Method

2.1. Sample

Following Institutional Review Board approval from the J. Iverson Riddle Developmental Center Committee for the Protection of Human Subjects and the University of Minnesota Committee for the Protection of Human Subjects, and parental/guardian consent, a convenience sample of 44 participants (52% male) with moderate (n = 6), severe (n = 3), and profound (n = 35) intellectual disabilities was recruited from a regional residential care facility. The participant mean age was 46 years (Range 28 – 67, SD = 9.87). Eight individuals had severely limited verbal abilities and 36 were completely non-verbal. Level of intellectual disability (i.e., mental retardation) was obtained via chart review and based on DSM-IV criteria. Thirty-two individuals were ambulatory, 8 were non-ambulatory, and 4 were partially ambulatory. Individuals with known chronic illness or similar conditions

considered to be painful or associated with discomfort (e.g. gastroesophageal reflux) were excluded as were individuals recently (within one week) having experienced acute illness (e.g., influenza) or injury severe enough to warrant medical attention (e.g., limb fracture).

The sample was also characterized by individuals with (N = 29) and without self-injurious behavior (N = 15). A long standing but little tested assumption is that individuals with severe and chronic forms of self-injury are pain insensitive, but conceptual and practical problems have limited detailed study of pain in this clinical group.[2, 33] We used the sample composition as an opportunity to address further the issue of sensory expression and self-injury among individuals with severe intellectual disability by comparing facial expression differences during sensory testing between individuals with and without self-injury. The presence of self-injurious behavior (SIB) was confirmed by (a) staff ratings - SIB occurred at least weekly as determined by the SIB subscale of the Behavior Problem Inventory,[28] (b) tissue damage - presence of bruising, wounding, or scarring related to SIB as determined by the Self-Injury Trauma Scale,[16] (c) chronicity (i.e., exhibited for at least 12 consecutive months), and (d) treatment refractory (SIB continued to require formal behavioral and or medical/psychiatric intervention due to rate or potential for injury). Chi-square tests showed no significant differences in the demographic (age, IQ, gender) or clinical (treatment with psychotropic medication) characteristics of individuals with and without SIB.

2.2. Sensory Testing Procedure

Each participant was tested individually. During testing participants were seated on a chair in front of a medical examining screen located in a clinic room at the facility (Figure 1a). To minimize stress or anxiety, all participants were familiarized with the testing room and the sensory testing staff. A trained clinician applied five different modalities of sensory stimuli (pinprick, warm, cool, deep pressure, and light touch – described in more detail below) to the participants' exposed back in random order through a small hole in the screen. Each trial consisted of three time segments audibly signaled aloud by a tape recorder ("stimulus #," "now," and "record."). Each time segment was five seconds in duration and ran consecutively (Figure 1b). Stimuli were applied immediately after the audible signal "now." Pin-prick was the only stimulus not applied for the full five seconds. It was applied immediately after the audible signal "now" and was in contact with the participant for approximately one second. The five stimulus modalities were selected based on prior tactile sensory evaluation work to be consistent with standard quantitative sensory protocols.[29] To ensure that behavioral coders were blind to the application of the stimulus, sham trials were included during which each trial continued to be signaled in the standardized audible format but no stimuli were applied. In total, there were ten trials (5 active, 5 sham) per participant with trial order randomized.

2.3 Sensory Stimuli

The five sensory modalities included pin-prick, warm, cool, deep pressure, and light touch. Pin-prick was applied with a Neuropen (Owen Mumford) with a sterile neurotip for each participant. The pin was pressed until the gauge on the side indicator reached the midpoint and then released as quickly as possible (within one second). Warm was applied with a test tube filled with water heated from an electronic thermos set to maintain a constant temperature (40.0°C). The temperature of the water in the tube was measured before application to ensure consistent temperature at the time of application. The bottom of the tube was dried quickly and placed lightly against the skin for 5 sec. Cool was applied with a Tip Therm pen (metal end; US Neurologicals). The metal side of the instrument was placed lightly against the skin so that entire tip was in contact with the skin for 5 sec. The tip end is a ¾ inch diameter circle. Because the tip maintains room temperature it was cooler than the participant's body temperature. Deep pressure was applied with an algometer (Wagner

model FPK). The algometer had an adhesive bandage on the tip to prevent the cool metal contacting the skin during application. The tip that contacted the back was a circle with a ¼ inch diameter. Approximately 1.8 kg (4 lbs) of consistent pressure was applied for the 5 sec period. Light touch was applied with a monofilament instrument VF-1 (2.0 g). The tip of the filament was pressed against the back until the filament bent. The filament was held in the bent position for 5 sec.

Because of the range and degree of cognitive and communicative impairments in this sample, stimulus trials were modified to be time limited and of short duration (≤ 5 sec.) and only conducted once per participant unlike conventional quantitative sensory testing in which participants indicate when the stimuli are perceived via successively increasing intensities of stimulation (i.e., 'ramps'). Research staff (N = 4) were initially tested using the stimuli and subjectively reported that the stimuli could be 'felt' but that they were non painful with the exception of the pin prick which was reported to be 'felt' and 'mildly painful'. It is important to note that the protocol as implemented was not considered a test of noxious (i.e., 'painful') versus non-noxious (i.e., 'non-painful') stimulation because there was no attempt to determine definitively pain thresholds in this non-verbal clinical sample. There are individual differences in pain thresholds in the normal population, and there is no reason to discount similar differences among individuals with intellectual disabilities. With the exception of pin prick, we selected stimulus intensities to be in the non-noxious range based on published standards and manufacturer specifications and thus serve as a more conservative test of the protocol (i.e., all things being equal, non-noxious stimuli would be less likely to be detected).

2.4 Behavioral Measurement

2.4.1. Facial action coding system—The Facial Action Coding System (FACS) is an anatomically based measurement system in which trained coders identify the presence/absence, intensity, and temporal features of well-defined, discrete facial action units (FAU). Each action unit represents the movement of a single facial muscle, or in a few cases, a group of muscle strands that move as a unit.[8] Originally developed in 1978 by Ekman and Friesen[11] to study emotion expression (e.g., anger, sadness), FACS has been applied to study FAUs in relation to pain and its expression. A number of study findings consistently show that there are specific FAUs that provide reliable information about reaction to pain and its expression including individuals with intellectual disabilities.[7, 17, 25] Sixteen FAUs were selected for this study based on Defrin et al.[9] and LaChapelle et al.[17] including AU4 (brow lowerer), AU5 (upper lid raise), AU6 (cheek raiser), AU7 (eye lid tightener), AU9 (nose wrinkler), AU10 (upper lip raiser), AU17 (chin raiser), AU18 (lip pucker), AU25 (lips part), AU26 (jaw drop), AU27 (mouth stretch), AU43 (eyes closed), and AU50 (vocalization).

To capture facial expression, a camera was set up approximately 2 m away orthogonal to and focused directly on the participant's face/head and upper body (see Figure 1a). Digital video was used and converted to DVD for subsequent coding using Pro-Coder for Digital Video (PCDV).[35] PCDV is a software program designed to facilitate the collection of different forms of observational data from digital media files and includes a key-board driven coding platform that enables coders to scroll through designated time windows with playback options available. Behavioral events are marked for later quantification and analyses. FAU frequencies were coded based on presence (coded as 1) and absence (coded as 0) of occurrence of the specific FAU during an observational period. Three certified FACS coders independently coded the videos of participants during the sensory testing procedure. To avoid dependency among the coders, they were assigned randomly in pairs

across subjects. The FACS coders were blinded to active vs. sham trials, specific stimulus modality, and self-injury status.

2.4.2. Gross motor head behavior—Four gross-motor head/face behaviors were included and coded based on the results of Defrin et al.[9] The four pain behaviors included 'head turn,' 'head down,' 'head back,' and 'freeze.' 'Head turn' was coded when the orientation of the head was right or left on a vertical axis as defined in the FACS manual. 'Head turn' did not include head tilt and eye movement. 'Head down' was coded when the chin was pressed down as in the FACS manual. 'Head back' was scored when the chin was lifted up as defined by Defrin et al.[9] The operational definition of 'freeze' was no facial and body movement during the entire time segment (5 consecutive seconds) based, in part, on Defrin et al.[9]

2.5. Inter-Rater Agreement & Reliability

Thirty percent of the FACS coding sessions were selected randomly and the inter-rater agreement among the three FACS coders was evaluated by dividing agreements over agreements plus disagreements (A/A+D). Each sensory testing session per participant included 10 trials (5 active, 5 sham) with three time segments per trial (pre, during, post-stimulus). Agreements for raw frequencies of coded AUs were determined at the time-segment level (i.e., pre, during, and post-stimulus). For example, if Rater 1 and Rater 2 recorded the same AUs occurring during the same time segment during the same trial for a given participant then an agreement was scored. If Rater 1 and 2 did not record the same AUs occurring during the same time segment during the same trial for a given participant, then a disagreement was scored. The inter-rater agreement on the frequencies of FAUs from 30% of the FAU data randomly selected was 76% (Range = 38 – 96%) among the three FACS coders, consistent with levels reported in comparable studies.[6, 19] The final passing 'gold standard' score to be certified as a FACS coder is 70%. Our lowest agreement scores (38%, 45%) occurred for two specific participants and each low agreement score related to each observer differing by one FAU code (AU 25 mouth open). For each of these two participants, the independent observers made different initial decisions regarding 'normative state' as per the FACS manual recommendations. The disagreements were resolved through consensus and further video review for both participants for that specific AU code.

Inter-rater reliability for FACS coding was evaluated by calculating intra-class correlation coefficients among the three certified FACS coders for the total AU frequencies across all time segments and all stimulus (active and sham) trials. The average intra-class correlation coefficient was 0.76 (Range = 0.71 – 0.78). Specific intra-class coefficients between the three certified FACS coders were: 0.78 (Coder 1 & 2); 0.78 (Coder 2 & 3); and 0.71 (Coder 1 & 3).

Thirty percent of the data on the frequencies of operationally defined gross motor behaviors were selected randomly and the inter-rater agreement among the three raters (same formula) were evaluated by dividing the total agreement in frequency between two coders by total agreement plus disagreement in frequency between two coders and averaged. The interrater agreement on the frequencies of operationally defined gross motor behaviors of 30% of the data randomly selected was 90% (Range = 81 – 98.3%) among the three coders.

2.6. Data Reduction & Analyses

The sensory testing behavioral data were analyzed with SPSS 14.0 software. The data were analyzed by repeated measures ANOVA to test for FAU and gross motor behavioral differences between the active and sham sensory trials, as well as differences between males and females and individuals with and without self-injury. The variables compared (i.e.

dependent variables) were mean frequencies of FAUs and mean frequencies of operationally defined gross motor behaviors. The factors were the three time segments consisting of 'baseline,' 'stimulus,' and 'recovery,' (i.e. "stimulus," "now," and "record" as audibly signaled, respectively) and modality of stimuli provided (i.e. pin-prick, warm, cool, deep pressure, light touch, and sham). Before testing overall FAU and gross-motor differences between active versus sham trials, the FAU and gross-motor data from active and sham trials were compared between participants with significant (i.e., severe/profound) versus moderate intellectual disability. No significant differences were found and we used the entire sample for all subsequent analyses. Before testing group differences, the data from the sham trials were evaluated and reduced (a mean of the 5 sham trials was derived). First, the FAU/gross motor frequencies of the five sham trials were compared across participants and tested by two-way repeated measures ANOVA to test for any significant differences among the five sham trials. The factors were three time segments (i.e. 'baseline,' 'stimulus,' and 'recovery') and five sham trials. Second, the mean FAU and gross motor frequencies from the five sham trials for each participant was then calculated to compare active versus sham stimulation.

3. Results

3.1. FACS scores – Active vs. sham sensory stimulation

First, following the data reduction procedures described above, there were no differences in FAU frequencies for the five sham trials which were then pooled for subsequent analyses. There was a significant within-subject effect for mean FAU frequencies across the three time segments ($F_{[2, 86]} = 4.71$, $p < .05$). Post-hoc analysis showed that there was a significant increase between baseline and stimulus segment ($t_{[43]} = -3.55$, $p < .001$) and a significant decrease between the stimulus and recovery segment ($t_{[43]} = 3.69$, $p < .001$) for the active but not the sham stimulation trials. There was a significant interaction effect for active vs. sham and the three time segments ($F_{[2, 86]} = 3.88$, $p < .05$) (Figure 2a). The results indicate that the mean FAU frequency for the stimulus segment during active trials (M = 1.78 SD = 1.43) was significantly greater than the stimulus segment during sham trials (M = 1.52, SD = 1.46). There were no significant differences among the five different modalities (i.e. pin-prick, warm, cool, deep pressure, and light touch) ($F_{[4, 172]} = .37$, $p = .83$) (Figure 2b). There were no significant interaction effects for the three time segments and the five modalities ($F_{[8, 344]} = 1.39$, $p = .21$). We conducted an additional set of analyses based on the relative frequency of individual FAUs and tested a subset of AUs that consistently seem to show the greatest ability to differentiate painful from non-painful conditions (4, 6, 7, 9, 10, 25, & 43[26]). Based on this combination of AUs, there remained a significant within-subject effects for FAU frequencies across the three time segments ($F_{[2, 86]} = 3.32$, $p < .05$). There was a significant interaction effect for active vs. sham and the three time segments ($F_{[2, 86]} = 3.55$, $p < .05$). The mean FAU frequency during the active stimulus for the second time segment was 1.33 [SD = 1.02] whereas the mean FAU frequency during the sham stimulus during the second time segment was 1.03 [SD = 1.07]. Conversely, we examined a subset of AUs that have been less frequently related to pain and sensory response (AUs 5, 17, 18, & 50) and found no significant main or interaction effects for stimulus type (active, sham) or time segment.

3.2. Gross Motor Head Behaviors

Four gross motor behaviors (i.e. head turn, head down, head back, and freeze) were coded based in part on the results reported by Defrin et al.[9] Total frequencies of head movement (i.e. head turn, head down, and head back) and freeze were analyzed. First, frequencies of head movement and freeze from the five sham trials from each participant were tested by two-way repeated measures ANOVA with two factors, three time segments (i.e. 'baseline,' 'stimulus,' and 'recovery') and five sham trials to examine whether there were any

significant differences among the five sham trials. There were no differences in head movement across the five sham trials but there was a significant difference in freeze across the five sham trials ($F_{[2, 43]} = 3.72$, $p = .05$). Post hoc analysis showed that there were significant increases in 'freeze' between the baseline (M = .03, SD = .11) and stimulus (M = .05, SD = .16) segments and decreases in 'freeze' between the stimulus (M = .05, SD = .16) and recovery (M = .01, SD = .07) segments during sham trials. Second, total head movement and freeze means from the five sham trials from each participant were compared between active and sham trials. There were no significant differences in head movement across three time segments ($F_{[2, 86]} = .08$, $p = .92$). There was a significant difference for total head movement between sham (M = 1.25, SD = .09) and active stimulation (M = 1.19, SD = .09) ($F_{[1, 43]} = 4.27$, $p = .05$). Head movement during sham trials was significantly higher than during active stimulation across the three time segments. There were no significant differences in within-subjects effects for freeze across the three time segments ($F_{[2, 86]} = 2.89$, $p = .06$). There were no differences in freeze between sham trials (M = .03, SD = .02) and active stimulation (M = .02, SD = .01) (F [1, 43] = .85, p = .36). Freeze during sham trails was higher than during stimulus and recovery segments during active stimulation. Frequency of 'freeze' (M = .03, SD = .1) was minimal compared to occurrence of head movement (M = 1.22, SD = .65) across the three segments during sham and active sensory. There were no interaction effects for stimulation (sham v. active) by time segment ($F_{[2, 86]} = .62$, $p = .54$) for head movement or freeze ($F_{[2, 86]} = .75$, $p = .42$).

3.3. FACS - Gender effects

There was a significant group effect for gender (Male M = 1.37, SD = 1.3; Female M = 2.22, SD = 1.9 ($F_{[1, 13]} = 21.32$, $p < .05$) for overall FAU activity. There were no significant interaction effects for gender by segment, stimulus modality, or segment by modality. Individual FAUs for female participants were higher on average than those for males except for AU 6 (cheek raiser) and 10 (upper lip raiser) which were non-significantly different (Figure 3a, b).

3.4. FACS – Self-injury

For overall FAU activity, there was a significant main effect for the presence/absence of SIB ($F_{[1, 13]} = 14.04$, $p < .05$) (M = 1.24, SD = 1.4 [no SIB], M = 2.1, SD = 1.7 [SIB]). There were no significant interaction effects of SIB/no SIB by segment, SIB/no SIB by modality, and SIB/no SIB by segment by modality. Individual FAU frequencies for the SIB group were consistently and significantly higher than those for the non-SIB group except for AU 18 (lip pucker) and 50 (vocalizations) for which there were no differences (Figure 4a, b). To check on whether there were differences specific to the application of the active stimuli relative to baseline, the 'recovery' time segment (Segment 3) was excluded from analyses and a paired-sample t-test was used to examine the difference in AU activity between the baseline and active stimulus time segments for each group. There was a significant difference in AU activity between baseline and the active stimulus time segment for the SIB group ($t_{(28)} = 3.59$, $p < .001$) (Baseline Time Segment M = 1.75, SD = 1.4 vs. Active Stimulus Segment M = 2.1, SD = 1.5) but not for the no SIB group ($t_{(14)} = 1.13$, $p > .25$) (Baseline Time Segment M = 1.11, SD = 1.1 vs. Active Stimulus Segment M = 1.24, SD = 1.1).

4. Discussion

This study was conducted to evaluate a sham-controlled modified sensory testing procedure in a sample of adults with significant intellectual disability and associated communicative impairments. There was significantly greater facial activity associated with the time interval corresponding to active versus sham stimulation providing initial evidence for the protocol's

validity. Females were more expressive (i.e., showed significantly more facial activity) than males and individuals with chronic self-injury were more expressive than comparable individuals without self-injury.

Individuals with intellectual and related developmental disabilities are a vulnerable group for whom the experience of pain has historically been denied or discounted[32] and for whom detection can be difficult.[20] The results reported here are contrary to conventional wisdom concerning sensory insensitivity and are consistent with the emerging literature[18] providing an empirical basis to question longstanding assumptions regarding sensory sensitivity and pain among individuals with intellectual disability. Notably, the results from Defrin et al.[10] showed that when reaction-time independent measures are used (method of levels; MLE) during quantitative sensory testing individuals with intellectual disability are more not less sensitive to pain (heat-pain). Increased pain expression (consistent with lower thresholds) was also reported by Nader et al.[19] from a sample of children with autism during a painful event (needle stick) compared with control-individuals further suggesting that the experience and expression of pain may be elevated not blunted.

The overall results from this study specific to facial activity are also consistent with previously reported findings on facial activity from LaChapelle et al.[17] and Defrin et al.[9] both of whom showed reliable differences in facial expression as acute pain behavior corresponding to a noxious event (i.e.,. needle injection). Defrin et al.[9] provided additional evidence showing differential effects based on the level of cognitive impairment. In their comparison of two evaluation tools – FACS based on detailed facial coding based and the Non-Communicating Children's Pain Checklist – Revised (NCCPC-R) based on observer ratings of broader sets of behavior – they found that FACS was less sensitive to the expression of pain for individuals with more severe and profound intellectual disability levels (the NCCPC-R was sensitive regardless of level of impairment). In this study sample, we found evidence that FACS was sensitive to facial changes associated with our sham-controlled sensory testing protocol for individuals with severe and profound intellectual disabilities. There are at least two possible reasons for the discrepancy between our results and those reported earlier by Defrin and colleagues. First, in our protocol we used a binary approach to coding frequencies of FAUs (1/0) whereas Defrin et al.[9] used the sum total of the intensity scores of the FAUs. Coding intensity provides graded information about a signal whereas we coded its presence/absence which could have led to different results (see Prkachin and Craig[27] for issues specific to coding facial pain expression). Second, and the more plausible of the two, is that the context in which the data were generated was different. Defrin and colleagues collected observational data in a real-world clinical context with a single instance of a noxious stimulus (i.e., needle stick) whereas our approach was based on a relatively controlled environment and set of procedures with multiple calibrated sensory stimuli held constant across participants.

A number of study-specific limitations regarding procedures should be summarized to further place the results in the correct context. First, the approach we developed was not a threshold detection procedure and so the inferences should be limited to facial expression differences related only indirectly to pain behavior (i.e., FAUs were selected based on pain expression studies but it does not necessarily follow that we were capturing an individual's pain experience) and not taken as evidence of threshold effects because we were not measuring or quantifying pain thresholds, per se. Foremost we were interested in investigating whether the protocol would create a sufficient set of blinds against observer bias to have confidence that any observed facial activity differences would be attributable to sensory stimulation not artifact. Second, the sample was one of convenience and so the results are necessarily limited to the individuals reported on. Because we were interested in individuals with the most significant levels of intellectual disability we over-sampled for

their disproportionate representation in the study sample. Whether we would obtain the same results with a more heterogeneous sample would have to be empirically tested. Third, the sensory stimuli as applied were probably sub-threshold for pain given that they were applied briefly (~ 5 s) and only once for each individual (a comparison of our average FACS scores with Defrin et al.'s,[9] seems to confirm this as ours were considerably lower). Thus, unlike conventional quantitative sensory testing in which an individual's tactile sensory and then pain threshold can be established by successive application of gradually increasing stimulus intensity levels (i.e., 'ramps'), we were limited in our approach to capturing any changes in expression specific to onset of the stimulus. By including both a pre-, and post-stimulus recording interval, however, we were able to ascertain some evidence for the sensitivity (i.e., time-locked facial activity) and specificity (i.e., occurring during active stimulus application only) of the procedure. Last, the results from the gross motor head movement recording and analyses produced inconsistent findings that were somewhat ambiguous and difficult to interpret suggesting additional study may be necessary to help determine their utility as sensory/pain indicators in this population.

There is virtually no empirical evidence concerning gender effects and pain among individuals with intellectual and related developmental disabilities. In their study comparing two approaches to quantitative sensory testing, Defrin et al.[9] reported a moderate gender difference in pain thresholds with male thresholds somewhat higher than females. No gender by group (intellectual disability/no intellectual disability) difference was observed. In our sample, we found evidence for a gender effect with females showing more facial activity than males during sensory testing. Although this would be consistent with males having higher thresholds than females our observation should not be interpreted as a threshold effect for reasons outlined above. Our findings are consistent, however, with an emerging research literature documenting sex-related differences in both experimental and clinical pain with women experiencing higher prevalence rates of chronic pain disorders and greater pain sensitivity.[12, 36]

The observation of increased facial activity occurring among individuals with chronic self-injury relative to comparable individuals (age, gender, intellectual level) without self-injury conflicts with models of self-injury in which individuals are considered hyposensitive to pain.[29, 33] Although the overall effect suggests a general between group difference, results from the analyses specifically comparing baseline to active stimulus application for each group are consistent with the possibility that the SIB group was more not less reactive to the range of stimuli tested. This novel observation is contrary to most clinical observations which suggest that self-injury is associated with decreased sensitivity to pain and at least one treatment approach (i.e., opiate antagonist) includes restoring pain sensitivity as part of its hypothesized mechanism of action.[31] To the degree that facial activity corresponding to sensory/tactile stimulation is related in some way to pain reactivity, these results suggest that a subset of individuals with chronic self-injury may be hyper- not hyposensitive. At the very least, these results indicate that additional attention toward pain and sensory thresholds in relation to self-injury would be warranted. In our previous work, we found evidence consistent with the possibility of distinct subtypes of self-injury in relation to the presence or absence of chronic pain and the frequency and location of self-injury.[5] Specifically, children with intellectual and related developmental disabilities and chronic pain problems who also self-injured tended to do so in much more localized body areas than comparable children with self-injury but without chronic pain. Notably, for both groups, pain expression was intact and not blunted.

In summary, the assessment of sensory expression in general and pain in particular among individuals with intellectual disability with associated communicative impairments is among the most challenging and difficult problems faced by clinicians and researchers. As a group,

individuals with severe intellectual disability are highly vulnerable and our efforts to improve health and quality of life outcomes should reflect this. Important advances in the clinical measurement of pain behavior among vulnerable groups have occurred in the past decade[14] but there remain a myriad number of unanswered questions germane to understanding the nature of the pain experience and response among individuals with severe intellectual impairments. Findings from controlled attempts to characterize sensory thresholds[10] and the detailed architecture of pain expression[9, 17, 18] are beginning to build an empirical basis for overturning a long standing assumption regarding pain insensitivity. The results reported here provide additional evidence that individual's with significant intellectual disabilities reliably react to tactile sensory stimulation regardless of their verbal ability. That there can be objective measurement of sensory expression in highly vulnerable populations showing that sensory events reliably lead to the same kinds of nonverbal expression seen in persons without intellectual and associated developmental disabilities suggests that there is little reason in precluding such individuals from sensory and pain research designed to improve clinical practice or from ignoring such assessment as a part of routine clinical health and rehabilitative care.

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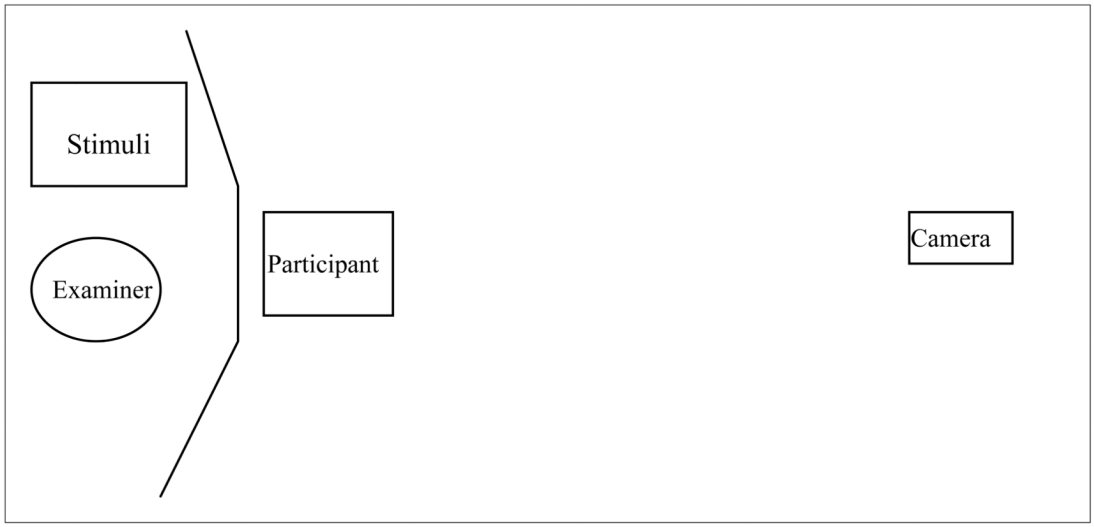
Perspective

This article presents a novel application of a modified approach to quantitative sensory testing for non-verbal adults with intellectual and developmental disabilities. This approach could be important in helping determine sensory issues related to tactile and nociceptive processes among a highly vulnerable group of individuals.

a.



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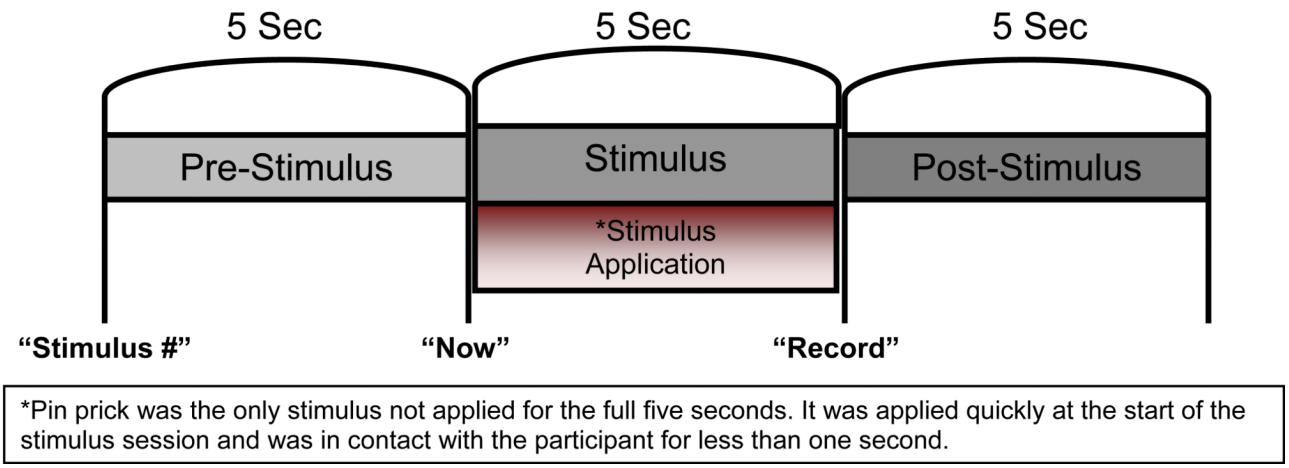
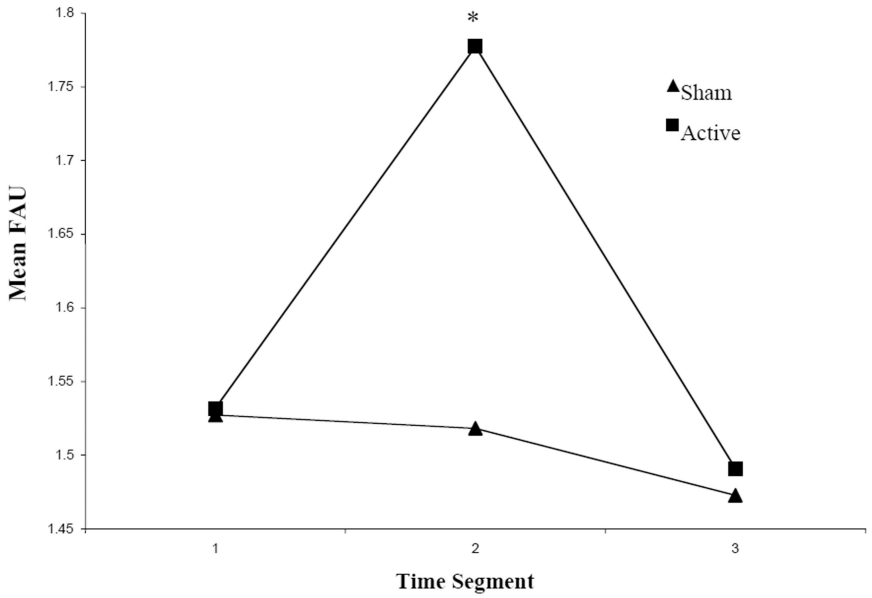


Figure 1.
(a). Physical arrangement during sensory testing protocol. (b). Schematic illustration of sensory protocol for each trial.

a.



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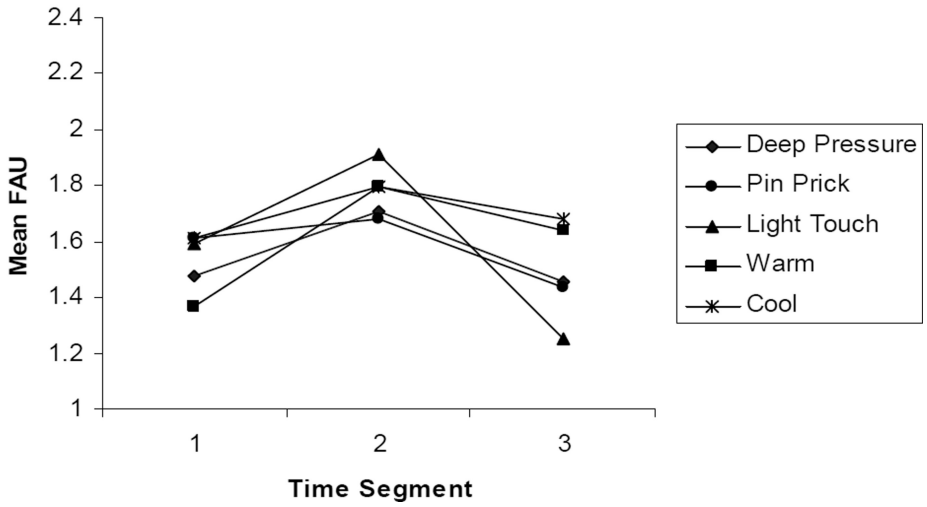

Figure 2.

(a) Mean FACS scores for active and sham trial time segments. FACS scores significantly increase during active but not sham stimulation (Time Segment 2) (* = $p < 0.05$). (b) Mean FACS scores for active trial time segments by stimulus modality.

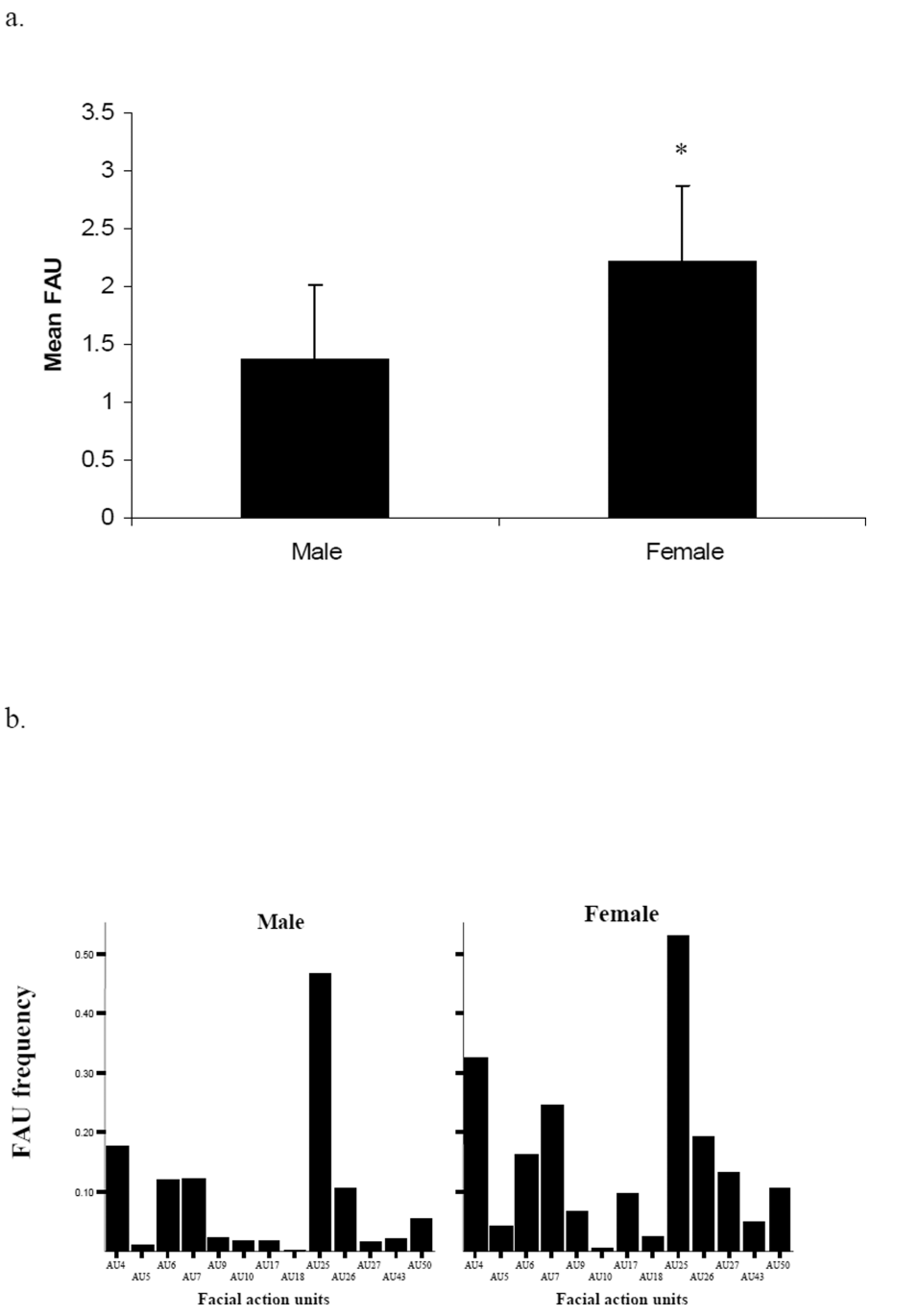

Figure 3.

(a) Mean FACS scores for active stimulus trials (segment 2) for males and females (* = $p < 0.05$). (b) Histogram showing frequency distribution of FACS AUs between males and females: AU4 (brow lowerer), AU5 (upper lid raise), AU6 (cheek raiser), AU7 (eye lid tightener), AU9 (nose wrinkler), AU10 (upper lip raiser), AU17 (chin raiser), AU18 (lip pucker), AU25 (lips part), AU26 (jaw drop), AU27 (mouth stretch), AU43 (eyes closed), and AU50 (vocalization).

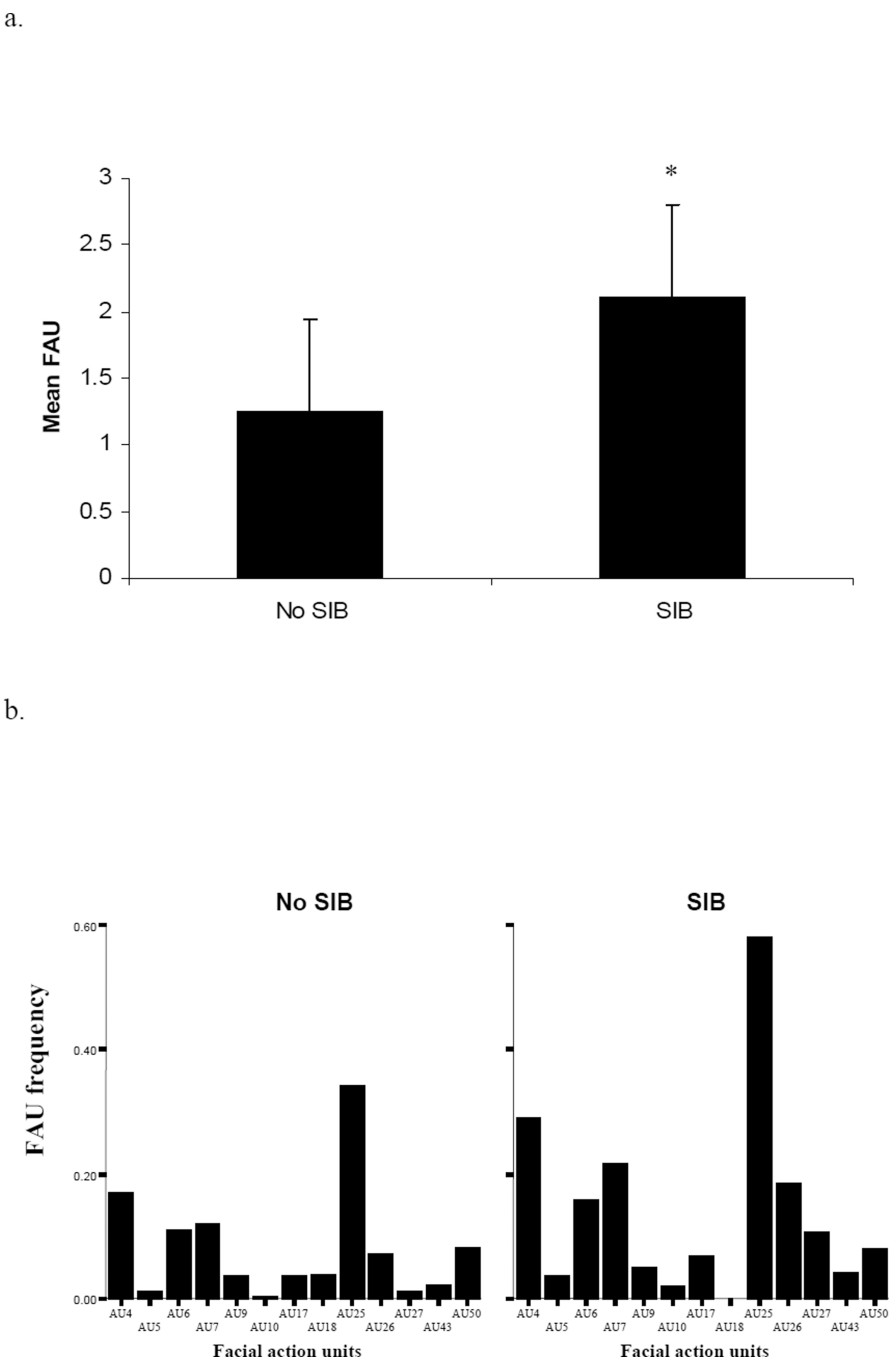

Figure 4.

(a) Mean FACS scores for active stimulus trials (segment 2) for individuals with and without chronic self-injury (* = $p < 0.05$). (b) Histogram showing frequency distribution of FACS AUs between individuals with and without self-injury: AU4 (brow lowerer), AU5 (upper lid raise), AU6 (cheek raiser), AU7 (eye lid tightener), AU9 (nose wrinkler), AU10 (upper lip raiser), AU17 (chin raiser), AU18 (lip pucker), AU25 (lips part), AU26 (jaw drop), AU27 (mouth stretch), AU43 (eyes closed), and AU50 (vocalization).