Extremity movements help occupational therapists identify stress responses in preterm infants in the neonatal intensive care unit: A systematic review

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Key words

- Stress ▪ Preterm infants ▪ Pain

Mots clés

- Stress ▪ Nouveaux-nés prématurés ▪ Douleur

Abstract

Background. *Accurate assessment and treatment of pain and stress in preterm infants in neonatal intensive care units (NICU) is vital because pain and stress responses have been linked to long-term alterations in development in this population.* **Purpose**. *To review the evidence of specific extremity movements in preterm infants as observed during stressful procedures.* **Methods**. *Five on-line databases were searched for relevant studies. For each study, levels of evidence were determined and effect size estimates were calculated. Each study was also evaluated for specific factors that presented potential threats to its validity.* **Results**. *Eighteen studies were identified and seven comprised the review. The combined sample included 359 preterm infants. Six specific movements were associated with painful and intrusive procedures.* **Clinical Implications**. *A set of specific extremity movements, when combined with other reliable biobehavioural measures of pain and stress, can form the basis for future research and development of a clinical stress scale for preterm infants.*

Résumé

Description. *L'évaluation et le traitement précis de la douleur et du stress chez les nouveaux-nés prématurés hospitalisés dans les unités de soins intensifs néonatals (USIN) sont essentiels, en raison des réactions à la douleur et au stress qui ont été associées à des altérations à long terme dans le développement de cette population.* **But**. *Cette étude avait pour but d'examiner des données probantes sur les mouvements spécifiques observés aux extrémités de nouveaux-nés prématurés pendant des interventions susceptibles de provoquer du stress.* **Méthodologie**. *Des recherches ont été effectuées dans cinq banques de données en ligne afin de cibler des études pertinentes. Pour chaque étude, les niveaux de preuve ont été déterminés et des estimations de la taille de l'effet ont été calculées. Des facteurs spécifiques pouvant mettre en doute la validité de chaque étude ont également été évalués.* **Résultats**. *Dix-huit études ont été ciblées et sept ont été retenues pour l'étude. L'échantillon combiné comprenait 359 nouveaux-nés prématurés. Six mouvements spécifiques ont été associés à des interventions douloureuses et intrusives.* **Conséquences cliniques**. *Lorsqu'ils sont combinés à d'autres mesures biocomportementales de la douleur et du stress, des série de mouvements spécifiques aux extrémités peuvent former la base de futures recherches et de l'élaboration d'une échelle du stress clinique chez les nouveaux-nés prématurés.*

Paediatric occupational therapists working in the neonatal intensive care unit (NICU) provide expertise in the assessment of complex and subtle movement patterns of the preterm infant. Before the development of feeding and play, these movements express the infant's first occupations, self-regulation, and purposeful engagement with the environment. Although traditionally these movements are evaluated as part of the neurological functioning of the infant, an ongoing need also exists for the accurate interpretation of these movements as indicators of pain and stress responses. Indeed, these stress responses represent markers of the mismatch between a preterm infant's developmental capacities and its environment. Accurate assessment and interpretation of these movements is critical so that occupational therapists apply appropriate interventions to reduce pain and stress responses.

Some occupational therapists have incorporated the concept of stress into their assessments by using the Newborn Individualized Developmental Care and Assessment Program (NIDCAP®) (Als, 1986). The NIDCAP® can be integrated easily with the Person-Environment-Occupation Model because the NIDCAP® is a transactional model that combines a detailed assessment of the infant's environment with the assessment of the infant's physiological and behavioural responses to various types of handling (Law et al., 1996). Movements observed using the NIDCAP® system are categorized as stress or stability indicators.

Despite the wide use of the NIDCAP®, not all occupational therapists are NIDCAP® certified because this program is both labour intensive (Cheng & Chapman, 1997; Tribotti & Stein, 1992) and expensive to implement (e.g., Petryshen, Stevens, Hawkins, & Stewart, 1997). Moreover, understanding the ways in which pain and stress may influence preterm infant development and how they are assessed currently is a highly specialized area of practice. Barriers such as these may limit occupational therapists from collaborating with other health care professionals in assessing pain and stress in preterm infants. Therefore, the purposes of this systematic review are as follows:

1. To summarize what is currently known about the effects of early exposure to pain/stress on development in preterm infants;
2. To review the literature on pain/stress measurement tools available for clinical use and the factors that may influence pain responses in preterm infants; and
3. To present preliminary evidence on a set of validated extremity movements which hold promise for future use in occupational therapy research and in clinical practice for the assessment of preterm infant stress and pain responses.

The long-term developmental effects of early pain exposure

Assessing and treating pain and avoiding the effects of stress are critical in the care of preterm infants because some investigators suggest that alterations in the brains of preterm infants observed at older ages, such as reductions in brain volumes in a variety of regions, may be directly attributable to pain and stressor exposure experienced in the NICU (Bhutta & Anand, 2002; Inder, Warfield, Wang, Hüppi, & Volpe, 2005; Isaacs et al., 2000; Nosarti et al., 2002; Peterson et al., 2003). Such a hypothesis is supported by research which indicates that early exposure to stressors may not only alter brain development (for review see Teicher et al., 2003), but also may change future responses to environmental stressors by resetting the hypothalamic-pituitary-adrenal (HPA) axis (e.g., Anisman, Zaharia, Meaney, & Merali, 1998; Grunau et al., 2005; Ladd et al., 2000). For example, animal and human studies reveal that specific stressors, such as early pain exposure, may alter nociceptive pathways (Anand, 2000; Anand, Coskun, Thrivikraman, Nemeroff, & Plotsky, 1999) and also may contribute to changes in other areas of development (e.g., difficulties in self-regulation learning, behavior, and motor problems) (Grunau, 2000, 2002, 2003). Investigators examining the long-term developmental effects of early, repetitive pain exposure in animal models have shown that neonatal rats exposed to tissue injury may have increased or decreased pain thresholds as adults (Anand et al., 1999; de Lima, Alvarez, Hatch, & Fitzgerald, 1999; Ren et al., 2004; Sternberg, Scorr, Smith, Ridgway, & Stout, 2005). Using inflammatory pain models, others have shown that noxious stimulation early in development produces long-lasting changes in pain pathways, such as long-term hyperexcitability and expansion of receptive field sizes in dorsal horn neurons in the spinal cord (e.g., Ruda, Ling, Hohmann, Peng, & Tachibana, 2000; Torsney & Fitzgerald, 2003). Importantly, early exposure to short-term inflammatory pain induces an increase in gene expression in the midbrain periaqueductal grey region, a region that is involved in both stress and pain modulation (Anseloni et al., 2005).

Changes in human infant pain processing as a result of early pain exposure have also been demonstrated. For example, following their discharge from the NICU, extremely low birth weight preterm infants (≤ 800 grams) assessed at four and eight months (corrected age) exhibited subtle differences when recovering from a finger lance applied to a pain-naïve site when compared to term controls (Grunau et al., 2001; Oberlander et al., 2000). Surprisingly, changes from four to eight months suggested that differences may become more marked over time. However, it is unknown whether these changes are clinically significant.

Further indirect evidence supports the notion that early pain exposure alters later development in extremely low birth weight preterm infants. Former preterm toddlers have lower pain sensitivity than term born controls, as described through parent report (Grunau, Whitfield & Petrie, 1994). At school age, former preterm children have increased somatization (Grunau, Whitfield, Petrie, & Fryer, 1994; Sommerfelt, Troland, Ellertsen, & Markestad, 1996) and greater affective responses to depicted medical pain (Grunau, Whitfield, & Petrie, 1998). They rated pictures of pain during recreation as higher in pain intensity than their term born peers. However, the difference in somatization does not persist into the middle school years (Whitfield, Grunau, & Holsti, 1997).

Not only does early pain exposure alter behavioural pain expression, but it also can change future responses to stress by resetting the stress-response system (i.e., the hypothalamic-pituitary-adrenal axis). Indeed, in the first study evaluating the hypothalamic-pituitary-adrenal reactivity in preterm infants following discharge from the NICU, researchers have found that preterm infants have altered cortisol levels both during baseline and in response to novel stimuli compared to full term controls (Grunau, Weinberg, & Whitfield, 2004).

Whereas examining the consequences of early pain exposure in preterm infants is crucial, other studies indicate that even routine, non-painful, caregiving tasks such as diaper changing can produce marked physiological and behavioural responses in preterm infants (e.g. Grunau, Holsti, Whitfield, & Ling, 2000; Hellerud & Storm, 2002; Holsti, Grunau, Oberlander, & Whitfield, 2005; Holsti, Grunau, Whitfield, Oberlander, & Weinberg, 2005). Thus, it is possible that the cumulative effects of painful and non-painful, yet stressful events have an impact on these infants.

Assessments of pain in preterm infants

For clinicians, the most accurate method to assess pain and stress responses in preterm infants is to use standardized measures whose psychometric properties have been rigorously established. To date, the development of measures of stress responses in preterm infants has focused almost exclusively on the development of pain assessments. Pain assessment in preterm infants is more complex than for term infants because their responses to pain may be dampened compared to term infants' responses (Craig, Whitfield, Grunau, Linton, & Hadjistavropoulos, 1993; Johnston & Stevens, 1996). Furthermore, preterm infants may display different behavioural pain cues than term infants because they are neurologically less mature. In addition, behavioural and physiological indicators of pain are often dissociated (Morison, Grunau, Oberlander, & Whitfield, 2001). Finally, no behavioural or physiological threshold marks specifically the presence or absence of pain.

Despite these complexities, investigators and clinicians have developed a number of unidimensional and multidimensional preterm infant pain measures (for reviews see Franck, Greenberg & Stevens, 2000; Franck & Miaskowski, 1997; Stevens & Gibbins, 2002). Unfortunately, because many of these tools have been derived from observations of term infants, they are less applicable to preterm infants. Furthermore, many lack complete assessments of their psychometric properties or clinical utility. In addition, although using a single pain index is easier for clinicians, the physiologic and behavioural responses of preterm infants to painful stimuli are often dissociated (Morison et al., 2001); therefore, using pain scales which combine physiological and behavioural pain responses into a single score may limit our understanding of the specificities of pain responses. To date, experts in preterm infant pain assessment have determined that changes in facial activity, shifts in infant sleep/waking state, and physiologic indices of heart rate and oxygen saturation are the most promising biobehavioural pain indicators (Franck et al., 2000; Grunau et al., 2000; Stevens, Johnston, & Grunau, 1995; Stevens, Johnston, Petryshen, & Taddio, 1996). Extremity movements as indicators of pain and stress have received relatively less attention.

Assessments of stress responses in preterm infants

Although a number of pain scales exist for use with preterm infants, they were not designed to assess stress responses during non-painful events. Moreover, many of the pain assessments do not include extremity movements. For example, the most well-researched pain scales for preterm infants, the Neonatal Facial Coding Systems (NFCS) (Grunau & Craig, 1987) and the Premature Infant Pain Scale (Stevens et al., 1996), both distinguish relative differences between painful and non-painful caregiving tasks. However, these tools include only facial reactivity as the behavioural indicator. Facial reactivity is considered the "gold standard" of behavioural pain cues in infants (Franck et al., 2000), yet in some preterm infants--particularly those at earlier gestational ages, those who are very ill, or those who are exposed to exogenous steroids--facial reactivity to pain may be dampened (Grunau, Oberlander, Whitfield, Fitzgerald, & Lee, 2001; Johnston & Stevens, 1996; Johnston et al., 1999; Stevens, Johnston, & Horton, 1994). Dampened facial response may lead clinicians to the erroneous conclusion that the infant is not feeling pain.

Unlike facial reactivity, extremity movements to pain in preterm infants are exaggerated because of differences in peripheral and central pain processing (Fitzgerald, 2000; Holsti, Grunau, Oberlander, & Whitfield, 2004; Holsti et al., 2005; Morison et al., 2003). Therefore, preterm infant extremity movements can provide complementary stress and pain cues when facial reactivity is dampened. With the exception of pain assessments, only one "distress" scale has been developed for use with infants (Sparshott, 1996). However, this scale was developed specifically for preterm infants who were ventilated, and its clinical utility has not been researched adequately.

Body movements as stress indicators in preterm infants

Using extremity movements as indicators of stress responses in preterm infants is not new. A number of researchers have used various groups of infant movements as indicators of stress, many deriving their stress response indicators from the NIDCAP®. Using specific behaviours from the NIDCAP® to assess pain and stress reactivity is appealing because it is an established tool, developmentally appropriate for preterm infants, and the coding descriptors of each of the infant movements are very specific. Moreover, the NIDCAP® can be applied to infants across varying gestational ages and during any caregiving tasks. Until recently, however, NIDCAP® behaviours used to identify stress responses in preterm infants had not been validated against other known biobehavioural indicators of pain or stress responses in preterm infants (Holsti et al., 2004).

In order to establish whether or not specific extremity movements are indicators of stress and pain responses in preterm infants, the most direct approach is to examine whether specific movements occur relatively infrequently when the infant is at rest and increase significantly during or following events which are stressful or painful (Grunau et al., 2000). Moreover, studies which group body movements as non-standardized measures of stress reactivity (e.g. Greneir, Bigsby, Vergara, & Lester, 2003) must include other known, valid and reliable indicators of pain and stress concurrently since failure to do so weakens the claim that the chosen behaviours measure the construct of stress. Furthermore, investigators should include physiological indicators along with behavioural indicators of stress (e.g. Peters, 1998, 2001; Stephens & Glazer, 1992; Van Cleve, Johnson, Andrews, Hawkins & Newbold, 1995), especially since researchers have shown only a modest correlation between physiological and behavioural indicators in studies of infant pain (e.g. Barr, 1998; Morison et al., 2001); thus one domain does not capture the whole spectrum of response. Finally, researchers must control for gestational age at assessment, baseline behavioural state, prior handling, and prior pain exposure because all these factors have been shown to alter preterm infant reactions to stressful procedures (Grunau et al., 2001; Holsti et al., 2005; Porter, Wolf & Miller, 1998; Stevens et al., 1994).

To contribute new knowledge of pain and stress reactivity in preterm infants, researchers have used the NIDCAP® in combination with physiological indicators and/or other valid behavioural measures of pain in preterm infants in the NICU during procedures of varying intensity (Grunau, Oberlander, Holsti, & Whitfield, 1998; Grunau et al., 2000; Holsti et al., 2004; Holsti, Grunau, Oberlander et al., 2005; Holsti, Grunau, Whitfield et al., 2005; Morison et al., 2001; Morison et al., 2003; Peters, 1998; Stephens & Glazer, 1992; Walden et al., 2001). We now present preliminary evidence of a number of extremity movements which, after more comprehensive research, occupational therapists can apply in clinical practice for the assessment of pain and stress responses in preterm infants.

Review method

Literature search

The following five online databases were searched electronically and manually: MEDLINE (1982-2004), Cumulative Index to Nursing and Allied Health Literature (1982-2004), EMBASE (1982-2004), Cochrane Database of Systematic Reviews and PsycINFO (1982-2004). Key words used in the search were *preterm infant, pain, assessment, stress, body movements, NIDCAP®*, and *developmental care*. Then, for each article found, the references were reviewed for additional studies.

Inclusion-exclusion criteria for studies

Only manuscripts published in English were included in this review. In addition, the study had to be published in a peer-reviewed journal no earlier than 1982, when Als' paper on the synactive theory of development was first published (Als, 1982). Furthermore, the study had to include preterm infants in the sample and report individual extremity movements observed during identified, individual NICU procedures. An individual study was excluded if:

1. It combined full-term and preterm infants in the sample;
2. The preterm infants had been exposed to analgesia or sedation within 72 hours of the observation; and
3. The study used broad categories of extremity movements rather than specific individual movements.

Evaluation of the studies

Levels of evidence (LOE) for each of the included studies were determined (Trombly & Ma, 2002). These levels of evidence were developed by the American Occupational Therapy Association Evidence-Based Practice Project. This rating system was chosen because it rates the study design and allows for separate ratings of internal and external validity. Using this system, each study design was rated from I to IV with randomized trials, the highest level of evidence, given ratings of I, and the lowest level of evidence (e.g. single-subject designs) given rating of IV, Then, each study was evaluated for its sample size. Studies including samples ≤ 50 were given an "A", those including 20-49 a "B" and those with less than 20 a "C". Internal validity was rated as "high =1, moderate =2 and low =3" and external validity as "high = a, moderate = b, low = c". For example, studies rated as having high internal validity were those for which no alternative explanation for the outcomes could be found and/or other threats to validity were controlled (e.g., loss of participants, lack of blinding of outcome evaluators or spontaneous recovery). On the other hand, studies having moderate internal validity were those which included one or two of the aforementioned threats to validity. Studies which did not fit into the high or moderate category were rated as having "low" internal validity. In addition to standard levels of evidence, the studies were reviewed for the following specific factors which threaten validity of studies evaluating pain and stress responses in preterm infants:

1. Using concurrent measures of pain and stress;
2. Including both behavioural and physiological measures of pain and stress;
3. "Blinding" assessors to procedures; and
4. Controlling for age at assessment, length of assessment time, baseline state, prior handling, and prior procedural pain exposure.

Finally, where possible, effect sizes estimates were calculated for the movements using change in frequencies of behaviours between the baseline and invasive procedure (e.g., lance phase of blood collection) (Rosenthal, Rosnow, & Rubin, 2000). One author provided extra information so that effect sizes could be calculated (Holsti et al., 2004; Holsti et al., 2005).

Results

Description of the studies

Eighteen studies were identified for potential inclusion in this review; only one used a randomized design. After more careful inspection, eleven studies were dropped from the review. One study reported individual body movements during the admission process to the NICU but did not report movements occurring during each individual procedure (Stephens & Glazer, 1992). A further two studies combined full-term and preterm infants in the sample; therefore, it was not possible to attribute specific movements to preterm infants

(Bozzette, 1993; Van Cleve et al., 1995). A fourth study did not exclude infants who were on sedation during the assessment period (Slevin, Daly, & Murphy, 1998). A fifth did not report the procedures which were observed (Pressler, Helm, Hepworth, & Wells, 2001). Six studies included the assessment of extremity movements to observe preterm infants in the NICU during procedures of varying intensities; however, each action was combined (e.g., flexion/extension) or grouped so that identification of individual movements could not be determined (Craig et al., 1993; Evans, Vogelpohl, Bourguignon, & Morcott, 1997; Goubet, Clifton & Shah, 2001; Peters, 1998; Peters, 2001; Sparshott, 1996). The remaining seven studies comprised this review (Grunau et al., 1998; Grunau et al., 2000; Holsti et al., 2004; Holsti et al., 2005; Morison et al., 2001; Morison et al., 2003; Walden et al., 2001).

Synthesis of the literature review

Six of the seven studies reviewed were non-randomized, controlled trials (levels of evidence III), where biobehavioural responses of preterm infants before, during, and/or after painful or intrusive NICU procedures were reported. The seventh study included infants observed before, during, and after a painful and a non-painful procedure; infants were randomized to procedure order. In all the studies, the infants were observed across three distinct time periods. First, infant movements were recorded for a baseline period during which the infants were left undisturbed immediately preceding the stressful stimulus. The *baseline period* allowed researchers and clinicians to observe how the infant responded during a "non-stressful" period. Next, infant movements were recorded during a heel lance for blood collection. The components of the blood collection, which were evaluated, were the *lance phase* when the skin was broken, and/or the *squeeze phase* during when the heel was squeezed to obtain the blood. Finally, infant movements were observed immediately after the blood collection for the *recovery phase*. All changes in movements during the lance phase or the recovery phase were evaluated against those occurring during the baseline period.

The combined sample of the seven studies included 359 preterm infants ranging in gestational age at birth from 23 to 32 weeks. Although combined analysis of the studies was not possible due to the differing study designs and analyses reported, calculating effect size estimates was possible for four of the studies. Table 1 presents the critical review of the seven studies. Based on this review, a set of extremity movements have been shown to be associated with acutely painful and/or intrusive events (see Figure 1).

General extremity actions

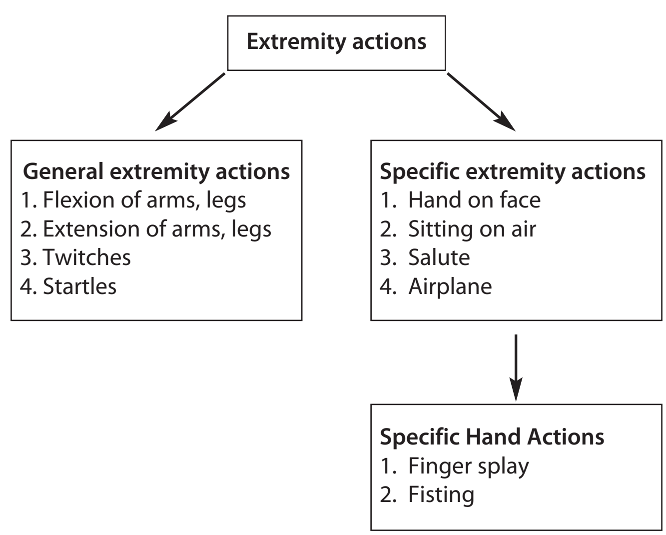
Flexion and extension of the extremities.

Although some researchers suggest that flexor activity of the arms and legs is associated with self-regulation (Als, 1984; Peters, 2001), others include knee or leg flexion in their pain index (Evans et al., 1997; Franck, 1986; Lawrence et al., 1993). These two interpretations appear contradictory; however, others have observed the coupling of flexor and extensor activity in preterm infants across procedures of varying intensities such as diaper changing, endotracheal suctioning and heel lance (Grunau et al., 2000; Holsti et al., 2004; Holsti et al., 2005; Morison et al., 2003,). In many contexts, the combined flexion and extension of the legs in preterm infants is likely the flexor withdrawal response rather than attempts to self-regulate. The flexor withdrawal response response in infants differs from the reflex in adults. In adults, this response is stimulated only by painful stimuli applied to the plantar aspect of the foot. In infants, however, this response can be elicited by both nociceptive and tactile stimulation. Even though the flexor withdrawal response has been used in a number of studies to assess the development of spinal pain processing in preterm infants and has been a reliable indicator of reactivity to varying intensities of stimulation (Andrews & Fitzgerald, 1999; Fitzgerald, Shaw, & McIntosh, 1988), the presence of these movements should not used be as sole indicators of stress in preterm infants. Other researchers have shown that they also occur frequently when preterm infants are at rest (Holsti et al., 2004; Holsti et al., 2005).

Specific extremity movements

Eloquently described by Als in the NIDCAP® program (Als, 1984), the following specific extremity actions have been associated with stressful and acutely painful procedures in the NICU (Holsti et al., 2004; Holsti et al., 2005; Morison et al., 2004;

FIGURE 1
Extremity movements associated with acute pain and stress in preterm infants in the neonatal intensive care unit.



*Twitches and startles in infants >30 weeks gestational age are associated with active sleep, but in infants < 30 weeks they may, in some contexts, be stress response cues (Holsti et al., 2004).

TABLE 1
Critical review of studies combining extremity movements with concurrent measures to assess pain and stress in preterm infants in the neonatal intensive care unit.

Study and level of evidence (LOE:Trombly & Ma, 2002)	Purpose	Study design (N)	Effect size† estimates for Extremity Movements	Threats to validity
Grunau et al., 1998 LOE: IIIB2a	To study the reliability, construct and concurrent validity of the full NFCS* at bedside.	Cohort (N=40) - Gestational age at birth: 26-30 weeks. - Infants assessed at 32 weeks PCA at bedside	N/A	- Sample size estimate not provided - Assessors not blind to procedure. - Influence of prior handling or pain exposure not examined
Grunau et al., 2000 LOE: IIIA2b	Preliminary study to explore the classification of NIDCAP® movements as stress and stability indicators in preterm infants in the NICU.	Convenience Sample (n=64) - Gestational age at birth: 23-32 weeks. - Infants assessed at 24-34 weeks PCA.	Finger splay: ø = 0.71 Facial grimace: ø = 0.81 Leg extension: ø = 0.73	- Conclusions limited to convenience sample. - Recommendations to study movements further. -Sample size estimate not provided - Assessor not blind to procedures. - Reliability not established between 2 assessors.
Holsti et al., 2004 LOE: IIIB2a	To determine whether NIDCAP® stress movements are associated with acute pain responses in preterm infants in the NICU.	Cohort (N = 44) - Gestational age at birth: 25-32 weeks. - Infants assessed at 32 weeks PCA	Flex arms: r = 0.25 Flex legs: r = 0.32 Extend arms: r = 0.22 Extend Legs: r = 0.19 Hand on face: r = 0.30 Finger splay: r = 0.23 Fisting: r = 0.33 Frown: r = 0.40 Twitch Face: r = -0.48 Twitch Body: r = -0.40 Twitch Extremities: r = -0.45 Mouthing: r= -0.23 Foot Clasping: r = -0.27	- Results specific to infants at 32 weeks gestational age at testing. - Assessors not blind to procedure for NIDCAP® coding. -Explored associations between prior handling, pain exposure, but not statistically controlled for data analysis.
Holsti et al., 2005. LOE: IB2a	To compare specific body movement responses to routine, non-skin-breaking handling with those during a painful procedure. Procedure order was randomized	Cohort (N=54) - Gestational age at birth: 27-32 weeks. - Infants assessed at 32 weeks PCA	Effect sizes for movements during Diaper changing: Flex arms: r = - 0.36 Flex legs: r = - 0.33 Extend arms: r = - 0.57 Extend legs: r = - 0.33 Finger splay: r = - 0.60 Salute: r = - 0.29 Airplane: r = - 0.20 Fisting: r = - 0.33 Sit on air: r = - 0.39 Hand on face: r = - 0.44 Movements during Pain: Flex arms: r = - 0.16 Flex legs: r= - 0.30 Finger splay: r = - 0.23 Salute: r = - 0.23 Sit on air: r = - 0.33 Hand on face: r = - 0.28	- Results specific to infants at 32 weeks gestational age at testing. - Assessors not blind to procedure for NIDCAP® coding. -Explored associations between prior handling, pain exposure, but not statistically controlled for data analysis.

Table 1, continued

Study and level of evidence (LOE:Trombly & Ma, 2002)	Purpose	Study design (N)	Effect size† estimates for Extremity Movements	Threats to validity
Morison et al., 2001 LOE: IIIA2a	To assess relations and concordance between behavioural and physiological reactivity to pain in preterm infants in the NICU stratified by gestational age at birth.	Cohort (N=136) - Infants stratified by gestational age at birth (23-26 weeks, n=48; 27-29 weeks, n=52; 30-32 weeks, n=36). - Assessed at 32 weeks PCA	- Finger splay and NFCS r = 0.23 for total sample during Lance. - 27-29 weeks GA r = 0.25 between finger splay and heart rate - 23-29 weeks GA between finger splay and NFCS r = 0.50 -30-32 GA between finger splay and NFCS r = 0.45	- Large sample, well described, appropriately analyzed, but sample size estimate not provided. - Assessors not blind to procedure. - Explored associations between prior handling, pain exposure.
Morison, et al., 2003	To examine preterm infants' reactions to acute pain in detail over prolonged time periods using full NIDCAP®.	Cohort (N=10) - Gestational age at birth: 27-32 weeks. - Infants assessed at 32 weeks PCA	N/A	- Pilot study, but conclusions not overstated. - Assessors not blind to procedure for NIDCAP® coding.
Walden, et al., 2001	- To identify physiological and behavioral responses of preterm infants during blood collection. - To determine how postconceptional age may influence these responses.	Convenience sample (N=11) - Gestational age at birth: 24-26 weeks. - Age at assessment: 27-32 weeks PCA	N/A	- Conclusions limited due to convenience sample. - Too small a number in each gestational age category to make conclusions regarding usefulness of NIDCAP® to assess pain in preterm infants. - Physiological data not digitally sampled. - Assessor not blind to procedure.

LOE - Level of Evidence
PCA: Post conceptual age
* NFCS; Neonatal Facial Coding System (Grunau & Craig, 1987)
† When continuous data analyses were reported, effect sizes (Cohen's r) were calculated using change in frequencies of behaviours between the baseline and invasive procedure (e.g. lance phase of blood collection). Where categorical analyses were reported, phi (ø) was calculated. Point bi-serial correlations were calculated when data types were mixed (i.e. dichotomous and continuous) (Rosenthal, Rosnow & Rubin, 2000).

Peters, 1998; Stephens & Glazer, 1992; Van Cleve et al., 1995):

1. *Hand on face*, a defensive-like action in which the infant places a hand on its face,
2. *Sitting on air*, in which the infant extends its legs fully into the air,
3. *Saluting*, an action in which the infant extends its arms into mid-air, and
4. *Airplane*, in which the infant extends its arms laterally.

Figure 2 provides drawings of these four movements. All these actions have been reported to occur concurrently with changes in physiological and other behavioural measures of pain and stress in responses of preterm infants (Holsti et al., 2004; Holsti et al., 2005; Morison et al., 2003; Peters, 1998).

Hand movements

Finger splay

The sudden backward extension of the fingers, *finger splay*, also appears to be a reliable indicator of stress in preterm infants (See Figure 3). In biobehavioural studies that examine acute pain responses in preterm infants at 32 weeks gestational age, the frequency of finger splay increased significantly during the painful procedure compared to the undisturbed baseline period (Grunau et al., 1998; Grunau et al., 2001; Holsti et al., 2004). Moreover, finger splay may be a developmentally-specific stress cue, since the infants who were born prior to 30 weeks gestational age had a higher frequency of finger splays to heel lance than those born after 30 weeks (Holsti et al., 2004). Not only did these earlier born infants show greater finger splays during lance/squeeze, but also during the Baseline Phase. This finding is consistent with previous studies that examined procedural stress and pain responses in this population (Grunau et al., 2000; Morison et al., 2003) and may indicate *sensitization* (lowered thresholds) which results from greater, early pain exposure (Fitzgerald, Millard & McIntosh, 1989; Andrews & Fitzgerald, 1994).

FIGURE 2
Specific extremity movements indicative of stress. a. hand on face. b. sit on air. c. salute. d. airplane

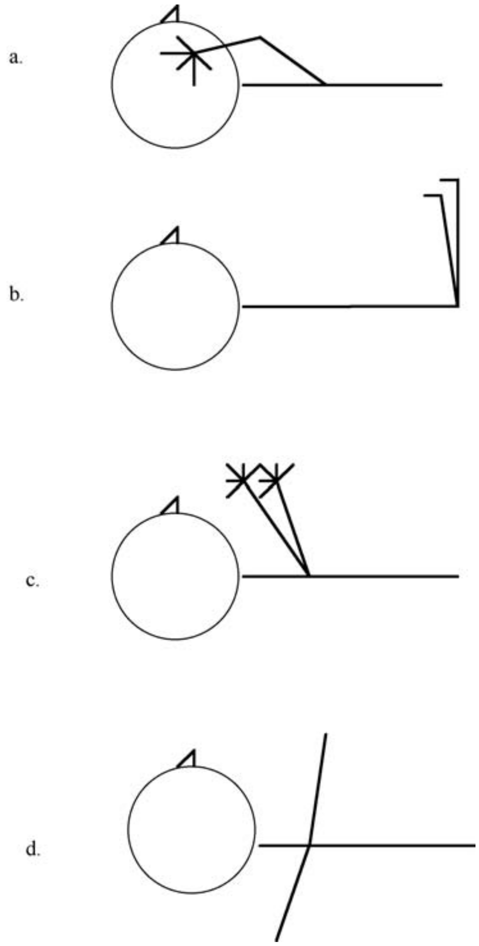


FIGURE 3
Photograph of an infant showing finger splay (a) and fisting (b)

(a) (b)

Photograph printed with parental permission.

While researchers have observed increased finger splays in preterm infants during painful procedures, such as heel lance and endotracheal suctioning (Grunau et al., 2000; Slevin et al., 1998), finger splay has also been observed in non-painful but stressful caregiving tasks including bathing, diapering, and chest physical therapy (Grunau et al., 2000; Holsti et al., 2005; Peters, 1998). Thus finger splay, although not pain specific, is an indicator of stress.

Fisting

According to the NIDCAP®, repeated fisting is usually interpreted as an indicator of stress (Als, 1984; Cheng & Chapman, 1997) and was also described in another study that examined preterm infant responses to pain (Bozzette, 1993). Furthermore, a recent study showed that fisting is considered by a majority of nurses to be a pain indicator (Howard & Thurber, 1998) (see Figure 3). Preterm infants, however, also showed fisting in response to diaper changing; thus fisting could be considered a non-pain specific stress cue (Holsti et al., 2005). In addition, fisting–like finger splay–appears to be a sensitive stress cue in infants born at an earlier gestational age (<30 weeks) and may be useful in identifying stress in those infants who are sicker early in postnatal life (Holsti et al., 2005).

Twitches and startles

Some clinicians consider *twitches and startles* to be indicators of stress in preterm infants (Als, 1984; Stephens & Glazer, 1992). Facial and body twitches have been associated with pain and stressful procedures in preterm infants who were more severely ill shortly after birth (Day 1 and Day 3), when they were assessed at 32 weeks gestational age (Holsti et al., 2004). Despite these findings, we would caution clinicians from relying on these specific behaviours as indicators of stress and pain because twitches and startles have been shown to decrease during painful or stressful procedures (Grunau et al., 2000; Holsti et al., 2004; Holsti et al., 2005; Morison et al., 2003). In fact, twitches may be related to the sleep cycle (Dipietro, Hodgson, Costigan, Hilton, & Johnson, 1996; Visser, 1992); for normal infant development, twitches may be necessary movement which influences neuron cell death, synapse elimination, and muscle fibre differentiation (Blumberg & Lucas, 1996). Thus, the reliability of these movements as stress indicators requires further study.

Conclusion

Accurate assessment of preterm infants' earliest occupations, self-regulation, and purposeful engagement with their environments require an understanding of how subtle movement patterns can be interpreted as stress and pain responses. These responses reflect the developmental mismatch between the infants' environment and its capacity to self-regulate. A set of specific movements now show promise as reliable and valid stress indicators in this population. Although these stress indicators have been observed in infants of varying gestational ages and across a variety of caregiving procedures, only one of the studies cited in this review was randomized. In addition, the studies did not include infants with significant intraventricular hemorrhage and/or parenchymal brain injury. Preterm infants with significant neurological insults have facial and heart rate reactivity during acute pain comparable to preterm infants without significant neurological insults (Oberlander, Grunau, Fitzgerald, & Whitfield, 2002). Further study is needed, however, to determine whether the two groups could be differentiated by their body movement responses.

Undoubtedly, it is not ideal to use these movements in a non-standardized and unstructured way to assess stress responses in preterm infants. We caution clinicians from attempting to provide comprehensive stress and pain assessments using their own non-standardized tools, particularly since interpretation of these movements is highly context-dependent; they can be observed even when infants are at rest, although at much lower frequencies. Such action may be viewed with scepticism by other members of the health care team. Moreover, further research is needed to examine these movements in randomized trials. Ongoing research is directed at integrating these movements along with other biobehavioural measures of pain and stress reactivity in the creation of a standardized stress response scale. A quantifiable tool would help to remove some of the subjective nature of the interpretation of the movements and provide a short and efficient tool for clinical use. In the interim, occupational therapists can continue to use developmental care approaches to minimize the deleterious effects of pain and stress associated with NICU procedures. For example, lowering lighting, reducing noise, and using non-nutritive sucking and facilitated tucking are interventions that reduce preterm infant stress responses. In this way, occupational therapists can contribute actively as collaborators in the treatment of pain and stress in these highly vulnerable infants.

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Book Marks ▪ Livres à la page

The Ludic Model: Play, Children with Physical Disabilities and Occupational Therapy, 2nd edition, (2005)

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The Ludic Model, 2nd edition, is based on the amended and expanded third French edition. As well as presenting the model again it includes updates that incorporate recent research, feedback from occupational therapists, possible parent and team member responses, and elements of the author's current additional book publications on play in children.

Francine Ferland systematically provides a historical and philosophical review of play, children and occupational therapy in the Canadian context to establish the model. Although the focus remains on children with physical disabilities, the application to children with intellectual disabilities, developmental delays, pervasive developmental disorders, mental health problems, and adults is briefly expanded with research results, occupational therapy practitioner feedback, and the author's reflections. In addition, the significant role of the parent is illustrated.

The development of the model thoughtfully challenges occupational therapy perceptions of child-centered practice to ensure play is not only the medium, nor only the objective but transparently the attitude of intervention. Discerning questions, visual figures and tables, a clinical case sample, and a methodical presentation of the model concepts and therapeutic implications all contribute to the reader's understanding and integration of the theory and practice of the model.

This book will be of interest to occupational therapists who work with children and their families in all settings. It supplies a clear and concise perspective on the Ludic premise and concepts, research results, occupational therapy practice applications and two useful assessment tools.

Christel K. A. Seeberger