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Does the Neonatal Facial Coding System Differentiate Between Infants Experiencing Pain-Related and Non-Pain-Related Distress?

Sara Ahola Kohut* and Rebecca Pillai Riddell*,†

*Department of Psychology, Faculty of Health, York University, Toronto, Ontario, Canada.
†Hospital for Sick Children, Toronto, Ontario, Canada.

Abstract: **The Neonatal Facial Coding System (NFCS) is widely accepted as a measure of infant pain-related distress in known pain-specific contexts. It has clearly shown the ability to distinguish between facial reactivity in no-pain and pain-related situations. The primary purpose of this study was to explore whether NFCS differentiates between pain-related and non-pain-related distress. Two groups of 35 infants (1 group was distressed before injection whereas the other group was not distressed before injection) were coded using NFCS before and after an immunization procedure. Within-group analyses of infants who were distressed before immunization suggested that NFCS was not able to discriminate between pain-related and non-pain-related distress. However, between-group analyses showed NFCS discriminated between potential gradations of distress in infants after immunization. Results suggest that NFCS has the ability to discriminate between intensities of distress but not between pain–related and non-pain-related distress.**

Perspective: ***Adding to the NFCS validity literature, this study suggests that while able to distinguish between no-distress and pain-related distress, facial actions of NFCS may not distinguish between pain-related and non-pain-related distress expressions. However, NFCS was able to discern infants presumed to have higher pain-related distress due to experiencing pre-needle distress.***

Key words: *Infant pain, pain expression, facial coding, Neonatal Facial Coding System.*

The Oxford English Dictionary generally defines distress as "extreme anxiety, sorrow, or pain"[6] and further describes pain as suffering of either a physical or mental nature.[22] Thus, although one may find it difficult to philosophically distinguish between pain of a more psychological versus physical cause, the pediatric facial expression literature states that there are specific facial constellations that appear in different types of distressful situations.[13,29] Moreover, there has been widespread agreement that there is a facial constellation specifically associated with pain.[4] In fact, infant pain-related distress faces are more consistent across babies than adult pain-related distress faces across adults.[4]

The Neonatal Facial Coding System (NFCS)[8,9] was designed to be used specifically with infant pain-related distress faces. Research has shown that the Baby Facial Action Coding System (Baby FACS)[21] demonstrated convergent validity with NFCS ($r = .79$) when compared across equivalent facial action units.[16] Moreover, using the constellation of facial actions used by NFCS, facial expression has been shown to be more consistent across infants than crying, heart rate, or infant body movements.[8,14]

Unlike the other facial coding systems, NFCS was developed specifically to code pain-related distress. NFCS diminishes subjectivity because it codes specific facial actions rather than using a coder's attributions of infant emotion.[8] Having an objective, valid and reliable assessment measure for nonverbal infants allows researchers and clinicians to measure pain-related distress without self-report. Facial coding has been shown to be 1 of the most sensitive and reliable methods by which to measure infant pain-related distress[7] therefore it is no surprise that facial coding has been described as an optimal measurement tool in pain-related distress measurement.[27]

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Address reprint requests to Ms Sara Ahola, York University, 4700 Keele Street, Toronto, Ontario, Canada M3J 1P3. E-mail: sahola@yorku.ca

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Given the assessment value of NFCS in known acute pain contexts (eg, immunizations, neonatal intensive care units),[8] an interesting area of inquiry would be to explore if NFCS distinguishes between different types of infant distress contexts (such as when distress is not linked to an acutely painful stimulus). Although NFCS does not purport to measure other facial expressions, from a psychometric perspective it is noteworthy if the constellation of NFCS facial action units is able to distinguish pain-related distress from non-pain-related distress. From a clinical perspective, it would be valuable to know if NFCS could help determine the presence of pain in unknown distress contexts (eg, a situation in which an infant is distressed but the assessor did not witness the precipitating event). To begin work in this direction, the primary aim of the current study was to investigate whether NFCS distinguishes between pain-related and non-pain-related distress. The primary analysis will be a within-sample comparison and will compare the NFCS scores before and after an immunization, for infants who experienced non-pain-related distress before their immunization. Given the lack of research in the area, it was decided to assume the null hypothesis and hypothesize that NFCS would not discriminate between the non-pain-related distress before the needle and the pain-related distress after the needle.

Finally, it has been suggested in the literature that infants who are already distressed will become more distressed with the application of another stressor.[12] Given the availability of 2 appropriate samples of infants (1 distressed before the needle and the other not distressed before the needle), the secondary analysis will be a between-groups analysis comparing the post-needle NFCS scores of infants who were distressed before the immunization to the post-needle NFCS scores of infants who were not distressed before the immunization. Because of previous work in the area with extremely low-birth-weight infants,[12] we hypothesize NFCS will be able to do this with healthy term infants.

Methods

Participants

Approval was obtained through both the York University Human Participants Review Committee and the Hospital for Sick Children Research Ethics Board. Two groups of 35 infants were selected from samples obtained from 4 pediatricians' clinics in Toronto, Ontario during routine immunizations (6 months: Pentacel, Prevnar and Hepatitis B; 12 months, Prevnar, and MMR; and 18 months, Varicella, Prevnar, MMR, Pentacel). A subset of 33 infants from the current sample had been previously analyzed.[24] To be recruited, infants had to be between the ages of 4 and 20 months with no suspected developmental delays or impairment, no chronic illnesses, have never been admitted to a neonatal intensive care unit, and been born no more than 2 weeks premature (>36 weeks' gestation at birth). Additionally, mothers were required to be able to speak and read English, and be the biological mother of the infant that was brought into the clinic.

The 2 groups of infants were created based on the presence or absence of distress pre-needle. It is believed that the pre-needle distress was due to the weighing or measuring procedures because infants are undressed and taken away from their mother's arms in an unfamiliar environment by a stranger. However, when executed diligently, as was the case in our clinics, none of these events are considered physically painful. Events that directly preceded the facial expressions of distress before the needle were noted and categorized into 4 groups: being swabbed by alcohol before needle stick (37.1%), during physical examination by doctor (31.5%), being weighed by the doctor (17.1%) and other (14.3%; eg, presence of doctor, mother talking to doctor).

Further bolstering the supposition that pre-needle distress in our analysis is not due to pain, all infants who experienced pre-needle distress did so for a short period of time directly after a non-painful event (as opposed to continuous distress reactivity during the pre-needle phase). Therefore, it was believed infants were not experiencing ongoing pain before the immunization (such as teething). Furthermore, another study examining infant reactions in a play context also found that approximately 34% of the infants demonstrated distress at some point during a play interaction[18] despite the absence of painful stimuli. This also suggests than infants can experience distress without experiencing physical pain and strengthens the validity of our non-pain-related and pain-related comparison.

Thus, this paper will move forward defining pain-related distress by the facial pain expression following a known physically noxious stimuli (ie, immunization needle) and non-pain-related distress by the facial expression immediately following a non-physically noxious event (such as separation from caregiver, being weighed by the doctor). A convenience sample of 70 was selected from an original pool of 175 infants. Of the 175 infants, 6 were excluded due to research assistant error (eg, spoiled ID number, video camera malfunction) which left 169 infants. Of those 169 infants, 35 infants demonstrated sufficient pre-needle distress (see Measures section) and were analyzed in the comparisons involving pain-related and non-pain-related NFCS scores. Of the remaining infants that had no pre-needle distress, 35 were chosen at random to be included in the comparisons involving post-needle NFCS scores (to ensure equal n's in the 2 groups). Infants showing pre-needle distress had a mean age of 9.98 months (SD, 4.83), were 46% male, had mothers with a mean age of 33.57 years (SD, 5.16) and had a Hollingshead Index score of 47.40 (SD, 14.51). Infants showing no pre-needle distress had a mean age of 12.40 months (SD 3.96), were 49% male, had mothers with a mean age of 31.69 years (SD, 4.16) and a Hollingshead Index score of 49.29 (SD, 7.99). The Hollingshead Index of Socio-Economic Status[11] is a measure of family socioeconomic status based on the education and occupation of the head(s) of household. Scores range from 8 to 66 and the lower the score the higher the status of that family. Scores falling in the 40 to 54 range indicate a placement within the social strata of: "me-

dium business, minor professional, technical."[11] *t* tests with a Bonferroni correction (family-wise error rate of .10), indicated no between-group differences on infant age, maternal age and socio-economic status.

Procedure

Mothers with infants receiving immunizations were given a flyer by the medical receptionist or research assistant. If they indicated interest, a research assistant would read the mothers an introductory script explaining the study. The research assistant then obtained written consent from the mother to participate and to be video taped. Once in the examination room, 2 video cameras were set up to capture a close-up "face" shot of the infant as well as a "wide" shot in order to obtain a full view of mother and child. The footage from the face shot camera was used in this study to code NFCS. If the infant's face was obstructed in the face shot, the wide shot was used as a secondary source to code NFCS. The research assistant videotaped the infant's face from the moment the infant was in the examination room to the moment that they had calmed and settled after the immunization. NFCS was coded for 10 seconds before the first needle and for 10 seconds immediately after the last needle. Unless otherwise requested by a mother, the entire clinic appointment was videotaped. The research assistant said "now" at the moment of infant skin puncture to ensure the exact time of needle was accurately recorded. Mothers received a $5 gift certificate to a donut shop and were later mailed a DVD copy of their child's appointment footage.

Apparatus

Immunizations were video recorded using 2 Canon-Elura 70 digital video recorders. A hand-held tripod was used for the face shot, whereas a floor-standing tripod was used for the wide shot.

Measures

The current study collected demographic data using a questionnaire, whereas NFCS[8,9] and the Modified Behaviour Pain Scale (MBPS)[28] were coded from video footage.

Demographics

Participant demographic data were collected on socio-economic status questions (parental education and occupation), as well as familial data such as age and number of children in the household and marital status of parents.

Infant Facial Expression

The facial expressions of infants were coded using NFCS[8,9] (See Table 1). NFCS scores the presence (1) or absence (0) of 10 distinct facial actions involved in the infant pain face; they include brow bulge, eye squeeze, nasolabial furrow, open lips, horizontal stretch mouth, vertical stretch mouth, lip purse, taut tongue, chin quiver, and tongue protrusion. However, as found in previous studies,[5,20,23] lip purse, chin quiver and tongue protrusion were not included in the analysis due to low frequency of these facial actions (less than 2% coded as present). Each of the remaining actions was coded for every 2 seconds of the video-recorded tape. Thus, for every 10 seconds of coding, each facial action receives a total score of 0 to 5, based on presence or absence of the facial coding for each of the 5 2-second epochs. Final scores were then summed together for a total score of 0 to 35. Higher scores on NFCS are indicative of higher levels of pain. Each infant, regardless of group, had 2 10-second epochs coded for NFCS: 1 that occurred pre-needle and 1 that occurred 10 seconds directly post-nee-

Table 1. Neonatal Facial Coding System: Neonatal Facial Coding System (NFCS): Grunau & Craig, 1987; 1990

Facial Action	*Description*
Brow bulge (BB)	Bulging, creasing and vertical furrows above and between brows occurring as a result of the lowering and drawing together of the eyebrows.
Eye squeeze (ES)	Identified by the squeezing or bulging of the eyelids. Bulging of the fatty pads about the infant's eyes is pronounced.
Nasolabial furrow (NLF)	Primarily manifested by the pulling upwards and furrow deepening of the naso-labial furrow (a line or wrinkle that begins adjacent to the nostril wings and runs down and outward beyond the lip corners).
Open lips (OL)	Any separation of the lips.
Vertical stretch mouth (VSM)	Characterized by a tautness at the lip corners (vertical) coupled with a pronounced downward pull of the jaw. Often stretch mouth is seen when an already wide open mouth is opened a fraction further by an extra pull at the jaw.
Horizontal stretch mouth (HSM)	Appears as a distinct horizontal pull at the corners of the mouth.
Lip purse (LP)	The lips appear as if an 'oo' sound is being made.
Taut tongue (TT)	Characterized by a raised, cupped tongue with sharp tensed edges. The first occurrence of taut tongue is usually easy to see, often occurring with a wide open mouth. After this first occurrence, the mouth may close slightly. Taut tongue is still scoreable on the basis of the still-visible tongue edges.
Chin quiver (CQ)	An obvious high-frequency, up-down motion of the lower jaw.
Tongue protrusion (TP)	Tongue visible between the lips extending beyond the mouth.

NOTE: Possible range of scores for individual facial actions is 0 to 5 for each 10-second epoch. Possible range of scores for total NFCS is 0 to 50 for each 10-second epoch.

dle. Coders were blinded to the study hypothesis and 25% of the data were double coded for inter-rater reliability (κ reliability range, .86 to .98).

General Level of Infant Distress

To facilitate a valid comparison of the pre- and post-needle distress (ie, non-pain-related versus pain-related distress), the current study wanted to control for general distress intensity, that is, only comparing infants who expressed equivalent distress intensity from both pain-related and non-pain-related stimuli. Research suggests that cry is not a good specific measure of infant pain.[8,19,25] However, cry is considered to be a useful and sensitive method of determining infant distress in general.[10] Therefore, infant cry was used to ensure that both pre- and post-needle distress were of a comparable intensity level.

The MBPS[28] cry subscale was used as the general measure of distress intensity. The cry subscale was coded by trained MBPS coders who were blinded to the hypothesis of the study (κ inter-rater reliability = .98). A score of 0 indicates laughing or giggling, 1 indicates not crying, 2 indicates moaning, quiet vocalizing, gentle or whimpering cry, 3 indicates a full lunged cry or sobbing, and 4 indicates a full lunged cry, more than baseline cry (only if infant crying during baseline). Infants in the primary within-group analysis were required to have a cry score of 3 on the infant cry subscale of the MBPS.[28] Infants who had no pre-needle distress (for the secondary analysis) were required to have a cry score of 0 or 1 before immunization.

Missing Data Management

Coding NFCS in an immunization context is challenging because facial actions are often obstructed for periods of time due to infant movement. To prevent the systematic bias inherent with deleting infants who moved during the immunization procedure, missing data were managed using a system that involved coders making conservative judgments regarding missing facial actions. When missing facial data was an issue, a blinded coder reviewed the video footage in order to determine the cause of the missing data (eg, infant turned away from camera, camera angle did not enable full coding). The blinded coder also coded whether 2 other common pain-related distress infant behaviors (eg, body movements, cry) remained constant.[28] The assumption was made that if all other behavioral indicators remained constant, it was highly probable that the infant's facial reactivity remained constant. However, this system was only used under the following circumstances: (a) facial data were available for a portion no less than 60% of the total 10 second epoch; (b) cry and body movement data were available and constant for the duration of the epoch; and (c) there were no other reasons why the coder should not reasonably infer that the infant's missing facial actions remained constant. If all conditions were met, the missing values were exchanged for the preceding value. If no preceding value was available, the next adjacent facial action value was used. If these assumptions could not be met, the infants were excluded from this current data analysis. Across discrete facial actions, 12% of data units were replaced with a constancy value. This procedure has been used in the past and has been subject to peer review.[24]

Results

Preliminary Demographic Analysis

Initial analyses contrasted demographic characteristics of the 4 different pediatrician's clinics used in data collection. After ensuring the assumptions were met, 3 1-way analyses of variance (ANOVA) were used to examine group differences in infant age, mother age, and distress. Results revealed no significant differences between clinics with respect to infant, mother age, or distress. A χ^2 test was used to determine group differences between clinics with regard to infant sex, and no significant differences were found. Therefore all analyses proceed with the data collapsed over the 4 clinics.

Primary Analyses Plan

During preliminary data screening it was determined that the sample data was not normally distributed. This was done on the NFCS data via an examination of histograms, calculating skewness and kurtosis as well as conducting Levene's test for homogeneity of variances. Therefore, nonparametric techniques were chosen as the most appropriate method in which to conduct the comparisons for this study.[3]

Each of the primary analyses was run on each individual facial action unit as well as the total NFCS scores. The family-wise error rate for the primary analyses was 10% (.013 for each comparison of the 7 individual facial action units and total score).

Primary Analysis

Infants With Pre-Needle Distress: Differences Between Pre- and Post-Needle NFCS Scores

To examine whether NFCS performed differently with pain-related and non-pain-related distress, a Wilcoxon matched-pairs signed-ranks test was conducted using the group of 35 infants who were distressed pre-needle (ie, comparing their pre-needle and post-needle NFCS scores). Overall, the NFCS total score was not significantly different during pain-related and non-pain-related distress. Among the 7 individual facial action units, none were significant and only 1 facial action approached significance. Vertical stretch mouth appeared to occur slightly longer during non-pain-related distress ($Z = -2.09$, $P = .04$). A post hoc power estimate based on the current sample size, reliability score, and desire to find a moderate effect size approaches .97 (based on a Monte Carlo study by Blair & Higgins [1985]).[1]

Table 2. Tests for Differences in Post-Needle Neonatal Facial Coding System (NFCS) Scores for Infants Distressed and Not Distressed Before Immunization (n = 70)

	Median	*Range*	*Mode*	*Z*	P
Brow bulge (BB)					
Pre-needle distress	5	2–5	5 (n = 30)	−2.28	.023
No pre-needle distress	5	0–5	5 (n = 22)		
Eye squeeze (ES)					
Pre-needle distress	5	2–5	5 (n = 29)	−2.14	.032
No pre-needle distress	5	0–5	5 (n = 22)		
Nasolabial furrow (NLF)					
Pre-needle distress	5	2–5	5 (n = 31)	−2.89	.004
No pre-needle distress	5	0–5	5 (n = 21)		
Open lips (OP)					
Pre-needle distress	5	2–5	5 (n = 34)	−2.22	.026
No pre-needle distress	5	0–5	5 (n = 28)		
Vertical stretch mouth (VSM)					
Pre-needle distress	4	0–5	5 (n = 14)	−1.87	.062
No pre-needle distress	2	0–5	0 (n = 17)		
Horizontal stretch mouth (HSM)					
Pre-Needle Distress	5	0–5	5 (n = 26)	−2.05	.040
No pre-needle distress	4	0–5	5 (n = 17)		
Taut tongue (TT)					
Pre-needle distress	4	0–5	5 (n = 16)	−3.07	.002
No pre-needle distress	0	0–5	0 (n = 23)		
Total NFCS score					
Pre-needle distress	31	13–35	35 (n = 10)	−2.86	.004
No pre-needle distress	25	0–35	30 (n = 6)		

Secondary Analysis

Differences in Post-Needle (Pain-Related) NFCS Scores: Infants With and Without Pre-Needle Distress

Using all 70 infants, a Mann-Whitney *U* test was conducted to compare the pain-related NFCS scores of infants with and without pre-needle distress (Table 2). Infants who were distressed pre-needle had significantly higher NFCS total scores during pain-related distress when compared with infants who had no pre-needle distress ($U = -2.86$, $P < .01$). A set of 7 Mann-Whitney *U* tests were also conducted on each individual facial action unit. Infants distressed pre-needle scored significantly higher on NFCS on nasolabial furrow and taut tongue. There was also a trend for infants with pre-needle distress to score higher on brow bulge, eye squeeze, open lips, and horizontal stretch mouth. There were no significant differences between scores on vertical stretch mouth.

Discussion

We acknowledge that NFCS is not designed to measure different types of distress. However, to demonstrate specificity to pain, it is reasonable to examine if NFCS performs differently in pain-related and non-pain-related distress. Our findings for the primary analysis showed that the facial action units of NFCS did not differentiate pain-related and non-pain-related distress. It is speculated that this may be due to several reasons. Pragmatically, NFCS is coded on a 2 second basis giving each facial action a score of present or absent. Although it has been validated for detecting the presence of pain-related distress versus no distress (ie, a comparison to baseline), a more finely grained system may be required for distinguishing between different types of distress. With premature infants and infants at risk for neurological impairment, who have less vigorous signaling, NFCS is coded every second.[15,26] This more detailed method may allow NFCS to pick up on the slight differences between types of distress, especially with certain facial actions that showed a trend toward significance in the current study (eg, a longer occurrence of vertical stretch mouth during non-pain-related distress). Alternatively, from an emotional development perspective, NFCS may be unable to distinguish between distress types because some researchers suggest infants in this age range have not yet developed the ability to express different negatively valenced emotions.[2,17]

In our secondary analyses, the current study goes beyond the current state of research by suggesting that NFCS is able to detect gradations of distress in healthy infants. This was demonstrated by the significantly higher post-needle NFCS scores of infants with pre-needle distress compared to the infants with no pre-needle distress. We suggest this difference is due to the higher intensity of pain-related distress contexts expression in the group of infants who were already distressed pre-immunization. Research has previously demonstrated that premature infants who are distressed before painful

stimuli express more pain-related facial expressions after the painful event.[12] However, one could argue that NFCS's ability to detect gradations of distress was not replicated in our initial within group analysis comparing the non-pain-related distress and the pain-related distress. However, our review of actual infant scores suggest that this may be because many infants had already hit their ceiling on NFCS scores pre-immunization, thus their NFCS scores post-immunization were not significantly different. Alternatively, it is possible that the effect we are attributing to NFCS's ability to detect gradations of distress may partly be due to the slight age difference of 2 months between the groups. However, as previously mentioned infant facial expressions of pain are remarkably consistent across infancy.[4] Further research involving a longitudinal cohort would help clarify this issue.

Limitations of the Study

This study contains several limitations to consider when interpreting the results. Due to the nature of these present analyses, infants not sufficiently distressed before needle had to be excluded from the primary within-group analysis. Thus, the results of the distress discrimination testing may only generalize to infants with a low threshold of distress. However, infants who were distressed pre-needle did not differ significantly from the infants who were not distressed on key demographics such as infant sex, mother age, and infant age. Therefore, these results have the potential to generalize to a broader range of infants.

A further limitation was the timing of the type of distress was not randomized. First, non-pain-related distress always preceded pain-related distress. Future studies may attempt to randomize infants to be coded for non-pain-related distress before or after the needle. However, unpublished parent report data from previous studies at our laboratory suggest that infants can feel pain-related distress up to 3 days post-immunization. Thus, randomization must take account of lingering pain post-immunization. Moreover, as mentioned earlier, whereas no infant included in this study cried throughout the pre-needle and post-needle phase, it is acknowledged that due to the lack of experimental control in this naturalistic setting, the within groups analysis could be comparing a non-pain-related distress face to a pain-related distress face that could have carry-over from the non-pain-related distress. However, regardless of this fact, NFCS did not distinguish between the 2 types of distress.

In addition, while future researchers should try to use the same stimuli to trigger the non-painful distress, attention must be paid to the fact that the same stimuli may not result in the same type of non-painful distress (eg, triggering fear versus anger) in the infants. Last, due to the large age range in the sample, these data encompass different developmental stages. However, regardless of age and stage, all infants were experiencing significant distress post-needle and NFCS has been validated with a broad age range of infants.[16]

Conclusions and Future Directions

The results suggest that NFCS, as it is currently coded in healthy infants, may not distinguish between different types of distress experienced in naturalistic medical settings. Perhaps more detailed coding may elicit differences in pain-related and non-pain-related distress or perhaps infants are not yet distinguishing between similar negative emotions. However, due to its sensitivity and potential ability to detect gradations of infant distress, NFCS continues to clearly demonstrate its use in known pain-related distress contexts. While NFCS did not distinguish between pain-related and non-pain-related distress in this observational study, future research should examine more systematically variables such as distress gradations, different time points, as well as different distressful stimulations for the infant.

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