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The Nature, Expression and Impact of Pain in Children With Severe Cognitive Impairments

By

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Submitted in partial fulfilment of the requirements for the degree of

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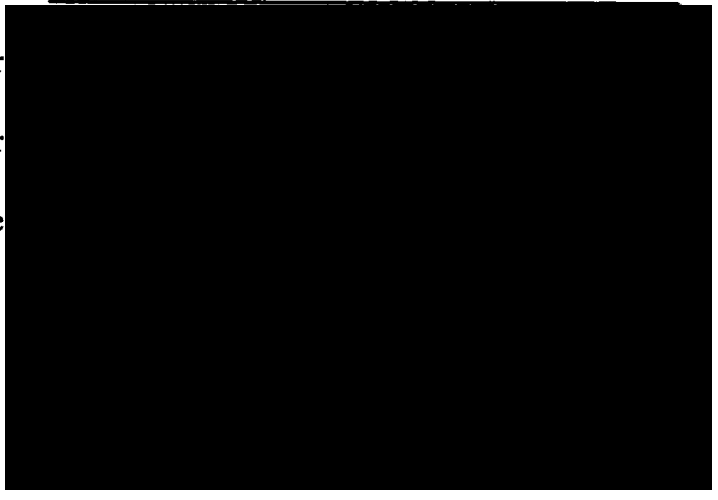

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DEDICATION

This thesis is dedicated to my husband, Pierre, and my two children, Genny and Michel André. This work could not have been accomplished without their love and support.

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ABSTRACT

This research investigated the pain of a cohort of 101 children aged 3 to 18.7 years (M = 10.3, SD = 4.4) who had severe cognitive impairments. Five studies are described, each included a subgroup of this cohort.

Study 1 examined four Pain Surveys administered to caregivers by telephone at 3-month intervals. During each, caregivers reported the features of their children's pain over the past week. They had, on average, over 9 hours of pain each week and 35% to 52% had pain each week. Most pain was due to chronic conditions or illnesses (82%).

Study 2 examined the effect of pain on 89 children's development of adaptive skills. Skills in four areas (communication, daily living skills, socialization, motor skills) were documented at entry to the study and one year later. Time with pain (hours, minutes), over four one-week periods at three-month intervals was summed and its effect on change in the four adaptive areas examined. Children with more pain gained significantly fewer adaptive skills overall. Univariate effects of pain were significant for communication, daily living skills and motor skills.

Study 3 investigated the psychometric properties of the Non-communicating Children's Pain Checklist-Revised (NCCPC-R) for everyday pain. As part of one-week Pain Diaries, 71 caregivers completed the NCCPC-R and rated the intensity of pain occurring during daily two-hour observations. Comparisons were made between scores during observations with and without pain. The NCCPC-R demonstrated excellent internal reliability (α = .93), total scores were significantly correlated with pain intensity ratings (r = .46), and total scores during pain (M = 21.1, SD = 15.9) differed significantly from scores when pain was absent (M = 4.8, SD = 5.2). Receiver operating characteristic (ROC) curves indicated a total score of 7/90 had 84% sensitivity and 68% specificity. For

Study 4, researchers and caregivers completed the Non-communicating Children's Pain Checklist-Postoperative Version (NCCPC-PV) before and after 24 children had surgery. Total postoperative scores were significantly greater (M = 12.2, SD = 10.9) than preoperative scores (M = 4.8, SD = 3.6) and ROC curves indicated a total score of 11/81 provided good sensitivity (75% - 88%) and specificity (63% - 81%). Inter-rater reliability of total postoperative NCCPC-PV scores was good (r = .72).

Study 5 examined the validity of the Child Facial Coding System (CFCS) for postoperative pain in 26 children. The 13 facial actions of the CFCS were coded for five 10-second time segments selected from film collected in the recovery room. Visual analogue scale (VAS) ratings of pain intensity and sedation were also made for each segment. Children with VAS pain ratings of 30 or more out of 100 displayed significantly more frequent and intense facial actions. Facial action was not related to sedation ratings. Categorical principle components analyses revealed a "pain face" (brow lower, nose wrinkle, nasolabial furrow, cheek raiser, horizontal mouth stretch and flared nostril) that was related to higher VAS pain ratings and analgesic administration.

The results of these studies suggest children with severe cognitive impairments have frequent pain that is most often due to chronic conditions and illnesses and that pain reduces development of adaptive skills. The results also suggest observational pain measures are viable with this group. The importance of these results to furthering research and improving clinical care for children with severe cognitive impairments is discussed.

LIST OF ABBREVIATIONS AND SYMBOLS

Abbreviations

NCCPC	Non-communicating Children's Pain Checklist
NCCPC-R	Non-communicating Children's Pain Checklist - Revised
NCCPC-PV	Non-communicating Children's Pain Checklist - Postoperative Version
ROC curve	Receiver Operator Characteristic Curve
VABS	Vineland Adaptive Behavior Scales
VAS	Visual analogue scale
VAS-Pain	Visual analogue scale of pain intensity
VAS-Sedation	Visual analogue scale of sedation intensity

Symbols

α	Coefficient alpha, or probability level
ANOVA	Analysis of Variance
χ^2	Chi Square Statistic
F	f-test statistic
M	Mean
Mdn	Median
n	Number of participants in a subsample
N	Number of participants in total sample
p	Probability level
SD	Standard Deviation
t	t-statistic
U	Mann Whitney U statistic

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This research could not have been completed without the help of many people. First, the caregivers and children who took part in these studies must be recognised. They contributed their time, effort and knowledge to this program over the course of years. They also gave of their hearts. Since this program began in 1999, I have had the good fortune to meet most of them, at the IWK Health Centre, at their homes, and at group homes and schools. These special people, who already carry a heavier burden than most, were eager to take part as fully as possible. Their commitment and perseverance set the standard high, and all who worked on this project were inspired by their example.

I would also like to acknowledge the tremendous assistance of three members of my committee who have been at my side since this project began. Dr. Patrick McGrath, my supervisor, taught me to dream big and not let obstacles obstruct my view of possibilities. His belief in the value of research to individual children set the tone for this research and provided incentive for excellence. His commitment to the scientist-practitioner model of psychology influenced my research and clinical work and has shaped my goals in this field. Dr. Carol Camfield also provided invaluable support. She opened my eyes to the world of paediatric neurology, something I now find intriguing, but once viewed with trepidation. Her ability to envision the everyday struggles of the caregivers and children who participated in these studies helped in making this research as real-world friendly as possible. Finally, her warm kitchen and home made bread provided a refuge when, as can happen, the demands of such a large program research seemed overwhelming. Dr. G. Allen Finley also contributed to making this project a

success. He demystified the world of surgery and translated into English the many drugs, procedures and tests that are part of that world so that it was possible to develop protocols for the surgical studies described here. He also provided moral support with his steady applause for the novelty, scope, and breadth of the program.

I would also like to acknowledge Drs. Dan Waschbusch, Peter Camfield, and Todd Mondor. Dr. Waschbusch was a member of my Dissertation Committee, but was not able to take part in the commencement of this research. However, his input at committee meetings and during the writing of this manuscript provided an outside view that helped to keep this thesis accessible to those outside of the field of paediatric pain research. Dr. Peter Camfield was not a member of the committee. However, over the course of this project he provided invaluable input into the design and manuscripts of the studies conducted. Again, his view from outside the field of paediatric pain research added to the depth of this research and this thesis. Dr. Mondor introduced me to the world of research. His glee over new puzzles to solve and exclamations of "there's nothing better than a day with data" helped set me on this path.

Because this research spanned several years, many others contributed to its completion. Beth Currie Shier was instrumental in the planning of the original protocol. Jill MacLaren provided hands on support as well as being a colleague with whom I could share ideas. Heather Wortman volunteered time to collect data for more than two years. However, this research would not exist if it were not for the commitment of Alyson Currie, the Study Co-ordinator. Alyson not only carried out the day-to day tasks that kept this project going, such as record keeping, data entry, organising visits to homes and

filming in the IWK Health Centre, she also, in many ways, became the "heart" of this project. Her deep caring for the children shone through to them and their caregivers. This project was clearly more than a "job" and her willingness to go the extra mile made that clear, from driving through storms to meet a families, writing personal notes to them, or spending hours in a windowless file room tracking down that one piece of information I really needed "yesterday".

Finally, I would like to thank my family. They have all stood behind me since I returned to university in 1993. They have also all shown a real interest in the research and the children that are described in this thesis. In a sense, they felt their support of me was their contribution to helping this special group of children. I agree. My husband Pierre made this work happen. His encouragement kept me going. His willingness to lend a hand, whether it was through gathering papers at the library or doing dishes, gave me the time. He was there to cheer me on during the "ups" and hold my hand during the "downs". He never doubted its success. Genny, now 17, only vaguely recalls a time when Mom was not in school. Over the past nine years she has cheered me on, lent a hand, and been there to share complaints about "homework". Her encouragement made me smile. Michel André, who is now 11, cannot remember when I was not in school. He has been an energetic coach and shown an understanding beyond his years when "the research" encroached on his life. He earnestly believed this research was important and that I could accomplish anything. His faith was comforting.

CHAPTER ONE: INTRODUCTION

> But by far the greatest obstacle to the progress of science and to the undertaking of new tasks and provinences therein, is found in this - that men despair and think things impossible. (Francis Bacon, Novum Organum, XCIX; as cited in Robinson (1986), p. 213)

This thesis describes a program of research that began as an attempt to measure the pain of children who cannot communicate verbally due to severe cognitive impairments. Out of that goal, many paths emerged. As the possibility of measuring their pain evolved from hope to reality, countless opportunities unfolded. Measuring pain opened the door to documenting the occurrence of pain and the impact of that pain on the children's lives. The success in measuring pain in one context led to exploration of measuring pain in others and the examination of other methods for measuring their pain. The richness of the pain behaviour displayed by these children led to questions regarding the factors that may influence their pain expression. Finally, the successes at measuring pain lead to questioning of previous assumptions that these children do not display pain behaviour. Could other factors, within the observer, be at play? These questions, and many more, were the basis of an "Experimenta Lucifera", described by Bacon and conducted in the spirit of discovery. In his words:

> Now experiments of this kind have one admirable property and condition; they never miss or fail. For since they are applied, not for the purpose of producing any particular effect, but only for discovering the natural cause

> of some effect, they answer the end equally well whichever way they turn out; for they settle the question. (Francis Bacon, Novum Organum, XCIX; as cited in Robinson (1986), p. 213)

Thus, the studies presented here were designed to uncover knowledge, not to test hypotheses. Some researchers have investigated the pain of children with cognitive impairments in relation to children without cognitive impairments (Leland, Garrard, & Smith, 1994; Gilbert-Macleod, Craig, Rocha, & Mathias, 2000), and this can provide useful information from a theoretical perspective. Others have conducted studies with a priori expectations of the way children with cognitive impairments will display pain (Oberlander, Gilbert, Chambers, O'Donnell, & Craig, 1999). This current research did neither. Instead, a deliberate attempt was made to design studies that would uncover the full nature of the pain these children experience and its concomitant behaviour, however unique it might be.

Paediatric Pain

According to the International Association for the Study of Pain, pain is an "unpleasant sensory and emotional experience associated with actual or potential tissue damage, or described in terms of such damage" (International Association for the Study of Pain & Task Force on Taxonomy, 1994). This assumes a correspondence between pain experience and pain report that can be expressed in a specific manner. This definition has been problematic for those who cannot provide self-report verbally (Anand & Craig, 1996) and is one reason research into children's pain has typically followed in the wake of advancements into adults' pain, because it relegates research into pain through other

methods, such as behaviour, to a "second tier" methodology. This definition also emphasises self-report of pain in terms of actual or potential tissue damage, something children may be unable to do at certain developmental stages.

It was only with Eland and Anderson's seminal paper on the differences in pain management afforded children and adults, that paediatric pain began to emerge as an independent field of enquiry (Eland & Anderson, 1977). They compared the analgesics given to 25 children in hospital for surgery to that given 18 adults with identical diagnoses. Only 12 of the children received any analgesia at any point during their hospitalisation. This totalled 24 doses. In contrast, the 18 adults were given 671 doses of analgesia. This discrepancy in the treatment given children and adults spurred researchers and clinicians alike to investigate paediatric pain further. Since that time, the number of studies into children's pain has increased (Guardiola & Banos, 1993), and the care provided for children has improved (Broome, Richtsmeier, Maikler, & Alexander, 1996; Schechter, 1997). However, there is clearly room for advancement.

Measurement has been the focus of most paediatric pain research. It is the cornerstone of effective management. It is also the foundation for research into the aetiology of pain, the consequences of pain, and the advancement of pain management. This is important because, in children, untreated pain can lead to destabilisation (Anand & Hickey, 1992), and sensitisation of the brain to future painful experiences (Grunau, Whitfield, Petrie, & Fryer, 1994; Taddio, Goldbach, Ipp, Stevens, & Koren, 1995). The experience of pain is also central in children's experience and view of hospitalisation (Woodgate & Kristjanson, 1996).

Because pain is a subjective experience, and because this is emphasised in the most accepted definition of pain (International Association for the Study of Pain, 1994), self-report is often considered the "gold standard" in pain measurement (McGrath & Unruh, 1987). There are many situations, however, in which it is not appropriate or available from children. There are questions about the reliability of self-reports of pain intensity by children under three years (Johnston, 1993). Children with severe cognitive impairments or who are unable to communicate verbally are also unable to supply self-reports of pain (McGrath, 1998a). Some authors have also suggested that children will resist reporting pain to avoid the discomfort associated with analgesia delivered by needle (Eland et al., 1977), and children suffering pain after surgery may not be in condition to report their pain (Beyer, McGrath, & Berde, 1990). Thus, observational methods of pain assessment are vital to paediatric pain management.

Research into paediatric pain has also taught us that each child's pain experience is unique. Although our knowledge is still incomplete, we do have enough information to believe that a child's personal characteristics, such as age (Goodenough et al., 1997), gender (Carr, Lemanek, & Armstrong, 1998) and temperament (Rocha, Prkachin, & Beaumont, 1998) may influence both their pain experience and their expression of pain. We also have growing confirmation that context (Sweet & McGrath, 1998) and social factors (Goodman, McGrath, & Forward, 1997) interact with these personal characteristics, creating a dynamic process. This means that pain assessment methods must be robust to individual differences if they are to be clinically useful. However, it has also meant that a great deal of paediatric pain research has been conducted on

homogeneous groups of children to simplify validation of measures or the testing of treatments. This has lead to exclusion of some children from research, especially those with severe cognitive impairments.

Pain in Children with Cognitive Impairments

In contrast to the main body of paediatric pain research, research aimed at understanding the pain of children with severe cognitive impairments has just begun and we know very little about their pain. These children are often referred to as "mentally challenged", "intellectually disabled" or "cognitively impaired", and those with additional physical impairments are sometimes referred to as "multiply disabled" or "multiply handicapped". Most of these children have severe limitations in their ability to function on a daily basis. Children who have severe cognitive impairments usually have suffered neurological injury prior to or at birth, have genetic syndromes, were extremely premature, or have suffered severe brain injury in early childhood. Thus, they usually have suffered brain damage that affects many areas of their functioning.

Because these children are usually not able to use speech to communicate, they have been excluded from most studies of paediatric pain in which children are asked about their pain. This exclusion also reflects the unspoken hypothesis that the very characteristics that set these children apart from most children are relevant to their pain experience and behaviour. That is, there is a belief that the neurological damage that underlies their cognitive impairments may also alter how they feel, interpret or react to pain (McMurray, 1955; Biersdorff, 1994). These children are also typically excluded from studies of specific painful conditions, such as pain associated with cerebral palsy or

pain due to gastrointestinal disorders, because they often have several medical conditions that may cloud investigation of pain due to one. They are also frequently blind or deaf, and many display autistic behaviours. This makes studies of their pain behaviour difficult, as their behaviour can be altered by these conditions (Curcio, 1978). The behaviour of individuals with cognitive impairments has been reported to be idiosyncratic, with facial expressions that may be viewed as exaggerated when they do not have pain (Trauner, Ballantyne, Chase, & Tallal, 1993). This may make it difficult for observers to interpret their behaviour (Maurer & Newbrough, 1987a; Maurer & Newbrough, 1987b). It also means that pain measurement tools based on "typical" pain behaviours shown by children without impairments may not be suitable. In summary, a number of factors have lead to this group being excluded from the bulk of paediatric pain research.

This exclusion has dire consequences, as this group of children is particularly vulnerable to pain. Because many have physical disabilities (Nordin & Gillberg, 1996), they may be at high risk for suffering pain due both to their underlying physical disorders and to treatments for those disorders. Their complex medical profile may also lead to pain going untreated for longer periods when it does occur, because adults are unaware they have pain or are challenged to find the cause of the pain. Those with severe cognitive impairments may also be at greater risk for fatality due to delayed diagnoses, because their pain is not noticed by caregivers (Jancar & Speller, 1994) and they may also be at greater risk for accidental injury. For example, one study found that children in

a day-care setting were more likely to suffer injury if they had cognitive impairments (Leland et al., 1994).

This combination of factors has lead to a situation in which children with severe cognitive impairments are more likely to suffer needlessly than do their peers without impairments. Added to these factors is the possibility that this group, as a whole, is undervalued by our society (McGrath, 1998b). In addition, children with multiple chronic medical conditions are increasingly more likely to survive due to advancements in medical technology (Nicholson & Alberman, 1992; Lorenz, Wooliever, Jetton, & Paneth, 1998). In some cases, the children's multiple medical conditions are so complex that complete relief from pain is unlikely. They may always have pain in their lives. Thus, a growing number of vulnerable children are at risk for suffering if advancement in the assessment and management of their pain is not forthcoming.

Questions Regarding the Pain Experience of Children with Cognitive Impairments

Because the study of pain in children with cognitive impairments is new, theories regarding specific aspects of their experience have not been proposed. However, the most frequently discussed issue regarding pain in individuals with cognitive impairments is that of whether reports that their pain is not detected (Roy & Simon, 1987; Jancar et al., 1994) or that observers' manage their pain differently (Fanurik, Koh, Harrison, & Conrad, 1996) are due to deficits in the communication of their pain or to the fact that they do not attempt to communicate their pain because of pain insensitivity or pain indifference. In this context, the term "pain insensitivity" is most often used to describe deficits in the sensory perception of pain, while the term "pain indifference" is used to refer to deficits

in an individual's evaluation or interpretation of the sensory experience of pain (Biersdorff, 1991).

Those who feel that pain insensitivity or pain indifference are important factors in poor pain detection suggest that individuals with cognitive impairments frequently do not generate signals to communicate their pain because their experience of pain is altered. The most direct discussion of this issue is presented by Biersdorff (Biersdorff, 1991). In effect, she suggests they may not attempt to communicate pain because they may not feel it (pain insensitivity) or they may not interpret what they feel as negative (pain indifference).

As Oberlander (Oberlander, 2001) points out, there is no definitive evidence to suggest that the neurological impairments that underlie the cognitive impairments of these children do lead to an altered pain experience. Although neurological damage could reduce pain sensation or lead to pain indifference, it is also possible that the neurological damage these children have suffered could lead to a heightened pain experience. However, those who have suggested cognitive impairments may be associated with pain insensitivity or indifference have not based their arguments on evidence of altered neurological structures that could be responsible for pain processing, but on a perceived lack of pain behaviour by individuals with cognitive impairments in situations where it was believed pain should have been experienced. It is important to note that these studies were conducted using informal retrospective reports by caregivers (Biersdorff, 1994) or pain measures that had not been validated with this group (Lu, 1981). Thus, this view posits that the lack of observed behaviour reflects an altered experience.

However, it is also possible that the apparent insensitivity and indifference displayed by individuals with cognitive impairments are the result of problems with the communication of pain. Craig suggests that factors within the individual sending signals of pain and factors within the individual receiving signals of pain can lead to poor communication of that pain (Craig, Lilley, & Gilbert, 1996). For example, some have suggested that individuals with cognitive impairments have idiosyncratic patterns of expressing pain (van Dongen, Abu-Saad, & Hamers, 2000a; van Dongen, Huijer Abu-Saad, Hamers, & Terstegen, 2000b) that make detection of behaviours reflecting pain difficult. This suggests a problem with the sender of signals in that those signals are not consistent. Others have argued that an emphasis on the verbal report of pain has led to under-recognition of pain in those who cannot use spoken language (Anand et al., 1996; Anand, Rovnaghi, Walden, & Churchill, 1999), such as infants and those with cognitive impairments, because observers are not aware of non-verbal communication of pain or discount the importance of non-verbal communication of pain. Still others have suggested that attitudes and beliefs regarding the pain experience of individuals with cognitive impairments held by observers could interfere with accurate detection or interpretation of pain behaviour (MacLaren, Breau, McGrath, Finley, & Camfield, 2001). These latter two suggestions reflect problems within the receiver of the pain signal. The unifying hypothesis underlying these various proposals, however, is that poor detection of pain is not due to the fact that pain signals are not sent. Rather, the breakdown lies in the process of communication of those signals from sender to receiver.

Three studies presented here addressed this issue because they examined the ability of structured pain tools to measure the pain of a cohort of children with severe cognitive impairments. Results indicate these tools have good psychometric properties and are sensitive to the pain of these children, providing evidence that the lack of valid, reliable tools for measuring pain in this group may have given the false impression that pain behaviour was absent. These results also suggest that their pain behaviour is not too heterogeneous to be measured using a standardised tool. Thus, the results suggest a breakdown in the communication of pain that most likely lies at the receiving end of the communication process. These results do not directly test whether pain insensitivity or indifference could also cause poor pain detection in some children with cognitive impairments. However, they do raise questions regarding the view that deficits in communication of pain necessarily reflect an altered pain experience. These results will also open the door to more rigorous investigation of the possibility that children with cognitive impairments have an altered pain experience, whether that be a reduced or increased experience.

Summary

Evidence suggests that pain may be a substantial problem for children with severe cognitive impairments. They may be more likely to suffer painful conditions and procedures and their pain may be inadequately managed. The literature review in the next chapter indicates research into their pain has emerged slowly. However, the review also reveals a shift towards the increasing use of scientific methodology in the study of pain. This trend will provide testable conclusions concerning many aspects of pain in this

group, from questions regarding the possibility that their pain experience is altered to validation of pain assessment and treatment strategies. The studies described in this thesis build on the literature described by providing information regarding the nature and impact of pain in these special children and through validation of pain measurement tools that will make further research possible.

CHAPTER TWO: PREVIOUS RESEARCH REGARDING PAIN IN CHILDREN WITH COGNITIVE IMPAIRMENTS

Early Work

The first study to investigate the pain of children with cognitive impairments was published in 1965 (Reynell, 1965). Reynell followed the reaction of 50 children with cerebral palsy after surgery, primarily orthopaedic. Eighteen of these children, aged 2 to 14 years, had an intelligence quotient (IQ) that was below 70, a level generally associated with a diagnosis of mental retardation (American Psychiatric Association, 1994). The children's behaviour was observed and recorded during physiotherapy sessions for up to nine months after surgery. This included behaviour that would indicate pain, including verbal or non-verbal complaints, flinching and withdrawal, and behaviour believed to reflect their emotional state, such as excitation, laughing or smiling, talking, quietness, whimpering or complaining and crying. The results indicate there was no relation between children's pain related behaviour or emotional behaviour and their level of intellectual functioning (IQ less than 70, IQ of 70 to 90, IQ greater than 90). This was the first evidence that children with multiple disabilities do experience pain under circumstances most children would be expected to have pain and that their pain behaviour is observable.

Although the assessment of pain has advanced rapidly for the general population since the time of Reynell's study (Reynell, 1965), advancements in the assessment of pain for individuals with severe cognitive impairments have been much slower in coming.

Over twenty years later, in 1988, the next study of pain in people with severe cognitive impairments appeared. Mette and Abittan reported on their attempts to discern pain in four children and two adults with severe multiple disabilities (Mette & Abittan, 1988). They asked five observers to make visual analogue scale ratings of the children's pain at a time when they were suspected of having pain and again several weeks later. They found the ratings provided by the observers distinguished between the observations with and without pain in all but one case.

A decade later, case series began appearing that provided rich accounts of individuals' reactions to pain (Collignon, Giusiano, Porsmoguer, Jimeno, & Combe, 1995). For example, Collignon and his co-authors describe a teen with severe cognitive impairments who was observed to cry and slap her face repeatedly for no known reason (Collignon, Porsmoguer, Behar, Combe, & Perrin, 1992). Upon examination, it was discovered she had suffered a fracture of her right clavicle. In another publication, this group describes a 10-year old child with multiple disabilities who reacted with disorganised agitation when examined after surgery (Collignon et al., 1995). They describe the child's reaction as similar to that of an infant. Although these reports provided insight into the clinical experience of working with these children, they did not advance development of validated, quantitative pain measurement tools.

Towards Quantitative Measurement of Pain

The first study to quantify the pain behaviour of people with severe cognitive impairments was published in 1995 (Giusiano, Jimeno, Collignon, & Chau, 1995). The

behaviour of 100 individuals aged 2 to 33 years (average 16 years) residing in a long-term care facility was used as the basis for developing a list of behaviours that occur during pain. These individuals could not speak, 45% had no voluntary movement of any limbs, and 70% were described as having a "chronic vegetative state". Fifty of the children had been referred for pain problems, while the remaining 50 were randomly selected from the remaining residents of the facility. A physician-generated checklist containing 22 items was used to record the behaviour observed by a physician or nurse during a physical examination. The observer's opinion of whether the individual had pain (yes/no) and their certainty in that opinion (0 to 2) were used as the criteria for determining if the behaviours observed were related to pain.

A neural network analysis was used by Giusiano and his colleagues (Giusiano et al., 1995) to examine the relationships between the observers' opinions of the presence of pain, the behaviours observed and the individual's degree of physical dependence (not very dependent, quite dependent, and very dependent). A painful expression during manipulations by the examiner was most related to pain, followed by lack of interest in surroundings, lack of interaction with the adult present, protection of the painful area, "calm autistic" behaviour and search for a less painful position. The behaviours most related to the individuals' degree of dependence were a painful expression during manipulations by the examiner, protection of the painful area, spontaneous painful expression, increased voluntary movements, search for a less painful position, and a guarding reaction when the painful area was brushed. It is notable that

only two of the five behaviours that were most related to pain were strongly related to the individual's degree of independence, a painful expression during manipulations by the examiner and protection of the painful area. However, the greatest contribution of this study was the fact that it provided the first indication that a set of common pain behaviours could be found for a group of people with severe cognitive and physical impairments. Unfortunately, this study only examined pain during a physical examination, and was limited to a group of children suffering from cerebral palsy, limiting the generalisability of the results. In addition, the items generated were based on change from "usual" behaviour. This makes it difficult for people not familiar with a child to use the instrument generated.

A similar effort was begun by McGrath and his colleagues shortly thereafter, in an effort to develop an objective observational measure of pain for these children (McGrath, Rosmus, Camfield, Campbell & Hennigar, 1998). They conducted semi-structured interviews with the primary caregivers of 20 individuals aged 6 to 29 years, 18 of whom had spastic quadriplegia or hemiparesis. All had severe or profound mental retardation and their average communicative age, based on caregivers' responses to the MacArthur Communicative Development Inventory (Fenson L. et al., 1993) was 8 to 15 months.

Thirty-one behaviours were extracted from the interviews by two authors who independently reviewed transcripts of the interviews. The transcripts were then coded for the 31 items by a researcher who was not involved in generation of the items. An additional researcher also coded eight of the transcripts to assess reliability. Reliability

between the two coders was .77, indicating good agreement. The 31 items were reported by 5% to 90% of caregivers. The items most frequently described by caregivers were those involving vocal expressions such as crying (90%) and moaning, whining or whimpering (80%). Only 11 items were described by fewer than 25% of the caregivers, while 10 were reported by at least 50%. These items were grouped into seven categories (Vocal, Eating/Sleeping, Social/Personality, Facial Expression, Activity, Body & Limbs, and Physiological). The least used behaviours were those belonging to the Activity and Physiological categories. Although specific behaviours varied amongst individuals, caregivers' reports indicated all individuals displayed some behaviour from each of the seven categories. This study confirmed the findings of Giusiano's group (Giusiano et al., 1995), that the pain behaviour of people with severe cognitive impairments was observable and that there was some consistency in the type of pain behaviours displayed by them during pain. This confirmation set the stage for further research specifically aimed at development and validation of pain measurement tools that could one day be used in clinical practice.

In a subsequent study, this group provided a preliminary validation of their pain measure, the Non-communicating Children's Pain Checklist (NCCPC), when used in a home setting (Breau, McGrath, Camfield, Rosmus & Finley, 2000). The caregivers of 33 individuals, aged 3 to 44, completed the NCCPC for four observations, two in which the individual with severe cognitive impairments had pain, one in which they were distressed but had no pain, and one during which they were calm. Despite the observations being

based upon a heterogeneous set of acute and longer-term pains, the test-retest reliability indicated there was consistency over time in the individuals' pain behaviour. Scores on the checklist also differentiated between observations with pain and when the individuals were calm. This study provided the first evidence of consistency in the pain behaviour of people with severe cognitive impairments over time. In a later study, it was shown that caregivers' retrospective report of a core set of pain behaviours from the NCCPC could significantly predict the display of those behaviours during subsequent pain episodes (Breau, Camfield, McGrath, Rosmus, & Finley, 2001a). This finding provided the first suggestion that caregivers' descriptions of what their child usually does during pain could be used to assess discrete episodes of pain. These studies indicated the pain behaviour of children with severe cognitive impairments was not idiosyncratic, was measurable, was consistent over time and was predictable.

The Scope of Research Widens

Several other studies appeared in the late 1990's investigating some aspect of pain in children with cognitive impairments. Each of these has added to what is known about the pain experienced by this particular group. In 1996, Fanurik, Koh, Harrison and Conrad reported a study of sixty-six children with cognitive impairments (Fanurik et al., 1996). They asked the parents of these children to describe their child's behaviours during a previous needle stick. The behaviours parents described included: verbalisation, localisation of the painful area, cry, behavioural or emotional changes, facial expression, body movement and self-abusive behaviour. A limitation of this study, however, was that

it was retrospective. In addition, only 41% of the children involved had severe cognitive impairments and 48% of these children did have verbal skills.

Fanurik and her colleagues also conducted a study of the self-report skills of children with borderline to profound cognitive impairment (Fanurik, Koh, Harrison, Conrad, & Tomerlin, 1998). Only 47 of these children were capable of completing the tasks involved in the assessment of their self-report ability. Of the 47, aged 8 to 17 years, only 10 showed an understanding of the concepts required to rate pain on the numerical score, and all had a diagnosis of borderline to mild mental retardation. In fact, only one child with severe mental retardation was able to arrange five blocks in order of size, a preliminary task in the protocol. No children with moderate to profound impairment could complete tasks in which they were required to indicate the order or magnitude of numerals. This study provided confirmation that, for most children with cognitive impairments, self-report of pain would not be a viable option.

Fanurik's group also interviewed the parents of children to collect information regarding the children's pain experience, expression and treatment (Fanurik, Koh, Schmitz, Harrison, & Conrad, 1999a). Fifty-nine percent of 145 parents expressed a belief that their child experiences pain differently than children without cognitive impairments, but this belief did not vary due to the child's level of cognitive impairment. When asked specifically about pain sensitivity and pain tolerance, 12% of parents stated their child was less sensitive to pain than children without cognitive impairments, 42% felt their child was more tolerant of pain, and 23% felt their child was both less

sensitive and more tolerant. Some parents described a lack of reaction to pain as the reason for this belief.

When asked how they know their child has pain, the parents in this study also described different pain behaviours for children with mild or moderate cognitive impairments than those with severe to profound impairments (Fanurik et al., 1999a). Whereas 57% of those with mild or moderate impairments were said to make direct verbal statements about pain, only 7% of children with severe to profound cognitive impairments were said to do so. In contrast, 22% of children with severe to profound cognitive impairments, but only 10% of children with mild to moderate cognitive impairments cried in response to pain. Similarly, 45% of parents of a child with severe to profound cognitive impairments said their child displays a combination of behaviours when in pain, while only 12% of parents of a child with mild or moderate cognitive impairments reported the same. These results suggested pain behaviour might vary with children's level of cognitive impairment.

Despite the fact that many parents in Fanurik et al.'s study believed their child experienced pain less intensely than children without pain, many also felt their child's pain was underestimated or under-treated by healthcare professionals (Fanurik et al., 1999a). Twenty-nine percent expressed a belief that their child's pain was treated differently because of their cognitive impairments.

Interestingly, this team of investigators also found, in a subsequent study, that healthcare professionals were not influenced by children's level of cognitive impairments

when making judgements about pain for children depicted in vignettes (Fanurik et al., 1999b). In that study, nurses' and physicians' estimates of a child's pain due to a posterior spinal infusion or a kidney biopsy did not differ in relation to the severity of the cognitive impairments depicted. However, only a global pain rating was given, based on vignettes, and the pain depicted was due to specific standardised procedures. Thus, the results may not reflect professional's beliefs regarding other types of pain experienced by children with cognitive impairments.

Others have investigated whether physiological factors can provide valid measurement of pain in children with severe cognitive impairments. Oberlander et al. examined the response to pain in eight adolescents with spastic quadriplegia (Oberlander et al., 1999). The adolescents were filmed and their heart rate was monitored while they received both a mock and a real injection in random order. The Facial Action Coding System (Ekman & Friesen, 1978) and the Child Facial Coding System (Chambers, Cassidy, McGrath, Gilbert, & Craig, 1996) were used to code the videotapes for facial movements. They report that there was no significant increase in heart rate or facial action during the injection. However, several weaknesses of that study suggest the results should not be taken as definitive. First, no control group was used. Thus, there is no way of knowing if the reaction shown by these adolescents differed in any way from that a group of peers without impairments would display. Second, the sample used was small and no power analyses were provided to confirm there was sufficient statistical power to detect differences. Third, the authors did not indicate the time between mock and real

injections. This is a problem, as some of the participants received a real injection first, and previous research using the Child Facial Coding System with children who are not impaired indicates facial response to injection pain may linger after insertion is complete (Breau et al., 2001b). Thus, it is possible the participants who received a mock injection after a real injection were still displaying a reaction to the actual insertion of a needle. Finally, these authors chose to analyse only facial action occurrence, through summing the occurrence of all actions over each period of time examined. Previous research suggests the intensity of facial action carries important information regarding pain, and inclusion of this parameter may have altered the results (Breau et al., 2001b).

While the studies described thus far have examined pain expression and treatment, another has also examined the cause of pain in children with cognitive impairments. Hunt et al. asked the parents of 120 people aged 1 to 25 years with static or progressive encephalopathies to describe the causes of pain that their child suffered (Hunt & Burne, 1995). The most frequently described cause was gastrointestinal disorders (56%), followed by musculoskeletal problems (54%), seizures (21%) and headaches (16%). In all, 70% of parents reported their child had pain. The most common sign of pain was change from usual postural movements and crying, shown by 96% of the children, facial expression, shown by 88%, and physiological changes shown by 58%. Sixty-five percent of the parents also reported their child's mood changed when they have pain.

Another group documented the occurrence of pain in a group of 34 children with cognitive impairments by having their caregivers complete two-week diaries (Stallard, Williams, Lenton, & Velleman, 2001). They report over 73% of children had pain at least once and that 52% had pain on four or more consecutive days. They also report that most pain episodes lasted longer than 10 minutes and that most pain of moderate to severe intensity occurred in the overnight hours. Unfortunately, they did not include description of the causes of the children's pain.

Summary

As the brevity of this review suggests, research into the pain of children with severe cognitive impairments is in its infancy. Since the program of research described in this thesis began, only six reports of research have been added to the literature. These have begun to lay a foundation, but are by no means sufficient to reach any conclusions regarding the nature, assessment, treatment, or impact of pain in this vulnerable group. The studies described in the next chapter represent a concerted effort to strengthen this foundation.

CHAPTER THREE: THE STUDIES

The studies reported in this thesis are part of a program of studies conducted with the cohort of children, families and caregivers described in the next chapter. Participants began the tasks of the protocol as they entered. The studies presented here are based on information collected over the first two years of the program, during which time most participants completed at least one year of the two-year protocol.

Five studies from this research program are discussed in this thesis. Each consists of a portion of the data collected from the participants and addresses a specific aspect of pain in this group of children. Because of the long-term nature of this research, and the varied circumstances of the families and children who took part, all did not complete every measure. For example, some families moved or went through family crises over the two years, and several children were hospitalised for several months for life-threatening medical conditions. Thus, there were some cases in which asking caregivers to comply with the study protocol would have created an excessive burden for them. In some cases, it was possible to postpone completion of measures, but in others, this was not possible. In addition, some children died before completion of the study and some families withdrew due to their child's or their own health status deteriorating to the point of becoming life-threatening (e.g., one parent was diagnosed with inoperable cancer). With the consent of their caregivers, the data collected concerning these children before their withdrawal or death was included wherever possible. Thus, almost every study described in this thesis includes a subset of the full cohort. Whenever possible, statistical analyses

were conducted to compare those who were and were not included in a specific study to provide an indication of the generalisability of the results to the full cohort.

Because many measures were used in more than one study, an overview of the recruitment, primary measures used, and sequencing of those measures is provided here. Chapters describing specific studies will include only additional information that is germane to that specific study.

Protocol for the Longitudinal Research Program

Ethical Approval

The Research Ethics Boards of the IWK Health Centre and of Dalhousie University approved the protocol for the two-year program. Written support for all studies involving health centre staff was obtained from Program Managers and Clinical Team Leaders of the IWK Health Centre whose staff might be involved in the protocol.

Support Personnel

This program of research was conducted with the assistance of Paediatric Pain Research Lab staff, undergraduate and medical students, and volunteers. In most cases, these research assistants assisted with specific portions of the program. In addition to the author of this thesis, however, one full-time Study Co-ordinator, Alyson Currie, assisted with all aspects of data collection throughout the program. Ms. Currie also supervised the students and volunteers who assisted with data collection and data entry at different points during the protocol.

Recruitment

Caregivers were recruited through physicians of the Division of Child Neurology at the IWK Heath Centre, Halifax, Nova Scotia. The IWK Health Centre is a tertiary paediatric care centre for the provinces of Nova Scotia, New Brunswick and Prince Edward Island. This centre's physicians follow virtually all children with cognitive impairments residing in these provinces. All paediatric neurologists in this region are located at this facility and provide an extensive travelling clinic system throughout Nova Scotia. In January 1999, the Medical Records Department, Division of Child Neurology, and the Occupational Therapy Department identified patients who had mental disabilities or were seen for adaptive seating equipment during 1996-1999. A physician in the Division of Neurology then reviewed the medical charts of the children they cared for, and those with moderate to profound mental retardation were selected for recruitment.

Caregivers were sent a description of the study by mail. The letter advised them that a researcher would be telephoning them to provide further information regarding the research program and to invite them to participate (Appendix A). If verbal consent was obtained over the telephone, two written consent forms were mailed to the caregiver. A second telephone call was made to them shortly after they received the consent forms to determine if they had further questions regarding the research program. If they were willing to participate at that time, they were asked to return one copy of the consent form and to retain the second for their records. Several caregivers were provided with the consent form and information regarding the research program in person, because they

informed the researcher during the first telephone contact that they would be in the IWK Health Centre in the immediate future.

A total of 185 children were selected as eligible for recruitment. Ten children's caregivers could not be reached by mail or telephone. Of the remaining 175 children, 7 (4%) had died since they had last been at the IWK Health Centre. An additional 22 (13%) children did not meet inclusion criteria because: (a) they were younger than three years or older than 18 years (6 children, 27%), (b) they could produce more than two-word utterances (13 children, 59%), (c) they were about to move outside the Maritime provinces (2 children, 9%), or (d) their caregiver could not speak English sufficiently well to complete written materials (1 child, 5%).

Of the remaining eligible 146 children, 104 caregivers (73%) agreed to take part in the program, while 42 (27%) did not. Of those who declined, 20 (48%) stated they had no time, 5 (12%) indicated they were not interested and 2 (5%) stated their child had no pain. The remaining 15 (36%) did not indicate a reason as to why they did not wish to participate. Information concerning each child, that could have determined eligibility, was not obtained during these telephone calls. Thus, it is possible that some children would not have met the criteria for entry.

After assessments were completed at entry to the program, three of the 104 children were deemed to have language abilities that exceeded the level required for participation. In all three cases, the children had no verbal speech, but could communicate close to their chronological age using assistive technology.

Children Withdrawn from the Research Program

Over the course of their first year of participation, 10 children (10%) were withdrawn from the program. Five children died and seven families withdrew from the study, due to the child becoming severely ill, a parent becoming severely ill, or a family move. Because these withdrawals occurred at different points during the study, partial data are available for some children and is included whenever possible. Caregivers were asked if they would like their child's completed measures withdrawn from the study at the time they withdrew. None wished to have the data already collected withdrawn.

Assessment at Entry to the Program

Measures

The Demographic Information and Medical History Sheet. The Demographic Information and Medical History Sheet (Appendix B) was created for this study. Caregivers provided information regarding their child's medical history and demographic information concerning the child and the caregiver. The IWK Research Ethics Board required that caregivers be told that providing information regarding their age, marital status, education, occupation and family income was optional.

The Non-communicating Children's Pain Checklist-Revised. The Non-communicating Children's Pain Checklist-Revised (NCCPC-R) is based on the Non-communicating Children's Pain Checklist created by McGrath's group (Breau et al., 2000). It consists of 30 items in 7 classes (Vocal, Eating/Sleeping, Social/Personality, Facial Expression, Activity, Body & Limbs, Physiological). An adult completes the checklist. The original version of the checklist asked caregivers to indicate whether each

item was present or absent during pain. It has displayed good psychometric properties when used by caregivers for the pain of children with severe cognitive impairments. Scores during pain correlated with numerical ratings of pain intensity, did not differ between two pain events, and differed significantly from scores when children were without pain and calm (Breau et al., 2000). Caregivers' reports using the original checklist can also predict pain behaviour during future episodes of pain (Breau et al., 2001a).

For the studies described here, two revised checklists were used, one for observations completed by caregivers at home and one for retrospective report of their child's usual pain behaviour. For the observations used at home, caregivers indicated how often their child displayed each item on the checklist when they had pain (Not at all, Just a little, Fairly often, Very often) as part of Pain Diaries. For retrospective report using the checklist at entry to the research program, they indicated how often their child usually showed each item on the checklist during pain in the past (Appendix B).

The Vineland Adaptive Behavior Scales. The Vineland Adaptive Behavior Scales (VABS; (Sparrow, Balla, & Cicchetti, 1984) is a standardised measure of functional adaptation (Appendix B). It assesses the degree to which a child is independent and socially responsible relative to his or her age (Kronenberger, 1996). It is administered as a structured interview to an adult who knows the child well. Administration takes approximately 60 minutes.

Standard scores (M = 100, SD = 15) and age equivalents are available from the VABS (Sparrow et al., 1984), and children receive a score in four domains: Communication, Daily Living Skills, Socialization, and Motor Skills.

Children diagnosed with mental retardation tend to score below 70 on all domains of this scale. The VABS shows excellent test-retest reliability when used with non-impaired children, with correlations for composite scores ranging from .83 to .93 depending upon the age of the children (Sparrow et al., 1984). A developmental progression in scores has also been documented for non-impaired children (Sparrow et al., 1984). In this program, children's age equivalents on the four domains were used to reflect their adaptive abilities in communicating to others, performing daily tasks such as self-care, socialising, and performing tasks requiring fine and gross motor skills.

The four Domains of the VABS include 36 to 92 items each. Within each domain, subdomains provide an indication of children's abilities in more specific areas of functioning. The Communication Domain includes three subdomains: Receptive, Expressive and Written Communication. The Daily Living Skills Domain also has three subdomains: Personal, Domestic and Community. The Socialization Domain includes Interpersonal Relationships, Play and Leisure and Coping Skills subdomains. The final Domain, Motor Skills, has two subdomains: Gross Motor and Fine Motor Skills.

Procedure

After written consent was obtained, a researcher contacted the caregiver by telephone to arrange a convenient time to conduct an interview with her by telephone. At the arranged time, the researcher administered the following measures, all of which are

contained in Appendix B: (a) the Demographic and Medical Information Sheet, (b) a retrospective version of a revised version of the Non-communicating Children's Pain Checklist-Revised (Breau et al., 2000) (c) the Vineland Adaptive Behavior Scales (Sparrow et al., 1984). All measures were completed in that order. The Vineland Adaptive Behavior Scales were administered last in all cases, as caregivers find this measure reassuring because of its focus on children's abilities, as opposed to disabilities.

After completion of the interview, caregivers were reminded of the protocol for the next year. They were also provided with information regarding the Postoperative Pain Studies and were told that participation in these additional studies was optional. They were asked to notify the researcher if their child was scheduled for surgery and they had an interest in taking part in the studies.

Completion of Measures over the First Year

Measures

The Pain Survey. The Pain survey consisted of a semi-structured interview administered to caregivers by telephone (Appendix C). It contains questions regarding the pain their child experienced in the previous week. To facilitate caregivers' recall, the interviewer systematically questioned him regarding pain episodes that had occurred, day by day, beginning six days previous to the call and moving forward to the day of the call. The caregiver was also asked to describe what he believed was the cause of each pain episode. The interviewer then asked him to rate the intensity of each pain episode from 0 (none at all) to 10 (worst pain ever) and to estimate how long each pain episode had lasted. He was also asked if they took any action to relieve his child's pain, to describe

any action he took, and to rate the effectiveness of that action from 0 (didn't help at all) to 10 (completely relieved the pain). If he took a second action to relieve the pain, he was asked again to describe it and rate its effectiveness on the 0 to 10 scale.

Pain Diaries. These diaries were personalised for each child (Appendix C). For each diary, caregivers conducted a daily two-hour observation of their child at the same time each day. For each observation, caregivers indicated whether their child had experienced pain and whether the pain was due to injury, chronic condition, illness, or medical procedure. They also indicated the duration of the pain (hours and minutes) and the intensity of the pain on a numerical rating scale anchored by 0 (none at all) and 10 (worst ever).

The 30 items of the Non-communicating Children's Pain Checklist-Revised were included for each day of the diary. Caregivers rated how often each item had been observed over the two-hour observation (Not at all, Just a Little, Fairly Often, Very Often). One exception was the Eating/Sleeping subscale. Since it was considered unlikely that these behaviours would be observable over a two-hour period, caregivers were asked to indicate how often they were observed over the day.

In order to record the adaptive functioning of children for each day a Pain Diary was completed, the five most advanced items previously performed by each child from each Domain of the VABS (Sparrow et al., 1984) was included (Communication, Daily Living Skills, Socialization, Motor Skills). This was based on the caregiver's report during administration of the VABS at entry to the program. Caregivers indicated whether their child displayed these abilities during the two-hour observation period. In some

cases, children had fewer than five abilities within a domain. In these cases, only those abilities they were capable of displaying were included. Thus, the total number of potential adaptive behaviours caregivers were asked to record for the two-hour observations ranged from 2 to 20 ($M = 16.9$, $SD = 4.1$).

Examples of behaviours most children were capable of included: (a) Communication: smiles in response to the presence of caregiver, smiles at a familiar person other than the caregiver, raises arms when caregiver says "up" or "come here"; (b) Daily Living Skills: opens mouth when food is presented, removes food from a spoon with mouth; (c) Socialization: responds to the voice of caregiver, shows affection to familiar people, plays interactive games such as "peek-a-boo"; (d) Motor Skills: holds up head for 15 seconds, sits with support for at least a minute, picks up objects.

Procedure

Caregivers completed four Pain Surveys during their first year of participation in the two-year study. Approximately one month after entry ($M = 4.8$ weeks, $SD = 3.3$), they were contacted by telephone to complete the first Pain Survey. Subsequent Pain Surveys were administered at three-month intervals.

Approximately 3 months after entry to the study ($M = 11$ weeks, $SD = 7$), caregivers were mailed a one-week diary which they completed and returned by mail. Subsequent Pain Diaries were mailed at three-month intervals. The timing of these was arranged such that a caregiver did not complete both a Pain Survey and a Pain Diary in the same month. Thus, data were collected for a one-week period during eight separate months of the first year of participation.

Assessment after the First Year

One year after they entered the program, a second telephone interview was conducted with caregivers. At that time, they completed all measures administered during the interview conducted at entry to the program, except the Demographic Information and Medical History Sheet. Caregivers' responses to the VABS (Sparrow et al., 1984) at this second administration were used to update the adaptive ability section of children's Pain Diaries for the second year of the program.

Completion of Measures over the Second Year

Caregivers completed Pain Diaries and Pain Surveys over the second year of the program in the same manner as described for the first year. At completion of the second year, the telephone interview was completed a third time. All measures were re-administered.

Protocol for the Postoperative Pain Studies

Two postoperative pain studies were conducted as part of this research program. Caregivers were informed of these studies when they entered the research program. They were also informed that their participation in these additional studies was optional and dependent upon them informing the research team that their child was scheduled to have surgery at the IWK Health Centre.

Measures

The Surgery Information Sheet

The researcher completed this sheet after the child left hospital. It documented information concerning the surgery, such as the procedures performed, length of time in surgery and in the recovery room, and analgesics given (Appendix D).

The Hospital Information Sheet

The researcher completed this sheet after the child left hospital. It documented basic information about the child's stay in hospital, such as the length of time the child was in hospital and the medications given to that child (Appendix D).

The Non-communicating Children's Pain Checklist-Postoperative Version

A researcher and a caregiver completed this version of the Non-communicating Children's Pain Checklist-Revised two times, once before the child had surgery and once after the child left the recovery room (Appendix D). This is identical to the Non-communicating Children's Pain Checklist-Revised, except that the Eating/Sleeping subscale is omitted.

Visual Analogue Scale of Pain

A visual analogue scale (VAS -Pain) was used to rate the intensity of children's pain during observations before and after surgery and to rate the pain of children from videotape collected while they were in the recovery room (Appendix D). Visual analogue scales are the most common type of instrument used to measure children's pain (McGrath et al., 1987). For this study, a 100-millimetre line was anchored at the left by the phrase "no pain at all" and at the right by "worst pain ever". This measure has been used to

visually discriminate between pain and absence of pain in non-verbal children with severe cognitive impairments (Mette et al., 1988).

Visual Analogue Scale of Sedation

A visual analogue scale of sedation (VAS-Sedation) was used to rate children's level of sedation from videotape collected while they were in the recovery room. This consisted of a 100 millimetre line anchored on the left by "no sedation at all" and on the right by "worst sedation ever".

The Child Facial Coding System

This coding system was used to code children's facial movements from the videotape collected during the time the child was in the recovery room. Further details about this measure are provided in Chapter Nine.

Procedure

When a caregiver contacted the Pediatric Pain Research Laboratory to notify a researcher that their child was scheduled for surgery, two copies of an informed consent form were mailed to her (Appendix D). Several days later, a researcher telephoned the caregiver and reviewed the protocol with her to ensure that she understood what was being asked of her. She was then asked to return one signed consent form if she agreed to participate. Verbal agreement for that child to participate was then obtained from the surgeon scheduled to perform the procedures.

On the day of surgery, a researcher met the child and caregiver in the surgery waiting room. At that time, the researcher collected the informed consent form and reviewed the protocol with the caregiver once more. The researcher, a caregiver and a

nurse then completed measures before the child was moved to the operating room. After notification from the recovery room staff that the child was in the recovery room, the researcher went to the recovery room and filmed the child for up to one hour. After the child left the recovery room, the researcher, a caregiver and a nurse completed the same measures a second time.

Statistical Analyses

Data for all studies described were analysed using SPSS® for Windows™ Version 10.0.7 (Anonymous. 2000) and power analyses were conducted with SamplePower® 1.2 (Anonymous. 1997) or were based on tables by Stevens (Stevens, 1996). All power computations were conducted a priori and were based on means available. When power computations were not available for multivariate procedures, power was computed for main effects. Small, medium and large effect sizes were based on guidelines by Cohen (Cohen, 1988; Cohen, 1992). In the case of comparisons of means these indicate a difference in means of .2 standard deviations comprises a small size effect, a difference of .5 standard deviations comprises a medium effect and a difference of .8 standard deviations comprises a large size effect. For tests of proportions, the size of effect is based on the proportion of two independent samples that are positive on the factor in question. A small size effect reflected a difference of 40% versus 50% positive in the two subgroups, a medium size effect reflected a difference of 40% positive versus 65% and a large size effect reflected a difference of 40% versus 78% of the two samples being positive for the factor in question or greater. For correlations, $\underline{r} = .1$ reflected a small

effect, $\underline{r} = .3$ a medium effect and $\underline{r} = .5$ a large effect compared to zero. Power computations were not available for some nonparametric procedures.

Wilks' lambda was used to assess significance for all multivariate analyses of variance (MANOVA's) and multivariate analyses of covariance (MANCOVA's). For repeated measures and mixed measures analyses of variance (ANOVA's), univariate tests were used except when Mauchley's tests indicated assumptions of sphericity were violated. In those cases, Wilks' lambda was used to assess significance.

Alpha was set at .05 for all analyses. Bonferroni corrections were applied to all sets of analyses to maintain alpha for the set at .05. These are computed by dividing alpha of .05 by the number of tests conducted within the set. This is a conservative method of adjusting for multiple tests (Norman & Streiner, 1994). However, it was felt reducing the risk of false positive results was important because there was no specific literature regarding pain in children with cognitive impairments with which to compare the results of the current studies to gauge their validity. When tests were not significant before or after corrections, they are designated "nonsignificant". Some tests were significant before corrections, but did not maintain that significance after corrections were applied. Because Bonferroni corrections are a conservative approach to controlling for multiple tests and because some have questioned the practice of arbitrary cut-offs for determining the presence of an effect (Cohen, 1990), this is stated in the text so that the reader will be aware that those particular results may have approached significance or have achieved significance had another approach to multiple comparison corrections been used. For this reason, raw $\underline{p}$ values are given for all tests.

Summary of the Studies Described in this Thesis

The studies described here contain information collected from various aspects of the research program. A brief description of the purpose and content of each study is given below. The first two studies address the occurrence of pain in this group of children and the impact of that pain. This is followed by three studies examining methods of pain measurement for this group.

The Incidence and Characteristics of the Pain of Children with Severe Cognitive Impairments

This study documents the amount and type of pain experienced by the cohort over four one-week periods to derive an estimate of the incidence of pain in this population. Data for this study were collected from the Pain Surveys administered by telephone. Information regarding children's characteristics was collected during interviews conducted at entry to the program and from medical records.

The Functional Impact of Pain on the Development of Adaptive Abilities by Children with Severe Cognitive Impairments over One Year

This study examines the change in adaptive functioning displayed by the children from the time they entered the study until one year later as a function of the amount of pain they experienced over a sample of time from that year. This study contains data collected as part of the first and second interviews completed by caregivers. The focus of this study is caregivers' reports of their children's adaptive functioning using the VABS (Sparrow et al., 1984). The pain reported by caregivers over the first year, in four Pain Surveys conducted by telephone, is used as a measure of the children's pain. Information

regarding children's characteristics was collected during interviews and from medical records.

Validation of the Non-communicating Children's Pain Checklist-Revised

This study investigates the psychometric properties of the NCCPC-R for everyday pain. Data for this study were collected from Pain Diaries. Four observations from the diaries were used, two with pain and two without. The data examined here include the first and second observation recorded by caregivers during which their child experienced pain. Information regarding children's characteristics was collected during interviews and from medical records.

Validation of the Non-communicating Children's Pain Checklist-Postoperative Version

This study investigates the psychometric properties of the NCCPC-PV for postoperative pain. The data collected from observations conducted before and after children had surgery is contained in this study. Information collected from surgical records is also included, as is information regarding children's characteristics collected during interviews and from medical records.

Using the Child Facial Coding System to Detect the Postoperative Pain of Children with Severe Cognitive Impairments

This study investigates whether the Child Facial Coding System (Chambers et al., 1996) can detect the pain these children experience after surgery. This study is based upon the coding made by researchers of the videotape collected while children were in the recovery room after surgery. Information collected from surgical records is also

included. Information regarding children's characteristics was collected during interviews and from medical records.

Summary

As this thesis was compiled, the program of research was underway and measures were being completed by caregivers on a daily basis. Although an attempt was made to postpone analyses until all caregivers had completed each measure, the fact that caregivers entered the program over a 15-month period meant that not all caregivers had completed the measures presented in this document when analyses were conducted.

In summary, the following chapters present data that, although substantive, reflects only part of that which will have been collected when this program is complete in May 2002. A strength of this program was that all studies contain information collected from one cohort of children. This meant measures did not have to be re-administered to new participants, increasing the efficient use of the data collected and reducing the burden on caregivers.

CHAPTER FOUR: THE COHORT

These studies were completed with the assistance of a cohort of 101 children and their caregivers, who entered the research program between March 1, 1999 and June 8, 2000. These children resided in Nova Scotia (n = 87), New Brunswick (n = 12), Prince Edward Island (n = 1), and Newfoundland and Labrador (n = 1). Thirty-two children lived in large urban areas or regional centres (population > 100,000), 4 in large towns (population >25,000) and 10 in small towns (population 10,000-25,000). The remaining 55 children lived in rural areas.

Demographic Characteristics of the Children

The children were aged 3 to 18.7 years at the time they entered the study (M = 10.3, SD = 4.4, Mdn = 9.2). Fifty-five of the children were boys and 46 were girls, a 1.2:1 ratio, similar to the range of 1.3:1 to 1.9:1 typically reported in the literature for children with any degree of mental retardation {Aicardi 1998 36 /id}. Eighty-nine children (88.1%) resided with their family, while the remaining 12 (11.9%) resided in group homes or residential centres.

Demographic Characteristics of the Caregivers

Seventy-seven caregivers (76.2%) were the mother of the child of interest, while 9 (9.0%) were their father (including one grandfather who was the primary caregiver of a child living in his home). Four (4.0%) caregivers were a child's foster mother. An additional 11 caregivers (10.9%) were the primary caregiver of a child who was in a

group home or residential centre and they had been that child's primary caregiver for at least 6 months prior to entry into the program.

Demographic information regarding the biological parents of children who resided in group homes or residential centres is not reported, as caregivers in these facilities could not always provide accurate information or details. Seventy-four of the caregivers (83.1%) of the 89 children who lived at home were married at the time they entered the studies. Seven caregivers (7.8%) were separated, divorced or widowed. The final eight caregivers (9.0%) were single parents. The mothers of the children who lived at home ranged in age from 20 to 66 years ($M = 38.6$, $SD = 7.5$). Fathers' ages were available for 77 children. These ranged from 28 to 70 years ($M = 40.8$, $SD = 8.0$).

The education level of the mothers and fathers and their employment status is displayed in Table 4.1. The number of children in the families ranged from one to five, as shown in Table 4.2. Of these 89 families, seven (7.9%) reported a family income of less than $10,000 Canadian, 59 (66.3%) reported an income of $10,000 to $50,000, and 18 (20.2%) reported a family income greater than $50,000. Five (5.6%) did not provide this information.

Medical Characteristics of the Children

Level of Diagnosed Mental Retardation

The children's level of diagnosed mental retardation was one characteristic documented in these studies, based on previous research suggesting that cognitive deficits may influence the amount and type of pain behaviour children typically show (Fanurik et al., 1999a; Breau et al., 2000). According to the Diagnostic and Statistical Manual of

Mental Disorders (DSM-IV) of the American Psychiatric Association, a diagnosis of mental retardation is only appropriate when an individual displays general intellectual functioning, as measured by a standardised instrument, that is significantly below average (American Psychiatric Association, 1994). The below average IQ must be accompanied by impairments in adaptive functioning and must also have had an onset before age 18 years.

Most children in this cohort were not capable of completing individually administered tests of their intellectual or adaptive abilities due to the severity of their cognitive impairments or physical impairments, such as blindness. Thus, children's medical charts were used to determine their level of mental retardation, as diagnosed by a physician with the Division of Neurology at the IWK Health Centre. Nine children (8.9%) had a diagnosis of moderate mental retardation, 66 severe (65.3%) and 18 profound (17.8%). No information regarding a diagnosis of mental retardation was given in the charts of 8 children (7.9%).

Timing of the Onset of Neurological Damage

The timing of the onset of the children's neurological damage was determined from their medical records. Neurological damage was deemed to be of prenatal origin if there was evidence the damage occurred before labour began. Neurological damage that occurred between the onset of labour and the 28th day of life was classed as perinatal (Stromme & Hagberg, 2000). Neurological damage occurring after the 28th day of life was classed as postnatal.

Seventy-one children (70%) had cognitive impairments of prenatal origin. This is identical to the proportion reported by Stromme et al. (Stromme et al., 2000) in their epidemiological study of 178 children with mild to severe mental retardation from a cohort of 30,037 children born in the Oslo area of Norway between 1980 and 1985. It is also within the 60% to 75% generally reported in the literature for all levels of mental retardation (Aicardi, 1998).

In contrast, a greater number of children in the current cohort suffered neurological damage of perinatal (13%) and postnatal (14%) onset than reported by Stromme et al., who found only 4% of their cohort had severe mental retardation of perinatal origin and 5% of postnatal origin (Stromme et al., 2000). Similarly, Matilainen et al. report 9% perinatal onset and 8% postnatal onset in their study of 77 children with severe mental retardation from a cohort of 12,882 children born between 1969 and 1972 in one province in Finland (Matilainen, Airaksinen, Mononen, Launiala, & Kaariainen, 1995). The greater proportion of children having impairments of perinatal and postnatal onset in the current cohort could reflect improvements in medical diagnostics and record keeping. In 18% of the cases described in the Norwegian study (Stromme et al., 2000) and 23% of the cases described in the Finnish study (Matilainen et al., 1995), the cause of mental retardation could not be determined based on the records available. In contrast, records were not available to determine timing of the neurological damage in only 3% of the cases in the current cohort. Thus, it is possible that many of the cases for which the previous studies could not determine the timing of neurological damage would have been

diagnosed as having an onset of perinatal or postnatal origin had more sophisticated diagnostic tools been used or had more information been available in the records.

Finally, both Stromme's group and Matilainen's group conducted studies of the incidence of mental retardation, while the current study provides an indication of the prevalence of mental retardation. Thus, children were accounted for in their research who died before the studies were conducted. Because the current studies were prospective, children in the region who died before the program began are not accounted for in the statistics.

Aetiology of Neurological Impairment

Most research distinguishes only between mental retardation of organic aetiology and that due to the environment. However, there are more than 200 recognised organic aetiologies of mental retardation and the cognitive abilities of children with different aetiologies may differ significantly (Burack, Hodapp, & Zigler, 1988). Thus, an attempt was made to ascertain the primary neurological diagnosis of the children in this cohort. In most cases, children had more than one neurological diagnosis. For example, some children with prenatal neurological anomalies experienced perinatal complications that also may have contributed to their cognitive impairments. Many children who were extremely premature or who had dysmorphic or chromosomal syndromes also had neonatal seizures. In cases in which more than one neurological diagnosis was recorded, the earliest was considered primary.

Following the classifications used by Aicardi (Aicardi, 1998) and Stromme and his colleagues (Stromme et al., 2000), the children were grouped into the following categories based on their primary neurological diagnosis: dysmorphic syndrome, chromosomal syndrome, neurodegenerative syndrome, complications of extreme prematurity, epileptic syndrome, intrauterine acquired condition, complications of hypoxia/ischemia, complications of infection, or traumatic brain injury.

Thirty children (29.7%) had a primary neurological diagnosis of a dysmorphic syndrome. This diagnosis indicates a child has one or more physical abnormalities in one or more external body sites. This may consist of one major anomaly or many minor anomalies, and there are frequently anomalies in the craniofacial area (Hall, 1994). Dysmorphic features may be congenital, indicating they are due to prenatal factors, or acquired, indicating they are due to perinatal or postnatal factors.

Some children in this group had identifiable chromosomal syndromes that were associated with dysmorphic features. For example, children in this group were diagnosed with Sanfilippo Syndrome, Smith Lemli-Opitz Syndrome, Linear Nevus Sebaceous Syndrome, Langer-Gideon Syndrome, Aicardi Syndrome, Proteus Syndrome, and Spina Bifida. However, in some cases, no specific syndrome had been identified, although there were obvious dysmorphic features. This is not unusual. In about 20% of cases of severe mental retardation, multiple anomalies are present in the absence of evidence of specific chromosomal abnormalities (Aicardi, 1998).

A second group of seven children (6.9%) had an identified chromosomal syndrome that was not necessarily accompanied by dysmorphic features. This included

tuberous sclerosis, which involves hamartomas, which are localised areas of faulty cellular development due to excessive calcium, in the cerebral cortex (Meyer, 1997), lissencephaly, which is a group of disorders that involve incomplete development of the brain, Angelman's Syndrome, frequently accompanied by mild cortical atrophy or ventricular enlargement, and Fucosidosis, a metabolic lysosomal storage disease in which mucopolysaccharide metabolism is altered.

Six children (5.9%) were diagnosed with a neurodegenerative disorder. Metachromatic Leukodystrophy is one example of this. It involves an accumulation of cerebroside sulfate in the nervous system, which leads to neuronal damage and myelin breakdown (Menkes, 2000). Like most neurodegenerative disorders, it occurs rarely.

Eleven children (10.9%) suffered neurological damage due to "prematurity" (born at or before 36 weeks gestation) or "extreme prematurity" (born before 28 weeks gestation). In many cases, these infants also met criteria for being classed as "Low Birth Weight" infants (less than 2,500 grams at birth), "Very Low Birth Weight" infants (less than 1,500 grams at birth), or "Extremely Low birth weight" infants (less than 1,000 grams at birth). In this cohort, the lowest birth weight was 630 grams.

Prematurity and low birth weight are associated with Respiratory Distress Syndrome. In one study, 60% of infants born at or less than 32 weeks gestation experienced this difficulty (Silverman, Watanabe, Marshall, & Baer, 1984). Typically occurring in the first two days of life, it and can lead to hypoxia (low oxygen content in body tissues), ischemia (low blood flow to body tissues) and acidosis (blood pH less than 7.3). Because the newborn's brain is still developing, children born at different gestational

ages will be susceptible to different types of brain injury when these problems occur (Amiel-Tison, 1996). Prematurity also makes these infants more susceptible to haemorrhage due to mechanical problems with labour, especially those involving the arachnoid and subdural spaces (Amiel-Tison, 1996).

In some cases, the site and extent of brain injury suffered by premature infants is easily detected. However, there may be neurological damage that is not immediately detectable, but which has severe long-term consequences in terms of the child's development. Intraventricular haemorrhage is the most common type of perinatal intracranial haemorrhage in infants who are premature, with an incidence of 29% to 49% in infants less than 2000 grams birth weight (Scheller & Koch, 1994). In 90% of cases this occurs within the first 3 days of life (Scheller et al., 1994) and hydrocephalus is a common result (Scheller et al., 1994). Hydrocephalus is the abnormal accumulation of cerebrospinal fluid in the ventricular systems of the brain (McCarthy & Land, 1992). This can be caused by obstructions to the normal circulation of the CSF, known as "obstructive hydrocephalus". Hydrocephalus may also be caused by excessive production of CSF or failure of the CSF to be absorbed, in which case it is called "communicating hydrocephalus". Acute hydrocephalus usually occurs within a few days of birth, subsequent to an intraventricular haemorrhage, when the circulation and absorption of cerebrospinal fluid is prevented by particulate blood from the haemorrhage (Miller & Clark, 1998).

Three children (2.9%) had an epileptic syndrome as their primary diagnosis. Two had infantile spasms, also known as West's Syndrome when a hypsarrhythmic

electroencephalogram (EEG) accompanies them and mental retardation (Vining & Gladish, 1992). Infantile spasms occur almost exclusively during the first year of life and are often refractive to typical anticonvulsant drugs. They are characterised by brief muscular contractions that usually occur in clusters, from a few at a time to more than 100, occurring 5 to 30 seconds apart (Aicardi, 1994b). Approximately two thirds of children with these seizures are "symptomatic", meaning they have a pre-existing developmental, genetic or metabolic abnormality (Vining et al., 1992). The remaining third are "cryptogenic"; meaning the children had no prior neurological abnormality that could be detected when the seizures began (Vining et al., 1992). This was the case for the two children in this cohort. Severe mental retardation is also more common in children with infantile spasms than in children with other types of infant epilepsy (Aicardi, 1994a).

The third child had Severe Myoclonic Epilepsy of Infancy. This syndrome is not associated with neurological or metabolic abnormalities, but there may be a genetic component. These children have long seizures (10 to 90 minutes) that are most often triggered by fever or infection at 4 to 11 months of age (Aicardi, 1994c). Although children may continue to develop normally immediately after onset, development usually stalls in the second to third year of life and extreme hyperactivity can complicate the picture (Aicardi, 1994c).

Eight children (7.9%) had neurological impairments that were classed as Intrauterine Acquired. This means neurological damage occurred between conception and birth. In many cases in this cohort, the foetus failed to develop physically, leading to the

diagnosis of "intrauterine growth retardation". Many teratogens or agents that can cause a defect in the embryo or foetus have been identified, including illicit drugs and medically prescribed drugs (Graham & Morgan, 1997), or environmental toxins such as radiation (Graham et al., 1997). Maternal malnutrition can increase risk for neural tube defects, hydrocephalus, and prematurity (DeLong, 1993). Maternal chronic illness such as diabetes, thyroid disease and hypertension and traumatic injury to the foetus can also increase risk for intrauterine difficulties (Graham et al., 1997). Intrauterine infections may also lead to abnormal neurological development or neurological damage in the foetus. The most common are toxoplasmosis, rubella, cytomegalovirus, herpes simplex virus and syphilis (Dormans & Pellegrino, 1998). Placental anomalies are also associated with prenatal neurological damage and lead to intrauterine growth retardation. The placental problems may include hemangiomas, placental infarcts, single umbilical artery, and small placental size (Prada & Tsang, 1998).

Nine children (8.9%) in this cohort suffered a traumatic brain injury after birth that led to permanent neurological impairment. In two cases, this was due to accidental head injury, while three children experienced non-accidental head injury. The remaining four children suffered neurological damage due to tumour, stroke, cardiac difficulties leading to severe hypoxia, and severe dehydration due to encephalitis.

For 12 children (11.8%), the primary aetiology of their neurological impairment was asphyxia or hypoxia/ischemia. This may occur before birth due to cord compression, abruptio placentae (premature detachment of the placenta), foetal haemorrhage, or maternal circulatory failure (Amiel-Tison, 1996). It may also occur during or after birth

due to severe Respiratory Distress Syndrome. Asphyxia can result in increased cardiac output to the brain, as much as a 30% to 175% increase, or can lead to reduced cerebral blood flow when the onset of asphyxia is sudden and severe, because of increases in cerebrovascular resistance (Menkes & Sarnat, 2000). These events lead to tissue necrosis (death) and edema (fluid accumulation), usually within 36 to 72 hours (Lupton, Hill, Roland, Whitfield, & Flodmark, 1988).

Four children (4%) had neurological impairment due to infection. Three children developed meningitis during the neonatal period. This involves an inflammation of the meninges and is most often due to Group B Streptococcus or Escherichia Coli (Palumbo, Davidson, Peloquin, & Gigliotti, 1995). Children who have had meningitis in the neonatal period experience a high rate of neurological abnormalities (41%) and of IQ's that are below 70 (38%) (Doctor et al., 2001). Outcome has not been improved by advancements in neonatal care (Polin & Harris, 2001). A third child was diagnosed with congenital cytomegalovirus, the most common congenital infection (Palumbo et al., 1995). This infection is typically transmitted from the mother to the foetus through the placenta, or is transmitted during delivery or through breast milk after birth. However, the most severe impairments occur if the foetus is infected during the first six months of gestation (Palumbo et al., 1995).

Associated Medical Conditions

To determine the medical conditions the children experienced, caregivers were administered a questionnaire upon entry to the study (Appendix B). The majority of children in this cohort had at least one medical condition. Forty-eight children (48%) had

a diagnosis of Cerebral Palsy and 68 children (67%) had a diagnosis of a seizure disorder. Of these, 36 (53%) experienced generalised seizures and 29 (43%) focal seizures. Three children were not experiencing seizures at the time they entered the study due to successful treatment for their seizure disorder.

Only 53 children (52.5%) were capable of eating solid food on a regular basis, while 17 children (16.8%) could eat only soft or pureed food. An additional 31 children (30.7%) were completely fed through a gastrostomy or jejunostomy tube. This was due to either an inability to chew and swallow food because of major motor impairment, or to recurrent gastrointestinal disorders such as gastrointestinal reflux.

Caregivers indicated only 24 children (24%) did not require regular medical monitoring for a medical or physical condition. In contrast, 44 (44%) required monitoring by a medical professional monthly, 11 (11%) required weekly monitoring, and 22 (22%) required daily monitoring. Seventy-nine children (79%) were taking medications at the time they entered the program. This averaged 3.5 medications regularly (SD = 2.6). Twenty-one children were taking one medication, 16 children were taking two, and 8 children were taking three. Thirty-four children took four to eleven medications on a regular basis. The medications being taken by more than one child are listed in Table 4.3. The most common medications were anticonvulsants. Fifty-eight children (57.4%) were taking at least one of these, 22 were taking two, and 6 children were taking three or more. This was followed by benzodiazepines, being taken by 20 children, with three children taking two medications in this category. Fifteen children were using antacid medications and eleven were using laxatives on a regular basis. Only one child was taking more than

one laxative. Thirteen children took bronchodilators or inhaled steroids, with 3 children using two of these medications regularly and one child using three. Finally, seven children were using analgesics regularly, with only one child using more than one analgesic.

Associated Physical Impairments

The children in this cohort had many physical impairments that accompanied their neurological impairment and associated medical conditions. Information about these was collected from caregivers at entry to the study (Appendix B). Eighteen children (17.8%) had spastic quadriplegia, meaning they had use of neither upper nor lower limbs. Of the remaining 83 children (Table 4.4), only 44 (53%) had full use of their arms and only 21 had full use of their legs (25.3%). Hearing impairment was also common. Nine children (8.9%) had no hearing and 7 (6.9) had only partial hearing. Even more children had a visual impairment. Twenty-six children (25.7%) were blind and 18 (17.8%) were partially blind.

Adaptive Functioning of the Children

In order to determine the ability of the children in the cohort to function on a daily basis, the Vineland Adaptive Behavior Scales (Sparrow et al., 1984) were administered to their caregivers at entry to the research program. Caregiver's responses were used to generate age equivalents for the children in four Domains of functioning: Communication, Socialization, Daily Living Skills and Motor Skills. The results are listed in Table 4.5.

As that table shows, although the children were aged 3 to 18 years, they functioned at much younger levels. Overall, the children were functioning at an average of 16 months of age, an average of nine years below their chronological age (SD = 4.4 years). However, their abilities differed depending on the area of functioning in question. Those in the areas of Communication, Socialization and Daily Living Skills were relatively similar, at approximately 13 months of age, but these differed markedly from their age equivalents for Motor Skills, which were, on average, at approximately 8 months of age. Table 4.6 lists some behaviours and tasks that most children in the normative sample of the Vineland Adaptive Behavior Scales had mastered by one year of age (Sparrow et al., 1984). It also lists the percentage of children in this current cohort who were capable of these particular behaviours or tasks.

Summary

As the data presented in this chapter indicate, the children in this cohort had severe cognitive impairments and, in a majority of cases, multiple disabilities. Many of the children also had a chronic medical condition that required ongoing medication and regular medical monitoring by a healthcare professional. In terms of daily functioning, the children were performing many years below levels expected for their chronological age. In many cases, the children were completely dependent on others for daily care.

CHAPTER FIVE: THE INCIDENCE AND CHARACTERISTICS OF THE PAIN OF CHILDREN WITH SEVERE COGNITIVE IMPAIRMENTS

Almost all children have pain. This can range from an occasional bump or bruise to the prick of an immunisation needle, to recurrent headaches or abdominal pain. For most children, pain is a frequent event, signalling a scraped knee, a stubbed toe, or a bruised elbow. Other times, a sore throat or a throbbing ear tells them it is time to get help from a grown-up.

There is very little research examining how often children experience pain in general. Most focuses on specific recurrent conditions, such as headache (Kristjansdottir & Wahlberg, 1993), migraine (Abu-Arafeh & Russell, 1994), back pain (Taimela, Kujala, Salminen, & Viljanen, 1997), recurrent limb pain (Abu-Arafeh & Russell, 1996) or recurrent abdominal pain (Bode, Rothenbacher, Brenner, & Adler, 1998).

In a review of the literature, Goodman and McGrath (1991) point out that most epidemiological studies of children's pain have been retrospective, have included only the occurrence or non-occurrence of pain, and have asked only about one pain type (Goodman & McGrath, 1991). Since that review, several studies have looked at combinations of pains such as headache, stomach pain and back pain (Kristjansdottir, 1997) or abdominal migraine and migraine headache (Abu-Arafeh & Russell, 1995).

Only one study has investigated the occurrence of pain in general (Perquin et al., 2000). It examined information collected from 1001 parents of pre-school children and 4459 children aged 4 to 18 in the Netherlands. This included information concerning any

pain experienced in the previous three months and the location of that pain. Information regarding the intensity, duration and frequency of the most "troublesome" pain was also collected. The authors report that 53.7% of the children had experienced pain in the previous three months, with 25% reporting pain that had been recurrent or continuous for more than three months and 24.2% reporting pain that had not. Approximately half of the children who had experienced pain had experienced it in more than one location, with the combination of headache and abdominal pain being most common. The percentage of children reporting pain also differed by body location, gender and age. The most common locations were head (0.2% to 4.6%), abdomen (1.6% to 4.6%), and limb (1.1% to 6.1%). Girls experienced abdominal pain and multiple pains more than boys. In contrast, boys experienced more limb pain and pain at other locations. There was also an increase in the percentage of children experiencing severe chronic pain with age, defined by the authors as pain that occurred weekly for more than three months and was rated greater than 50 on a 100 millimetre visual analogue scale of pain intensity. However, the authors do not report the cause of the pain experienced by the children, or whether the pain experienced at each site was due to a chronic or recurrent condition, temporary condition, illness or injury. The study was also based on retrospective report, and research suggests that the retrospective report of children for pain over a two-week period alone may differ substantially from that reported by their caregivers (Peterson, Moreno, & Harbeck-Weber, 1993). Although this does not mean that the children are necessarily incorrect in their recollections, it does raise concerns about the accuracy of the recall of both them and their caregivers for pain experienced in the past.

Only one study could be found that examined pain due to injury in the home setting (Peterson et al., 1993). As part of a study examining recall of painful events by children and their mothers, children and mothers were interviewed every two weeks for a six-month period. During each call, they were asked questions concerning injuries in the previous two weeks. These authors report that children recalled approximately 10 injuries per interview (SD = 3.1), while mothers recalled 6.5 (SD = 2.2). They also report that consensus between child and mother recall was greatest when the injury required medical treatment or when the child had contacted the mother. Unfortunately, the study did not examine the intensity or duration of the pain associated with the injuries.

No large-scale study of the incidence of pain has included children with severe cognitive impairments. However, several studies have suggested that, although they may experience some pain that is similar to that of other children, they may also experience additional pain that is related to medical conditions. The injuries they experience may also differ from those of children without impairments.

For example, caregivers reported in one study conducted at children's homes that children had experienced injuries, such as falls, burns, insect stings and pinched fingers, but also experienced pain due to medical interventions, such as intravenous insertions, irritations caused by prostheses, and surgery (Breau et al., 2000). The children also experienced pain that was longer in duration, such as pain due to sinus, throat and urinary tract infections, and pain due to severe burns, menstrual cramps and constipation. In a second study, the medical conditions of children with neurodegenerative diseases admitted to a hospice were investigated (Hunt et al., 1995). They found that 16 of 45

children admitted in one year had pain, with muscle spasms being the most frequent source, followed by joint pain. They also report pain caused by constipation, gastroesophageal reflux, ear and throat infections and menstruation. Finally, a study of ambulatory children with cognitive impairments at a day care found that the type of injuries experienced by the 36 children differed from those experienced by the 209 children without impairments over a 12 month period (Leland et al., 1994). Of the children with cognitive impairments, 72% had at least one injury, while only 25% of children without cognitive impairments had at least one injury. Children with cognitive impairments were also less likely to have injuries while running, playing on playground equipment, or due to falls, while they were more likely to have suffered a "purposive" injury by another person.

Two recent studies have report on the incidence of pain in children with cognitive impairments. In the first, Lenton et al. report that 59% of 123 children with non-malignant life-threatening conditions in the area surrounding Bath, United Kingdom had pain that caregivers believed was not being managed effectively (Lenton, Stallard, Lewis, & Mastroyannopoulou, 2001). No further details about that pain are provided. The same group documented the occurrence of pain in a group of 34 children with cognitive impairments (Stallard et al., 2001). The participants were caregivers of 18 boys and 16 girls over age 2 who had non-malignant, life-threatening illnesses and were served by a community nursing service in Bath, United Kingdom. The caregivers completed a diary, documenting the presence, severity (mild, moderate, and severe) and duration of pain observed during 5 daily time periods. Twenty-five children (73.5%) had pain at least

once, 21 (62%) on 5 or more days, and 8 (23.5%) on 12 or more days. On average, pain occurred on 6 consecutive days. This suggests pain is much more frequent for these children than for the children without impairments described by Perquin et al. (Perquin et al., 2000). Of the 415 episodes reported, 59.9% were rated as mild, 38.6% as moderate and 11.5% as severe. In addition, the authors report that 44.1% of the episodes lasted less than 10 minutes, 38.1% 10 to 30 minutes and 17.8% more than 30 minutes. Moderate to severe pain and longer lasting pain tended to occur between 12:00 am to 8:00 am. Unfortunately, this study did not document the cause of the children's pain.

Documenting the nature of the pain experienced by children with severe cognitive impairments is important. Incidence rates can help clinicians attempting to diagnose the source of pain. Knowledge about the typical duration of pain due to different sources may also help caregivers and clinicians narrow down potential causes for a child's pain. Information about the typical intensity of painful conditions may also assist in making judgements concerning pain management. This information is also important for further research. Understanding which causes of pain are most common in this population can help researchers focus their energies where the most impact can be made. Knowing the typical duration and intensity of pain can also help in research into possible treatment strategies for pain.

This study represents an effort to collect information regarding the incidence and nature of pain in the cohort of children with severe cognitive impairments. Over the course of a year, caregivers of the children completed four one-week Pain Surveys by telephone. For each survey, they reported on each painful event their child had

experienced in the previous week, its cause, its intensity, and its duration. From these surveys, descriptive statistics were generated to provide a picture of the pain experienced by this group of children. Analyses were also conducted to explore whether the pain experienced differed due to child and family characteristics.

Method

Participants

The full cohort of 101 children participated in this study. However, three children died before their caregivers had completed all measures for this study. Three additional caregivers also withdrew from the longitudinal study before completing all measures due to life-threatening child or parent illness or family move and one caregiver completed only two of the four Pain Surveys. These seven children were excluded from these analyses. One additional caregiver completed 3 of the 4 Pain Surveys. This child's data were retained. Because their caregiver reported they had experienced no pain during the first three Pain Surveys, they were given that score for the fourth. Thus, 94 children formed the final sample for this study, 41 girls and 53 boys. They ranged from 3 to 18.7 years of age (M = 10.0, SD = 4.4). Because of the small number of children excluded, it was not possible to conduct statistical comparisons of those children from the cohort who were and were not included in these analyses.

The cause of the 94 children's cognitive impairments included: dysmorphic syndromes, 28; chromosomal syndromes, 7; extreme prematurity, 11; asphyxia at birth, 10; traumatic brain injury, 9; neurodegenerative disorders, 6; intrauterine acquired condition, 6; infection, 4; and epileptic syndrome, 2. The cause of impairment was

unknown for seven children and information was unavailable for four. Eighty-three of the children resided with their family, while the remaining 11 resided in group homes or residential centres. These children's mothers ranged from 20 to 66 years of age (M = 38.7, SD = 7.7) and their fathers ranged from 28 to 70 years (M = 40.8, SD = 8.2, n = 72). Further demographic information concerning the children and their families is displayed in Table 5.1. Information regarding the children's medical conditions and physical impairments is displayed in Table 5.2 and the children's chronological and adaptive ages are shown in Table 5.3.

Measures

Demographic Information and Medical History

This questionnaire (Appendix B) was administered to caregivers during their first telephone interview when they entered the research program. It is described in detail in Chapter Three.

The Vineland Adaptive Behavior Scales (VABS)

This semi-structured interview was administered as part of the first interview at entry to the research program (Sparrow et al., 1984). It is described in detail in Chapter Three.

The Non-communicating Children's Pain Checklist-Revised (NCCPC-R)

The Non-communicating Children's Pain Checklist-Revised (Appendix B) was administered to caregivers during the first interview at entry to the research program. It is described in detail in Chapter Three. In this study, the NCCPC-R was used

retrospectively as a measure of the amount of pain behaviour children typically display while in pain.

Pain Surveys

These were administered to caregivers by telephone (Appendix C). They are described in detail in Chapter Three. To facilitate caregivers' recall, the interviewer systematically questioned the caregiver regarding pain episodes that had occurred, day by day, beginning with six days previous to the call and moving forward to the day of the call. The caregiver was also asked to describe what they believed was the cause of each pain episode. The interviewer then asked them to rate the intensity of each pain episode from 0 (none at all) to 10 (worst pain ever) and to estimate how long each pain episode had lasted.

Procedure

Approximately one month after entry into the research program ($M = 4.8$ weeks, $SD = 3.3$), caregivers were contacted by telephone to complete the first Pain Survey. The second Pain Survey was completed approximately three months later ($M = 13.4$ weeks, $SD = 4.2$). Approximately three months elapsed between the second and third Pain Surveys ($M = 13.1$ weeks, $SD = 2.7$) and the third and fourth Pain Surveys ($M = 13.6$ weeks, $SD = 2.7$).

Statistical Analyses

Coding of the Pain Episodes

Because some children experienced pain more than once on a specific day or experienced pain that lasted more than one day, "episodes" were used as a unit of

analysis. A pain episode consisted of one continuous period in which the child had pain. Thus, an episode could last from a few moments to several days.

The descriptions and causes of the pain episodes provided by caregivers were used to classify the episodes by cause. These causes are listed in Table 5.4. "Post-accidental" pain consisted of pain due to an injury on a previous day. Pain that was described as being due to "gas" or "gastrointestinal" without elaboration of cause was coded as "digestive". Pain described as being in the child's bowels, but not specifically described as due to constipation was coded as "bowels". Pain due to viral infections or bacterial infections that were not in the throat or chest were designated as "other infection/illness".

Causes were then grouped into two types, "accidental" (includes accidental and post-accident) and pain that was "non-accidental" (pain due to all causes except: accident, needle, surgery), to investigate pain related to specific events and pain related to conditions. The majority of episodes of accidental pain were due to fall, bumps, and bruises experienced around the home. In some cases these involved altercations with siblings. Pain episodes were also grouped into the following categories: gastrointestinal (digestive, gastroesophageal reflux, bowels, constipation), musculoskeletal (muscle spasms/tightness, orthopaedic), common childhood pains (headache, menstrual pain, teething pain), infection (throat infection, chest infection, other illness/infection), medical intervention (procedural pain, post-surgical pain, feeding tube irritations), recurrent (ear pain without infection, pain due to seizures, diaper rash), other (pain due to: cast,

chemical burn resulting from intravenous line, self-injurious behaviour, tumour), and unknown (caregiver could not discern cause of pain).

Missing Data

Descriptions were provided for all 409 episodes, but cause of pain could not be determined from three descriptions provided by caregivers. These three episodes were excluded. Of the 406 episodes of pain described by caregivers, pain intensity ratings on the 0-10 scale were not provided for 22 (5%) and duration was not provided for 68 (17%).

Statistical Procedures

Descriptive statistics were generated for each Pain Survey and for the four surveys combined. Because all children were under observation for the same time period, incidence rates were the ratio of children who experienced at least one episode of pain to those who did not (Kelsey, Whittemore, Evans, & Thompson, 1996).

Comparisons of children who experienced accidental or non-accidental pain and those who did not on continuous demographic factors were made using independent t-tests, comparisons on ordinal factors using Mann Whitney U tests, and comparisons of categorical factors using Pearson's chi-square tests. Power for these tests to detect large effects ranged from .92 to .97 and to detect medium effects, power was .59 to .69.

Chi-square tests were used to examine differences in the proportion of children who experienced categories of pain by gender and age group (3 to 7 years, 8 to 12 years, 13 years or more). Power to detect a medium difference in proportions for gender was approximately .67, while power to detect a large difference in proportions was .97. Power

to detect a medium differences in proportions due to age was approximately .59, while power to detect a large difference in proportions was .87.

Comparisons of adaptive age equivalents between children who had or had not experienced accidental or non-accidental pain were made using multivariate analyses of variance (MANOVA's) on children's four age equivalents from the VABS (Sparrow et al., 1984). Power was approximately .90 to detect a medium multivariate difference. Power to detect large univariate effects was approximately .82.

Pearson correlations were used to examine the relations among children's scores on the NCCPC-R and the number of episodes of pain they experienced. This was used as an indication of whether children's typical display of pain behaviour had led to some caregivers reporting more pain than others. Pearson correlations were also used to examine the relations among children's scores on the NCCPC-R, their adaptive age equivalents on the VABS and number of types of pain they experienced over the four weeks. Power was .94 to detect medium size correlations.

Results

Descriptive Statistics of the Children's Pain

Twenty-one children (22%) did not experience any pain over the four weeks covered by the Pain Surveys. The remaining 73 children (78%) experienced at least one episode of pain over that time, with a total of 406 episodes being reported. Of the 2632 days reported on, children had pain on 470 (18%). Only two children had more than one type of pain at any given time. Their primary pain was used in all analyses.

Pain by Week of Survey

Table 5.4 lists the number of pain episodes reported and the number of children experiencing each type, category and cause of pain for each week surveyed. Each week, 35% to 52% of children had pain. Accidental pain was the most common type of pain, with 7% to 15% of children experiencing this each week, followed by gastrointestinal pain, experienced by 5% to 13% of children each week.

Table 5.5 displays the number of days on which children had pain, on average, for each week. It also includes the amount of time children were in pain each week. These are provided for the group as a whole, to provide an approximation of the incidence of pain for this population, and for only children who experienced pain at least once during each week. This suggests children in this population are experiencing pain, on average, once per week and that it lasts over 9 hours when it does occur. For those children who had experienced pain at least once over these four weeks surveyed, however, an average of 2 days per week included some pain and pain lasted over 19 hours per week on average.

Pain Incidence, Intensity and Duration Over Four Weeks

Table 5.6 includes the number of children who experienced each pain type and each category of pain over the four weeks surveyed. That table also lists the incidence of each pain type and category over the four weeks and the mean intensity and duration for each pain type and category. Thirty percent of the children experienced accidental pain at least once during the four weeks, while 62% experienced non-accidental pain. Pain of unknown cause was the most intense, although rare. This was followed by digestive pain

and pain that was due to recurrent conditions. Accidental pain was least intense. The pain that lasted longest was due to throat infections, followed by constipation and teething. Accidental pain had the shortest duration, less than one hour on average.

Relation between Child Characteristics and Pain Experienced

Pain by Gender and Age

Table 5.7 lists the number and percentage of children who experienced each pain category and cause at least once over the four-week period surveyed by gender. Although the difference for musculoskeletal pain, which was experienced at least once by 7% of girls and 28% of boys, appeared large, this was not significant after corrections for multiple comparisons.

Table 5.8 lists the number and percentage of children who experienced each pain category and cause at least once over the four-week period surveyed by age group. Although no differences were significant after corrections for multiple tests, some differences between groups appeared. For example, there was a tendency for more children aged 3 to 7 to have accidental pain than children aged 8 to 12. In contrast, there was a tendency for fewer children aged 3 to 7 to have musculoskeletal pain than children aged 8 to 12. Finally, children aged 8 to 12 tended to have more recurrent pain than children aged 13 and older.

Table 5.9 provides the number and percentage of children, by gender and age group, grouped by number of non-accidental pain types and episodes over the four weeks. Chi-square tests indicated girls and boys did not differ in the number of non-

accidental types of pain they experienced (χ^2 (2) = 4.0, p = .13). However, a significantly greater proportion of boys (34%) experienced 5 or more episodes of pain than girls (20%; χ^2 (2) = 7.2, p = .03), before corrections for multiple tests. There were no significant differences in the proportion of children aged 3 to 7 and 8 to 12 by number of types of pain (χ^2 (2) = 4.9, p = .08), or between those aged 3 to 7 and those aged 13 to 18 (χ^2 (2) = 3.2, p = .20), or those aged 8 to 12 and those aged 13 and over (χ^2 (2) = 2.0, p = .37).

The proportion of children experiencing no non-accidental pain, 1 to 4 episodes, or 5 or more episodes also did not differ between children aged 3 to 7 and children aged 8 to 12 (χ^2 (2) = 3.0, p = .23), or children aged 8 to 12 and children aged 13 to 18 (χ^2 (2) = 2.9, p = .23). But the proportion of children did differ between the 3 to 7 age group and the 13 to 18 age group (χ^2 (2) = 8.1, p = .02) before corrections for multiple tests. Although an equal percentage of children in each group experienced no non-accidental pain, more children aged 3 to 7 experienced 1 to 4 episodes, while more children aged 13 to 18 experienced 5 or more episodes of non-accidental pain.

Relation between Family Demographic Characteristics and Pain Experienced Accidental Pain

A set of analyses was conducted to explore whether having had accidental pain was associated with demographic characteristics of the children's families. Children who experienced at least one episode of accidental pain over the 4 weeks surveyed (n = 28) did not differ from those who experienced no accidental pain (n = 66) in the proportion who lived with their family (41% versus 57%; χ^2 (1) = 0.3, p = .61). For those who lived

with their families, mothers and fathers of children who did or did not experience accidental pain also did not differ in terms of education ($U = 474.0$, $p = .04$ and $U = 450.0$, $p = .64$, respectively) or age ($t(81) = 0.5$, $p = .60$ and $t(70) = 0.8$, $p = .41$, respectively) after corrections for multiple tests. Nor did their families differ in income ($U = 576.0$, $p = .45$) or in the number of children in the family ($U = 692.5$, $p = .87$).

Non-accidental Pain

Children who experienced at least one episode of non-accidental pain over the 4 weeks ($n = 58$) did not differ from those who experienced no non-accidental pain ($n = 36$) in the proportion who lived with their family (88% versus 89%; χ^2 (1) = 0.02, $p = .89$). Mothers and fathers of children who did or did not experience non- accidental pain did not differ in terms of education ($U = 718.0$, $p = .64$ and $U = 463.5$, $p = .11$, respectively) or age ($t(81) = -0.3$, $p = .80$ and $t(70) = 0.6$, $p = .55$, respectively). The families of the two groups of children also did not differ in income ($U = 666.5$, $p = .50$) or in the number of children in the family ($U = 684.5$, $p = .20$).

Differences in Adaptive Abilities In Relation to Pain Experienced

Accidental Pain

A MANOVA comparing children who had or had not experienced accidental pain in regard to their age equivalents on the four domains of the VABS (Sparrow et al., 1984) revealed a significant multivariate effect($F(4,89) = 2.8$, $p = .029$) due to pain. Univariate tests indicated children who had experienced accidental pain had higher age equivalents for Motor Skills ($M = 12.5$, $SD = 8.4$) than children who had not ($M = 7.0$, $SD = 9.5$; $F(1,92) = 7.0$, $p = .009$). Children who had experienced accidental pain did not differ

significantly from those who had not in age equivalents for Communication (M = 13.8, SD = 10.8 versus M = 13.8, SD = 8.0; F(1,92) = 0.002, p = .97), Daily Living Skills (M = 13.1, SD = 12.9 versus M = 16.0, SD = 8.0; F(1,92) = 1.2, p = .27) or Socialization (M = 14.2, SD = 14.0 versus M = 13.7, SD = 10.1; F(1,92) = 0.03, p = .86). Thus, children who had at least one accident over the four weeks had higher overall adaptive abilities, but these were primarily due to their greater motor skills.

Non-Accidental Pain

A second MANOVA compared children who had or had not experienced non-accidental pain over the four weeks in regard to their age equivalents on the four domains of the VABS (Sparrow et al., 1984). The multivariate effect of having experienced pain was significant (F(4,89) = 3.7, p = .007). Univariate tests indicated children who had experienced non-accidental pain had significantly lower age equivalents than those who had not for Communication (M = 11.5, SD = 8.5 versus M = 17.6, SD = 11.5; F(1,92) = 9.0, p = .004), Daily Living Skills (M = 10.8, SD = 7.9 versus M = 19.1, SD = 14.8; F(1,92) = 12.3, p = .001), Socialization (M = 11.5, SD = 10.3 versus M = 18.2, SD = 15.6; F(1,92) = 6.3, p = .01) and Motor Skills (M = 6.2, SD = 7.5 versus M = 12.7, SD = 11.0; F(1,92) = 11.5, p = .001). Thus, children who experienced non-accidental pain at least once over the four weeks surveyed had lower adaptive abilities in all areas.

Relation between Adaptive Ability and Pain Experienced

The previous set of analyses examined whether the mere presence of accidental or non-accidental pain was associated with children's adaptive functioning. A second set of analyses was conducted to explore whether the amount of pain experienced was

associated with their level of adaptive abilities. Table 5.10 lists the Pearson correlations between children's adaptive age equivalents and the number of pain episodes and the number of types of pain episodes they experienced. Although many achieved significance, only four were significant after corrections for multiple tests. Having lower age equivalents for Daily Living Skills was associated with more episodes of pain of all types. As children's age equivalents for Communication and Daily Living Skills decreased, the number of types of pain they experienced also increased. In addition, decreasing age equivalents for Daily Living Skills were associated with more episodes of non-accidental pain.

Pain and NCCPC-R Score

Pearson correlations indicated children's total scores on the NCCPC-R, a measure of typical pain behaviour, were not related to the total number of pain episodes reported ($r = .20$, $p = .055$). Thus, caregivers' reports were not biased in favour of children who typically exhibit more pain behaviour. Total scores on the NCCPC-R were also not related to the number of types of pain experienced ($r = .11$, $p = .28$). However, children's scores on the NCCPC-R were significantly related to the number of accidental ($r = -.23$, $p = .03$) and non-accidental types of pain they experienced ($r = .23$, $p = .03$), before corrections for multiple tests, but did not achieve significance after corrections were made. As the number of pain behaviours children typically exhibit increased, their number of episodes of accidental pain tended to decrease and their number of episodes of non-accidental pain tended to increase.

Discussion

This is the largest and most detailed study to document the incidence and characteristics of pain experienced by children with cognitive impairments. The results indicate pain is a frequent experience for this special group. Pain was experienced on 18% of days over a four-week period, and 35% to 52% of children experienced pain each week. This contrasts with the results of the Perquin et al.'s study, in which just over 50% of children without impairments experienced pain over a three month period (Perquin et al., 2000). However, it is closer to the 73% of children who experienced pain at least once in a two-week period in Stallard's et al.'s study of children with non-malignant life-threatening conditions (Stallard et al., 2001).

The pain experienced by the children was not only frequent, but also significant in nature. Only post-accidental pain received a mean intensity rating below 4 on the 0 to 10 scale, and the average intensity for all episodes was 5.7. In addition, pain was not short lasting; children who had pain at least once spent 19 to 29 hours week in pain. The results also suggest that most of this pain was not due to accidental injury or medical procedures. Of the 94 children, 62% had at least one episode of pain that was not due to accident or medical procedures, and 82% of pain episodes were not due to injury or medical procedures. Non-accidental pain was also more intense than accidental, with mean intensity ratings of 6.1 and 3.8, respectively. It also lasted longer, six hours on average, compared to forty-five minutes for accidental pain. Together, these results suggest that pain is a frequent and substantial problem for children with severe cognitive impairments,

and that pain due to causes other than injury or medical procedures is most severe in frequency, intensity and duration.

The results also provide information regarding the causes of pain for children with severe cognitive impairments. The greatest percentage of children (30%) had at least one episode of pain that was due to an accident. This was followed by gastrointestinal pain, which was experienced by 22% of children and represented 25% of all pain episodes reported. Although it has been suggested that children with severe cognitive impairments may suffer a great deal of pain due to medical procedures, such as surgery and injections (McGrath, Rosmus, Camfield, Campbell, & Hennigar, 1998), only 13% of these children experienced medical pain over the four weeks of the study, and these represented only 8% of all pain episodes reported. Even more surprisingly, most of this was not due to surgery, or to injection, but was due to ongoing problems related to leakage at feeding tube sites. It is likely that the children still have more medical pain than children of similar ages who do not have cognitive impairments. However, data of similar detail must be collected regarding children without cognitive impairments to confirm this.

The analyses of differences in family demographic characteristics revealed no differences between children who did or did not have accidental pain or non-accidental pain. However, analyses did indicate that children with greater motor kills were more likely to have accidental pain. Given that the children who did have accidental pain did not have significantly greater age equivalents than those who did not for the Communication, Socialization, and Daily Living Skills domains of the VABS (Sparrow

et al., 1984), this result suggests that a combination of greater mobility without a concomitant increase in other abilities may underlie this difference.

There were few differences in the pain experienced by girls and boys. However, boys did tend to have more musculoskeletal pain than girls, and tended to have more episodes of non-accidental pain over the four weeks surveyed. Similarly, there were few differences in the pain experienced by children due to their age. Younger children tended to have more accidental pain than older children, while older children tended to have a greater number of pain episodes. Children aged 8 to 12 had more recurrent pain than older children and more musculoskeletal pain than children aged 3 to 7. This may represent the fact that chronic painful conditions develop in mid childhood, but that management improves with time and age. Further research would be needed to verify this hypothesis.

The results regarding children's amount of pain, their typical pain behaviour and their adaptive abilities are intriguing. The fact that the number of pain behaviours typically displayed by children tended to decrease with the number of accidental pain episodes experienced, but tended to increase with the number of non-accidental pain episodes, could reflect three possible phenomena. First, it could be that children who experience multiple injuries reduce their pain behaviour over time, as they become accustomed to their daily bumps and bruises. Second, children who experience more non-accidental pain may display more pain behaviour over time, opening the possibility that they become sensitised to pain. This is supported by the marginal increase in pain behaviour with increasing number of all pain episodes. Third, it is possible the children

who experienced more pain in this study were less able to produce pain behaviour due to having greater physical impairments. This coincides with recent research indicating that daily pain in elderly persons who are institutionalised is greater with increasing dependency (Finne-Soveri & Tilvis, 1998). The finding that adaptive abilities, especially in communication and daily living skills, decreased with the number of types of pain and the number of non-accidental pain episodes experienced also supports this possibility. However, these correlational analyses do not indicate the direction of causality. Thus, one cannot forget the possibility that more pain experiences may also have contributed to the reduced abilities of these children, as recent studies suggest that repeated pain may have long-term consequences for children's behaviour (Porter, Grunau, & Anand, 1999; Whitfield & Grunau, 2000). Prospective research examining the adaptive abilities and pain behaviour of children at baseline, and following them over time to observe the effect of subsequent pain on those abilities and pain behaviour, is needed to unravel these complex relationships.

There were several limitations to this study. First, caregivers' provided retrospective reports for one-week periods. Although the interview used was designed to encourage accurate recall, the reports given may be underestimates of the number of episodes of pain the children experienced (Peterson et al., 1993). Second, caregivers were used as the sole source of information regarding the cause of their child's pain. Although these caregivers have developed an intimate knowledge of their child's medical conditions and reactions to pain due to different sources, they may not have been accurate in all cases. For example, for 5% of episodes, caregivers could not discern the cause of

their child's pain. However, these episodes were also rated most highly in terms of pain intensity, suggesting signs that the child was in pain were strong despite the caregiver being unable to detect the cause. Third, the number of children who experienced each category of pain was small. Thus, comparisons of children who had categories of pain on factors such as age and gender may not have had sufficient power to detect differences. Finally, these results are based on only four weeks of recordings by caregivers, and, therefore, should be taken as preliminary. Nevertheless, the fact that these represent four weeks over the course of a year strengthens the validity of the findings.

In summary, these results indicate the burden of pain in these children is considerable and appears to be greater than that of children without cognitive impairments. The majority of the children in this study experienced pain weekly, and most of that pain was not due to injury, but to illness and chronic conditions. These findings highlight the importance of further research into the management of pain in this special group. Studies to uncover possible risk factors for pain are needed to help caregivers and healthcare professionals to diagnose pain when it occurs. Research into treatment strategies are also crucial to develop management approaches that may help reduce the frequency, intensity and duration of the pain in this vulnerable group.

CHAPTER SIX: THE IMPACT OF PAIN ON THE DEVELOPMENT OF ADAPTIVE ABILITIES BY CHILDREN WITH SEVERE COGNITIVE IMPAIRMENTS OVER ONE YEAR

As more knowledge is gained concerning the nature and frequency of pain in childhood, researchers are turning to investigations of how this pain might affect children's functioning and development. Most studies of the impact of paediatric pain have examined the effect of chronic pain on psychological (Thompson, Gil, & Keith, 1994; Varni et al., 1996) or psychosocial functioning (Timko, Stovel, Moos, & Miller, 1992; Aasland, Flato, & Vandvik, 1997). Others have examined school attendance (Shapiro et al., 1995; Sturge, Garralda, Boissin, Dore, & Woo, 1997) or general quality of life (Langeveld, Koot, & Passchier, 1997; Ruperto et al., 1997). However, no study could be found that investigated the effects of pain on children's development of abilities to perform everyday tasks.

Longitudinal studies are the only method of evaluating change in individuals and provide valuable information regarding causality because they incorporate the temporal order of events (Bijleveld et al., 1998). The present study investigated the long-term impact of pain on the development of adaptive functioning in children with severe cognitive impairments. The amount of time children spent in pain over four one-week periods was determined. These weeks were spaced over a one-year period and provided time samples of their pain over the year. Children's adaptive functioning was assessed at the beginning and the end of the year using the Vineland Adaptive Behavior Scales

(Sparrow et al., 1984), a standardised measure of adaptive abilities. Because the children had many co-morbid medical conditions that could make them ill, but not cause pain, the effect of their medical stability was accounted for as well.

As the data described in Chapter Five suggest, children with severe cognitive impairments experience pain frequently, and some experience many hours of pain each week. This study was designed to provide a preliminary exploration of that impact of their pain over a one-year period. It was hoped that the information gained might provide an indication of the areas of functioning most susceptible to the negative effects of pain.

Methods

Participants

The full cohort of 101 children participated in this study. However, four children died before completion of the first year of the study. Seven additional caregivers withdrew from the longitudinal study due to severe child or parent illness or family move. In addition, one caregiver was unable to complete the second administration of the Vineland Adaptive Behavior Scales (Sparrow et al., 1984). One child, whose caregiver had not returned two of the four pain surveys at the time the analyses for the previous chapter were conducted, was included in these analyses because his/her pain surveys were received before these analyses were conducted. Thus, 89 children formed the final sample for this study, 40 girls and 49 boys. The small number of children who were excluded precluded statistical comparison of the groups who did and did not complete the measures required for this study.

The cause of the children's cognitive impairments included: dysmorphic syndromes, 29; chromosomal syndromes, 6; extreme prematurity, 11; asphyxia at birth, 9; traumatic brain injury, 8; neurodegenerative disorders, 5; intrauterine acquired condition, 5; infection, 4; and epileptic syndrome, 2. The cause of impairment was unknown for seven children and information was unavailable for three. The children ranged in age from 3 to 18.9 years (M = 10.0, SD = 4.4). Seventy-eight children lived with their family and 11 in group homes or residential centres. The mothers of the children living with their family were 20 to 66 years old (M = 38.9, SD = 7.5, n = 78) and their fathers were 28 to 70 years old (M = 41.2, SD = 8.1, n = 69).

Information regarding the families of children who lived in the family home is listed in Table 6.1. Further information regarding the children's medical conditions is displayed in Table 6.2.

Measures

Demographic Information and Medical History

This questionnaire (Appendix B) was administered to caregivers during their first telephone interview when they entered the research program and one year later. It is described in detail in Chapter Three. The frequency with which children required medical monitoring by a healthcare professional (none, monthly, weekly, daily) was used as a measure of Medical Stability.

The Vineland Adaptive Behavior Scales (VABS)

This semi-structured interview was administered as part of the first interview at entry to the research program (Sparrow et al., 1984). It is described in detail in Chapter Three.

Pain Surveys

These were administered to caregivers by telephone (Appendix C). They described in detail in Chapter Three. To facilitate caregivers' recall, the interviewer systematically questioned the caregiver regarding pain episodes that had occurred, day by day, beginning with six days previous to the call and moving forward to the day of the call. The caregiver was also asked to describe what they believed was the cause of each pain episode. The interviewer then asked them to rate the intensity of each pain episode from 0 (none at all) to 10 (worst pain ever) and to estimate how long each pain episode had lasted.

Procedure

Approximately one month after entry into the research program ($M = 4.8$ weeks, $SD = 3.3$), caregivers were contacted by telephone to complete the first Pain Survey. The second Pain Survey was completed approximately three months later ($M = 13.4$ weeks, $SD = 4.2$). Approximately three months elapsed between the second and third Pain Surveys ($M = 13.1$ weeks, $SD = 2.7$) and the third and fourth Pain Surveys ($M = 13.6$ weeks, $SD = 2.7$). Caregivers completed the VABS a second time approximately one year after entering the study ($M = 58.8$ weeks, $SD = 9.9$).

Statistical Analyses

Coding of the Pain Episodes

Over the four weeks children had 369 episodes of pain. Among the causes of the episodes were accidental injury (14%), orthopaedic conditions (10%), gastrointestinal conditions (21%), infections (17%), muscle spasms or tightness (3%) and teething (3%). Caregivers' estimates of the duration of each pain episode were used to compute the total time each child spent in pain over the four weeks surveyed in hours and minutes (Table 6.3). This was then divided by quartiles for use as a between-subjects factor in the primary analyses to avoid empty cells.

Adaptive Behaviour Scores

Children's age equivalents on the Vineland Adaptive Behavior Scales (Sparrow et al., 1984) for the 4 domains and 11 subdomains were generated for descriptive purposes. Age equivalent difference scores were also computed, by subtracting children's age equivalents at entry to the program from their age equivalents one year later. However, difference scores for age equivalents were skewed and did not meet normality assumptions (skewedness: -0.6 - 4.2; kurtosis: 2.6 - 26.3). Thus, raw scores were used to compute raw difference scores for use in analyses.

Missing Data

Duration was not provided for 51 (13.8%) pain episodes. To provide a conservative estimate of the total duration of pain over the four weeks, these missing values were replaced with zero before summing the time each child spent in pain over the four weeks.

Statistical Procedures

Pain duration over the four weeks. A repeated measures analysis of variance (ANOVA) was used to determine if pain duration differed across the four weeks. Wilks' lambda was used to test significance, as Mauchley's test indicated the assumption of sphericity was not met. Power exceeded .80 to detect medium size differences.

The effect of baseline abilities on change in adaptive functioning. To determine if children's change in scores on the four adaptive behaviour domains was related to their level of functioning at entry to the study, and whether raw scores at entry should be included as a covariate in analyses examining change, Pearson correlations were generated between children's composite age equivalent at entry, and their raw difference scores. Power to detect a medium size correlation was .83.

Change in adaptive functioning. Before examining the impact of pain on development, analyses were conducted to determine if children had developed skills during the year of the study and whether their level of medical stability influenced this. A repeated measures analysis of covariance (ANCOVA) was conducted on the four domain total raw scores at entry and one year later, with children's level of Medical Stability (monitored daily, weekly, monthly, not at all) included as a covariate. Mauchley's tests indicated sphericity assumptions were not met, so Wilks' lambda was used to test significance. Power to detect a small effect, without accounting for the covariate, exceeded .80.

The impact of time in pain on change in adaptive functioning. A multivariate analyses of covariance (MANCOVA) was then conducted on raw difference scores for

the four VABS domains, including quartiles of pain duration (Pain) as a between-subjects factor. Frequency of medical monitoring (Medical Stability) was included as a covariate (Girden, 1992). Power to detect large univariate effects was .88, accounting for the effect of the covariates. Descriptives of change in age equivalents by quartile of pain were also generated to demonstrate effects found in these analyses because of their greater ease of interpretation.

Additional MANCOVA's were then conducted on subdomain raw difference scores when univariate effects for a domain in the preceding MANCOVA were significant. In each case, children's Medical Stability was included as a covariate. Power to detect large univariate effects was .90, accounting for the effect of the covariate.

Results

Pain Experienced by the Children

Pain duration over the four weeks. Table 6.3 lists the pain experienced by children. This ranged from 0 to 146.5 hours (more than 6 days). Of the 2492 days reported on for the 89 children, pain occurred on 428 days (17%) and only 20 children (23%) had no pain over the four weeks surveyed. The children were assigned to four groups based on the total duration of pain they experienced over the four weeks surveyed. The mean duration of pain for each group is listed in Table 6.3. The range of pain duration experienced by the groups was as follows: Quartile One: no pain, Quartile Two: less than 1 minute to 2 hours, Quartile Three: 2 hours, 1 minute to 30 hours, Quartile Four: 48 hours to 343 hours, 10 minutes. A repeated measures ANOVA indicated there

was no significant difference in the duration of pain during the four weeks ($F(3,86) = 0.8$, $p = .97$).

Adaptive Functioning of the Children

The effect of baseline abilities on change in adaptive functioning. Pearson correlations indicated the change in children's raw scores over the year was not related to their abilities at baseline for the four domains after corrections for multiple tests: Communication ($r = .17$, $p = .10$), Daily Living Skills ($r = -.06$, $p = .57$), Socialization ($r = .01$, $p = .91$) and Motor Skills ($r = .24$, $p = .02$). Thus, composite adaptive age at entry was not included as a covariate in subsequent analyses.

Change in adaptive functioning. Table 6.4 lists the raw scores and age equivalents for the children on the VABS domains and subdomains at entry to the study and one year later. On the whole, children gained skills in all adaptive areas except Expressive Communication. A repeated measures ANCOVA on domain raw scores revealed a main effect for Time ($F(1,87) = 2.8$, $p = .10$) that approached significance, and a significant effect of Domain ($F(3,85) = 5.3$, $p = .002$). The effect of the covariate Medical Stability was also significant ($F(1,87) = 6.2$, $p = .01$). The interaction between Time and Domain ($F(3,85) = 3.5$, $p = .02$) was significant, while the interaction between Time and Medical Stability was not ($F(1,87) = 0.08$, $p = .78$). The interaction between Domain and Medical Stability was nonsignificant ($F(3,85) = 2.0$, $p = .12$) and the three way interaction approached significance ($F(3,85) = 2.2$, $p = .24$). Thus, children gained abilities over the year, but this varied in relation to Domain. Medical Stability also affected children's

performance of adaptive abilities, but did not affect the change in functioning over the year.

The Impact of Time in Pain on Change in Adaptive Functioning

A MANCOVA on difference scores for adaptive functioning, using quartiles of time in pain (Pain) as the between subjects factor and including Medical Stability as a covariate, indicated Pain significantly affected change in adaptive functioning ($F(12,214.6) = 2.7$, $p = .002$). Children who had more time in pain showed less development across the four adaptive domains (Table 6.5). The multivariate effect of the covariate Medical Stability was nonsignificant ($F(4,81) = 1.4$, $p = .25$). Univariate tests revealed, after corrections for multiple tests, the effect of Pain was significant for Communication ($F(3,84) = 4.1$, $p = .01$), Daily Living Skills ($F(3,84) = 4.8$, $p = .004$) and Motor Skills ($F(3,84) = 3.9$, $p = .01$). The univariate effect of Pain was nonsignificant for Socialization ($F(3,84) = 1.9$, $p = .14$). The univariate effects of Medical Stability on Communication ($F(1,84) = 0.03$, $p = .87$), Socialization ($F(1,84) = 0.6$, $p = .44$) and Motor Skills were nonsignificant ($F(1,84) = 0.7$, $p = .41$), although the effect on Daily Living Skills ($F(1,84) = 3.1$, $p = .08$) approached significance. These results indicate that as time spent in pain increased overall development was reduced across the four areas of functioning. Development in communication, daily living skills and motor skills were significantly impacted by the amount of pain experienced (Table 6.5). However, development of social skills was not affected significantly by time spent in pain. Finally, Medical Stability had no significant impact on children's change in abilities over the year.

Three additional MANCOVA's examined the impact of pain duration on development in Communication, Daily Living Skills and Motor Skills. The mean change in age equivalents for the subdomains is listed in Table 6.5. The first MANCOVA indicated the multivariate effect of Pain was significant across Communication subdomains ($F(9,199.7) = 2.0$, $p = .05$). The multivariate effects of the covariate Medical Stability, was nonsignificant ($F(3,82) = 0.1$, $p = .96$). Univariate tests revealed the effect of Pain was significant for Receptive Communication ($F(3,84) = 4.6$, $p = .005$), but only approached significance for Expressive Communication ($F(3,84) = 2.3$, $p = .08$) and was nonsignificant for Written Communication ($F(3,84) = 1.0$, $p = .40$). The univariate effects of Medical Stability were nonsignificant for Receptive ($F(1,84) = 0.6$, $p = .80$), Expressive ($F(1,84) = 0.2$, $p = .65$) and Written ($F(1,84) = 0.0$, $p = .99$) Communication. Thus, time in pain primarily affected communication through its impact on children's ability to understand language, although it had a tendency to cause some impact on expressive communication. Medical Stability did not influence children's development of communication skills.

The second MANCOVA indicated the effect of Pain across Daily Living Skills subdomains only approached significance ($F(9,197.3) = 1.7$, $p = .10$). The multivariate effect of Medical Stability was nonsignificant ($F(3,82) = 1.6$, $p = .19$). The univariate tests revealed a marginal effect of Pain on the Personal subdomain after corrections for multiple tests ($F(3,84) = 3.2$, $p = .026$). However, the univariate effect for the Domestic subdomain only approached significance ($F(3,84) = 2.3$, $p = .08$) and the effect of Pain on Community Skills was nonsignificant ($F(3,84) = 1.8$, $p = .15$). The univariate effect of

Medical Stability was nonsignificant for the Personal ($F(1,84) = 2.2$, $p = .14$), Domestic ($F(1,84) = 0.1$, $p = .79$) and Community subdomains ($F(1,83) = 3.2$, $p = .08$). Thus, time in pain had some effect on daily living tasks within the personal care subdomain, such as eating and personal hygiene, although this did not achieve significance after corrections were made for multiple tests. Time spent in pain had no effect on children's ability to function in the community or handle domestic or household tasks (Table 6.5).

The final MANCOVA produced a significant multivariate effect of Pain across the Motor Skills subdomain ($F(6,166) = 2.4$, $p = .03$), accompanied by a significant univariate effect for Fine Motor Skills ($F(3,84) = 3.8$, $p = .01$), but a nonsignificant effect for Gross Motor Skills ($F(3,84) = 1.4$, $p = .24$). The multivariate effect of Medical Stability and the univariate effect of medical Stability on Fine Motor Skills approached significance ($F(2,83) = 4.0$, $p = .10$ and $F(1,84) = 3.6$, $p = .06$, respectively). The univariate effect of Medical Stability on Gross Motor Skills was nonsignificant ($F(1,84) = 1.0$, $p = .31$). Thus, pain affected development of motor skills, and this was due primarily to its effect on fine motor abilities. Medical Stability affected overall motor skills and fine motor skills to some extent, but not gross motor skills.

Discussion

These results indicate the amount of time spent in pain over four weeks significantly affected the development of these children with severe cognitive impairments. Increasing time in pain significantly reduced overall development of adaptive abilities over the year, and this was especially true for Communication, Daily

Living Skills and Motor Skills. These differences were significant even when medical stability was controlled for indicating pain had effects independent of possible reductions due to illness or health conditions.

The effect of pain also varied across domains and subdomains of the adaptive areas examined. Within the Communication domain, development of receptive communication was most affected by time in pain. This result suggests pain had effects on attention or cognitive functioning, as understanding language does not require physical abilities or development. Pain did not affect written communication. However, there was very little variability in these scores, as few children could perform any tasks relevant to written communication. For example, children could perform an average of 7 items from the Receptive subdomain ($\underline{M} = 6.9$, $\underline{SD} = 3.2$) and over five items from the Expressive subdomain ($\underline{M} = 5.7$, $\underline{SD} = 5.1$), but less than one from the Written subdomain ($\underline{M} = 0.2$, $\underline{SD} = 0.9$) Thus, it is likely the nonsignificant effect reflects this lack of variation among scores.

Within the Daily Living Skills domain, personal care skills were also most reduced by pain. An examination of change in the Daily Living Skills subdomains indicated 34% of children lost skills in this area, while only 6% lost domestic skills and only 16% lost community skills. One possible reason for the difference among these subdomains could be that children had more personal skills to lose. At entry, children could perform an average of eight tasks in the personal subdomain ($\underline{M} = 8.0$, $\underline{SD} = 6.5$) but less than one in the domestic and community subdomains ($\underline{M} = 0.7$, $\underline{SD} = 2.2$ and $\underline{M} = 0.9$, $\underline{SD} = 2.1$, respectively). Thus, the difference in the effect of pain across these

subdomains could reflect the fact that little reduction in skills was possible on the latter two subdomains.

Within the Motor Skills domain, time in pain most affected fine motor skills. It is also interesting that this is also the only domain on which children's overall change was still in the positive direction, regardless of pain. Children were also capable of slightly more gross motor ($\underline{M} = 4.4$, $\underline{SD} = 3.9$) than fine motor skills ($\underline{M} = 3.2$, $\underline{SD} = 3.5$) at entry. One possible reason for the difference could be the nature of the children's physical impairments. That is, the severity of their physical limitations may have attenuated development of gross motor skills such as walking, resulting in less variability due to the pain experienced. This is supported by the data, which suggest that, regardless of pain, the children advanced only 1.3 months in gross motors skills over the year, while they advanced 3.7 months in fine motor skills. However, further research should examine the process of pain's impact on motor skills to clarify these differences further.

Finally, within the Socialization domain, time in pain did not significantly affect development. A look at the development in this area suggests only the children who experienced the greatest amount of pain did not make gains in this area. Thus, it may be that social functioning is more robust than other areas of pain, and decreased by only the most unremitting pain. It is also possible that the behaviours in this subdomain were still performed, but in an altered form, because they are less specific than those in other domains. For example, behaviours within the Socialization Domain, such as showing affection, showing an interest in others and taking part in an activity with others are more adaptable than many behaviours within other domains, such as feeding oneself with a

fork, using person's names or sitting up without help. Thus, it may be that the children still performed these social tasks, but their performance of more complex versions of the tasks was attenuated. Further research should examine the factors that may make social functioning more robust in the face of pain than other areas of development and whether the complexity of children's social behaviours is affected by pain.

There were several limitations to this study. First, caregivers provided retrospective reports for one-week periods. Although the interview used was designed to encourage accurate recall, the reports given may be underestimates of the time the children spent in pain (Peterson et al., 1993). Second, these results are based on only four weeks of recordings by caregivers over a one-year period and the results could differ if caregivers had been surveyed regarding their child's pain for the full 52 weeks of the year. Nevertheless, the fact that these represent four weeks over the course of a year strengthens the validity of the findings.

Third, the wide variety of causes of pain prevented analyses based on pain of specific types. Nor did the sample size allow for analyses examining the effects of pain on subgroups of children. Future research should investigate whether certain types of pain or intensities of pain have a greater impact on development of these skills or whether children with specific characteristics or who have reached certain milestones of development are more or less susceptible to the negative impact of pain.

Fourth, the measure of medical stability provided only an estimate of the children's medical fragility based on caregivers' report of how frequently the children

needed medical monitoring. This may not reflect the level of medical stability the children would be deemed to have were a physician to make that judgement. Further studies should incorporate more comprehensive measures of medical status to confirm that medical factors do not modulate the impact of pain on development of adaptive abilities.

The study also had several strengths. Data was collected over four one-week periods, resulting in 28 days of information concerning pain for each child. These one-week periods were spaced over the course of a full year to increase the likelihood that the samples of pain experienced were random and not associated with temporary conditions, time of year, or other transient factors. Analyses also suggest the children's time in pain was stable across these four weeks. Thus, the sample of pain experience documented appears to be representative of that the children experience over the long-term. The measure of adaptive functioning used was also comprehensive and allowed an examination of eleven specific areas of functioning. Thus, the information regarding children's adaptive abilities at entry to the study and at the end of the year provided a rich description of their skills and development of those skills over a year.

These results suggest pain may hinder development in children who are already burdened with severe cognitive impairments. Although pain had some effect on most areas of development examined, the impact was greatest in relation to receptive communication, personal care skills and fine motor skills. These areas of functioning are vital to everyday functioning. The fact that the effect of pain was evident over only one year, and based on only a small sample of pain experienced over that year, suggests that

over the course of childhood pain may have serious consequences for the children's development in these areas. This suggests good pain management might do more than reduce suffering, it may help these children, who are already burdened with physical and cognitive impairments, reach their fullest potential. It also suggests additional interventions may be needed for some children, especially in cases where no validated pain management techniques are available or where pain is difficult to manage.

CHAPTER SEVEN: VALIDATION OF THE NON-COMMUNICATING CHILDREN'S PAIN CHECKLIST-REVISED[1]

The results of the previous two chapters indicate children with severe cognitive impairments experience pain frequently and that that pain has an impact on their long-term development. This situation cannot be rectified without valid, reliable methods of measuring their pain. Pain measurement is essential for successful pain management and is also the foundation of research into the causes of pain. Over the past two decades there has been substantial growth in the number of tools for measurement of children's pain. However, there are still no standardised measures available for clinical assessment of the pain experienced by children with severe cognitive impairments. This chapter is the first of three describing studies designed to validate pain measures for this special group of children.

The previous two chapters included report from caregivers of whether or not their child had pain, their judgement of the cause of pain, their estimates of the duration of that pain, and their visual analogue ratings of the intensity of that pain. The study described in this chapter examines the validity of a structured pain measure used by the caregivers for pain episodes. It investigated the psychometric properties of the Non-communicating

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[1] The data presented in this chapter included: Breau, L.M., McGrath, P.J. , Camfield, C.S. & Finley, G.A. (2001). Psychometric Properties of the Non-communicating Children's Pain Checklist-Revised. Manuscript submitted for publication.

Children's Pain Checklist-Revised (NCCPC-R), a pain measurement tool specifically designed for children with severe cognitive impairments.

In an effort to develop an objective observational measure of pain for these children, McGrath's group conducted semi-structured interviews with the primary caregivers of 20 individuals with severe or profound mental retardation (McGrath et al., 1998). Thirty-one behaviours were extracted from the interviews. These were reported by 5% to 90% of caregivers. Although specific behaviours varied amongst individuals, caregivers' reports indicated all individuals displayed some behaviour from each of the seven categories. In a subsequent study, this group provided a preliminary validation of their pain measure, the Non-communicating Children's Pain Checklist (NCCPC), when used in a home setting (Breau et al., 2000). The caregivers of 33 individuals, aged 3 to 44, completed the NCCPC for four observations, two in which the individual with severe cognitive impairments had pain, one in which they were distressed but had no pain, and one during which they were calm. The NCCPC could differentiate between the behaviour of the children during the painful, calm, and distressful events, and was internally consistent and reliable over time (Breau et al., 2000). In a second study, these authors found that the occurrence of a subset of items from the NCCPC during discrete episodes of pain could be predicted by caregivers' report that those signs were commonly displayed during pain by their children (Breau et al., 2001a).

All pain assessment tools require repeated evidence of their validity and reliability before they can be recommended for clinical use. Given the wide range of observed mental and physical abilities and limitations in children with severe cognitive

impairments (van Dongen et al., 2000a), and the large number of pain behaviours caregivers of these children have reported occur during pain (Abu-Saad, 2000), it is especially important that tools developed for this group demonstrated acceptable psychometric properties.

To be clinically useful, a pain measurement tool must show several psychometric properties. First and foremost, the tool must be shown to measure pain. This is often described as "validity". Concurrent validity is one aspect of validity and it is assessed by investigating how closely the tool corresponds to pre-existing measures of pain (Johnston, 1998). Discriminative ability is also one aspect of validity. This refers to how well the tool discriminates between observations during which a child has pain and does not have pain. Finally, a tool's sensitivity and specificity refers to how accurately a tool can distinguish between individuals with and without pain. A pain tool must also be internally reliable or consistent. This indicates all items of the tool measure some component of pain, but are not redundant (Streiner & Norman, 1995b).

A pain measurement tool must also be reliable or stable over time. This is often referred to as test-retest reliability or as intra-rater reliability. Thus, an individual should achieve a similar score across similar episodes of pain (Streiner & Norman, 1995a). This characteristic is key for children with severe cognitive impairments, as reports that pain behaviour is heterogeneous in this group (van Dongen et al., 2000a) suggest these children may display quite variable behaviour across episodes of pain which could make measurement of pain based on their behaviour difficult. In addition, it is also critical that items from the pain tool are consistently absent when pain is absent. This aspect is often

not investigated in development of pain tools, but is essential for measures of pain in children with severe cognitive impairments because they may show pain-like behaviours when not in pain (McGrath et al., 1998).

This study was designed to provide evidence of the soundness of the NCCPC-R's psychometric properties by investigating its concurrent validity, internal consistency, test-retest reliability and discriminative ability using a different sample of children than those used in the development or preliminary validation of the original measure. Caregivers completed the NCCPC-R as part of Pain Diaries. They also recorded information regarding the nature of the pain their children experienced for the times the NCCPC-R was completed.

Method

Participants

The full cohort of 101 children participated in this study. However, only 71 children experienced pain at least once during the first 20 months of the research program. These children were included in these analyses, while children who had not experienced pain during that period were not.

The cause of the children's neurological impairments included: dysmorphic syndromes, 21; chromosomal syndromes, 6; extreme prematurity, 9; asphyxia at birth and traumatic brain injury, 8 each; neurodegenerative disorders, 5; intrauterine acquired condition, 4; infection, 2; and epileptic syndrome, 1. The cause of impairment was unknown for four children and information was unavailable for three.

Sixty of the children resided with their family, while the remaining 11 resided in group homes or residential centres. The mothers of children who lived with their families ranged from 20 to 54 years of age ($M = 38.0$, $SD = 6.8$) and their fathers ranged from 28 to 68 years ($M = 41.2$, $SD = 7.6$, $n = 53$). Further demographic information concerning the families is displayed in Table 7.1, information regarding the children's medical conditions and physical impairments is displayed in Table 7.2, and the children's chronological ages and adaptive age equivalents are listed in Table 7.3.

Representativeness of the Sample

Analyses were conducted to determine if the children from the cohort who were included in these analyses differed from those who did not. Power analyses suggested there was .95 or greater power to detect a large difference in means or proportions between the two groups. Independent t-tests revealed there was no difference in age ($t(99) = .054$, $p = .96$) or number of medications being taken ($t(99) = -2.2$, $p = .3$) after correction for multiple tests. Chi-square tests also indicated the two groups of children did not differ in terms of gender ($\chi^2 (1) = .02$, $p = .88$), whether they had cerebral palsy ($\chi^2 (1) = .30$, $p = .58$), or seizures ($\chi^2 (1) = .70$, $p = .40$), or whether they lived with their family ($\chi^2 (1) = 2.98$, $p = .08$). Mann Whitney U tests revealed there were no differences in the children's upper limb limitations ($U = 945.0$, $p = .34$), lower limb limitations ($U = 1043.5$, $p = .87$), level of diagnosed mental retardation ($U = 839.0$, $p = .46$), or how frequently they required medical monitoring ($U = 910.0$, $p = .22$). Finally, for those children who lived with their families, chi-square tests revealed that there was no significant difference in the caregivers' marital status ([$\chi^2 (4) = 4.4$, $p = .36$]), t-tests

indicated no difference in the age of mothers ($t(87) = -0.5$, $p = .62$) or fathers ($t(75) = -0.4$, $p = .69$), and Mann Whitney tests indicated no significant differences in income ($U = 666.0$, $p = .29$), number of children in the family ($U = 864.5$, $p = .96$), or the education achieved by mothers ($U = 789.0$, $p = .73$) or fathers ($U = 555.0$, $p = .61$). Thus, the results of this sub-sample of children can be generalised to the full cohort.

Measures

Caregivers completed Pain Diaries (Appendix C) as described in Chapter Three. In order to investigate the sensitivity of the NCCPC-R to pain, caregivers completed the Pain Diaries in the same manner regardless of whether their child had pain during each observation period.

For each diary, caregivers conducted a daily two-hour observation of their child at the same time each day. For each observation, caregivers indicated whether their child had experienced pain and whether the pain was due to injury, a chronic condition, an illness, or a medical procedure. They also indicated the duration of the pain (hours and minutes) and the intensity of the pain on a numerical rating scale anchored by 0 (none at all) and 10 (worst ever). The 30 items of the Non-communicating Children's Pain Checklist-Revised were included for each day of the diary. Caregivers were asked to rate how often each item had been observed over the two-hour observation (Not at all, Just a Little, Fairly Often, Very Often). One exception was the Eating/Sleeping subscale. Since it was considered unlikely that these behaviours would be observable over a two-hour period, caregivers were asked to indicate how often they were observed over the day.

Procedure

Approximately 3 months after entry into the research program (M = 11 weeks, SD = 7), caregivers were mailed a Pain Diary which they completed and returned by mail. Subsequent diaries were mailed at three-month intervals. The Pain Diaries used here included those completed within 20 months of the commencement of the research program.

Statistical Analyses

Selection of Observations for Analysis.

Four observations from the diaries were used, two with pain and two without. The data examined here include the first and second observation recorded by caregivers during which their child experienced pain. The criterion for selection of the first pain observation (Pain Episode 1) was that this was the first pain incident recorded in a diary since the child entered the study. The data revealed 38% of the episodes involved pain related to an injury (e.g., falls, self-injury), 32% to a chronic condition (e.g. gastrointestinal reflux, constipation), 24% to an illness (e.g. sore throat/ear during flu, chest congestion) and 6% to a medical procedure (e.g. surgery, enema). The pain the 71 children experienced ranged in duration from 1 minute to 24 hours (M = 3 hours, SD = 6 hours, 12 minutes) and the intensity, as recorded by caregivers, ranged from 1 to 10 (M = 5.1, SD = 2.4) on the 0-10 numerical rating scale of pain intensity. These observations occurred, on average, 18.5 weeks after entry to the research program (SD = 15.4 weeks).

The criterion for the second pain observation (Pain Episode 2) was that there had been at least two days during which no pain had occurred since the first pain episode or

that the cause of the second pain episode clearly differed from that of the first. Pain Episode 2 also included pain due to an injury (15%), a chronic condition (44%), an illness (28%) and a medical procedure (13%). The pain experienced by the 55 children during this observation ranged in duration from 1 minute to 48 hours (M = 6 hours, 48 minutes, SD = 10 hours) and the intensity, as recorded by caregivers, ranged from 1 to 9 (M = 4.9, SD = 2.1). These observations occurred, on average, 37.2 weeks after entry to the research program (SD = 20.4 weeks).

The data also include observations for two days on which the child did not have pain (No Pain Episodes 1 and 2). The criteria for the observations without pain were: a) that this was the first (No-Pain Episode 1) or second (No-Pain Episode 2) day an observation was made that there was no pain, and b) that the day had been preceded and followed by at least one day in which there was no pain recorded. The observations for No-Pain Episode 1 occurred, on average, 15.2 weeks after entry to the research program (SD = 11.0 weeks). The observations for No-Pain Episode 2 occurred, on average, 30.6 weeks after entry to the research program (SD = 13.1 weeks).

Time Interval between Observations

To ensure that differences between scores on the NCCPC-R during the selected observations were not due to the fact there were long time intervals between observations, the time since entry to the study was computed for all observations for comparison. Power was .98 to detect a medium effect using matched sample t-tests. These indicated that the number of weeks since entry to the study did not differ significantly between Pain Episode 1 (M = 19.1, SD = 15.6) and No-Pain Episode 1 (M =

22.9, SD = 63.6; t (66) = -.48, p = .635) or between Pain Episode 2 (M = 37.0, SD = 21.0) and No-Pain Episode 2 (M = 31.5, SD = 13.7; t(51) = 1.48, p = .145). Thus, differences in observed behaviours were not due to the observations being conducted at different times. Similarly, to ensure that consistencies found between NCCPC-R scores for Pain Episodes 1 and 2 were not due to those observations being conducted closely in time, a matched-sample t-test of weeks since entry was conducted. The results indicated there was a significant difference in the number of weeks since entry for Pain Episode 1 and 2 (t(54) = -8.4, p < .001). Thus, consistency between these two observations was not due to them being conducted close in time.

Time of Day of Observations

Matched sample t-tests, with power greater than .95 to detect a large size difference, were conducted on the mean start times of the episodes rounded to the nearest hour and beginning with 12:00 a.m. (0 hours) and ending with 11:00 p.m. (23 hours). These indicated the mean start time of the observation periods did not differ significantly between Pain Episode 1 and No-Pain Episode 1 (t(70) = .54, p = .59) or between Pain Episode 2 and No-Pain Episode 2 (t(54) = -1.60, p = .116). Thus, differences in NCCPC-R scores between observations in which pain was or was not identified would not be due to differences in the time of day the observations were made.

Missing Data

An examination of the data revealed only 2% of the items were not completed for the 71 children who had an observation for Pain Episode 1 and the 55 children whose caregivers documented a second episode of pain. However, 21% of the scores for the

item "eats less, not interested in food" were missing for Pain Episode 1 and 12.7% for Pain Episode 2. This was primarily due to caregivers of children who were tube fed noting that the item was "not applicable". To examine whether the Eating/Sleeping subscale scores of children who were tube fed differed from those of children who were not, two independent t-tests were conducted on children's scores for the Eating/Sleeping subscale for Pain Episode 1 and 2. Power analyses indicated there was .88 power to detect a large size effect. These indicated Eating/Sleeping subscale scores did not differ for Pain Episode 1 ($t(69) = -0.2$, $p = .88$) or Pain Episode 2 ($t(69) = 0.9$, $p = .38$). Thus, no action was taken to prorate the scores of children who were tube fed, and children whose caregivers' indicated the item was "not applicable" were given a score of "not at all". A second set of independent t-tests was conducted to explore whether children with full use of their arms received higher scores on the Body /Limb subscale than children with only partial or no use of their arms. Power was .91 to detect a large size effect. These indicated Body/Limb subscale scores did not differ for Pain Episode 1 ($t(69) = 0.6$, $p = .54$) or Pain Episode 2 ($t(69) = -1.3$, $p = .18$). Thus, scores were not prorated for this factor.

An examination of the data also revealed that 3% of the responses for No-Pain Episode 1 and less than 1% of responses for No-Pain Episode 2 were missing. This was primarily due to the fact that three children (4%) had no day during which they did not experience pain and one child had only one observation without pain. These children's missing values for the No-Pain Episodes were replaced by the median score of the remaining children for each item and each episode.

Scoring of the NCCPC-R

Subscale and total scores on the NCCPC-R were computed for each child for each of the four observations. To do so, applicable items were summed (Table 7.4). Mean subscale and total scores were also computed. Mean subscale scores were used for repeated measures analyses to avoid significant effects for subscale that were an artefact of the difference in possible scores for the seven subscales.

Statistical Procedures

Internal consistency. The internal consistency of the NCCPC-R was determined with Cronbach's alpha.

Reliability. The test-retest reliability of the NCCPC-R was determined through two separate repeated measures Analyses of Variance (ANOVA), one on mean subscale scores for the two pain episodes and one on mean subscale scores for the two episodes without pain. Power exceeded .80 to detect medium size effects. The reliability was also examined through a matched sample t-test comparing the number of items present for the two pain episodes. Power was .95 to detect medium size effects. As a final test of the stability of NCCPC-R scores over time, analyses were conducted to determine what proportion of children had scores that were stable over time. Using a procedure recommended by Brophy (Brophy, 1986), true scores were computed for each child using the equation: True Score = $\underline{r}\underline{X} + (1-\underline{r})\underline{M}$, where $\underline{r}$ is the Pearson correlation between scores for Pain Episodes 1 and 2, $\underline{X}$ is each child's total NCCPC-R score, and $\underline{M}$ is the mean score for all children for Episode 1. To determine whether each child's NCCPC-R total score for Pain Episode 2 differed from their true score more than would be expected

due to measurement error, confidence intervals for each child's re-test score were computed using the standard error of estimate for the group (SE_Y; $SE_Y = SD_X(1-r^2)^{1/2}$). First, 68% confidence intervals, which should include a re-test score two out of three times when the score is reliable, were computed as a stringent test of reliability based on suggestions by Brophy (Brophy, 1986). Then, larger 95% confidence intervals, which should include reliable re-test scores 95 times out of 100, were computed.

Validity. The concurrent validity of NCCPC-R scores during pain was assessed by computing Pearson correlations between total scores for Pain Episode 1 and Pain Episode 2 and the numerical ratings of pain intensity provided for the same pain episodes by caregivers. Power was .74 to detect a medium size correlation for Pain Episode 1 and .63 for Pain Episode 2.

The discriminative validity of the NCCPC-R was examined with two additional repeated measures ANOVA's. These compared mean subscale scores for Pain Episode 1 to those of No-Pain Episode 1 and for Pain Episode 2 to No-Pain Episode 2. Power for these exceeded .80 to detect medium size effects. Post-hoc comparisons were conducted with matched sample t-tests. Power was .98 to detect medium size effects. Matched sample t-tests were then used to compare total NCCPC-R scores for the two pain episodes to the two no-pain episodes. A second set of matched sample t-tests was used to compare the number of items present for the two pain episodes to the two episodes without pain. These had power of .95 to detect medium size effects.

Sensitivity and specificity. Receiver Operator Characteristic (ROC) Curves, based on the bi-negative exponential assumption, were used to determine cut-off points that

offered the best combination of sensitivity and specificity for the presence of pain. To do so, a random subsample of the 31 children with scores for Pain Episode 1 was generated. The scores of these 31 children during Pain Episode 1 were compared to those of the remaining 40 children for No-Pain Episode 1. To replicate this, a second random sample of the 55 children who had valid observations for Pain Episode 2 was generated and the scores of these 25 children during Pain Episode 2 were compared to the scores of the remaining 30 children for No-Pain Episode 2.

Results

Internal Consistency

Cronbach's alpha (α) was .93 for NCCPC-R scores obtained for both Pain Episode 1 and Pain Episode 2. The corrected item-total correlations and the percentage of caregivers who reported the presence of that item for Pain Episode 1 and Pain Episode 2 are listed in Table 7.4. Only one item, "gesturing to/touching part of body that hurt" had extremely low item-total correlations for both episodes. However, deletion of this item would only increase α by .002 for both episodes. In addition, 28% of caregivers reported this item for Pain Episode 1 and 24% for Pain Episode 2. In contrast, only one child displayed this behaviour during each of the two no-pain episodes. Thus, it was retained. The items "decreased appetite" and "increased sleep" had low item-total correlations only for Pain Episode 2. However, α was only increased .003 by removal of either item. In addition more than 25% of caregivers reported the presence of each item for the two pain episodes, while only 4% to 9% of children exhibited these items during the two no-pain episodes. Thus, these items were also retained for subsequent analyses. Overall, these

analyses indicate that the 30 items of the NCCPC-R measure the same construct, and are not redundant.

Reliability

Test-retest Reliability of Behaviour during Pain

The test-retest stability of the NCCPC-R during pain was examined through comparison of the scores achieved for Pain Episode 1 and Pain Episode 2. A 2 (episode) by 7 (subscale) repeated measures ANOVA was conducted on mean NCCPC-R subscale scores for the 55 children who had records for two episodes. Wilks' Lambda revealed nonsignificant effects for Episode ($F(1,54) = .001$, $p = .978$), but a significant effect of Subscale ($F(6,49) = 8.03$, $p < .001$) and a significant Episode by Subscale interaction ($F(6,49) = 3.19$, $p = .01$). To explore the interaction, a set of matched-sample t-tests was conducted on subscale scores. These revealed only the Social subscale changed significantly from Pain Episode 1 to Pain Episode 2 (Table 7.5). These analyses indicate that the NCCPC-R is stable across episodes of pain.

Test-retest Reliability of Behaviour when Pain is Absent

To investigate whether the behaviour displayed by the children in this study was variable when they did not have pain, a second analysis was conducted on mean NCCPC-R subscale scores obtained by the children for the two observations when they had no pain (Table 7.5). The 2 (episode) by 7 (subscale) repeated measures ANOVA revealed a nonsignificant Wilks' Lambda for Episode ($F(1,54) = .08$, $p = .785$), a significant effect of subscale ($F(6,49) = 4.74$, $p = .001$), and a nonsignificant interaction between the two ($F(6,49) = 1.03$, $p = .420$). Thus, when not in pain, the children in this study displayed

differing levels of behaviour from the seven NCCPC-R subscales, but did not differ in the number of behaviours they showed during different observations, suggesting their behaviour while not in pain was also stable. The combination of stable NCCPC-R scores when pain is absent, as well as when pain is present, suggests the NCCPC-R is specific to pain and that differences between scores when pain is present or absent should also be stable.

Reliability of Number of Pain Behaviours Displayed during Pain

Comparisons were made of the number of items observed by caregivers during the two episodes of pain to determine if the size of children's repertoire of pain behaviours changed over time. The matched sample t-test indicated the number shown during Pain Episode 1 ($M = 12.7$, $SD = 6.7$) did not differ significantly from the number shown during Pain Episode 2 ($M = 14.0$, $SD = 6.3$; $t(54) = -1.7$, $p = .09$). Thus, the number of items from the NCCPC-R displayed by children was stable over time and the total NCCPC-R scores obtained during pain were not due to a high frequency of only a few items of the NCCPC-R.

Proportion of Children with Reliable NCCPC-R Scores

As a final test of the stability of NCCPC-R scores over time, analyses were conducted to determine what proportion of children had scores that were stable over time using a procedure recommended by Brophy (Brophy, 1986). The results indicated that, 75% of children's total NCCPC-R scores for Pain Episode 2 fell within the 68% confidence interval, suggesting that they did not differ beyond that expected due to variations in testing conditions. Using 95% confidence intervals, 93% of children's scores

fell within the range expected by different test conditions. Thus, the vast majority of children's individual NCCPC-R total scores were stable over time using the stringent criterion suggested by Brophy and using more common confidence limits of 95%, almost all children's scores were stable.

Validity

Concurrent Validity

Pearson correlations between caregivers' numerical pain ratings and NCCPC-R total scores were generated. These were significant for Pain Episode 1 ($r = .46$. $p < .001$, $n = 71$) and Pain Episode 2 ($r = .46$, $p < .001$, $n = 55$). Thus, NCCPC-R scores and numerical pain ratings were highly related for both episodes of pain, indicating the NCCPC-R is measuring pain.

Discriminant Validity

Two separate 2 (episode) by 7 (subscale) repeated measures ANOVA's were used to examine the ability of the NCCPC-R to discriminate between observations during which there was and was not pain.

The first, conducted on scores for Pain Episode 1 and No-Pain Episode 1, revealed significant Wilks' Lambda's for Episode ($F(1,70) = 64.7$, $p < .001$), Subscale ($F(6,65) = 16.6$, $p < .001$) and the interaction between them ($F(6,65) = 5.2$, $p < .001$). To explore the interaction, a set of matched sample t-tests was used to compare subscale scores for Pain Episode 1 and No-Pain Episode 1. The results indicated all subscales differed significantly between the two episodes (Table 7.5). A final matched sample t-test

was used to examine the total scores for the two episodes. It also revealed a significant difference between the NCCPC-R total score obtained for Pain Episode 1 and No-Pain Episode 1 (Table 7.5).

The second repeated measures ANOVA was conducted on mean subscale scores for Pain Episode 2 and No-Pain Episode 2. It revealed significant Wilks' Lambda's for Episode ($F(1,54) = 70.3$, $p < .001$) and Subscale ($F(6,49) = 4.8$, $p = .001$) and a significant interaction between them ($F(6,49) = 3.6$, $p = .005$). To explore this interaction, a set of matched sample t-tests was used to compare subscale scores for Pain Episode 2 and No-Pain Episode 2. The results indicated all subscales differed significantly between the two episodes (Table 7.5). A final matched sample t-test was used to examine the total scores for the two episodes. Again, a significant difference was found (Table 7.5).

As a final test of the NCCPC-R's discriminant validity, the number of items observed on Pain Episode 1 and No-Pain Episode 1 were compared using a matched sample t-test. The results indicated there was a significant difference ($t(70) = 10.3$, $p < .001$) in the number displayed during Pain Episode 1 ($M = 12.7$, $SD = 6.7$) and No-Pain Episode 1 ($M = 3.7$, $SD = 3.8$). As well, a significant difference also found between Pain Episode 2 ($M = 14.0$, $SD = 6.3$) and No-Pain Episode 2 ($M = 3.2$, $SD = 3.9$; $t(54) = 11.7$, $p < .001$). Thus, not only total scores incorporating the number and frequency of items differed between pain and the absence of pain, but the absolute number of items also differed significantly.

Sensitivity and Specificity

To determine cut-off scores appropriate for establishing the presence or absence of pain, a random subsample of the 31 children with scores for Pain Episode 1 were generated. The scores of these 31 children during Pain Episode 1 were compared to those of the remaining 40 children for No-Pain Episode 1. A Receiver Operating Characteristic curve (ROC) was generated and indicated that a total NCCPC-R score of 7 or greater had the best combination of sensitivity (84%) and specificity (68%) for pain. To replicate this, a second random sample of the 55 children who had valid observations for Pain Episode 2 was generated. The scores for Pain Episode 2 of these 25 children were compared to the No-Pain Episode 2 scores of the remaining 30 children. A second ROC curve was conducted of these scores and indicated, again, that a total NCCPC-R score of 7 had the best combination of sensitivity (84%) and specificity (77%). Thus, a total NCCPC-R score of 7 appears to be an indicator that pain is present in this group of children.

Discussion

These analyses provide evidence that the Non-communicating Children's Pain Checklist-Revised (NCCPC-R) has excellent psychometric properties. The NCCPC-R exhibited excellent internal consistency across two separate episodes of pain observed by caregivers. NCCPC-R total scores were also significantly correlated with caregivers' numerical ratings of their children's pain intensity, indicating the NCCPC-R does measure pain.

The scores obtained by these children were also stable over time. The analyses indicate that neither total NCCPC-R scores or subscale scores differed significantly between the two pain episodes. The one exception to this was the Social subscale. The children received lower scores for the second episode of pain observed. It is unclear why scores for this one subscale was not consistent over time. However, it is interesting that this was the same type of behaviour for which development was not impacted by pain in the previous chapter. Thus, it may be that social behaviour has a complex relation to pain or is more easily adapted when pain is present. Further research is needed to investigate this phenomenon more comprehensively.

The results also indicate the number of items displayed by the children did not differ across the two pain episodes. In addition, the number of signs and behaviours displayed when children did not have pain was also consistent. This is an important finding because it suggests their typical daily behaviour does not resemble their pain behaviour and suggests there is a low baseline occurrence of the signs and behaviours of the NCCPC-R when pain is absent that is stable across time. Finally, calculation of confidence intervals for total NCCPC-R re-test scores suggests the vast majority of children had scores that are stable from one time to another. Using a 68% confidence interval, 75% of children had scores for Pain Episode 2 that were within the range expected due to intrinsic differences in testing situations. When the confidence interval was increased to 95%, only 7% of children had scores for Pain Episode 2 that were more variable than that expected due to measurement error. This result confirms the NCCPC-R

is reliable over time for a wide array of individual children with severe cognitive impairments.

The most important quality of any pain assessment measure is the ability to discriminate between the presence and absence of pain. These results suggest that the NCCPC-R can do just that. Analyses conducted here indicate that scores differed between observations during which children had pain and did not have pain. The number of behaviours and signs displayed also differed significantly between times they had pain and times they did not. Because the comparisons of the first pain episode were replicated with Pain Episode 2, the evidence that the NCCPC-R can discriminate pain is further strengthened. In addition, ROC curves indicated that a total score of 7 on the NCCPC-R was most sensitive and specific to pain. Again, this was replicated with a second set of pain observations, strengthening the finding.

This study had several limitations. First, children were observed in their home and not in structured settings with a standardised pain stimulus. Therefore, the pain they experienced varied in both quality and intensity and the results are not specific to any one type of pain experience. On the other hand, this demonstrates the validity and reliability of the NCCPC-R in a natural setting, which enhances its generalisability. Second, the same observer made the four observations for each child. Therefore inter-rater reliability could not be established. Finally, few observations were conducted during a medical procedure. Although this means the array of pain observed was likely a true sample of the types of daily pain experienced by children with severe cognitive impairments on a daily basis, based on indications in Chapter Five that medical pain represents less than 10% of

pain these children experience, these results cannot be generalised to situations in which these children experience pain in a medical setting. Further research with the NCCPC-R for pain due to medical interventions, such as injection and surgery, should be undertaken to determine if similar psychometric properties could be demonstrated in these specific situations. Finally, the concurrent validity of the NCCPC-R was assessed using numerical ratings provided by the same observers who completed the checklist. This may have lead to a higher correlation between the two measures than if different observers had completed the two measures. Future studies should examine the relation between NCCPC-R scores and pain intensity ratings provided by independent observers.

In summary, these results indicate the Non-communicating Children's Pain Checklist-Revised is a valid, reliable pain measurement tool for children with severe cognitive impairments. Although further research is needed before this tool should be used exclusively to detect pain in these children, the results indicate use of the NCCPC-R as a supplement to clinical judgement at this time may be of assistance to caregivers attempting to detect pain in children with severe cognitive impairments.

CHAPTER EIGHT: VALIDATION OF THE NON-COMMUNICATING CHILDREN'S PAIN CHECKLIST-POSTOPERATIVE VERSION[1]

Despite the escalation of research on paediatric pain in recent years (Guardiola et al., 1993), the literature suggests many children are still not appropriately medicated for postoperative pain. In 1983, Mather and Mackie (Mather & Mackie, 1983) reported 31% of the children they studied were not given any analgesic postoperatively. The same rate was reported by Kart et al. in 1996 (Kart, Rasmussen, Horn, & Wested, 1996). Further, a recent study (Gauthier, Finley, & McGrath, 1998) found 51% of 48 children surveyed postoperatively did not receive sufficient analgesics to keep their pain levels below their personal treatment threshold.

One reason for continued inadequate management of postoperative pain could be the lack of validated pain assessment tools. For example, Goddard and Pickup report that implementation of structured pain assessment contributed to significant improvements in the prescription and administration of analgesia for children (Goddard & Pickup, 1996). However, for some groups of children who are unable to provide valid self-report of pain, valid observational measures for postoperative pain are still needed. For example, pain assessment tools for pre-school children have only recently appeared (e.g. FLACC:

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[1] The data presented in this chapter is from Breau, L.M., Finley, G.A., McGrath, P.J. & Camfield, C.S. (2002).Validation of the Non-Communicating Children's Pain Checklist - Postoperative Version. Anesthesiology, 96 (3), 528-535.

(Merkel, Voepel-Lewis, Shayevitz, & Malviya, 1994); Pain Observation Scale for Young Children (POCIS): (Boelen-van der Loo, Scheffer, de Haan, & de Groot, 1999); COMFORT: (van Dijk et al., 2000).

Children with severe cognitive impairments are also unable to provide reliable self-reports of pain due to both cognitive and physical limitations. Yet, at present, no tools have been validated for their postoperative pain. Only one study has investigated the possible use of measures designed for children without impairments for the postoperative pain of children with cognitive impairments (Koh & Fanurik, 1997). They found that nurses could not use the Children's Hospital of Eastern Ontario Pain Scale (McGrath et al., 1985) effectively for measuring pain in children with borderline or severe cognitive impairments because of the children's idiosyncratic behaviours and physical limitations. Given that this instrument's utility for postoperative pain assessment in non-impaired children has been questioned (Beyer et al., 1990), this result is not surprising. Similarly, only one study has investigated the use of instruments designed for children without impairments for children with physical disabilities (Schade, Joyce, Gerkensmeyer, & Keck, 1996). They found questionable inter-rater reliability for the Nursing Assessment of Pain Intensity (Stevens, 1990) and the Postoperative Pain Score (Barrier, Attia, Mayer, Amiel-Tison, & Shnider, 1989) and a lack of discriminative ability for the Postoperative Pain Score for a subgroup of 20 infants with cerebral palsy. Only an adapted version of the Riley Infant Pain Scale (Joyce et al., 1994) showed satisfactory results for reliability and discriminative ability with the subgroup of physically impaired children.

The current study investigated whether a pain instrument designed specifically for children with severe cognitive impairments could be used to detect their postoperative pain. The Non-communicating Children's Pain Checklist (NCCPC) (Breau et al., 2000) is an observational measure of pain designed for children with severe cognitive impairments. Previous research suggests the NCCPC is valid in the home setting. It can differentiate between the behaviour of children with severe cognitive impairments and no speech during painful and calm periods, and is internally consistent, reliable over time, and sensitive to pain (Breau et al., 2000). The previous chapter indicates a newer version, the Non-communicating Children's Pain Checklist-Revised (NCCPC-R) also displays excellent psychometric properties when used in the home setting by caregivers. However, the pain of children with severe cognitive impairments at home is frequently due to injury and of a short-sharp nature. Therefore, the pain behaviour shown at home may differ from that shown due to surgery, especially pain experienced after the immediate postoperative period (McGrath, 1998a). Consequently, empirical evidence of the scale's sensitivity to pain due to surgery is needed. The validation of the NCCPC (Breau et al., 2000) and the validation of the NCCPC-R described in the previous chapter were also based entirely on observations by children's primary caregivers. Therefore, it was also felt that an investigation of the instrument when used by adults who are less familiar with the children having pain would provide further evidence of its clinical utility.

To accomplish these tasks, caregivers and researchers completed the Non-communicating Children's Pain Checklist-Postoperative Version (NCCPC-PV) for preoperative and postoperative observations of 25 children undergoing surgery at a

tertiary children's health centre. Because some children were under orders not to eat, either before or after surgery and because both anaesthesia (Kart et al., 1996) and analgesics (Cohen, 1993) can cause sleepiness and nausea, this version of the checklist does not include items from the Eating/Sleeping subscale.

Method

Participants

All caregivers who informed the Pediatric Pain Research Lab that their child was scheduled for surgery at the IWK Health Centre took part in the study, with the exception of one child for whom the surgeon did not provide consent. A total of 25 children from the cohort took part, 6 girls and 19 boys. No caregivers who contacted the Pediatric Pain Research Laboratory declined participation after they were informed of the protocol. The data from one child was omitted. This autistic toddler was extremely frantic before surgery because he had not eaten and was experiencing hunger pains. Thus, it was believed a preoperative observation of this child's behaviour would be invalid. Nine children suffered cognitive impairments due to dysmorphic or chromosomal syndromes, 3 due to traumatic brain injury, 5 due to asphyxia at birth. Of the remaining children, 2 were extremely premature and one each had a neurodegenerative syndrome and intrauterine acquired condition. The cause of impairment was unknown for one child, and information was unavailable for two. Twenty (83%) of the children lived with their family. The mothers of these children were aged 33 to 66 years ($M = 40.1$, $SD = 8.3$) and their fathers were aged 35 to 68 years ($M = 44.1$, $SD = 9.4$, $n = 15$). Further demographic characteristics of these families are listed in Table 8.1. The medical conditions and

physical impairments of the children are listed in Table 8.2 and their chronological and adaptive ages are shown in Table 8.3. At the time of this surgery, the children in this study had experienced from 0 to 18 previous surgeries ($M = 7.1$, $SD = 4.6$).

Representativeness of the Sample.

Analyses were conducted to determine if the children from the cohort who took part in this study differed from those who did not. Power analyses suggested there was .92 power to detect a large difference in means or proportions between the two groups. Independent t-tests revealed there was no difference in age ($t(99) = -1.1$, $p = .28$) or number of medications being taken ($t(99) = -1.1$, $p = .28$). Chi-square tests indicated, after corrections for multiple tests, the two groups of children did not differ in terms of gender ($\chi^2 (1) = 5.4$, $p = .02$), whether they had cerebral palsy ($\chi^2 (1) = 4.6$, $p = .03$), or seizures ($\chi^2 (1) = 1.2$, $p = .28$), or whether they lived with their family ($\chi^2 (1) = 0.7$, $p = .41$). Mann Whitney U tests revealed there were no differences in the children's level of diagnosed mental retardation ($U = 745$, $p = .68$), upper limb limitations ($U = 786$, $p = .24$), lower limb limitations ($U = 749.5$, $p = .15$), or how frequently they required medical monitoring ($U = 815$, $p = .36$). There was also no difference in their number of previous day surgeries (1-3, 4-6, 7-10, >10; $U = 858$, $p = .57$) or hospitalisations including surgeries (1-3, 4-6, 7-10, >10; $U = 904.5$, $p = .87$). Finally, for those children who lived with their families, chi-square tests revealed that there was no significant difference in the caregivers' marital status ([$\chi^2 (4) = 3.1$, $p = .54$], t-tests indicated no difference in the age of mothers ($t(87) = -1.0$, $p = .32$) or fathers ($t(75) = -1.8$, $p = .08$), and Mann Whitney tests indicated no significant differences in income ($U = 541.0$, $p = .20$), number of

children in the family (U = 561.0, p = .19), or the education achieved by mothers (U = 642.50, p = .77) or fathers (U = 433.5, p = .82). Thus, the results of this sub-sample of children can be generalised to the full cohort.

Medical Procedures the Children Experienced

The types of procedures the children underwent are listed in Table 8.4 The surgeries lasted from 12 to 225 minutes (M = 74.3, SD = 64.9) and the children stayed in the recovery room from 0 to 325 minutes (M = 107.9, SD =69.7). Eight children (33%) were given intravenous opioids during surgery and 11 (46%) were admitted to hospital after the surgery.

Measures

The Non-communicating Children's Pain Checklist-Postoperative Version

The Non-communicating Children's Pain Checklist-Postoperative Version was used in this study (Appendix D). This is identical to the Non-communicating Children's Pain Checklist-Revised, except that it does not include the three items from the Eating/Sleeping subscale. Total scores are derived by summing the scores for each of the 27 items (Not at All = 0, Just a Little = 1, Fairly Often =2, Very Often =3). Total scores may range from 0 to 81.

Visual Analogue Scales of Pain

Visual Analogue Scales of Pain (VAS's) were also used to rate the intensity of children's pain during the observation periods (Appendix D). A 100-millimetre line was anchored at the left by the phrase "no pain at all" and at the right by "worst pain ever".

Surgical Information Sheet

A researcher completed this after the child was released from hospital (Appendix D). It was used to abstract information regarding the surgical procedures the child experienced.

Hospitalisation Information Sheet

A researcher completed this after the child was released from hospital (Appendix D). It was used to abstract information regarding the child's stay in hospital after surgery.

Procedure

Caregivers of children in the cohort contacted a researcher if their child was scheduled to have surgery. A copy of an informed consent form was mailed to the caregiver (Appendix D) and a telephone call was placed to them several days later to ascertain whether they had questions regarding what was being asked of them. If they agreed to take part, they were asked to return the consent form by mail. Verbal agreement from the surgeon scheduled to perform the surgery was then obtained. Because this study spanned two years, consent forms were updated based on requirements from the IWK Health Centre Research Ethics Board.

On the day of surgery, a caregiver and a researcher observed each child for the same 10 minutes a) approximately 30 minutes before surgery and b) 30 to 60 minutes after the child left the recovery room. They completed NCCPC-PV for each 10 minute period indicating how frequently each behaviour or sign had occurred during the observation (Not at All = 0, Just a Little = 1, Fairly Often =2, Very Often =3). They also made a visual analogue scale (VAS) rating of the child's pain intensity for the same

observation period. Caregivers were told not to discuss their observations or ratings with the researcher also conducting an observation. A nurse also completed a VAS of the child's pain intensity during each observation. Time constraints precluded having the nurse complete the NCCPC-PV. After the child was released from the hospital, a researcher abstracted information regarding the surgery and the child's stay in hospital using the Surgery Information Sheet and the Hospitalisation Information Sheet.

Statistical Analysis

Missing Data.

Scores on the NCCPC-PV by a researcher after surgery were not available for one child. Two additional item scores were missing from a researcher after surgery. These were replaced by the median response for the applicable item. VAS ratings were also missing from one caregiver before surgery and one researcher after surgery. These were replaced by the mean score for that item at that observation time (pre-operative, postoperative). VAS ratings from nurses were missing for 6 children before surgery and 4 children after surgery. These were replaced by the mean for the remaining children at that observation time (pre-operative, postoperative), and analyses were conducted with and without the replaced data. The percentage of children who displayed each item of the NCCPC-PV at least "a little" before and after surgery according to caregivers and researchers is shown in Table 8.5.

Examination of VAS Ratings

The same nurse did not make pre-operative and post-operative VAS ratings for 14 children. Thus, a multiple regression was performed to determine whether this affected

postoperative VAS pain ratings before further tests were conducted. The R of .37 indicated having the same or a different nurse make postoperative VAS pain ratings accounted less than 14% of the variance in scores ($F(1,22) = 3.48$, $p = .08$). Thus, no adjustments were made. The adjusted R^2 indicated having the same or a different nurse make postoperative VAS pain ratings accounted for only 10% of the variance in those pain ratings.

To investigate the relation among the 100-mm VAS pain ratings of the three observers (caregiver, researcher, and nurse), Pearson Correlations were conducted. Power analyses indicated there was .77 power to detect a large size correlation. Matched sample t-tests were also used to determine whether VAS ratings differed from before to after surgery. Power calculations indicated there was .96 power to detect a large difference in means.

Internal and Inter-rater Reliability of the NCCPC-PV

The internal reliability of the NCCPC-PV was determined using Cronbach's alpha. Two-way mixed effects intraclass correlation coefficients for consistency (Shrout & Fleiss, 1979), which correct for chance agreement, were used to assess inter-rater agreement.

Validity of the NCCPC-PV

The validity of the NCCPC-PV was examined in several ways. First, concurrent validity was assessed by determining the relation between VAS ratings and NCCPC-PV total scores using Pearson correlations. Power for these exceeded .80. Second, change in NCCPC-PV scores over time was assessed. Total NCCPC-PV scores before and after

surgery were compared using matched sample t-tests. Power was .96 to detect a large size difference. Comparison of subscale scores on the NCCPC-PV from before and after surgery was conducted with a Repeated Measures Analysis of Variance (ANOVA). Because sphericity assumptions were not met, Wilks' Lambda was used to test significance. In addition, mean subscale scores were used because total possible scores on the six subscales differ and determining whether the change from before to after surgery differed by subscale was of interest. Power exceeded .80 to detect large size main effects. Third, caregivers' VAS ratings of the children's pain were used to class the children as having experienced mild or moderate severe/pain. Independent t-tests were then used to compare the total NCCPC-PV scores of the two groups. Power was .96 to detect a large size difference. Caregivers' VAS ratings were used to group the children based on evidence their ratings are typically most related to children's self-report (Tarbell, Cohen, & Marsh, 1992; Schneider & LoBiondo Wood, 1992). Children who were assigned a score of 30 or greater were classed as experiencing moderate pain or greater (Collins, Moore, & McQuay, 1997). The average VAS score of the 16 children with mild pain was 11.1 (SD = 10.1) and for the 8 with moderate/severe pain it was 53.5 (SD = 17.3).

Sensitivity and Specificity

Receiver Operator Characteristic (ROC) Curves, based on the bi-negative exponential assumption, were used to determine cut-off points for caregivers and researchers that offered the best combination of sensitivity and specificity. Caregivers' VAS scores were used to define groups of children with mild versus moderate/severe pain for both ROC curves so that the cut-off value found for each observer would be

relative to the same VAS score. This was also based on evidence that caregivers' VAS ratings are typically most related to children's self-report (Tarbell et al., 1992; Schneider et al., 1992).

NCCPC-PV Scores as a Function of the Characteristics of Children and their Surgeries

Children's pain scores should not vary with their personal characteristics such as gender, as there is no evidence of systematic variation of self-report ratings of postoperative pain in children based on these. Only age has been found to be inconsistently associated with children's postoperative pain, with older children reporting less pain in some studies (Palermo & Drotar, 1996), while others have reported no difference (Chambers, Reid, McGrath, & Finley, 1996). Similarly, there is no evidence that pain varies reliably with the type of surgical procedure performed. For example, studies have found no relation between surgery severity ratings, based on several aspects of the procedures performed (Palermo et al., 1996), or types of surgery (orthopaedic, ear/nose/throat, abdominal, genitourological) (Kart et al., 1996). Because there is no accepted classification of surgery severity, length of surgery, length of time in the recovery room, opioid administration intraoperatively and admission to hospital were used as markers for surgery severity to determine if NCCPC-PV scores differed. Before doing this, tests were conducted to determine if analgesic administration or VAS ratings varied due to these factors. A categorical regression, using optimal scaling techniques, was used to predict analgesic administration (none, non-opioid, opioid) after surgery (Van de Geer, 1993a; Van de Geer, 1993b) with length of surgery, length of time in the recovery room, opioid administration intraoperatively and admission to hospital included

as predictors. Multiple linear regressions were used to predict VAS ratings with these factors. Independent t-tests were then used to examine the effect of dichotomous demographic and surgical factors on children's total NCCPC-PV scores and Pearson correlations were used to examine the relation between continuous and ordinal demographic and surgical characteristics on NCCPC-PV scores. Power exceeded .80 for these tests.

Predicting Nurses' VAS Ratings

To explore how nurses assessed pain, a multiple regression was conducted on their postoperative VAS ratings. Because nurses' VAS ratings appeared to be most related to researchers' ratings and NCCPC-PV scores, researchers' subscale scores on the NCCPC-PV were entered as predictors.

Results

Visual Analogue Scale Pain Ratings

Change over Time

Matched sample t-tests revealed caregivers' and researchers' VAS pain ratings (Table 8.6) were significantly higher after surgery ($t(23) = -3.885$, $p = .001$ and $t(23) = -5.52$, $p < .001$, respectively), while nurses' VAS pain ratings were not significantly higher after surgery ($t(23) = -1.46$, $p = .16$). However, a second matched sample t-test of nurses' VAS ratings without replacement of missing values indicated nurses' ratings did differ marginally ($t(15) = -2.07$, $p = .06$). The mean scores for all observers are listed in Table 8.6.

Relations among VAS Ratings

A set of Pearson correlations was conducted to examine the relations among VAS ratings by caregivers, researchers and nurses before and after surgery (Table 8.7). They indicated caregivers' VAS pain ratings were significantly related to researchers' ratings before and after surgery. In addition, nurses' ratings before surgery were related significantly to both caregiver and researcher ratings. However, after surgery, nurses' ratings were not significantly related to caregivers' ratings but were related to researchers', although this was not significant after corrections for multiple tests. A re-analysis, using nurses' ratings without replacing missing data did not change the significance of the relationship between nurses' and caregivers' VAS ratings after surgery ($r = .09$, $p = .71$, $n = 20$) but the correlation between nurses' and researchers' postoperative VAS ratings achieved significance ($r = .73$, $p < .001$, $n = 20$).

NCCPC-PV Scores

Internal Reliability

To examine the internal reliability of the NCCPC-PV, Cronbach's alpha was conducted for both caregiver and researcher responses to the 27 items after surgery. These indicated caregivers' scores on the items had excellent internal consistency ($\alpha = .91$), and that researchers' had satisfactory internal consistency ($\alpha = .71$). The proportion of children displaying each behaviour before and after surgery is depicted in Table 8.5.

Inter-rater Reliability

To assess the relation between scores provided by caregivers and researchers, the Intraclass Correlation Coefficient (ICC) was computed for subscale and total scores after

surgery. These were .77 for the Vocal subscale, .48 for Social, .81 for Facial, .61 for Activity, .45 for Body and Limb and .63 for Physiological Signs. The ICC for total scores were .82 prior to surgery and .78 after surgery. Because of the small sample size, the ICC for individual items was not computed.

Change in NCCPC-PV Scores

A set of matched-sample t-tests was conducted to investigate the change in caregiver and researcher total NCCPC-PV scores from before to after surgery. These revealed both caregiver ($t(23) = -3.36$, $p = .003$) and researcher scores ($t(23) = -3.72$, $p = .01$) were significantly greater after surgery. Mean total NCCPC-PV scores for caregivers and researchers are depicted in Table 8.6.

Caregiver and researcher subscale total scores are depicted in Table 8.8. To examine the change from before to after surgery and the effect of observer on subscale scores, a 2 (Time) X 2 (Person) X 6 (Subscale) Repeated Measures ANOVA was conducted on mean subscale scores for before and after surgery. Wilks' Lambda indicated there was a significant effect for Time and for Subscale. No other effects or interactions were significant (Table 8.9). Thus, scores were higher after surgery than before, regardless of observer, and scores differed among subscales, regardless of time or observer.

Convergent Validity

A set of Pearson correlations was conducted to examine the relation between VAS pain ratings and NCCPC-PV total scores before and after surgery (Table 8.7). They indicated significant relationships between almost all caregiver and researchers measures.

One exception was the relation between researchers' VAS pain ratings and caregivers' NCCPC-PV scores before surgery. No correlations between nurses' VAS ratings and the VAS ratings or NCCPC-PV scores of caregivers and researchers were significant after corrections for multiple tests. A re-analysis of nurses' data without replacement of missing values did not change the significance of the relationships between nurses' VAS ratings and NCCPC-PV scores of caregivers and researchers before surgery ($r = .43$, $p = .08$, $n = 18$ and $r = .54$, $p = .02$, $n = 20$, respectively) or after surgery ($r = .17$, $p = .49$; $n = 18$ and $r = .49$, $p = .03$; $n = 20$, respectively)

Sensitivity and Specificity

To further examine the sensitivity of the NCCPC-PV, a comparison of the scores obtained by children experiencing pain that was moderate to severe and those experiencing milder pain was conducted. To do so, caregivers' VAS ratings were used as a measure of the severity of pain. Children who were assigned a score of 30 or greater on the 100 mm VAS were classed as experiencing moderate pain or greater based on research indicating VAS scores of this magnitude are considered moderate or greater by adults (Collins et al., 1997). Children with scores less than 30 were classed as experiencing mild pain.

Two independent t-tests were conducted to examine whether the total NCCPC-PV scores provided by caregivers and researchers for the 16 children with mild pain differed significantly from those of the 8 children with moderate to severe pain. The results of both were significant. Caregivers' and researchers' scores for children with mild pain were lower than their scores for children with moderate pain ($t(22) = -3.67$, $p = .001$ and

$t(22) = -2.39$, $p = .03$, respectively). The mean total NCCPC-PV scores of children with mild pain were 7.56 (SD = 4.86) for caregivers and 9.09 (SD = 6.09) for researchers, while the mean score for children with moderate to severe pain was 21.50 (SD = 13.85) for caregivers and 15.31 (SD = 5.82) for researchers.

A Receiver Operator Characteristic (ROC) curve was then constructed to determine which caregiver cut-off score provided the best sensitivity and specificity for separating mild from moderate to severe pain by total NCCPC-PV Scores. A score of 11 or greater provided the best combination of sensitivity (.88) and specificity (.81). Only 1 child with moderate to severe pain would be classed as having mild pain and only 3 children with mild pain would be classed as having moderate to severe pain. A second ROC curve was constructed to investigate which cut-off score was appropriate for researchers. The results suggested that a score of 11 or greater had the best sensitivity (.75) and specificity (.63). However, the results also suggested that researchers' were less adept at distinguishing those children whom caregivers rated as having moderate to severe pain with the NCCPC-PV. Two children who were rated as having moderate to severe pain based on caregivers' VAS ratings would not be classed as having pain based on researchers' NCCPC-PV scores and 6 children who had mild pain would be classed as having moderate to severe pain based on researchers' NCCPC-PV scores. Thus, based on both researchers' and caregivers' scores on the NCCPC-PV, a score of 11 or greater appears to be a good indication that a child is experiencing pain that is at least moderate in intensity.

Total NCCPC-PV Scores by Child Characteristics

To investigate whether children's postoperative scores on the NCCPC-PV differed due to their demographic characteristics, three independent t-tests were conducted. These indicated total caregivers' total NCCPC-PV score did not differ due to the child's gender ($t(22) = 0.24$, $p = .81$), whether they had cerebral palsy ($t(22) = -1.82$, $p = .08$), or whether they lived with their family or in a residential centre ($t(22) = -1.33$, $p = .27$). Similar results were found for researchers' postoperative NCCPC-PV scores. Total NCCPC-PV score did not differ due to the child's gender ($t(22) = -0.11$, $p = .92$), whether they had cerebral palsy ($t(22) = -0.74$, $p = .47$), or whether they lived with their family or in a residential centre ($t(22) = -0.46$, $p = .67$).

To investigate the relation between total scores on the NCCPC-PV and children's chronological and adaptive ages, Pearson correlations were generated (8.10) These indicated that only the children's Communication age equivalents from the VABS (Sparrow et al., 1984) were related to the total NCCPC-PV scores of researchers after corrections for multiple tests. To examine the relation between total NCCPC-PV scores and children's required frequency of medical monitoring and upper and lower limb use, Spearman correlations were calculated. These revealed no significant effects after correction for multiple tests. Together, these results indicate children's scores on the NCCPC-PV, by either caregivers or less familiar observers, are not related to their demographic characteristics.

Total NCCPC-PV Scores and Surgery Characteristics

Analgesic administration could not be predicted by any surgical factors. The nonsignificant multiple R for the categorical regression of .36 indicated all factors together predicted a nonsignificant proportion (13%) of the variation in analgesic administration ($F(4,19) = 0.7$, $p = .60$). Similarly, all factors together predicted less than 5% to 9% of the variation in caregivers' ($R = .21$; $F(4,19) = 0.3$, $p = .91$), researchers' ($R = .26$; $F(4,19) = 0.3$, $p = .84$) and nurses' ($R = .30$; $F(4,19) = 0.5$, $p = .80$) postoperative VAS ratings.

Independent t-tests revealed neither caregivers' nor researchers' total NCCPC-PV scores after surgery differed due to the whether the child received opioids intravenously during surgery ($t(22) = -1.3$, $p = .22$, and $t(22) = -0.1$, $p = .92$, respectively or were admitted to hospital after surgery ($t(22) = -0.1$, $p = .95$ and $t(22) = -1.0$, $p = .32$, respectively). Correlations between NCCPC-PV scores and length of surgery and length of time in the recovery room were also nonsignificant (Table 8.10).

Predicting Nurses' VAS Ratings

A multiple regression indicated that only the Facial subscale of researchers' NCCPC-PV significantly predicted nurses' VAS ratings ($R = .64$, $F(1,18) = 12.2$, $p = .003$).

Discussion

These results provide evidence the Non-communicating Children's Pain Checklist- Postoperative Version has good psychometric properties when used with children who have severe cognitive impairments. The NCCPC-PV was internally

consistent. Inter-rater reliability was good for total scores and very good for some subscales, such as the Facial and Vocal subscales. Lower inter-rater reliability for the Social subscale and Body and Limb subscale were most likely because caregivers were more familiar with the children's abilities to interact socially and to use their body and limbs voluntarily. Caregivers observed all items from these subscales more frequently than researchers, with the exception of "seeking comfort" and "difficult to distract". Therefore, caregivers may have noted subtle actions or attempts at actions that researchers did not.

NCCPC-PV total and subscale scores were significantly higher after surgery than before. It is important that there was no differences due to observer, suggesting familiarity with these children may not be required to assess their postoperative pain and that, with further refinement, this scale may be clinically useful for healthcare professionals. It is also important that all subscales changed, indicating all have some ability to discriminate pain due to surgery.

ROC curves indicated a score of 11 by caregivers and researchers correctly classified 88% and 75% of the children with moderate to severe pain, respectively. Although false positives and false negatives were greater for researchers the two children who were falsely classed as having mild pain had scores of 7 and 8. Of the six children who were false positives for researchers, all but one had scores of 17 or less. Thus, these children did not have scores that were extremely removed from their correctly classified peers within the possible NCCPC-PV range of 0 to 81. The fact that sensitivity was better than specificity for both caregiver and researcher scores is appropriate for a pain scale

designed to supplement clinical judgement and to alert healthcare professionals to the possibility that a child has moderate to severe pain. Future research should examine whether training or information about the children could improve the sensitivity and specificity of unfamiliar observers.

NCCPC-PV total scores were also significantly correlated with caregivers' and researchers' VAS ratings of the children's pain. Scores from each observer also correlated significantly with the VAS ratings of the other observer, adding to the evidence that familiarity with these children may not be necessary to assess their pain. However, nurses' VAS ratings did not change significantly from before to after surgery or correlate significantly with caregivers' and researchers' VAS ratings or NCCPC-PV scores. One reason for this could be that completing the NCCPC-PV provided caregivers and researchers with training in what to look for and that their VAS ratings were altered by this training. Due to time constraints, nurses were not given this opportunity. This coincides with previous research that indicates nurse and researcher pain ratings are more closely related when nurses use a standardised instrument rather than a global assessment (Colwell, Clark, & Perkins, 1996). This might also be explained by different nurses having rated the pain before and after surgery. However, the regression conducted indicated that only 10% of the variance in nurses' VAS pain ratings after surgery were related to whether nurses had also completed the pre-operative VAS pain rating. Thus, this latter explanation is unlikely.

One other explanation, explored in this study, is that nurses had a priori expectations of how the children would show pain based on their experience with

children without cognitive impairments and that these children did not show the reactions they were expecting. Previous studies indicate nurses use factors other than children's behaviour to make pain judgements, such as the type of surgery, time since surgery, and time since last analgesic administration (Caty, Tourigny, & Koren, 1995). Notably, these factors did not predict any pain ratings or analgesic administration in this study. There is also consistent evidence that nurses' put a great deal of weight on verbal and vocal signs of pain (Caty et al., 1995) (Hamers, Abu-Saad, van den Hout, Halfens, & Kester, 1996). Children in our study could not produce verbal signs, and they showed few vocal signs. According to researchers, only 50% of children displayed any vocal behaviour and according to caregivers only 67%. Furthermore, both reported that only 12% of children displayed any vocal behaviour "fairly often" or "very often". Thus, the lower ratings given by these nurses may well be because they were looking for vocal reactions to pain and few of the children showed these.

It is noteworthy that facial subscale had the highest inter-rater agreement and, that it was also the only subscale to predict nurses' VAS ratings. In studies of children without impairments, inter-rater reliability for facial expressions of pain have been relatively low (Merkel et al., 1994; van Dijk et al., 2000). However, studies of adults with dementia have found that facial reaction to pain is greater in those with impairments than those without (Porter et al., 1996; Hadjistavropoulos, LaChapelle, MacLeod, Snider, & Craig, 2000). As well, facial expression in response to noxious stimuli has been reported to be greater in children with developmental disorders than in children without (Soussignan, Schaal, Schmit, & Nadel, 1995) and children with language impairments may use

exaggerated facial expressions to convey their emotions (Trauner et al., 1993). Thus, it could be that children with severe cognitive impairments show more facial reaction to pain than children without impairments, or observers may be more sensitive to facial activity in people who cannot speak. Further research is needed to explore these possibilities.

The limitations of this study include the fact that the children here experienced a wide range of surgeries, from dental surgery to orthopaedic surgery. The small number of children did not permit comparison of NCCPC-PV or VAS scores on the basis of surgery type. However, the fact that significant pre-surgery to post-surgery results were found despite the wide range of surgeries included adds to the external validity of the results, suggesting that the NCCPC-PV may be clinically useful for pain from a variety of surgical procedures.

Another limitation of the current study was that scores from Eating/Sleeping subscale of the NCCPC-R were not included. It would have been preferable to include these items so that comparisons of scores achieved by children for postoperative pain and pain experienced in other situations could be made. However, use of the item "eating less" was not considered appropriate because many children were under orders not to eat either before or after surgery. Similarly, sedation is a common side effect of analgesics and general anaesthetics. Consequently, the two items regarding sleep included in the NCCPC-R were not included in these analyses as it was believed that any change in the children's sleep from before to after surgery might not be specific to pain.

The sample of children observed in this study was small. Thus, analyses of individual items were not possible. The small sample size also suggests that the results, especially those concerning cut-off scores for inferring the presence of moderate to severe pain, should be taken as preliminary. Validation of any pain assessment instrument requires repeated evidence of its validity and reliability across samples and observers.

Nonetheless, these results provide the first evidence that the postoperative pain of children who have severe cognitive impairments can be assessed using a formal pain assessment measure designed for this population. Even more importantly, adults who were unfamiliar with these children were able to detect signs of pain using the NCCPC-PV. The results also indicated that all adults who observed the children used facial expressions and that they may be the most consistently used cue the children presented. The next chapter describes a study that investigated the possibility that facial movement may be another alternative method of assessing children's pain after surgery.

CHAPTER NINE: USING THE CHILD FACIAL CODING SYSTEM TO DETECT THE POSTOPERATIVE PAIN OF CHILDREN WITH SEVERE COGNITIVE IMPAIRMENTS

Assessing the postoperative pain of children with severe cognitive impairments can be difficult and self-reports from these children are usually not possible. In a recent study examining the ability of children with cognitive impairments to self-report pain, Fanurik and her colleagues investigated the ability of 120 children scheduled for elective surgery to use a 0 to 5 pain rating scale (Fanurik et al., 1998). Only 47 of these children were capable of completing the tasks involved in the assessment of their self-report ability. Of the 47, aged 8 to 17 years, only 10 showed an understanding of the concepts required to rate pain on the numerical score, and all had a diagnosis of borderline to mild mental retardation. In fact, only one child with severe mental retardation was able to arrange five blocks in order of size, a preliminary task in the protocol. Further, 53% of the children could not complete the task despite the fact that their nurse predicted they would be capable of it. This study highlights the difficulty in assessing postoperative pain in children who have severe cognitive impairments and the need for alternate methods of assessing their postoperative pain.

This paper examines the effectiveness of the Child Facial Coding System (CFCS) (Chambers et al., 1996), an observational pain measure, for assessing postoperative pain in children with severe cognitive impairments. Facial expression has been recognised as a method for measuring internal states for some time. This began with studies of adults

(LeResche, 1982) using Ekman and Friesen's Facial Action Coding System (FACS; Ekman et al., 1978). The Neonatal Facial Coding System (NFCS; Grunau & Craig, 1987) was developed from a subset of facial actions that reflect pain in adults. The Child Facial Coding System (CFCS;Chambers et al., 1996), used here, was derived from both the FACS and the NFCS to detect pain in toddlers and school age children. It includes 13 facial actions and has been used in studies of acute procedural pain (Breau et al., 2001b) and post-operative pain (Gilbert et al., 1999).

There is reason to believe that facial activity may be a viable method of pain assessment for this special group of children. Even children with severe physical limitations may be able to display this behaviour. Furthermore, the study of postoperative pain in children with severe cognitive impairments described in the previous chapter found that facial response was the only significant predictor of nurses' pain ratings of children and had the highest inter-rater agreement of the subscales of the NCCPC-PV (Breau, Finley, McGrath, & Camfield, 2002).

In addition, several studies have suggested that the facial action of individuals with cognitive impairments, measured using the FACS (Ekman et al., 1978), does change with pain and can be accurately interpreted by observers. One study of response to aversive stimuli found that the facial action of children with pervasive developmental disorders was more accurately classed as reflecting pleasant, neutral or negative stimuli than the facial action of a control group of children (Soussignan et al., 1995). The authors suggest this result may reflect the fact that children with pervasive developmental disorders do not acquire the ability to moderate their facial response in accordance with

social norms. Although this may pose difficulties for these children in some situations, this deficit may make assessment of their pain through facial activity more feasible.

A recent study of institutionalised adults with mental retardation receiving intramuscular injections found pain-related facial activity did not vary due to ability to self-report pain and was significantly higher during insertion of the needle (LaChapelle, Hadjistavropoulos, & Craig, 1999). The intensity of the facial action displayed by these individuals was also the only significant predictor of observers' ratings of their pain. Another study of elderly individuals with and without dementia also suggests individuals with cognitive impairments may display greater facial responsiveness to pain than those without impairments (Porter et al., 1996). This group found elderly participants with dementia displayed greater facial action in response to venipuncture pain than a group of similar elderly individuals without dementia.

Together, these studies suggest facial activity may provide a useful method of assessing postoperative pain in children who have severe cognitive impairments. The purpose of this study was to examine whether the CFCS (Chambers et al., 1996), developed specifically for children, could detect the pain of a group of children with severe cognitive impairments immediately following surgery.

Methods

Participants

Twenty-six children from the cohort took part in this study, 8 girls (30.8%) and 18 boys (69.2%). Twenty-three of these children also took part in the study validated the

NCCPC-PV described in the previous chapter. One child who was scheduled for surgery did not take part because the surgeon did not consent.

Ten children suffered cognitive impairments due to dysmorphic or chromosomal syndromes, 3 due to traumatic brain injury, 6 due to asphyxia at birth, 2 due to extreme prematurity and 2 due to a neurodegenerative syndrome. One child suffered cognitive impairments due to an intrauterine acquired condition, while the cause of impairments was unknown for one child, and information was unavailable for another. Twenty-three (88.5%) of the children lived with their family. The mothers of these children were aged 33 to 66 years ($\underline{M}$ = 39.9, $\underline{SD}$ = 7.9, $\underline{n}$ = 23) and their fathers were aged 30 to 68 years ($\underline{M}$ = 43.0, $\underline{SD}$ = 9.2, $\underline{n}$ = 18). Further demographic characteristics of these families are listed in Table 9.1. The medical conditions and physical impairments of the children are listed in Table 9.2 and their chronological ages and adaptive age equivalents are shown in Table 9.3. At the time of this surgery, the children in this study had experienced from 0 to 18 previous surgeries ($\underline{M}$ = 7.2, $\underline{SD}$ = 4.7, $\underline{n}$ = 19).

Representativeness of the Sample

Analyses were conducted to determine if the children from the cohort who took part in this study differed from those who did not. Power analyses suggested there was .92 power to detect a large difference in means or proportions between the two groups. Independent t-tests indicated there was no difference in age ($\underline{t}(99) = -1.1$, $\underline{p} = .25$) or the number of medications being taken ($\underline{t}(99) = -1.5$, $\underline{p} = .13$). After corrections for multiple tests, chi-square tests indicated there was no relation between participation in the study and being male (χ^2 (1) = 3.1, $\underline{p} = .08$), not living with the child's family (χ^2 (1) = 0.0, $\underline{p}$ =

.95), having seizures (χ^2 (1) = 0.1, p = .81), or having cerebral palsy (χ^2 (1) = 4.5, p = .03). Mann-Whitney U tests also revealed no difference between the children who did and did not take part in terms of the frequency of their medical monitoring (U = 781.50, p = .11), their level of mental retardation (U = 652.00, p = .09), or ability to use their arms (U = 710.00, p = .03) or legs (U = 688.500, p = .02) after corrections for multiple tests. Finally, for those children who lived with their families, chi-square tests revealed that there was no significant difference in the caregivers' marital status ([χ^2 (4) = 2.3, p = .69]), t-tests indicated no difference in the age of mothers (t(87) = -0.9, p = .35) or fathers (t(75) = -1.3, p = .19), and Mann Whitney tests indicated no significant differences in income (U = 610.5, p = .36)], number of children in the family (U = 633.0, p = .22), or the education achieved by mothers (U = 716.0, p = .84) or fathers (U = 475.5, p = .63). Thus, this subsample did not differ significantly from the remainder of the cohort in relation to their demographic characteristics.

Medical Procedures the Children Experienced

The procedures the children underwent included: dental extractions (n = 6), G-button insertions and removals (n = 6), heelcord/tendon lengthening (n = 3), other orthopaedic surgery (n = 2), endoscopies and biopsies (n = 2), fundoplication (n = 2), subcutaneous venous access device insertion (n = 1), bilateral myringotomy tube insertion (n = 1), strabismus repair (n = 1) skin graft (n = 1), and mole removal (n = 1). Seven of the children had two procedures, with dental extractions being the most frequent procedure conducted in combination with others. Surgeries lasted from 8 minutes to 3 hours 45 minutes (M = 74 minutes, SD = 64) and length of stay in the recovery room

ranged from none to 5 hours 25 minutes (M = 1 hour 45 minutes, SD = 67 minutes). Nine children (35%) received opioids intraoperatively, three children (12%) were given non-opioid analgesics in the recovery room and six (23%) were given a combination of opioids and non-opioids in the recovery room. Thirteen (50%) were admitted to hospital.

Measures

The Child Facial Coding System (CFCS)

The Child Facial Coding System (Chambers et al., 1996) was used to code the discrete facial movements of the children from videotape. This system includes 13 facial actions (Appendix E). Ten facial actions are coded for intensity (0-2; absent, slightly present, distinctly/maximally present). Three facial actions (blink, flared nostril, and open lips) are coded as present or absent (0-1). The Time Segment Selection Sheet was used to record 10-second segments of the videotape to be coded (Appendix E). Coders used the CFCS Coding Sheet to record their coding (Appendix E).

Visual Analogue Scale of Pain (VAS-Pain)

A Visual Analogue Scale (VAS-Pain) was used to rate the intensity of children's pain from videotape. The 100-millimetre line was anchored at the left by the phrase "no pain at all" and at the right by "worst pain ever" (Appendix E). This measure has been used to visually discriminate between pain and absence of pain in non-verbal children with severe cognitive impairments (Mette et al., 1988).

Visual Analogue Scale of Sedation (VAS-Sedation)

A Visual Analogue Scale of Sedation (VAS-Sedation) was also used to rate the amount of children's sedation from videotape. The 100-millimetre line was anchored at the left by the phrase "no sedation at all" and at the right by "worst sedation ever"(Appendix E).

Surgical Information Sheet

A researcher completed this after the child was released from hospital (Appendix E). It was used to abstract information regarding the surgical procedures the child experienced.

Hospitalisation Information Sheet

A researcher completed this after the child was released from hospital (Appendix E). It was used to abstract information regarding the child's stay in hospital after surgery.

Procedure

Caregivers of children in the cohort contacted a researcher if their child was scheduled to have surgery. A copy of an informed consent form was mailed to the caregiver (Appendix E) and a telephone call was placed to them several days later to ascertain whether they had questions regarding what was being asked of them. If they agreed to take part, they were asked to return the consent form by mail. Verbal agreement from the surgeon scheduled to perform the surgery was then obtained. Because this study spanned two years, consent forms were updated based on requirements from the IWK Health Centre Research Ethics Board.

On the day of surgery, a researcher filmed the child in the recovery room for up to 60 minutes from the time they were deemed "reactive" by nursing staff. This entailed the child showing positive signs across five areas of functioning including respiration, energy, alertness, circulation and temperature. This occurred 15 minutes to 4 hours 5 minutes after surgery was completed (M = 1 hour, 12 minutes, SD = 47 minutes). One child was sent immediately from the operating room to the intensive care unit because of pre-existing medical conditions. Filming for that child was conducted there.

Filming continued for 60 minutes or until the child left the recovery room. Children spent from 50 minutes to 5 hours, 25 minutes in the recovery room, excluding the one child who went directly from the operating room to the intensive care unit (M = 2 hours, 4 minutes, SD = 1 hour, 9 minutes). Information regarding the length of surgery, administration of analgesics in the operating room and recovery room and length of stay in the recovery room was abstracted from children's charts after they were released from hospital using the Surgery Information Sheet and Hospitalisation Information Sheet.

Coding of Videotapes

Videotapes were divided into 10-minute segments using the Time Segment Selection Sheet. From each 10-minute each segment, the first 10-second interval in which the child's face was clearly visible was selected for coding. A trained researcher coded each segment for the 13 facial actions of the CFCS using the CFCS Coding Sheet. A second trained researcher coded 7 (27%) of the tapes to assess reliability. Each segment was then independently rated for pain intensity using the 100-mm VAS-Pain by a third researcher and by a nurse who had not coded the videotapes using CFCS and had not

been involved in filming the children. Several months later, the same nurse, and the second researcher independently coded each segment for sedation using the 100-mm VAS-Sedation.

Inter-rater reliability for CFCS ratings were computed with the formula recommended by Ekman & Freisen (Ekman et al., 1978). This provides a conservative index of agreement because it does not take into account agreement between observers on the absence of facial actions and regards disagreements in intensity values as errors. Reliability for the 13 facial actions ranged from to .65 to .97 (M = .80, SD = 8.8), as can be seen in Table 9.4. The inter-rater reliability of the two VAS-Pain ratings provided by the researcher and the nurse and their two VAS-Sedation ratings was computed using Pearson correlation coefficients. Power was .81 to detect a large size correlation. These are shown in Table 9.5. Overall, the analyses indicated good to excellent reliability for these measures.

Statistical Analysis

Missing Data

Twenty children were filmed for the full 50 minutes, but only 15 for the full 60 minutes. Thus, analyses were conducted on the first 50 minutes of film for each child. Three children had no film available during one of the five segments and three children had no film available during two of the segments, a total of 6.9% of the segments. This was primarily due to view of the child being blocked as staff provided care or as caregivers comforted the child. All missing data were replaced with the median value for each facial action for the remaining children during that second and that segment. This

amounted to replacement of 8% of data for segments two and four, 4% of segment three and 15% of segment 5. Missing VAS-Pain and VAS-Sedation scores for these segments were replaced with the mean rating for that segment by the same rater.

Statistical Procedures

Descriptive Statistics. The total number of seconds and percentage of seconds that each facial action was present during each segment was computed. The mean intensity for each of the 10 facial actions coded for intensity was also computed for each of the five segments. To compute facial action intensity summary scores for use in the categorical principle components analyses, the mean intensity of each facial action across all 50 seconds was computed and this was multiplied by the number of seconds in which that action was present. To equate the weighting of binary facial actions with that of facial actions receiving an intensity score (0, 1 or 2) for use in multivariate procedures, the summary scores for the binary facial actions were computed using the mean of the value for absent (0) and present (2).

Examination of VAS-Pain and VAS-Sedation ratings. To examine whether VAS-Pain or VAS-Sedation ratings changed over time, two repeated measures analyses of variance (ANOVA's) were conducted on the nurse's ratings for the five segments. These had power greater than .80 to detect a medium size effect. The nurse's ratings were used because they were highly correlated with the researchers' ratings, but it was believed these were most generalisable to other professionals in this setting.

Relation between facial action scores and VAS-Pain and VAS-Sedation ratings. Partial correlations were conducted between facial action total frequency scores and

mean intensity scores and the VAS-Pain ratings for each time segment. The effect of VAS-Sedation ratings was partialled out of these correlations. Power was .81 to detect large size correlations, without accounting for the effect of the partialling out of VAS-Sedation ratings. Partial correlations were conducted between facial action total frequency scores and mean intensity scores and the VAS-Sedation ratings for each time segment. The effect of VAS-Pain ratings was partialled out of these correlations. Power was .81 to detect large size correlations, without accounting for the effect of the partialling out of VAS-Pain ratings.

Change in facial action over time. To examine the pattern of facial action over the 5 segments coded, a 5 (segment) x 10 (mean facial action intensity) and a 5 (segment) x 13 (total facial action frequency) mixed measures analysis of covariance (ANCOVA) were conducted. The first examined the mean intensity of the 10 actions coded for intensity. The second examined the total frequency of all 13 facial actions across the five segments. To include VAS-Pain ratings as a between-subjects factor, children were grouped as having "high" or "low" pain through splitting their mean VAS-Pain scores at the median. This resulted in 13 children in the "high" pain group with a mean VAS-Pain score of 34.3 from the researcher (SD = 17.7) and 10.5 (SD = 4.1) from the nurse. The "low" pain group had a mean VAS-Pain score of 13.0 from the researcher (SD = 12.9) and 2.7 (SD = 2.4) from the nurse. Children's mean VAS-Sedation score from the two coders was included as a covariate. Power computations indicated there was just under .80 power to detect a medium main effect.

The statistical configuration of facial actions To investigate the organisation of the facial actions, their meaning, and their relation to child characteristics, categorical principal components analyses (PCA), using optimal scaling techniques, were used (Van de Geer, 1993b). The facial action summary scores for each segment were first standardised. This allowed use of the individual scores for each child's five segments while avoiding the confounding of time due to repeated measures (Bijleveld et al., 1998). Variable principal normalisation was used for both analyses. For the first categorical PCA, standardised facial action summary scores were entered. In addition, demographic variables were entered as supplementary variables to explore the relation between these and the facial action components. Thus, they were not active in the derivation of components, but their loading with the facial action components were computed. In order to validate the meaning of the facial action components, a second categorical PCA was conducted following a procedure recommended by Kline (Kline, 1994) and used in previous analyses of CFCS data (Breau et al., 2001b). This entailed entering the extracted components plus "marker" variables that were believed to reflect the meaning of the components. The component scores for each child for the facial action components extracted in the first categorical PCA were entered. Mean VAS-Pain and VAS-Sedation ratings for the 5 segments were also standardised and entered, as were surgery factors.

Results

Descriptive Statistics

The total frequency of the 13 facial actions and the mean intensity of the 10 facial actions that are not binary across the five segments coded are shown in Table 9.6. The

most frequent facial actions were those involving the mouth, such as open lips, and upper lip raise. Upper lip raise, vertical mouth stretch and horizontal mouth stretch were also the most intense, but were followed by eye squeeze and nasolabial furrow.

Table 9.7 lists the total frequency of all facial actions per segment and the mean intensity of all facial actions per segment. There was not much variation across segments.

Examination of VAS-Pain and VAS-Sedation Ratings.

A repeated measures ANOVA on the nurse's VAS-Pain ratings for the five segments resulted in a Wilks' lambda that indicated there was no significant change due to segment ($F(4,22) = 1.1$, $p = .38$). As can be seen in Table 9.7, VAS-pain ratings did not change over time. A second repeated measures ANOVA on the nurse's VAS-Sedation ratings for the five segments resulted in a Wilks' lambda that indicated VAS-Sedation ratings did change over the five time segments ($F(4, 22) = 3.4$, $p = .025$). As can be seen in Table 9.7, VAS-sedation ratings became lower over time.

Relation between Facial Action and VAS-Pain and VAS-Sedation Ratings

The partial correlations between facial action frequency and intensity for each segment and the VAS-Pain and VAS-Sedation ratings are listed in Table 9.7. Almost all correlations between facial action and VAS-Pain ratings were significant after corrections for multiple tests. The exception was the correlation between facial action frequency and VAS-Pain ratings in the first segment, and the correlations between facial action frequency and intensity and VAS-Pain ratings in the final segment. No correlations between facial action frequency or intensity and VAS-Sedation ratings were significant.

Change in Facial Action over Time.

A 2 (high/low pain) x 5 (segment) mixed measures analysis of covariance conducted on total frequency scores for the five segments revealed a nonsignificant main effect for Segment ($F(4,92) = 1.5$, $p = .21$), the covariate Sedation ($F(1,23) = 1.7$, $p = .21$), and the interaction between the two ($F(4,92) = 2.0$, $p = .11$). However, the main effect of Pain (high/low) was significant ($F(1,23) = 8.6$, $p = .007$) and it did not interact with Segment ($F(4,92) = 1.7$, $p = .16$). Children in the high VAS-Pain rating group displayed significantly more frequent facial action across segments ($M = 52.9$, $SD = 22.8$) than children in the low VAS-Pain group ($M = 33.7$, $SD = 14.3$). In summary, facial action did not vary across the segments, nor was it affected by sedation, but it was affected by pain.

A second analysis on mean intensity scores for the 10 facial actions coded for intensity for the five segments revealed a nonsignificant main effect for Segment ($F(4,92) = 0.3$, $p = .86$), the covariate Sedation ($F(1,23) = 0.4$, $p = .54$), and the interaction between the two ($F(4,92) = 0.4$, $p = .76$). However, the main effect of Pain (high/low) was again significant ($F(1,23) = 10.5$, $p = .004$) and it did not interact with Segment ($F(4,92) = 0.9$, $p = .45$). Thus a similar pattern was found as for frequency. Facial action intensity was greater for children in the high VAS-Pain group ($M = 0.64$, $SD = 0.37$) than children in the low VAS-Pain group ($M = 0.29$, $SD = 0.15$), but it did not vary by segment or due to sedation.

The Statistical Configuration of the Facial Actions

The first categorical PCA examined the organisation of the facial action summary scores. The best solution consisted of three independent components, accounting for 67% of the variance. The eigenvalues of the three components, variance accounted for, and loadings by the 13 facial actions are shown in Table 9.8. The first component appeared to reflect pain. Facial actions that loaded onto it most were those previously associated with a "pain face" in children (Breau et al., 2001b), such as brow lower, nose wrinkle, nasolabial furrow, cheek raiser, horizontal mouth stretch and flared nostril. Eye squeeze and vertical mouth stretch, previously associated with an expectation of pain by children (Breau et al., 2001b), also loaded onto this component to some extent. The second component was loaded onto most highly by open lips, followed by vertical mouth stretch and upper lip raise. This component appeared to reflect an open mouth and it was hypothesised it might pertain to children who had had dental procedures. The third component was loaded onto most highly by squint and blink and appeared to reflect eye movements only. It was hypothesised that this component might reflect awakening.

The relation between demographic characteristics and facial action components.

Children's demographic characteristics were also entered into this analysis as supplementary variables, to explore the relation between their characteristics and the facial action components. The loadings of these variables with the three components are shown in Table 9.9. As can be seen there, few child characteristics were strongly related to the facial action components. Children's age, adaptive skills age equivalents and gender were not related to the facial action components. There was a tendency for

component 1, reflecting the "pain face" to be associated with greater upper limb impairment, more severe mental retardation and less ability to eat, suggesting children with more severe physical and cognitive impairments displayed more of these facial actions.

Validation of the facial action components. To determine the meaning of the facial action components, a second categorical PCA was conducted on children's scores for the three facial action components and the characteristics of their surgeries. To ascertain whether Facial Action Component 1 reflected pain, children's VAS-Pain ratings by the nurse were entered, as were the intravenous opioids given intraoperatively and analgesics given in the recovery room. To determine if the Facial Action Component 2 reflected having had dental procedures conducted, a variable was entered to indicate whether the child had had any dental procedures performed. To determine if the Facial Action Component 3 reflected awakening, VAS-Sedation ratings by the nurse were entered. In addition, length of time in surgery and length of time in the recovery room were entered.

The best solution involved three components accounting for 59.8% of the variance. The eigenvalues and loadings of the facial action components and surgical variables onto these validation components are shown in Table 9.10. As these loadings indicate, Validation Component 1 is loaded onto most highly by the presence of dental surgery and long length of surgery. The facial action component that loads highest onto this validation component is Facial Action Component 2, consisting of facial actions representing an opening of the mouth. Facial Action Component 3, reflecting squinting

and blinking, also loads fairly highly onto this validation component, but in the negative direction. Thus, this set of facial actions appears to represent the open mouth of children who had had dental procedures performed and to reflect to some extent their grogginess after lengthy surgeries.

Facial Action Component 1 and the VAS-Pain ratings loaded onto validation Component 2 very highly, supporting the hypothesis that these facial actions reflect a "pain face". Analgesic administration in the recovery room also loaded with this component, suggesting the physicians who were caring for children with this facial action believed they had pain. The VAS-Sedation ratings also loaded negatively onto this component, suggesting these facial actions were most prevalent in children who were not experiencing continued sedation due to anaesthetics.

Validation Component 3 was loaded onto most highly be the VAS-Sedation ratings, followed by intravenous opioids provided intraoperatively. Two facial action components loaded negatively onto this validation component, Facial Action Component 2, reflecting the open mouth, and Facial Action Component 3, reflecting blinking and squinting. It is likely that these loadings represent the fact that children who were more groggy produced less facial action than their counterparts who were not still experiencing the residual effects of anaesthesia.

In summary, this analysis suggests that the facial actions contained in Facial Action Component 1 do reflect a "pain face". The facial actions contained in Facial Action Component 2, on the other hand, seem to relate to having experienced oral surgery. Finally, the facial actions contained in Facial Action Component 3, which loaded

in the opposite direction as sedation on all three validation components appears to reflect the absence of sedation.

Discussion

The purpose of this study was to determine if the CFCS (Chambers et al., 1996) could detect pain in children with severe cognitive impairments. The results indicate the overall frequency of facial action and intensity of facial action was greater in children rated as having higher pain by observers. However, facial action did not change over the course of the 50 minutes of time in the recovery room examined here. This is similar to the findings of Gilbert et al. (Gilbert et al., 1999) who also reported facial action did not change over time for children without impairments who had experienced surgery.

The analyses also indicate only a subset of the CFCS facial actions may reflect pain. Of the three components extracted from the CFCS summary scores, only one was strongly related to the nurse's VAS-Pain ratings and administration of analgesics. The facial actions that loaded most were highly onto this component are similar to the "pain face" reported in children during immunisation (Breau et al., 2001b), such as brow lower, nose wrinkle, nasolabial furrow, cheek raiser, horizontal mouth stretch and flared nostril. To a lesser extent, facial actions associated with an expectation of pain by children (Breau et al., 2001b), such as eye squeeze and vertical mouth stretch, also loaded onto this component. The similarity in the structure of the components found in this study and those observed during immunisation suggest that a core set of facial actions may be shown by children during various types of pain, a finding reported for adults (Prkachin, 1992), and that the set of facial actions shown by children with severe cognitive

impairments may not differ substantially from those shown by children without impairments. However, direct comparison of children with and without impairments undergoing the same type of pain would be needed to confirm this.

The results also indicate, however, that some facial actions contained in the CFCS (Chambers et al., 1996) reflected aspects of the children's surgical experience other than pain. The second component extracted was most closely related to whether children had had dental procedures performed. This set of facial actions appeared to represent a wide-open mouth. Having had a lengthy surgery and sedation were also associated with this set of facial actions. Thus, this set of facial actions appears to be specific to dental surgery. A pots-hoc comparison of the duration of surgeries indicated children who had dental procedures completed were in the operating room longer than children who did not ($\underline{M}$ = 2 hours, $\underline{SD}$ = 1 hour, 8 minutes and $\underline{M}$ = 39 minutes, $\underline{SD}$ = 30 minutes, respectively; $\underline{t}(24) = 3.7$, $\underline{p} = .003$), supporting the relation between length of surgery and having had dental procedures performed.

The final component represented blinking and squinting. It was hypothesised this component reflected awakening. Upon awakening in the recovery room, children are met by bright lights and an array of sounds, scenes and people. This was supported by the fact that the VAS sedation ratings provided by the nurse loaded negatively with this set of facial actions.

These results differ from those reported by Gilbert et al. (Gilbert et al., 1999). Whereas they report one component, three were found here. There are several possible reasons for this. First, in conducting the PCA, they do not report rotating their solution,

which could lead to extraction of a single component reflecting within-subject similarity (Kline, 1994). Second, it is not clear whether they used a 0-2 coding for computing summary scores for the three facial actions that are binary. Failing to do so would underweight these facial actions relative to the other facial actions coded for intensity and could alter the solution.

It is also possible the differences in results are due to the nature of the surgeries the children in the two studies underwent. The children who were observed for Gilbert et al.'s (Gilbert et al., 1999) study appear to have had their pain well managed. This is suggested by the authors as one reason that they did not find any relation between facial action and pain medication administered. However, they do not report the VAS ratings of pain conducted from videotape, so this cannot be confirmed. It is also notable that only 17% of the children in that study received no analgesic either intraoperatively or in the recovery room. In sharp contrast, 65% of the children in this study received no intravenous opioids intraoperatively and 65% received no opioid or non-opioid analgesics in the recovery room. Although the number of children receiving general anaesthetics that could have had analgesic properties was not recorded in either study, there is no reason to belief the numbers were not comparable. Thus, it is probable that the children in Gilbert's study may have experienced less pain due to the higher frequency of analgesic administration. In this current study analgesic administration was also related to the "pain face" found, something Gilbert et al. did not find. Thus, the differences could reflect the fact that the children in this study experienced more pain than the children in Gilbert et al.'s study.

The lower correlations among VAS pain ratings and facial actions scores reported by Gilbert et al. (Gilbert et al., 1999) than the ones found here may also reflect a lower pain level for those children than the ones observed here, or a reduced range of scores. The correlation reported by them ranged from .15 to .73, while those of the current study ranged from .47 to .81 with the effects of sedation accounted for. Only four facial actions were present more than 10% of the time in their sample, and the highest frequency for a facial action they report related to pain was less than 14%. In contrast, only two facial actions in this study, blink and lip corner puller, were present less than 10% of the time. The remaining facial actions were coded as present 14% to 82% of the time, with the six loading .80 or greater onto the "pain face" component present 14% to 68% of the time.

In summary, it is likely that the differences found between the results reported by Gilbert et al. and those found here reflect both statistical factors and the fact that the children in this study may have had more intense pain during the observation, leading to higher pain ratings and more facial actions. However, it is also possible that the children in this study displayed more facial activity in response to pain because of their severe cognitive impairments. This would coincide with the findings of Soussignan and his colleagues (Soussignan et al., 1995), who found children with pervasive developmental disorders displayed more facial reaction to aversive stimuli than children without impairments. This would also coincide with the results of Porter and her colleagues (Porter et al., 1996), who found that elderly individuals with dementia displayed greater facial reaction to a venipuncture than elders without dementia. Future research with

children who do and do not have cognitive impairments who are experiencing the same pain stimulus is needed to confirm this possibility.

This study had several limitations. The children who participated underwent a wide variety of procedures and it was not possible to examine each type of procedure separately. However the results did indicate that facial action might reflect some aspects of the procedures experienced, such as their duration and whether oral procedures were performed. This should be investigated further. Second, samples of the facial action displayed by the children before they underwent surgery were not collected for comparison with their post-surgical facial action. This is needed to provide confirmation that factors related to surgery, other than pain, are not responsible for the results found here. Although the facial action found here did correlate highly with pain ratings provided by a trained nurse and did not correlate with sedation ratings, it is possible that unknown factors were at play.

Nevertheless, the results suggest that facial action may be a viable method of assessing the pain of children with severe cognitive impairments after surgery. A subset of facial actions was found that is similar to those seen in children without impairments during immunisation and after surgery. The facial action of the children in this study also did not vary substantially due to their physical limitations. The Child Facial Coding System (Chambers et al., 1996) is a research tool at this point, and clinical use is not feasible. Coding must be conducted from videotape and coding of one minute of videotape can take up to one hour. However, these results suggest that research aimed at refining the CFCS into a clinically feasible measure, perhaps through development of

aggregate facial expressions based on derivation of components, is warranted and may assist those who manage the postoperative pain of this vulnerable group of children.

CHAPTER TEN: DISCUSSION

The preceding chapters describe research that began as an attempt to measure the pain of children who have severe cognitive impairments. This expanded to a set of studies investigating many aspects of the children's pain, several of which are described here. Two studies included here address the most basic aspects of children's pain, its nature and its impact. From there, three studies explore methods of measuring that pain. Together, these studies represent the foundation of the research program.

When Eland and Anderson's seminal paper on the differences in pain management afforded children and adults was published, paediatric pain began to emerge as an independent field of enquiry (Eland et al., 1977). Since that time, few studies have been specifically targeted at the pain of children with severe cognitive impairments. In essence, pain researchers have largely overlooked the predicament of this group.

This research represents a first step towards rectifying this situation. The results will form a foundation for further research and may prompt greater awareness of this group's pain in clinical settings. The following sections highlight the central findings reported here.

Summary of Major Findings

The Nature and Impact of Pain

Two studies investigated these basic aspects of pain in the cohort. Both were based on four weeks of information over a one-year period.

The Incidence and Characteristics of Pain in Children with Cognitive Impairments

This is the largest and most comprehensive study documenting the incidence and characteristics of pain experienced by children with severe cognitive impairments. The results indicate pain is a frequent experience for these children, with 35% to 52% of children experiencing pain each week. The pain experienced by the children was also significant in nature. The average intensity for all episodes was 5.7 on a 10-point scale. Pain also lasted long when it occurred. Children who had pain at least once over the four weeks examined spent, on average, 19 to 29 hours per week in pain.

The results of this study also provide information regarding the causes of pain for children with severe cognitive impairments. Although it has been suggested that children with severe cognitive impairments may suffer a great deal of pain due to medical procedures, such as surgery and injections (McGrath et al., 1998), only 8% of the episodes were related to medical pain over the four weeks of the study. Even more surprisingly, most of this was not due to surgery, or to injection, but was due to ongoing problems related to leakage at feeding tube sites. The greatest percentage of episodes (82%) was due to chronic conditions or illnesses, while only 15% of episodes were due to an accident.

In summary, this study indicates the majority of children with severe cognitive impairments experience pain weekly, that that pain is severe, and that a large part of that pain is due to illness and chronic conditions. Taken together, these results suggest that pain is a frequent and substantial problem for children with severe cognitive impairments.

The Functional Impact of Pain

The results of this second study indicate pain hinders the development of children with severe cognitive impairments. Increasing time in pain significantly reduced overall development of adaptive abilities over the year, and this was especially true for adaptive skills in the areas of Communication, Daily Living Skills and Motor Skills, as measured by the Vineland Adaptive Behavior Scales (Sparrow et al., 1984). These differences were significant even when medical stability was controlled for; indicating pain had effects independent of the effects of illness or health conditions.

Pain had its greatest effect on communication in the area of receptive abilities. This result suggests pain may affect attention or other areas of cognitive functioning, as understanding language does not require physical abilities or development. This is not surprising in light of experimental studies with adults indicating pain interferes with cognitive functioning (Eccleston & Crombez, 1999) and that this interference does not disappear with repeated pain (Crombez, Eccleston, Baeyens, & Eelen, 1997). However, this is the first indication that pain may actually slow cognitive development, suggesting it may have more than just transient effects in developing minds.

Within the Motor Skills domain, time in pain most affected fine motor skills. This may be because these skills require focussed attention and practice. It may also be because gains in gross motor skills are less possible, even for children who experienced little or no pain, and this is a simple reflection of the severity of the children's physical limitations. In essence, a ceiling effect on gross motor development may have prevented

detection of the effects of pain in this area. More comprehensive assessment of motor development may be necessary to do so.

Within the domain of Daily Living Skills, personal care skills were most attenuated by pain. This may also reflect a ceiling effect, as even the most basic skills within the domestic and community living areas of functioning, such as putting away possessions and helping with chores, require a level of independence and motor ability these children do not possess.

Only development within the Socialisation domain was unaffected by pain. A look at the development in this area suggests only the children who experienced the greatest amount of pain did not make gains in this area. Thus, it may be that social functioning is more robust than other areas of functioning, and was decreased by only the most unremitting pain. The children who displayed no gains experienced an average of 6 days in pain over the four weeks, almost one quarter of the time surveyed. It is also possible that the social behaviours documented were more easily adapted to pain so that they could still be performed, but in an altered fashion. Further research should examine why pain had less impact on social functioning for these children, and whether the factor that underlies this robustness could provide insight into how to reduce impacts in other areas.

These results suggest pain may hinder development in children who are already burdened with severe cognitive impairments. The areas of functioning examined are vital to everyday functioning. The fact that the effect of pain was evident over only one year, and based on only a small sample of pain experienced over that year, suggests that over

the course of childhood, pain may have serious consequences for children's development in these areas and lessen the chance they will reach their fullest potential.

Measuring Pain

Three studies here examined possible methods of measuring the pain of this special group of children. Two focussed on validation of revised versions of the Non-communicating Children's Pain Checklist (Breau et al., 2000). The third investigated the potential of the Child Facial Coding System (Chambers et al., 1996) to detect pain in children after surgery.

The Psychometric Properties of the Non-communicating Children's Pain Checklist-Revised

This study provided evidence that the pain of children with severe cognitive impairments can be measured validly and reliably in a home setting. The results show the Non-communicating Children's Pain Checklist-Revised has excellent psychometric properties. It exhibited excellent internal consistency across two separate episodes of pain observed by caregivers and total scores were also significantly correlated with caregivers' numerical ratings of their children's pain intensity. The scores obtained by these children were also stable over time and the number of items displayed by the children did not differ across the two pain episodes or the two episodes without pain. The latter is an important finding because it suggests their typical daily behaviour does not resemble their pain behaviour. Finally, the vast majority of children had scores that were stable from one time to another; suggesting individual children show consistent patterns of pain behaviour.

This study confirms the findings of two earlier studies with the original Non-communicating Children's Pain Checklist (Breau et al., 2000; Breau et al., 2001a) that indicated children with severe cognitive impairments do show measurable reactions to pain. This also adds the cumulating evidence that caregivers can provide valid, reliable reports of their child's pain.

Validation of the Non-communicating Children's Pain Checklist-Postoperative Version

This study provided evidence the Non-communicating Children's Pain Checklist-Postoperative Version (NCCPC-PV) has good psychometric properties when used with children who have severe cognitive impairments. The NCCPC-PV was internally consistent and inter-rater reliability was good for total scores. NCCPC-PV total and subscale scores were significantly higher after surgery than before surgery, and they were significantly correlated with the observers' visual analogue ratings of pain. These results confirm that the NCCPC-PV measures pain.

Perhaps the most important finding of this study was that there was no differences due to observer, suggesting familiarity with these children may not be required to assess their postoperative pain and that, with further refinement, this scale may be clinically useful for healthcare professionals. Intriguingly, nurses' VAS ratings did not change significantly from before to after surgery or correlate significantly with caregivers' and researchers' pain ratings or NCCPC-PV scores.

It is possible that completing the NCCPC-PV provided caregivers and researchers with training in what to look for when making their pain ratings. However, it appears more likely that nurses had a priori expectations of how the children would show pain

based on their experience with children without cognitive impairments and that these children did not show the reactions they were expecting.

In summary, these results provide the first evidence that the postoperative pain of children who have severe cognitive impairments can be assessed using a formal pain assessment measure designed for this population. Even more importantly, adults who were unfamiliar with these children were able to detect signs of pain using this instrument. This opens the door to accepting the report of observers who are unfamiliar with these children in other settings and with other instruments.

Using Facial Action to Detect Pain in Children with Severe Cognitive Impairments

This study investigated whether the Child Facial Coding System (Chambers et al., 1996) could detect postoperative pain in children with severe cognitive impairments. The results indicate the overall frequency and intensity of facial action was greater in children rated as having more intense pain by observers. Facial action scores were also significantly related to the visual analogue ratings of pain, but not to visual analogue ratings of sedation. The latter result is particularly interesting, as it suggests sedation may not preclude use of facial action to detect pain after surgery.

The results also indicate only a subset of the CFCS facial actions may reflect pain. The facial actions shown by the children formed three components, one representing the "pain face" reported in children during immunisation (Breau et al., 2001b), a second related to whether children had had dental procedures performed, and a third component reflecting wakefulness. Another important finding was that the facial action of the children in this study also did not vary substantially due to their physical limitations.

Overall, the results suggest that facial action may be a viable method of assessing the pain of children with severe cognitive impairments after surgery. The fact that physical limitations did not significantly impact scores means this method may be beneficial to those children who are most severely physically impaired, such as those with quadriplegia. Although the Child Facial Coding System (Chambers et al., 1996) is a research tool at this point, these results suggest that research aimed at refining the CFCS into a clinically feasible measure would be worthwhile.

Conclusions and Future Directions

This set of studies has laid a foundation for continued research in this area. At the time this research program began, only a handful of studies existed that focussed on the pain of this vulnerable group. Most of these suggested that individuals with cognitive impairments may be insensitive or indifferent to pain because they do not display what was believed are typical pain reactions (McMurray, 1955; Lu, 1981; Biersdorff, 1991; Biersdorff, 1994). The remaining studies were primarily qualitative in design and provided no starting point for further research (Mette et al., 1988; Collignon et al., 1992; Collignon et al., 1995).

The studies described in this thesis move the literature forward from this point. They confirm that children with severe cognitive impairments display a measurable pain reaction. This finding alone is pivotal. Measurement is crucial, not only to advancement of science, but to advancement in the treatment of individual children who are suffering.

This finding also addresses the debate over whether poor detection of pain in these children is due to the fact that they have an altered pain experience or to difficulties

in the communication of their pain. Although these results cannot provide evidence that the pain experience is not altered, they do suggest that previous findings, indicating individuals with cognitive impairments do not react to pain (Lu, 1981; Biersdorff, 1994), may have been due in part to the fact that those studies did not use validated measurement tools.

The studies reported here indicate that pain could be measured with two revised versions of the Non-communicating Children's Pain Checklist (Breau et al., 2000) and with the Child Facial Coding System (Chambers et al., 1996). Thus, the children observed here showed identifiable changes in behaviour in situations in which most people would experience pain and this pain behaviour could be quantified using two instruments, one based on gross behaviour and physiological signs and the other based on fine-grained facial movements. In addition, the analyses indicate children's pain behaviour at home was relatively consistent over time and that there was consistency between two separate observer's reports of pain behaviour after surgery.

These results question proposals that the pain reaction of individuals with cognitive impairments is too idiosyncratic to be detected using standardised instruments (van Dongen et al., 2000a; van Dongen et al., 2000b). They also suggest that the breakdown in the process of communicating pain may be in the observer, rather than in the child sending signals of pain. Thus, these results provide support for the view that poor detection of pain in this group of children is due to problems with the communication of that pain, but they also provide a method for future research to investigate the possibility of altered pain experience more stringently.

Further studies must be undertaken to refine the measures used here. Studies of children with differing levels of cognitive impairment should be conducted. The children here were among those with the most severe limitations and it is not known whether these measures will also be successful with children with less severe and less global deficits. Given the finding of Fanurik and her colleagues, that only children with relatively mild cognitive impairments are capable of self-report (Fanurik et al., 1998), observational pain measures are required for children functioning at moderate levels of impairment.

Studies must also be conducted to investigate the ability of these measures to detect pain of differing intensity and differing causes. The inability of these children to provide information regarding all aspects of their pain means that measures that can delineate the nature of their pain could help in clinical care. Research indicating specific behaviours or the frequency of behaviours are indicative of pain from different sources will be invaluable to diagnosis. Research showing pain behaviour varies with pain intensity or duration will also help in both diagnosis and treatment.

Studies must also be conducted to investigate the validity of these measures in different settings and with different observers. This is especially important for this particular group of children. They have many physical disabilities and medical conditions that result in frequent stays at hospitals. Surgeries and other painful procedures are also often a regular part of their lives. This means professionals, who are not necessarily familiar with the children, are often in situations where they must judge their pain. These

children may also reside in residential centres where staff changes can mean they are frequently cared for by caregivers who are not familiar with them.

Finally, studies should be conducted to investigate the validity and reliability of these measures with adults who have severe cognitive impairments. Although a literature is evolving concerning pain in adults with dementia (Farrell, Katz, & Helme, 1996; Weiner, Peterson, & Keefe, 1999; Morrison & Siu, 2000), no study could be found specifically addressing the pain of adults with severe cognitive impairments of childhood onset. Research indicates that infants at risk for cognitive impairments are living longer than previously (Lorenz et al., 1998) and the number of adults who have cognitive impairments of prenatal origin is increasing (Chaney & Eyman, 2000). Studies also suggest that adults with cognitive impairments suffer high rates of mortality due to conditions for which diagnosis may be delayed without self-report of pain, such as intestinal obstruction (Roy et al., 1987), respiratory infections (Chaney et al., 2000) and postoperative complications (Bernstein & Offenbartl, 1991). Thus, the children studied here may be at high risk for fatal illnesses as they age, especially if they are cared for at home, where both children and adults with cognitive impairments are at higher risk for mortality (Strauss, Eyman, & Grossman, 1996; Strauss, Kastner, & Shavelle, 1998).

In summary, the studies described in this thesis that focussed on pain measurement are only a foundation. Further research is needed along many paths to expand our current knowledge and our expertise in assessing the pain of all people with severe cognitive impairments. Research must follow that provides caregivers and

healthcare professionals with practical, feasible methods of using pain assessment to aid in the management of an individual's health.

This thesis also includes a study indicating pain is frequent in this vulnerable group of children. This finding provides compelling evidence of the need to move knowledge concerning pain assessment forward. The children who were studied here spent up to half their time in pain over four weeks. From a humane perspective, this amount of suffering is totally unacceptable and is reason enough to push research forward.

However, the most disturbing results of this program of research to date may be those revealing the effect of pain on the children's adaptive abilities. It appears that pain disrupts their ability to develop basic skills such as being able to lift a cup, understanding what is being said, and picking up a toy. This is particularly disturbing because development for most of these children is tenuous to begin with and the skills not being developed are necessary for daily functioning. Pain not only causes short-term suffering, but it may also contribute to making these children more dependent in the long-term.

All paediatric pain research focuses on one goal, to reduce children's pain. The final step in that journey is the development of pain management strategies. The research presented here will assist in this effort. Documentation of the nature and impact of the pain this special group of children experiences establishes the gravity of their situation. Evidence that their behaviour reflects their pain and is measurable provides the tools needed to advance research aimed at alleviating their pain.

Closing Remarks

No researcher can take part in a long-term research project with so many children without developing a sense of the individuals involved. This project has spanned several years and involved repeated, close contact with the children, their families and their caregivers. Throughout this, the spirit of the children has emerged as a reason for persevering. Despite physical and cognitive limitations that most would consider devastating, these children manage to project their individuality. Without words, they convey their hope that their situation will change, that the adults around them will make things better. This faith is contagious. It quashes any doubt that real change is possible. It is hoped that the research presented in this thesis is just the beginning of that reality.

TABLES

Table 4. 1: Educational and Employment Characteristics of the Caregivers whose Children Live with Them

	Mothers		Fathers	
Characteristic	No.	%	No.	%
Education				
Less than high school	19	21.3	20	22.5
High school	23	25.8	22	24.7
Post-secondary	22	24.7	17	19.1
University	23	25.8	16	18.0
Information not provided[a]	2	2.2	14	15.7
Employment				
Homemaker	43	48.3	1	1.1
Part-time Employment	2	2.2	0	0.0
Full-time Employment	36	40.4	62	69.7
Self-employed	5	5.6	4	4.5
Retired	1	1.1	4	4.5
Unemployed due to Disability	1	1.1	3	3.4
Unemployed not due to disability	-	-	1	1.1
Information not provided[a]	1	1.1	14	15.7

Note. Information not included for children residing in group homes or residential centres ($n = 12$); therefore $n = 89$. [a] Ethical guidelines of the IWK Health Centre Research Ethics

Board required that participants be notified that providing the information in this table was optional. Some caregivers chose not to provide the information requested and some children belonged to single-parent families.

Table 4. 2: Number of Children in the Families and Birth-order of the Children

	Number of Children in Family		Birth-order of Participating Child	
	No.	%	No.	%
1	18	20.0	34	38.2
2	31	34.8	30	33.7
3	29	32.6	17	19.1
4	9	10.1	6	6.7
5	2	2.2	2	2.2

Note. Information not included for children residing in group homes or residential centres ($n = 12$); therefore $n = 89$.

Table 4. 3: Medications Taken by Children on a Regular Basis

Drug Class/Group	Medication	Children	
		n	%
Analgesic, non-opioid	acetaminophen	4	4.0
Analgesic, opioid	codeine	2	2.0
	hydromorphone	2	2.0
Antacid	ranitidine hydrochloride	15	14.9
Anticonvulsant	carbamazepine	17	16.8
	clobazam	28	27.7
	divalproex	3	3.0
	ethosuximide	2	2.0
	lamotrigine	20	19.8
	phenobarbitol	4	4.0
	vigabatrin	3	3.0
	topirmate	2	2.0
	valproic acid	15	14.9
Antidepressant	amitriptyline	2	2.0
Anticholinergic	glycopyrrolate	2	2.0
Anti-inflammatory	naproxyn	3	3.0
Anti-reflux	cisapride	4	4.0
Antispasmodic	baclofen	3	3.0
Benzodiazepine	clonazepam	2	2.0
	diazepam	5	5.0
	lorazepam	7	6.9
	nitrazepam	9	8.9
Bronchodilators/antiasthmatic	budesonide	5	5.0
	ipratropium bromide	2	2.0
	salbutamol	11	10.9

Drug Class/Group	Medication	Children	
		n	%
Gastric antisecretory agent	omeprazole	6	5.9
Histamine H1 receptor antagonist	loratidine	3	3.0
Hormone	melatonin	2	2.0
Laxative	biscodyl suppository	3	3.0
	lactulose	7	6.9
	magnesium hydroxide	2	2.0
Stool softener	docusate	5	5.0

Note. N = 101. Medications taken regularly by only one child are not listed; some children were being administered more than one medication, these may have been from the same drug class/group.

Table 4. 4: Ability of Children to Use their Limbs

Level of Limb Use	All Children (N = 101)		Children without Quadriplegia (n = 83)	
	n	%	n	%
Upper limb use				
None	25	24.8	7	8.4
Partial	32	31.7	32	38.6
Full	44	43.6	44	53.0
Lower limb use				
None	42	41.5	24	28.9
Partial	38	37.6	38	45.7
Full	21	20.7	21	25.3

Table 4. 5: Adaptive Functioning of the Children in the Cohort

Adaptive Behaviour Domain/Subdomain	Age Equivalent in Months				Years Less than Chronological Age	
			Range			
	M	SD	Min.	Max.	M	SD
All Domains[a]	16.0	16.3	<1.0	102.0	9.0	4.4
Communication Domain	13.2	9.7	<1.0	58.0	8.9	4.4
Receptive	14.5	12.8	<1.0	94.0		
Expressive	12.9	8.6	<1.0	51.0		
Written	21.1	11.0	18.0	67.0		
Daily Living Skills Domain	13.2	10.7	<1.0	75.0	8.9	4.4
Personal	11.9	9.3	<1.0	46.0		
Domestic	20.1	14.0	16.0	135.0		
Community	12.4	12.1	5.0	81.0		
Socialization Domain	13.5	12.8	<1.0	93.0	8.9	4.3
Interpersonal relationships	12.0	10.7	<1.0	63.0		
Leisure and Play	13.3	18.1	<1.0	147.0		
Social Coping	16.2	13.4	11.0	85.0		
Motor Skills Domain	8.4	9.2	<1.0	44.0	9.3	4.5
Gross Motor Skills	8.7	8.2	<1.0	44.0		
Fine Motor Skills	9.5	11.7	<1.0	57.0		

Note. N = 101. As determined by the Vineland Adaptive Behavior Scales (Sparrow et al., 1984). [a]Based on mean age equivalents for the domains; for children over age 6 years, the Motor Skills Domain age equivalent is not included in the computation.

Table 4. 6: Percentage of Children in the Cohort Capable of Displaying Selected Behaviours and Tasks Typically Achieved by Age One Year

Behaviour or Task	Children	
	n	%
Communication Domain		
Turn eyes or head towards sounds	85	84.2
Smile at someone other than primary caregiver	96	95.0
Understand the word "no"	59	58.4
Imitate adults' sounds immediately after hearing them	34	33.7
Understand at least 10 words	75	74.5
Daily Living Skills Domain		
Open mouth when food is presented	70	69.3
Suck or chew on crackers	57	56.4
Eat solid food	62	61.4
Socialization Domain		
Respond to caregiver's voice	91	90.1
Reach out for some one familiar	61	60.4
Distinguishes caregiver from others	90	89.1
Play simple interaction games	60	59.4
Imitate simple adult movements, like waving	40	39.6
Motor Skills Domain		
Sit, with support, for at least one minute	81	80.2
Pick up small objects	53	52.5
Transfer objects from one hand to another	23	22.8
Crawl across the floor	26	25.7

Note to Table 4.6. N = 101. Items from the Vineland Adaptive Behavior Scales (Sparrow et al., 1984). Percentage of children who display behaviours/tasks does not include children who show these inconsistently or partially.

Table 5. 1: Demographic Characteristics of Families

Characteristic		n	%
Marital Status	Married	69	83.1
Mother's Education	Less than high school	18	21.7
	High school	23	27.7
	Post-secondary	18	21.7
	University	22	26.5
	Information not provided [a]	2	2.4
Father's Education	Less than high school	19	22.9
	High school	20	24.1
	Post-secondary	16	19.3
	University	15	18.1
	Information not provided [a]	13	15.7
Number of Children	1	17	20.5
	2	28	33.7
	3	27	32.5
	4 or more	11	13.2
Family Income	< $10,000	7	8.4
	$10,000 to $50,000	53	63.9
	> $50,000	18	21.7
	Information not provided [a]	5	6.0

Note. Information concerning parents of children residing outside the family home not included (n = 11); therefore n = 83. [a] Ethical guidelines of the IWK Health Centre Research Ethics Board required that participants be notified that providing the

information in this table was optional. Some caregivers chose not to provide the information requested and some children belonged to single-parent families.

Table 5. 2: Medical Conditions and Physical Impairments of the Children

Characteristic		n	%
Onset of Neurologic Impairment	Prenatal	67	71.3
	Perinatal	11	11.7
	Postnatal	13	13.8
	Information not Available	3	3.2
Level of Diagnosed Mental	Moderate	9	9.6
Retardation	Severe	61	64.9
	Profound	16	17.0
	Information not Available	8	8.5
Cerebral Palsy:		44	46.8
Primary Seizure Type:	Generalized	31	33.0
	Focal	28	29.8
Upper Limb Use:	Full	43	45.7
	Some	39	41.5
	None	12	12.8
Lower Limb Use:	Full	21	22.3
	Some	42	44.6
	None	31	33.0
Visual Impairment	Partial	18	19.1
	Full	23	24.5
Hearing Impairment	Partial	7	7.4
	Full	8	8.5
Requires Medical Monitoring:	None	22	23.4
	Monthly	43	45.7
	Weekly	10	10.6
	Daily	19	20.2
Tube Fed		25	26.6

Characteristic		n	%
Number of Regular Medications	None	20	21.3
	One	21	22.3
	Two	16	17.0
	Three	7	7.4
	Four	9	9.6
	Five or More	21	22.3

Note. N = 94.

Table 5. 3: Chronological Ages and Adaptive Age Equivalents of the Children

Measure	Range	Mean	SD
Chronological Age	3.0 years -18.7 years	10.1 years	4.3
Communication[a]	<1 month-56 months	13.8 months	10.0
Daily Living Skills[a]	<1 month-83 months	14.0 months	11.7
Socialization[a]	<1 month-90 months	14.0 months	12.9
Motor Skills[a]	<1 month-44 months	8.7 months	9.5

Note. N = 94. [a]Age Equivalents on the Vineland Adaptive Behavior Scales (Sparrow et al., 1984).

Table 5. 4

Number of Children Who Experienced at Least One Episode and Number of Episodes of Pain Reported Each Week by Category and Cause of Pain

Pain Category and Cause	Week 1			Week 2			Week 3			Week 4		
	Children		Episodes	Children		Episodes	Children		Episodes	Children		Episodes
	n	%	n	n	%	n	n	%	n	n	%	n
Accidental	14	15%	16	15	16%	21	10	11%	14	7	7%	10
Immediate	13		15	14		17	9		13	6		9
Post-accident	1		1	1		4	1		1	1		1
Gastrointestinal	11	11%	25	13	13%	35	9	10%	20	5	5%	21
Digestive	5		12	5		21	5		12	4		15
Reflux	1		2	1		1	0		0	0		0
Bowels	3		9	4		10	4		8	1		6
Constipation	2		2	3		3	0		0	0		0
Musculoskeletal	6	6%	12	5	5%	11	7	7%	11	3	3%	19
Muscle	3		3	2		2	1		1	1		5
Orthopedic	3		9	3		9	6		10	2		14

Pain Category and Cause	Week 1			Week 2			Week 3			Week 4		
	Children		Episodes	Children		Episodes	Children		Episodes	Children		Episodes
	n	%	n	n	%	n	n	%	n	n	%	n
Common Childhood	6	6%	12	3	3%	9	4	4%	6	5	5%	6
Teething	5		11	0		0	1		2	0		0
Menstruation	1		1	3		9	3		4	4		5
Headache	0		0	0		0	0		0	1		1
Infection	7	7%	9	7	7%	11	5	5%	22	7	7%	21
Chest infection	2		2	1		1	1		7	0		0
Throat infection	2		2	1		1	0		0	0		0
Other	3		5	5		9	4		15	7		21
Recurrent	4	4%	4	3	3%	15	5	5%	6	6	6%	8
Ears	3		3	3		15	2		2	5		7
Seizures	1		1	0		0	2		3	1		1
Diaper rash	0		0	0		0	1		1	0		0
Medical	5	5%	11	0	0%	0	7	7%	13	2	2%	7
Needle	1		1	0		0	4		4	0		0
Postoperative	1		1	0		0	0		0	1		3

Pain Category and Cause	Week 1			Week 2			Week 3			Week 4		
	Children		Episodes	Children		Episodes	Children		Episodes	Children		Episodes
	n	%	n	n	%	n	n	%	n	n	%	n
Feeding tube irritation	3		9	0		0	3		9	1		4
Other *	4	4%	4	0	0%	0	0	0%	0	1	1%	7
Unknown	1	1%	9	4	4%	9	0	0%	0	1	1%	2
Total Per Week	49	52%	102	44	47%	111	38	40%	92	33	35%	107

Note. N = 94. Percentages rounded. *cast, tumor, chemical burn, self-injurious behaviour.

Table 5. 5

Mean Number of Days with Pain and Mean Time Spent with Pain Per Week

	All Children						Children Who Experienced Pain at Least Once That Week					
	Days with Pain			Time in Pain (hours:minutes)				Days with Pain			Time in Pain (hours:minutes)	
	Mean	SD	Range	Mean	SD	Range	n*	Mean	SD	Range	Mean	SD	Range
Week 1	1.1	1.7	0-7	9:54	32:01	0:00-168:00	49	2.2	1.8	1-7	19:03	42:42	0:01-168:00
Week 2	1.3	2.1	0-7	9:12	27:15	0:00-168:00	44	2.9	2.1	1-7	19:42	37:20	0:01-168:00
Week 3	1.1	1.9	0-7	10:06	32:30	0:00-168:00	38	2.8	2.0	1-7	25:00	47:36	0:01-168:00
Week 4	1.3	2.4	0-7	10:30	35:24	0:00-168:00	33	3.8	2.6	1-7	29:45	52:20	0:01-168:00

Note. N = 94. * Does not include cases in which intensity or duration were not provided.

Table 5. 6: Incidence, Mean Intensity and Mean Duration of Pain by Type, Category and Cause Over Four Weeks

Pain Category	Children (N = 94)		Episodes (N = 406)		Intensity			Duration (Hours:Minutes)		
	n	%	n	%	Mean	SD	Range	Mean	SD	Range
Accidental	28	30%	61	15%	3.8	2.1	1-10	0:46	3:30	0:01-24:00
Immediate	26	28%	54	11%	3.8	2.1	1-10	0:06	0:11	0:01-1:00
Post-accident	3	3%	7	2%	3.9	1.7	1-5	4:36	8:36	0:01-24:00
Non-accidental	58	62%	333	82%	6.1	2.2	1-10	6:06	16:11	0:01-168:00
Gastrointestinal	21	22%	101	25%	7.5	2.0	1-10	3:48	9:03	0:02-72:00
Digestive	11	12%	60	15%	7.5	2.3	1-10	2:01	2:12	0:05-9:00
Reflux	2	2%	3	<1%	6.7	1.2	6-8	2:54	18:48	0:45-4:00
Bowels	9	10%	33	8%	7.6	1.2	5-10	4:42	7:54	0:15-24:00
Constipation	5	5%	5	1%	6.2	0.8	5-7	24:30	33:30	0:02-72:00
Musculoskeletal	18	19%	53	13%	5.1	1.7	2-10	1:06	3:42	0:01-24:00
Muscle	6	6%	11	3%	5.2	1.0	4-8	0:44	0:22	0:02-60:00
Orthopaedic	12	13%	42	10%	5.0	1.8	2-10	0:64	4:24	0:01-24:00
Recurrent	12	13%	33	8%	6.0	1.4	4-8	7:05	14:24	0:01-72:00
Ears	8	9%	27	7%	6.2	1.3	4-8	6:30	15:12	0:03-72:00
Seizures	3	3%	5	1%	5.4	1.5	4-8	6:00	10:12	0:01-24:00

Pain Category	Children (N = 94)		Episodes (N = 406)		Intensity			Duration (Hours:Minutes)		
	n	%	n	%	Mean	SD	Range	Mean	SD	Range
Diaper rash	1	1%	1	<1%						
Common Childhood	10	11%	33	8%	5.0	1.7	2-8	9:01	19:15	0:01-96:00
Teething	5	5%	13	3%	5.2	1.9	2-8	18:30	35:20	0:13-96:00
Menstruation	4	4%	19	5%	4.7	1.6	2-7	5:54	8:30	0:01-24:00
Headache	1	1%	1	<1%						
Infection	19	20%	63	16%	5.6	2.4	2-10	11:42	25:42	0:01-168:00
Chest infection	3	3%	10	2%	4.6	2.3	2-9	4:24	9:42	0:01-24:00
Throat infection	3	3%	3	<1%	5.3	1.5	4-7	32:00	13:54	24:00-48:00
Other	14	15%	50	12 %	5.8	2.5	2-10	11:20	27:12	1:00-168:00
Medical	12	13%	31	8%	3.8	2.1	1-10	0:46	3:30	0:01-24:00
Needle	5	5%	5	1%	4.4	0.9	3-5	16:00	13:48	0:02-24:00
Postoperative	2	2%	4	1%	5.0	0.0	5-5	12:45	23:30	1:00-48:00
Feeding tube irritation	5	5%	22	5%	4.4	1.5	2-7	0:58	1:09	0:00-2:30
Other *	5	5%	11	3%	6.8	0.7	5-8	3:20	8:24	0:20-24:00
Unknown	5	5%	20	5%	8.7	1.1	6-10	11:30	10:36	0:30-24:00

Note. N = 94. Percentages rounded. *cast, tumor, chemical burn, self-injurious behaviour.

Table 5. 7: Number of Children Who Experienced Each Pain Type, Category and Cause at Least Once Over Four Weeks by Gender

Pain Type and Category	All (N = 94)		Girls (n = 41)		Boys (n = 53)		Girls vs. Boys	
	n	%	n	%	n	%	χ^2	p
Any Pain	73	78%	32	78%	41	77%	0.0	.94
Accidental	28	30%	13	32%	15	28%	0.2	.72
Non-accidental	58	62%	28	68%	30	57%	1.3	.25
Gastrointestinal	21	22%	10	24%	11	21%	0.2	.68
Musculoskeletal	18	19%	3	7%	15	28%	6.6	.01
Common Childhood	10	11%	5	12%	5	9%	0.2	.67
Infection	19	20%	9	22%	10	19%	0.1	.71
Recurrent	11	12%	7	17%	4	8%	2.0	.15
Medical	14	15%	5	12%	9	17%	0.4	.52
Other[a]	4	4%	1	2%	3	6%		
Unknown[a]	5	%	3	7%	2	4%		

Note. Percentages rounded. No comparisons significant after corrections for multiple tests. [a]Insufficient numbers per cell to conduct statistical tests.

Table 5. 8: Number of Children Who Experienced Pain by Type, Category and Cause at Least Once Over Four Weeks by Age Group

Pain Type and Category	Age 3-7 (n = 32)		Age 8-12 (n = 31)		Age 13-18 (n = 28)		Age 3-7 vs. age 8-12		Age 3-7 vs. age 13-18		Age 8-12 vs. age 13-18	
	n	%	n	%	n	%	χ^2	p	χ^2	p	χ^2	p
Any Pain	26	81%	24	77%	21	75%	0.4	.55	0.5	.46	0.0	.95
Accidental	15	47%	5	16%	8	29%	7.2	.007	2.7	.10	1.6	.20
Non-accidental	19	59%	22	71%	15	54%	0.6	.44	0.3	.64	1.7	.20
Gastrointestinal	7	21%	6	19%	8	29%	0.1	.76	0.5	.47	0.9	.34
Musculoskeletal	2	6%	9	29%	5	18%	4.7	.03	0.7	.40	1.3	.26
Common Childhood	3	9%	3	10%	4	14%	0.0	1.0	0.7	.40	0.4	.53
Infection	9	28%	5	16%	4	14%	0.8	.38	1.2	.28	0.2	.68
Recurrent	3	9%	7	23%	1	4%	1.9	.17	0.9	.33	4.1	.04
Medical	7	22%	3	10%	4	14%	1.9	.17	0.8	.38	0.4	.53
Other[a]	0	0%	1	3%	3	11%						
Unknown[a]	0	0%			2	7%						

Note. N = 94. Percentages rounded. No comparisons significant after corrections for multiple tests. [a]Insufficient numbers per cell to conduct statistical tests.

Table 5. 9

Number of Non-accidental Pain Categories and Number of Pain Episodes Experienced by Children Over Four Weeks

Pain	All (N = 94)		Girls (n = 41)		Boys (n = 53)		Age 3-7 (n = 32)		Age 8-12 (n = 31)		Age 13-19 (n = 28)	
	n	%	n	%	n	%	n	%	n	%	n	%
Number of Categories												
0	36	38%	13	32%	23	43%	13	41%	10	32%	13	46%
1	33	35%	19	46%	14	26%	15	47%	10	32%	8	29%
2+	25	27%	9	22%	16	30%	5	16%	13	42%	7	25%
Number of Episodes												
0	36	38%	13	32%	23	43%	13	41%	10	32%	13	46%
1-4	32	34%	20	49%	12	23%	15	47%	12	39%	5	18%
5+	26	28%	8	20%	18	34%	5	16%	11	35%	10	36%

Note. Percentages rounded.

Table 5. 10

Pearson Correlations Between Children's Adaptive Age Equivalents and The Amount of Pain Experienced

	Communication		Daily Living Skills		Socialization		Motor Skills	
	r	p	r	p	r	p	r	p
Number of Pain Episodes	-.27	.009	-.33	.001*	-.20	.049	-.23	.028
Number of Types of Pain	-.34	.001*	-.33	.001*	-.29	.005	-.24	.020
Number of Accidental pain Episodes	.02	.820	.12	.235	.01	.919	.22	.036
Number of Non-accidental Pain Episodes	-.26	.011	-.33	.001*	-.20	.054	-.28	.007

Note. N = 94. Age Equivalents on the Vineland Adaptive Behavior Scales (Sparrow et al., 1984). * Significant after corrections for multiple tests.

Table 6. 1

Demographic Characteristics of the Families

Characteristic		n	%
Marital Status	Married	67	85.9
Mother's Education	Less than high school	18	23.1
	High school	22	28.2
	Post-secondary	16	20.5
	University	20	25.6
	Information not provided [a]	2	2.6
Father's Education	Less than high school	19	24.4
	High school	20	25.6
	Post-secondary	15	19.2
	University	13	16.7
	Information not provided [a]	11	14.1
Number of Children	1	16	20.5
	2	27	34.6
	3	24	30.8
	4 or more	11	13.1
Family Income	< $10,000	6	7.7
	$10,000 to $50,000	49	62.8
	> $50,000	18	23.1
	Information not provided [a]	5	6.4

Note. Information concerning parents of children residing outside the family home not included (n = 11); therefore n = 78. [a] Ethical guidelines of the IWK Health Centre Research Ethics Board required that participants be notified that providing the information in this table was optional. Some caregivers chose not to provide the information requested and some children belonged to single-parent families.

Table 6. 2

Medical Characteristics of the Children

Characteristic		n	%
Onset of Neurologic Impairment	Prenatal	64	71.9
	Perinatal	11	12.4
	Postnatal	12	13.5
	Information not Available	2	2.2
Level of Diagnosed Mental	Moderate	9	10.1
Retardation	Severe	59	66.3
	Profound	13	14.6
	Information not Available	2	2.2
Cerebral Palsy:		43	48.3
Primary Seizure Type:	Generalized	29	32.6
	Focal	26	29.2
Upper Limb Use:	Full	40	44.9
	Some	37	41.5
	None	12	13.5
Lower Limb Use:	Full	20	22.5
	Some	40	44.9
	None	29	32.6
Visual Impairment	Partial	18	20.2
	Full	22	24.7
Hearing Impairment	Partial	7	7.9
	Full	7	7.9
Requires Medical Monitoring:	None	22	24.7
	Monthly	41	46.1
	Weekly	8	9.0
	Daily	18	20.2

Characteristic		n	%
Tube Fed			
Number of Regular Medications	None	19	21.3
	One	21	23.6
	Two	14	15.7
	Three	7	7.9
	Four	8	9.0
	Five or More	20	22.5

Note. N = 89.

Table 6. 3

Pain Experienced by the Children By Pain Survey and By Quartile

	Time in Pain (hours:minutes)	
	Mean	SD
By Survey		
Survey 1	9:54	32:01
Survey 2	9:12	27:15
Survey 3	10:06	32:30
Survey 4	10:30	35:24
Total	39:12	71:18
By Quartile		
Quartile 1	0:0	0:0
Quartile 2	0:32	0:40
Quartile 3	11:00	7:48
Quartile 4	146:36	71:42

Note. N = 89.

Table 6. 4

Raw Scores and Adaptive Age Equivalents for the Children (N = 89) at Entry to the Research Program and One Year Later

Domain/Subdomain	Mean Raw Scores				Mean Age Equivalent (months)			
	Entry		One Year Later		Entry		One Year Later	
	M	SD	M	SD	M	SD	M	SD
Communication Domain	23.7	15.9	25.7	19.3	13.8	10.0	15.5	14.9
Receptive	12.9	6.6	13.7	7.6	15.1	13.1	20.4	24.1
Expressive	10.5	9.6	11.4	11.3	13.4	8.9	13.3	10.4
Written	0.4	1.5	0.6	2.1	21.6	11.6	23.2	13.6
Daily Living Skills Domain	18.3	18.0	19.5	18.8	13.8	10.9	14.4	11.1
Personal	14.9	12.1	15.8	12.9	12.5	9.4	13.1	9.9
Domestic	1.2	3.8	1.7	3.6	20.5	14.8	21.9	12.2
Community	1.8	3.8	2.0	3.6	12.9	12.6	13.7	13.2

	Mean Raw Scores				Mean Age Equivalent (months)			
Socialization Domain	28.7	15.1	31.9	17.4	14.0	13.3	16.5	15.9
Interpersonal relationships	17.7	7.0	18.7	18.4	12.3	11.0	15.2	17.7
Play and Leisure	9.6	5.5	11.1	5.4	13.8	18.9	16.0	16.7
Social Coping	1.4	3.9	2.1	5.1	16.8	14.2	18.7	18.6
Motor Skills Domain	14.2	13.5	17.2	15.3	8.5	9.4	10.7	11.7
Gross Motor Skills	8.5	7.6	9.3	8.2	8.7	8.3	10.0	10.4
Fine Motor Skills	5.8	6.6	7.7	7.8	9.7	11.9	13.4	15.1

Note. N = 89.

Table 6. 5

Change in Adaptive Age Equivalents (months) Over One Year by Duration of Pain

	Time in Pain Over Four Weeks Surveyed[a]							
Adaptive Behaviour Domain/Subdomain	Quartile One (n = 22)		Quartile Two (n = 22)		Quartile Three (n = 23)		Quartile Four (n = 22)	
	M	SD	M	SD	M	SD	M	SD
Communication Domain	4.1	15.6	3.0	6.1	1.5	5.3	-1.7	4.1
Receptive	8.2	19.7	10.1	19.7	4.4	22.7	-1.8	4.9
Expressive	0.9	6.9	1.3	6.0	-.03	3.8	-2.0	4.3
Written	3.8	13.1	4.0	11.5	0.0	2.0	-1.4	6.4
Daily Living Skills Domain	0.2	4.7	2.7	3.5	0.2	3.3	-0.5	3.4
Personal	0.8	3.8	2.0	3.3	-0.3	3.1	-0.1	2.3
Domestic	-0.8	12.4	4.8	5.8	1.1	5.4	0.6	3.0
Community	-0.1	7.3	3.4	9.6	1.0	6.0	-1.1	6.9

Adaptive Behaviour Domain/Subdomain	Time in Pain Over Four Weeks Surveyed[a]							
	Quartile One (n = 22)		Quartile Two (n = 22)		Quartile Three (n = 23)		Quartile Four (n = 22)	
	M	SD	M	SD	M	SD	M	SD
Interpersonal relationships	6.6	14.9	2.2	12.2	3.5	10.4	-0.9	5.4
Leisure and Play	2.6	5.7	3.9	6.1	3.4	9.8	-1.0	17.7
Social Coping	1.2	12.2	4.1	9.6	3.1	13.4	-0.7	12.1
Motor Skills Domain	3.9	6.2	3.7	3.3	0.5	2.5	0.5	2.1
Gross Motor Skills	3.0	10.2	1.1	2.4	0.1	2.8	0.5	2.3
Fine Motor Skills	5.0	6.8	6.6	6.5	1.6	6.1	1.5	3.5

Note. N = 89. [a]Quartile One: no pain; Quartile Two: less than 1 minute to 2 hours; Quartile Three: 2 hours, 1 minute to 30 hours; Quartile Four: 48 hours to 343 hours, 10 minutes.

Table 7. 1

Demographic Characteristics of the Families

Characteristic		n	%
Marital Status	Married	52	86.7
Mother's Education	Less than high school	11	18.3
	High school	18	30.0
	Post-secondary	14	23.3
	University	16	26.7
	Information not provided [a]	1	1.7
Father's Education	Less than high school	12	20.0
	High school	16	26.7
	Post-secondary	14	23.3
	University	10	16.7
	Information not provided [a]	8	13.3
Number of Children	1	13	21.7
	2	20	33.3
	3	19	31.7
	4 or more	8	13.3
Family Income	< $10,000	4	6.7
	$10,000 to $50,000	40	66.7
	> $50,000	14	23.3
	Information not provided [a]	2	3.3

Note. Information concerning parents of children residing outside the family home not included (n = 11); therefore n = 60. [a] Ethical guidelines of the IWK Health Centre Research Ethics Board required that participants be notified that providing the information in this table was optional. Some caregivers chose not to provide the information requested and some children belonged to single-parent families.

Table 7. 2

Medical Conditions and Physical Impairments of the Children

Characteristic		n	%
Onset of Neurologic Impairment	Prenatal	50	70.4
	Perinatal	9	12.7
	Postnatal	10	14.1
	Information not Available	2	2.2
Level of Diagnosed Mental	Moderate	6	8.5
Retardation	Severe	45	63.4
	Profound	14	19.7
	Information not Available	6	8.5
Cerebral Palsy:		35	49.3
Primary Seizure Type:	Generalized	24	33.8
	Focal	21	29.6
Upper Limb Use:	Full	32	45.1
	Some	28	39.5
	None	11	15.5
Lower Limb Use:	Full	15	21.1
	Some	32	45.1
	None	24	33.8

Characteristic		n	%
Visual Impairment	Partial	16	22.5
	Full	17	23.9
Hearing Impairment	Partial	6	8.5
	Full	9	12.7
Requires Medical Monitoring:	None	15	21.1
	Monthly	33	46.5
	Weekly	9	12.7
	Daily	14	19.7
Tube Fed		24	33.8
Number of Regular Medications	None	14	19.7
	One	13	18.3
	Two	13	18.3
	Three	5	7.0
	Four	6	8.5
	Five or More	20	28.2

Note. N = 71.

Table 7. 3

Chronological Ages and Adaptive Age Equivalents of the Children

Measure	Range	Mean	SD
Chronological Age	3.0 years -18.0 years	10.0 years	4.4
Communication[a]	<1 month-58 months	13.4 months	9.5
Daily Living Skills[a]	<1 month-52 months	13.2 months	9.8
Socialization[a]	<1 month-61 months	13.7 months	11.1
Motor Skills[a]	<1 month-44 months	8.8 months	9.1

Note. N = 71. [a]Age Equivalents on the Vineland Adaptive Behavior Scales (Sparrow et al., 1984)

Table 7. 4

Item-total Correlations and Percentage of Caregivers Reporting each Item of the Non-communicating Children's Pain Checklist-Revised for Pain Episode 1 (n = 71) and Pain Episode 2 (n = 55)

Item	Item-total Correlation		Percentage of Children Displaying	
	Pain Episode 1	Pain Episode 2	Pain Episode 1	Pain Episode 2
Vocal Subscale				
Moaning , whining, whimpering (Fairly soft)	.44	.53	73	78
Crying (Moderately loud)	.53	.66	59	46
Screaming/yelling (Very loud)	.46	.56	41	29
A specific sound or word for pain (for example: a word, cry, or type of laugh)	.52	.51	59	64
Eating/Sleeping Subscale				
Eating less, not interested in food	.42	.14	28	36
Increase in sleep	.31	-.03	25	29
Decrease in sleep	.34	.58	14	26

Item	Item-total Correlation		Percentage of Children Displaying	
	Pain Episode 1	Pain Episode 2	Pain Episode 1	Pain Episode 2
Social Subscale				
Not co-operating, cranky, irritable, unhappy	.74	.68	62	78
Less interaction with others, withdrawn	.75	.64	49	56
Seeking comfort or physical closeness	.33	.45	61	75
Being difficult to distract, not able to satisfy or pacify	.71	.70	49	66
Facial Subscale				
A furrowed brow	.49	.49	55	66
A change in eyes, including: squinching of eyes, eyes opened wide; eyes frowning	.66	.71	65	62
Turning down of mouth, not smiling	.79	.75	62	73
Lips puckering up, tight, pouting or quivering	.80	.78	44	53
Clenching or grinding teeth, chewing or thrusting tongue out	.52	.47	44	36
Activity Subscale				
Not moving, less active, quiet	.26	.38	44	53
Jumping around, agitated, fidgety	.57	.47	27	27

Item	Item-total Correlation		Percentage of Children Displaying	
	Pain Episode 1	Pain Episode 2	Pain Episode 1	Pain Episode 2
Body/Limb Subscale				
Floppy	.33	.21	25	26
Stiff, spastic, tense, rigid	.57	.47	49	64
Gesturing to or touching part of the body that hurt	.15	.06	28	24
Protecting, favouring or guarding part of the body that hurts	.45	.47	32	33
Flinching or moving the body part away, being sensitive to touch	.46	.45	41	42
Moving the body in specific way to show pain (e.g. head back, arms down, curls up etc.)	.65	.46	32	62
Physiological Signs Subscale				
Shivering	.46	.50	10	16
Change in colour, pallor	.62	.73	41	42
Sweating, perspiring	.56	.49	28	26
Tears	.66	.76	54	62
Sharp intake of breath, gasping	.52	.77	47	36
Breath holding	.47	.68	25	20

Table 7. 5: Non-communicating Children's Pain Checklist-Revised Subscale Scores for Four Observations

Subscale	Pain Episodes				No-Pain Episodes				Test-Retest Reliability		Discriminant Validity	
	1		2		1		2		P1 vs.P2	NP1 vs. NP2	P1 vs. NP1	P2 vs.NP2
	(n = 71)		(n = 55)		(n = 71)		(n = 55)		(n = 55)	(n = 55)	(n = 71)	(n = 55)
	M	SD	M	SD	M	SD	M	SD	t	t	t	t
Vocal	3.7	2.8	3.6	3.0	0.8	1.3	0.7	1.2	0.8	0.6	7.9**	7.0**
Eating/Sleeping	1.2	1.7	1.5	1.5	0.5	1.0	0.3	0.9	-1.4	0.8	3.3*	6.0*
Social	3.8	3.2	2.6	2.5	1.2	1.5	1.2	2.1	3.8**	0.2	6.0**	3.5**
Facial	4.5	4.0	4.6	3.7	0.8	1.4	0.8	1.8	-0.3	-0.7	7.5**	7.1**
Activity	1.1	1.2	1.3	1.2	0.5	0.8	0.4	0.8	-0.9	0.7	4.6**	5.9**
Body and Limbs	3.6	3.3	4.2	3.2	0.6	1.2	0.8	1.5	-1.3	-1.2	7.4**	7.7**
Physiological Signs	3.2	3.4	3.5	4.4	0.3	0.8	0.5	1.3	-0.7	-1.3	7.2**	5.3**
Total Score	21.1	15.9	21.2	15.2	4.8	5.2	4.7	6.5	-.04	-0.3	9.5**	8.2**

Note. P1 = Pain Episode 1, P2 = Pain Episode 2, NP1 = No-Pain Episode 1, NP2 = No-Pain Episode 2. Matched sample t-tests: *p = .001; **p < .001.

Table 8. 1

Demographic Characteristics of the Families

Characteristic		n	%
Marital Status	Married	15	75
Mother's Education	Less than high school	5	25
	High school	5	25
	Post-secondary	5	25
	University	5	25
Father's Education	Less than high school	5	25
	High school	3	15
	Post-secondary	4	20
	University	3	15
	Information not provided[a]	5	25
Number of Children	1	4	20
	2	3	15
	3	10	50
	4	3	15
Family Income	< $10,000	2	10
	$10,000 to $50,000	16	80
	> $50,000	2	10

Note. Information concerning parents of children residing outside the family home not included (n = 4); therefore n = 20. [a]Ethical guidelines of the IWK Health Centre Research Ethics Board required that participants be notified that providing the information in this table was optional. Some caregivers chose not to provide the information requested and some children belonged to single-parent families.

Table 8. 2

Medical Conditions and Physical Impairments of the Children

Characteristic		n	%
Onset of Neurologic Impairment	Prenatal	15	62.5
	Perinatal	4	16.7
	Postnatal	4	16.7
	Information not Available	1	4.2
Level of Diagnosed Mental	Moderate	2	8.3
Retardation	Severe	15	62.5
	Profound	5	20.8
	Information not Available	2	8.3
Cerebral Palsy:		16	66.7
Primary Seizure Type:	Generalized	3	12.5
	Focal	9	37.5
Upper Limb Use:	Full	9	37.5
	Some	10	41.7
	None	5	20.8
Lower Limb Use:	Full	4	16.7
	Some	8	33.3
	None	12	50.0

Characteristic		n	%
Visual Impairment	Partial	5	20.8
	Full	7	29.2
Hearing Impairment	Partial	1	4.2
	Full	2	8.3
Requires Medical Monitoring:	None	3	12.5
	Monthly	13	54.2
	Weekly	2	8.3
	Daily	6	25.0
Tube Fed		11	45.8
Number of Regular Medications	None	4	16.7
	One	4	16.7
	Two	2	8.3
	Three	2	8.3
	Four	4	16.7
	Five or More	8	33.3

Note. N = 24

Table 8. 3

Chronological and Adaptive Ages of the Children

Measure	Range	Mean	SD
Chronological Age	3.47 years - 18.7 years	10.9 years	4.4
Communication[a]	1 month-31 months	13.0 months	7.3
Daily Living Skills[a]	<1 month-75 months	12.7 months	15.0
Socialization[a]	3 months-93 months	17.2 months	20.2
Motor Skills[a]	<1 month-40 months	7.5 months	10.8

Note. N = 24. [a]Age Equivalents on the Vineland Adaptive Behavior Scales (Sparrow et al., 1984)

Table 8. 4

Procedures Performed for the Children

Procedures Performed	No.	%
Dental extractions only	5	20%
Dental extractions in addition to other procedure(s)	3	13%
G-Button change only	2	8%
G-Button change/insertion in addition to other procedure(s)	6	25%
Bilateral myringotomy tubes	2	8%
Heelcord/tendon lengthening	2	8%
Orthopedic surgery back/hip	3	13%
Port-a-cath insertion	1	4%
Fundoplication	1	4%
G-tube insertion/removal	3	13%
Endoscope and biopsies	2	8%
Strabismus repair	1	4%
Skin Graft	1	4%

Note. N = 24. Percentages sum to greater than 100%; 7 children had more than one procedure performed while they were under anesthetic.

Table 8. 5

Percentage of Children who Displayed Each Item of the NCCPC-PV at Least "A Little" Before and After Surgery According to Caregivers and Researchers

NCCPC-PV Item	Before Surgery (%)		After Surgery (%)	
	Caregiver	Researcher	Caregiver	Researcher
Vocal Subscale				
Moaning , whining, whimpering (Fairly soft)	25	37	58	50
Crying (Moderately loud)	4	4	25	12
Screaming/yelling (very loud)	4	8	8	4
A specific sound or vocalization for pain	4	4	33	8
Social Subscale				
Not co-operating, cranky, irritable, unhappy	25	8	46	42
Less interaction, withdrawn	17	0	33	17
Seeks comfort or physical closeness	29	33	54	66
Difficult to distract, not able to satisfy or pacify	11	17	29	29

NCCPC-PV Item	Before Surgery (%)		After Surgery (%)	
	Caregiver	Researcher	Caregiver	Researcher
Change in eyes, including: squinching, eyes opened wide, eyes frown	33	21	54	71
Turn down of mouth, not smiling	17	21	42	37
Lips pucker up, tight, pout, or quiver	8	8	29	17
Clenches/grinds teeth, chews, thrusts tongue out	25	33	17	37
Activity Subscale				
Not moving, less active, quiet	12	17	46	42
Jumping around, agitated, fidgety	37	46	37	54
Body and Limbs Subscale				
Floppy	25	33	29	25
Stiff, spastic, tense, rigid	25	21	33	29
Gestures to or touches part of body that hurts	0	4	21	17
Protects, favors or guards part of body that hurts	0	0	37	25
Flinches or moves away part of body that hurts	4	4	46	37
Moves in specific way to show pain	0	0	37	34

NCCPC-PV Item	Before Surgery (%)		After Surgery (%)	
	Caregiver	Researcher	Caregiver	Researcher
Protects, favors or guards part of body that hurts	0	0	37	25
Flinches or moves away part of body that hurts	4	4	46	37
Moves in specific way to show pain	0	0	37	34
Physiological Signs				
Shivering	4	4	21	12
Change in color, pallor	4	4	37	33
Sweating, perspiring	0	0	4	8
Tears	0	0	33	17
Sharp intake of breath, gasping	8	8	17	8
Breath holding	4	4	8	0

Note. N = 24. NCCPC-PV: Non-Communicating Children's Pain Checklist-Postoperative Version. From (Breau et al., 2002). Used with Permission.

Mean Visual Analogue Scale Pain Ratings and Total NCCPC-PV Scores of Caregivers, Researchers and Nurses Before and After Surgery

Observer	Before Surgery		After Surgery	
	Mean	SD	Mean	SD
VAS[a] Pain Ratings				
Caregiver	3.99	10.40	25.24	24.20
Researcher	2.13	7.22	21.68	16.57
Nurse[b]	5.20	12.02	10.90	15.08
Nurse[c]	1.75	2.72	11.06	17.92
NCCPC-PV[d] Total Scores				
Caregiver	4.83	3.56	12.21	10.90
Researcher	5.37	3.94	11.17	6.60

Note. N = 24. [a] Visual Analogue Scale, 100 mm. [b]With missing replaced. [c]Without missing replaced, n = 16. [d]Non-Communicating Children's Pain Checklist-Postoperative Version. From (Breau et al., 2002). Used with Permission.

Table 8. 7

Correlations Among NCCPC-PV Total Scores and VAS Pain Ratings Before and After Surgery

	Before Surgery				After Surgery			
	VAS[a] Pain Ratings			NCCPC-PV[b] Total	VAS[a] Pain Ratings			NCCPC-PV[b] Total
	Caregiver	Researcher	Nurse	Caregiver	Caregiver	Researcher	Nurse	Caregiver
VAS[a] Pain Ratings								
Caregiver								
Researcher	.91**				.62**			
Nurse[c]	.94**	.92**			.08	.49*		
NCCPC-PV[b] Total								
Caregiver	.46*	.39	.40		.84**	.64**	.09	
Researcher	.50**	.53**	.53**	.71**	.72**	.69**	.42*	.72**

Note. [a]Visual Analogue Scale. [b]Non-communicating Children's Pain Checklist -Postoperative Version. [c]n = 19 before surgery, n = 20 after surgery. * p < .05. ** Significant after corrections for multiple tests. From (Breau et al., 2002). Used with Permission.

Table 8. 8:

Mean Caregiver and Researcher NCCPC-PV Subscale Total Scores Before and After Surgery

Subscale	Possible Range	Before Surgery				After Surgery			
		Caregiver		Researcher		Caregiver		Researcher	
		Mean	SD	Mean	SD	Mean	SD	Mean	SD
Vocal	0-12	0.50	0.89	0.63	1.14	1.46	1.72	1.00	1.41
Social	0-12	1.21	1.41	0.88	1.36	2.33	2.48	2.21	1.56
Facial	0-15	1.13	1.36	1.38	1.61	2.67	3.17	3.38	3.10
Activity	0-6	0.88	1.12	1.04	1.12	1.38	1.38	1.54	1.19
Body and Limbs	0-18	0.83	1.24	1.17	1.09	2.92	3.12	2.00	1.72
Physiological Signs	0-18	0.29	0.75	0.29	0.69	1.46	1.72	1.04	1.37
Total NCCPC Score	0-81	4.83	3.60	5.38	3.94	12.21	10.90	11.17	6.60

Note. N = 24. NCCPC-PV: Non-Communicating Children's Pain Checklist-Postoperative Version. From (Breau et al., 2002). Used with Permission.

Table 8. 9

Repeated Measures Analysis of Variance of NCCPC-PV Subscale Scores Made by Caregivers and Researchers Before and After Surgery

Source	df	F	p
Time	1,23	15.1	.001
Person	1,23	0.00	.99
Subscale	5,19	9.7	<.001
Time x Person	1,23	0.5	.49
Time x Subscale	5,19	1.5	.25
Person x Subscale	5,19	2.1	.11
Time x Person x Subscale	5,19	0.8	.54

Note. N = 24. NCCPC-PV: Non-communicating Children's Pain Checklist-Postoperative Version.

Table 8. 10

Correlations between Total NCCPC-PV Postoperative Scores, Child Characteristics and Surgical Factors

Demographic Characteristic	Total NCCPC-PV Score	
	Caregiver	Researcher
Chronological Age (months)[a]	.30	.29
Communication [a,b]	.26	.55*
Daily Living Skills [a,b]	.06	.36
Socialization [a,b]	.01	.41
Motor Skills [a,b]	.14	.38
Required Regular Medical Monitoring [c]	.10	-.19
Upper Limb Impairment [c]	-.13	-.17
Lower Limb Impairment [c]	-.31	-.38
Time in Surgery (minutes) [a]	.15	.06
Time in Recovery Room (minutes) [a]	-.20	-.23

Note. N = 24. NCCPC-PV: Non-Communicating Children's Pain Checklist - Postoperative Version. [a] Pearson Correlations. [b] Age equivalent on the Vineland Adaptive Behavior Scales[15]. [c] Spearman correlations. *Significant after corrections for multiple tests. From (Breau et al., 2002). Used with Permission.

Table 9. 1

Demographic Characteristics of Families

Characteristic		n	%
Marital Status	Married	18	78.3
Mother's Education	Less than high school	6	26.1
	High school	5	21.7
	Post-secondary	6	26.1
	University	6	26.1
Father's Education	Less than high school	6	26.1
	High school	4	17.4
	Post-secondary	5	21.7
	University	3	13.0
	Information not provided[a]	5	21.7
Number of Children	1	4	17.4
	2	5	21.7
	3	11	47.8
	4	3	13.0
Family Income	< $10,000	2	8.7
	$10,000 to $50,000	17	73.9
	> $50,000	3	13.0
	Information not provided[a]	1	4.3

Note. Information concerning parents of children residing outside the family home not included ($n = 3$); therefore $n = 23$. [a]Ethical guidelines of the IWK Health Centre Research Ethics Board required that participants be notified that providing the information in this table was optional. Some caregivers chose not to provide the information requested and some children belonged to single-parent families.

Table 9. 2

Medical Conditions and Physical Impairments of the Children

Characteristic		n	%
Onset of Neurologic Impairment	Prenatal	17	65.4
	Perinatal	5	19.2
	Postnatal	4	15.4
Level of Diagnosed Mental	Moderate	1	3.8
Retardation	Severe	15	57.7
	Profound	7	26.9
	Information not Available	3	11.5
Cerebral Palsy:		17	65.4
Primary Seizure Type:	Generalized	5	19.2
	Focal	10	38.5
Upper Limb Use:	Full	8	30.8
	Some	11	42.3
	None	7	26.9
Lower Limb Use:	Full	4	15.4
	Some	7	26.9
	None	15	57.7
Visual Impairment	Partial	6	23.1
	Full	9	34.6
Hearing Impairment	Partial	1	3.8
	Full	3	11.5
Requires Medical Monitoring:	None	3	16
	Monthly	14	53.8
	Weekly	3	11.5
	Daily	6	23.1

Characteristic		n	%
Tube Fed		12	46.2
Number of Regular Medications	None	4	15.4
	One	5	19.2
	Two	2	7.7
	Three	3	11.5
	Four	3	11.5
	Five or More	9	34.6

Note. N = 26.

Table 9. 3

Chronological and Adaptive Ages of the Children

Measure	Range	Mean	SD
Chronological Age	3.7 years -19.6 years	11.6 years	4.3
Communication[a]	1 month-31 months	12.6 months	6.9
Daily Living Skills[a]	<1 month-753 months	11.9 months	14.5
Socialization[a]	3 months-93 months	15.8 months	19.6
Motor Skills[a]	<1 month-40 months	6.9 months	10.6

Note. N = 26. [a]Age Equivalents on the Vineland Adaptive Behavior Scales (Sparrow et al., 1984)

Table 9. 4

Reliability of the CFCS Facial Actions

Facial Actions	Reliability
Brow Lower	.73
Squint	.84
Eye Squeeze	.85
Nose Wrinkle	.68
Nasolabial Furrow	.75
Cheek Raiser	.81
Upper Lip raise	.91
Lip Corner Pull	.72
Vertical Mouth Stretch	.85
Horizontal Mouth Stretch	.81
Blink	.83
Flared Nostril	.65
Open Lips	.97

Note. CFCS = Child Facial Coding System (Chambers et al., 1996).

Table 9. 5

Visual Analogue Scale Ratings of Nurse and Researchers

Segment	VAS Pain Ratings[a]					VAS-Sedation Ratings[a]					VAS-Pain and VAS-Sedation Ratings[a,b]
	Nurse		Researcher			Nurse		Researcher			
	Mean	SD	Mean	SD	r	Mean	SD	Mean	SD	r	r
1	7.9	11.0	20.2	15.1	.73*	37.3	22.8	40.0	24.3	.44	-.21
2	6.5	11.9	18.1	20.5	.89*	42.1	21.5	42.6	23.0	.74*	-.05
3	7.9	12.0	24.0	24.6	.83*	33.9	23.6	35.3	23.8	.81*	-.14
4	6.9	12.3	23.2	27.9	.85*	32.2	23.8	35.6	24.6	.75*	-.17
5	8.5	14.7	23.1	25.1	.72*	30.1	22.6	31.9	23.9	.84*	.03

Note. N = 26. * p < .005, significant after corrections for multiple tests. [a]VAS = Visual Analogue Scale. [b]Based on Nurse's Ratings.

Table 9. 6

Total Facial Action Frequency and Mean Facial Action Intensity Across Five Segments

Facial Action	Total Frequency		Percentage of Seconds Present		Mean Intensity	
	Mean	SD	Mean	SD	Mean	SD
Brow Lower	12.0	12.4	27	26	.39	.45
Squint	13.7	13.1	18	19	.28	.32
Eye Squeeze	9.0	9.7	38	29	.44	.38
Nose Wrinkle	18.6	14.6	14	21	.22	.36
Nasolabial Furrow	7.0	10.5	26	24	.42	.44
Cheek Raiser	12.8	12.2	24	25	.35	.41
Upper Lip raise	34.0	18.6	68	37	1.04	.64
Lip Corner Pull	2.3	4.3	01	01	.06	.11
Vertical Mouth Stretch	29.2	17.7	58	35	.80	.59
Horizontal Mouth Stretch	24.0	16.8	48	34	.67	.54
Blink*	2.4	3.5	01	01	-	-
Flared Nostril*	7.7	11.1	15	22	-	-
Open Lips*	40.8	15.4	82	31	-	-

Note. N = 26. * Not coded for intensity.

Table 9, 7

Total Facial Action Frequency and Mean Facial Action Intensity Per Segment and Partial Correlations Between Facial Action Frequency and Intensity and Visual Analogue Scale Ratings of Pain and Sedation

Segment	Total Frequency[a]		Mean Intensity[b]		Partial Correlation with VAS Ratings[c]			
					Total Frequency		Mean Intensity	
	Mean	SD	Mean	SD	Pain[d]	Sedation[e]	Pain[d]	Sedation[e]
1	44.6	26.9	.48	.40	.55	.14	.60**	.14
2	40.2	22.6	.44	.37	.71**	.06	.81**	.16
3	43.0	30.1	.48	.45	.78**	-.33	.82**	-.12
4	42.7	29.5	.46	.45	.81**	-.28	.89**	-.30
5	43.4	27.2	.48	.43	.47	.00	.51	-.03

Note. N = 26. **Significant after corrections for multiple tests, $p < .003$. [a] Total for 13 facial actions. [b] Mean of 10 facial actions coded for intensity. [c]Nurse's 100-mm visual analogue scale ratings. [d]Effects of VAS-Sedation ratings partialled out. [e] Effects of VAS-Pain ratings partialled out.

Table 9. 8

Categorical Principal Components Analysis of Facial Action Scores Across Five Segments

Variables Entered	Component Loadings		
	1	2	3
Brow Lower	.83		
Squint		.36	.72
Eye Squeeze	.48	-.32	-.48
Nose Wrinkle	.86	-.31	
Nasolabial Furrow	.88		
Cheek Raiser	.83		
Upper Lip raise	.60	.54	
Lip Corner Pull		.37	
Vertical Mouth Stretch	.55	.55	
Horizontal Mouth Stretch	.82		
Blink*			.75
Flared Nostril*	.87	-.31	
Open Lips*	.36	.68	
Eigenvalue	5.4	1.8	1.5
Percentage of Total Variance Accounted For	41.7	13.9	11.7

Note. N = 26. Facial actions with loadings ≥ .30 shown.

Table 9. 9

Loadings of Supplemental Child Characteristic Variables onto Facial Action Components

Supplemental Variables Entered	Component Loadings		
	1	2	3
Age (months)	-.08	.04	.08
Gender	.17	-.19	-.05
Level of Diagnosed Mental Retardation	.24	.12	-.02
Communication Age Equivalent (months)	-.17	.03	.20
Daily Living Skills Age Equivalent (months)	-.11	.00	.00
Socialization Age Equivalent (months)	-.08	.06	.00
Motor Skills Age Equivalent (months)	-.16	-.07	.08
Cerebral Palsy	-.03	.18	-.15
Seizures	.00	.14	-.26
Upper Limb Impairment	.37	.26	-.04
Lower Limb Impairment	.14	.04	.01
Tube Feeding	.27	.07	.06
Frequency of Medical Monitoring	.18	-.02	-.12

Note. N = 26.

Table 9. 10

Validation Categorical Principal Components Analysis of the Facial Action Components and Surgical Factors

Variables Entered	Components Extracted		
	1	2	3
Facial Action Component 1	.19	.84	.24
Facial Action Component 2	.43	.08	-.59
Facial Action Component 3	-.47	.25	-.32
VAS Pain Rating	.17	.86	.30
VAS Sedation Rating	.33	-.37	.66
Intravenous Opioids Intraoperatively	-.44		.61
Analgesic in Recovery Room (none, non-opioid, opioid)	-.07	.52	-.26
Length of Surgery (minutes)	.81	.00	.11
Length of Time in Recovery Room (minutes)	.28	-.12	.15
Any Dental Surgery	.90	.04	-.11
Eigenvalue	2.3	1.9	1.5
Percentage of Total Variance Accounted For	23.1	19.5	15.2

APPENDIX A: MEASURES AND MATERIALS USED FOR RECRUITMENT

Sample Recruitment Letter to Families Who had Participated in a Previous Study Conducted by the Pediatric Pain Lab

Date

Mr. and Mrs. Murphy
101 Maple Street
Your Town, Nova Scotia

Dear Mr. and Mrs. Murphy;

Thank you for your help last year with our study of how nonverbal children show they are in pain. With your help, we learned that our pain checklist helps parents and caregivers know if their nonverbal child is in pain. We are now starting a new study to learn if the checklist also helps adults know when nonverbal children have pain in the hospital or during needles. As part of this new study, we also want to learn how pain inteferes the normal activities of children like Johnny, and how often they are in pain. We are asking families who visit the IWK Grace Neurology Clinic to take part in this study.

The people involved in the study include: myself, a Clinical Psychology PhD student, Dr. Carol Camfield of the Child Neurology Division, Dr. Patrick McGrath, a psychologist with the Pain Service, and Dr. Allen Finley, an anesthesiologist who also works with the Pain Service. The doctors of the Neurology Division have also agreed to help us.

In this study in which we learn from watching Johnny, and no treatments are involved. Taking part would mean letting us call you to complete an interview about the things Johnny does in day-to-day activities. Then, during a visit to the Neurology Clinic, you would be asked to fill out questionnaires and let us test Johnny's verbal and mental ability. When this is done, we would mail a diary to you every few months that requires checking off behaviours Johnny might have shown each day. This would take about 20 minutes each day. Finally, we would call you every few months and ask about the 2-3 most recent times Johnny was in pain.

Please consider joining our study. I will be calling to to answer any questions you might have and to ask if you would like to take part. If you would like to know more before I contact you, call me at: 902-494-1938 (please call collect if it is long distance) or you may also contact Dr. Carol Camfield at: 428-8479.

Lynn Breau,
Clinical Psychology Ph.D. Student
Dept. of Psychology, Dalhousie University

Carol Camfield, MD
Professor of Pediatrics
Division of Child Neurology
IWK Grace Health Centre

Note. Originals printed on IWK Health Centre Letterhead.

Recruitment Letter to Families Who were Contacted Previously

Regarding Participation in Another Study Conducted by the Pediatric Pain Lab

Date

Ms. Susan Smith
22 Forest Street
Blue Bay Nova Scotia

Dear Ms. Smith;

We are contacting you because you expressed an interest in a study we conducted two years ago, into how children with speech and mental problems show they are in pain. With the help of many families from the IWK Grace Neurology Clinic, we learned that our pain checklist helps parents and caregivers know if their child is in pain. We are now starting a new study to learn if the checklist also helps adults know when children with little speech have pain in the hospital or during needles. As part of this new study, we also want to learn how pain inteferes the normal activities of children like Jill, and how often they are in pain. We are asking families who visit the IWK Grace Neurology Clinic to take part in this new study. We hope this information will help highlight the fact that these children may experience pain often, and that help in recognizing their pain is important to their quality of life.

The people involved in the study include: myself, a Clinical Psychology PhD student, Dr. Carol Camfield of the Child Neurology Division, Dr. Patrick McGrath, a psychologist with the Pain Service, and Dr. Allen Finley, an anesthesiologist who also works with the Pain Service. The doctors of the Neurology Division have also agreed to help us.

In this study, we will learn from watching Jill, and no treatments are involved. Taking part would mean letting us call you to complete an interview about the things Jill does in day-to-day activities. Then, during a visit to the Neurology Cli nic, you would be asked to fill out questionnaires and let us test Jill's verbal and mental ability. When this is done, we would mail a diary to you every few months that requires checking off behaviours Jill might have shown each day. This would take about 20 minutes each day. Finally, we would call you every few months and ask about the 2-3 most recent times Jill was in pain.

Please consider joining our study. I will be calling to to answer any questions you might have and to ask if you would like to take part. If you would like to know more before I contact you, call me at: 902-494-1938 (please call collect if it is long distance) or you may also contact Dr. Carol Camfield at: 428-8479.

Lynn Breau,
Clinical Psychology PhD Student
Dept. of Psychology, Dalhousie University

Carol Camfield, MD
Professor of Pediatrics
Division of Child Neurology
IWK Grace Health Centre

Note. Originals printed on IWK Health Centre Letterhead.

Sample Recruitment Letter to Families Had Never been Contacted Regarding Participation in a Study Conducted by the Pediatric Pain Lab

Date

Bill and Linda Wilson
Pine Drive
Lakeside, P.E.I., C2B 7P1

Dear Mr. & Mrs. Wilson

I am writing on behalf of the Pediatric Pain Lab, to let you know about a study that is beginning with children who are seen at the IWK Grace Neurology Clinic. The study is being carried out to learn how children who have problems communicating by speech show parents and caregivers that they are in pain. Absolutely no pain will be inflicted upon the children in the study. It involves us asking questions about what children do when something painful happens at home or when they are in the hospital.

The people involved in the study include: myself, a Clinical Psychology PhD student, Dr. Carol Camfield, of the Child Neurology Division, Dr. Patrick McGrath, a psychologist with the Pain Service, and Dr. Allen Finley, an anesthesiologist who also works with the Pain Service. Our research team also includes: Alyson Currie, Heather Wortman and Jill MacLaren. The doctors of the Neurology Division have also agreed to help us, and have given us the names of children's families who might be interested in taking part in the study.

In this study we would learn from watching Chris, and no treatments are involved. Taking part would mean letting us call you to complete an interview about the things Chris does in day-to-day activities. Then, during a visit to the Neurology Clinic, you would be asked to let us test Chris's verbal and mental ability, if Chris is able to get across "yes" and "no" in some way (speech or gestures). When this is done, we would mail a diary to you every few months that requires checking off behaviours Chris might have shown each day. This would take approximately 20 minutes each day. Finally, we would call you every few months and ask about the latest times Chris was in pain.

We are hoping that you might think about taking part in our study. This letter to let you know that Alyson or I will be Calling to let you know more and to ask if you would like to take part. If you would like to know more before we contact you, please call Alyson or myself at: 902-428-2702 (Collect if necessary). You may also contact Carol Camfield at: 902-428-8479.

Lynn Breau
Clinical Psychology PhD Student
Dept. of Psychology, Dalhousie University
IWK Grace Health Centre

Note. Originals printed on IWK Health Centre letterhead.

APPENDIX B: MEASURES AND MATERIALS USED AT ENTRY TO THE LONGITUDINAL PROGRAM

Informed Consent Form Used by Caregivers

STUDY TITLE: LONGITUDINAL STUDY OF THE PAIN EXPRESSION OF COGNITIVELY IMPAIRED CHILDREN

INVESTIGATORS:

Lynn Breau, Clinical Psychology PhD. Student, Dalhousie University
Dr. Patrick McGrath, Psychologist, IWK Grace Pediatric Pain Service
Dr. Carol Camfield, Pediatrician, IWK Grace Division of Neurology
Dr. G. Allen Finley, Anesthetist, IWK Grace Dept. Of Anesthesiology

INTRODUCTION: You and your child are being asked to take part in a study to describe the way children with mental and verbal deficits show that they are in pain. This will be done by having caregivers record how their child behaves during naturally occurring painful events.

PURPOSE OF THIS STUDY: This study may provide us with basic information that does not currently exist such as: how do children who have no ability to speak show their pain, how often are these children in pain, how does their pain affect their daily life?

STUDY DESIGN: This is an observational study. No treatments are involved.

WHO CAN PARTICIPATE IN THIS STUDY:

You and your child are able to be a part of this study if:

Your child is a patient of the Neurology Clinic.
Your child is between the ages of 3-18 years.
Your child has cognitive impairments and speech deficits.
You have been the primary caregiver of your child for at least 6 months.
You speak English.

PROCEDURES OF THE STUDY:

This is a 2 year study. If you agree to take part you will be asked to do the following when you enter the study:

1. To complete an information sheet concerning your child's background and health history and to allow your physician to provide the researcher with your child's diagnosis. The information sheet includes demographic questions, such as your

education and general income range. These are included because they are sometimes related to how a child shows pain, and for us to know if the group of caregivers who are taking part are representative of the general public. However, you are free to skip any questions you are uncomfortable answering.

2. To complete an inventory over the phone that measures your child's ability to communicate. This will take about 30 minutes.
3. To complete an interview over the phone concerning your child's life skills. This will take about 60 minutes.
4. If you and the researcher judge that your child is capable, to have your child complete a test of verbal ability and a test of cognitive ability at this hospital or Dalhousie University. These 2 tests each take about 15 minutes to complete. If at all possible, this will be done when you already have plans to be at the hospital. If you cannot arrange to be at the hospital, a researcher may visit your home to do the tests.

Once you are enrolled in the study, you will be asked to:

1. Let a researcher phone you every 3rd month (8 times over 2 years). They will ask you questions about any pain your child has experienced in the previous week.
2. Complete a one-week diary every third month (8 times over 2 years). This will be mailed to you and will take approximately 20 minutes per day to complete. You will be supplied with a stamped envelope to mail it back once it is finished.
3. Let the researcher who calls you ask if your child is scheduled for surgery or vaccination in the near future. If so, let them tell you about additional studies you may wish to participate in.

After one year you will be asked to:

1. Let an interviewer conduct the phone interview about your child's life skills and verbal abilities again. This should take about 60 minutes.
2. Have your child repeat the tests of verbal and cognitive abilities if they completed them when they entered the study.

RISKS AND DISCOMFORTS: There are no risks or discomforts for you or your child if you agree to take part in this study.

POSSIBLE BENEFITS: There are no direct benefits for your child if they take part in this study. However, the information gained from this study may help caregivers and health professionals provide better pain management for cognitively impaired children in the future.

COMPENSATION: There will be no costs to your family for being in this study.

CONFIDENTIALITY: Neither you nor your child will be named in any reports or publications based on this research. All information collected from you will be stored in a locked cabinet at the Pediatric Pain Laboratory, Psychology Department, Dalhousie University. Only staff immediately involved in the research will have access to the information you give us.

QUESTIONS OR PROBLEMS: If you have any questions about this study, please feel free to contact Lynn Breau at 494-1938. You may also contact the IWK Grace Research Services Office at 428-8765 if you would like additional information from an outside source.

OTHER PERTINENT INFORMATION: You are free to withdraw from this study at any point without it affecting the care your child receives. You are also free to ask for the results of any of the tests, interviews, diaries concerning your child. At completion of this project, a summary of the results will be sent to everyone who took part.

I, _____ agree to take part in the study entitled: **LONGITUDINAL STUDY OF THE PAIN EXPRESSION OF COGNITIVELY IMPAIRED CHILDREN** being conducted by: Lynn Breau, Dr. Patrick McGrath, Dr. Carol Camfield, and Dr. G. Allen Finley. I also give consent for my child _____ to take part in this study. I have been informed of the purpose and requirements of the study. I understand that I may withdraw at any point without it affecting my child's care.

Signature: _____ Date: _____

Note. Originals printed on IWK Health Centre letterhead.

Demographic Information and Medical History Sheet

Child's Name:

Date sheet completed:

Child's date of birth: Day: _____ Month: _____ Year: _____

Child's gender: ☐ male ☐ female

Previous Hospitalizations including General Anesthesia: ☐ 1-3 ☐ 4-6 ☐ 6-10 ☐ >10

Previous Day Surgeries: ☐ 1-3 ☐ 4-6 ☐ 6-10 ☐ >10

Is your child currently taking any medications: ☐ Yes ☐ No

If yes:

Medication	Dose	Prescribed for

Is your child currently experiencing pain on a regular basis? ☐ Yes ☐ No

If yes, is this pain acute or chronic? ☐ Acute ☐ Chronic

Does your child have any physical conditions which require medical monitoring on a:

☐ monthly basis ☐ weekly basis ☐ daily basis ☐ not at all

Does your child have (please check the phrase that best describes your child):

- ☐ full upper limb function
- ☐ limited upper limb function requiring brace/prosthesis
- ☐ use of one upper limb
- ☐ use of neither upper limb

Does your child have (please check the phrase that best describes your child):
- ☐ full lower limb function
- ☐ limited lower limb function requiring ambulatory aids, brace, prosthesis
- ☐ limited lower limb function requiring use of a wheelchair but no assistance
- ☐ no lower limb function

Does your child have a hearing impairment (please check the phrase that best describes your child)?
- ☐ Partial hearing loss (please describe):
- ☐ Total Hearing loss

Does your child have a visual impairment (please check the phrase that best describes your child)?
- ☐ Partial visual loss(please describe):
- ☐ Total visual loss

Does your child have motor deficits which impair his/her speech? ☐ Yes ☐ No

If yes, please describe the deficit and how it affects his/her speech:

Does your child use a nonverbal means of communication: ☐ Yes ☐ No
If yes, please describe:

Family Information:

Parents' Marital Status: ☐ Married ☐ Separated

	Mother	Father
Age:		
Occupation:		
Education:	☐ Less than Highschool ☐ Highschool completed ☐ Post-Secondary ☐ University	☐ Less than Highschool ☐ Highschool completed ☐ Post-Secondary ☐University

Approximate Family Income:

☐ Less than $10,000 ☐ $10,000 - $50,000 ☐ more than $50,000

Number of Children in the Family: ☐ 1 ☐ 2 ☐ 3 ☐ 4+

This child is the (please circle one): ☐ 1st ☐ 2nd ☐ 3rd ☐ 4th+

Non-communicating Children's Pain Checklist-Revised: Retrospective Report

Name of child : _____Completed by: _____

How often has your child usually shown these behaviours when they have been in pain in the past? Please circle a number for each behaviour.

	Not at all	Just a little	Fairly often	Very often
Moaning , whining, whimpering (Fairly soft)	0	1	2	3
Crying (Moderately loud)	0	1	2	3
Screaming/yelling (Very loud)	0	1	2	3
A specific sound or word for pain (for example: a word, cry, or type of laugh)	0	1	2	3
Eating less, not interested in food	0	1	2	3
Increase in sleep	0	1	2	3
Decrease in sleep	0	1	2	3
Not co-operating, cranky, irritable, unhappy	0	1	2	3
Less interaction with others, withdrawn	0	1	2	3
Seeking comfort or physical closeness	0	1	2	3
Being difficult to distract, not able to satisfy or pacify	0	1	2	3
A furrowed brow	0	1	2	3
A change in eyes, including: squinching of eyes, eyes opened wide; eyes frowning	0	1	2	3
Turning down of mouth, not smiling	0	1	2	3
Lips puckering up, tight, pouting or quivering	0	1	2	3
Clenching or grinding teeth, chewing or thrusting tongue out	0	1	2	3
Not moving, less active, quiet	0	1	2	3
Jumping around, agitated, fidgety	0	1	2	3
Floppy	0	1	2	3
Stiff, spastic, tense, rigid	0	1	2	3
Gesturing to or touching part of the body that hurt	0	1	2	3
Protecting, favouring or guarding part of the body that hurts	0	1	2	3

	Not at all	Just a little	Fairly often	Very often
Flinching or moving the body part away, being sensitive to touch	0	1	2	3
Moving the body in specific way to show pain (e.g. head back, arms down, curls up etc.)	0	1	2	3
Shivering	0	1	2	3
Change in colour, pallor	0	1	2	3
Sweating, perspiring	0	1	2	3
Tears	0	1	2	3
Sharp intake of breath, gasping	0	1	2	3
Breath holding	0	1	2	3

APPENDIX C: MEASURES USED AS PART OF THE LONGITUDINAL PROGRAM

Telephone Survey Script & Record Sheet

To be completed by Researcher:

Date: Time of call: Survey #:
Name of child: Name of Researcher:
Name of caregiver responding:

Hi, I'm _____*(Researcher's name)* calling from the Pediatric Pain Lab.
I've called to administer the _____ *(1^{st}, 2^{nd},...)* Pain Survey to you.

Is this a convenient time for you? *(If "yes" proceed. If "no" arrange another time.)*

OK. I'm going to ask you some questions about the past 7 days only. So that would be as far back as last _____ *(Monday, Wednesday,...)*.

Has _____ had any pain since then? ☐ Yes ☐ No

If "no"	go to "Thank you" section. ☺
If "yes"	Could you please tell me about it?
(If more than once)	Could you please tell me about the first time?

What day did it happen? Date:
Could you describe it to me?

What do you think caused it?
How long did it last? Hours_____ Minutes _____

I'd like you to think about how strong the pain was for _____. If zero means "no pain at all" and 10 means the "worst pain ever" How strong do you think that his/her pain was? Answer: _____

Did you try to do anything to relieve his/her pain? ☐ Yes ☐ No

If "no"	*go to "Any other painful incidents" section* ☹
If "yes"	Could you tell me about that? What did you do?

I'd like you to think about how helpful that might have been for relieving _____'s pain. If 0 means it "didn't help at all" and 10 means it "completely relieved the pain", how helpful do you think that was? Answer _____

Did you try anything else? ☐ Yes ☐ No

If "no" *go to "any other painful incidents" section.* ☹

If "yes" Could you tell me about that? What did you do?

I'd like you to think again about how helpful that might have been for relieving _____'s pain. If 0 means it "didn't help at all" and 10 means it "completely relieved the pain", how helpful do you think that was? Answer _____

☹ Do you remember any other times _____ had pain since last _____*(day of week)?*

If "no" *go to "Thank you" section.* □
If "yes" *go to Supplementary Sheets.*

☺ Thank you very much for your time. Is _____ scheduled for surgery or a vaccination in the next 3 months?

If yes tell about relevant satellite study.

If "no" Have you any questions about this survey or the diaries you have been receiving in the mail? *(Answer appropriately)*

(If child has not completed second assessment).

Do you recall the date of your second assessment for _____? *(If "no" remind him/her of correct date).*

Thank you again for your time. We'll call you back and ask these questions in 3 months. If you have any questions in the meantime, please feel free to call.

Supplementary Pain Survey Sheet

Sheet # ___ of ___
To be completed by Researcher:

Date:
Time of call: Survey #:
Name of child:
Name of Researcher:
Name of caregiver responding:

Could you describe it to me?

What do you think caused it?

How long did it last? Hours_____ Minutes _____

I'd like you to think about how strong the pain was for _____. If zero means "no pain at all" and 10 means the "worst pain ever" How strong do you think that his/her pain was? Answer: _____

Did you try to do anything to relieve his/her pain? ☐ Yes ☐No

If "no" Do you remember any other times _____ had pain since last _____*(day of week)?*

If :no" *go to "Thank you" section.* ☺
If "yes" *go to next Supplemental Sheet.*

If "yes"
Could you tell me about that?

What did you do?

I'd like you to think about how helpful that might have been for relieving _____'s pain. If 0 means it "didn't help at all" and 10 means it "completely relieved the pain", how helpful do you think that was? Answer _____

Did you try anything else? ☐ Yes ☐No

*If "no"*Do you remember any other times _____ had pain since last _____*(day of week)?*

If no" *go to "Thank you" section☺*
If "yes" *go to next Supplemental Sheet.*

If "yes" Could you tell me about that? What did you do?

I'd like you to think again about how helpful that might have been for relieving _____'s pain. If 0 means it "didn't help at all" and 10 means it "completely relieved the pain", how helpful do you think that was? Answer _____

Do you remember any other times _____ had pain since last _____*(day of week)?*

If no" *go to "Thank you" section. ☺*
If "yes" *go to additional Supplementary Sheets.*

☺ Thank you very much for your time. Is _____ scheduled for surgery or a vaccination in the next 3 months?

If yes *tell about relevant satellite study.*

If "no" Have you any questions about this survey or the diaries you have been receiving in the mail? *(Answer appropriately)*

(If child has not completed second assessment).

Do you recall the date of your second assessment for _____? *(If "no" remind him/her of correct date).*

Thank you again for your time. We'll call you back and ask these questions in 3 months. If you have any questions in the meantime,
please feel free to call.

Sample Pain Diary Cover Letter and Day

PAIN IN COGNITIVELY IMPAIRED CHILDREN

Dear Parent or Caregiver:

This booklet contains a checklist to be completed every day for 7 days. Please pick a 2 hour period and use that same period for each day you complete the checklist. For example, if you complete the checklist for the period beginning at 5:00 pm and ending at 7:00 pm, please use that same period each day. A space is provided below to record the 2 hour period you have chosen:

The period of time I completed this booklet for was:

From: _____ To: _____

Your child may not have pain each day you complete the checklist, but please complete it anyway. If, for some reason, you forget or are unable to complete the checklist for a day, do not try to fill it in the following day. Just move on to the next day.

Thanks!

DAY 1

Today is: Monday Tuesday Wednesday Thursday

Friday Saturday Sunday

Section 1

Did Patty have any pain today? ☐Yes ☐No

What was this pain due to:
☐ injury ☐ chronic condition ☐ illness ☐ medical procedure

How long did this pain last? ____hours ____minutes

How strong was this pain? Please circle one:

0	1	2	3	4	5	6	7	8	9	10
None At all										Worst possible

SECTION 2

Please complete this section even if Patty had NO pain today.

How often did Patty show these behaviours today? Please circle a number for each behaviour.

	Not at all	Just a little	Fairly often	Very often
Moaning , whining, whimpering (Fairly soft)	0	1	2	3
Crying (Moderately loud)	0	1	2	3
Screaming/yelling (Very loud)	0	1	2	3
A specific sound or word for pain (for example: a word, cry, or type of laugh)	0	1	2	3
Not co-operating, cranky, irritable, unhappy	0	1	2	3

	Not at all	Just a little	Fairly often	Very often
Less interaction with others, withdrawn	0	1	2	3
Seeking comfort or physical closeness	0	1	2	3
Being difficult to distract, not able to satisfy or pacify	0	1	2	3
A furrowed brow	0	1	2	3
A change in eyes, including: squinching of eyes, eyes opened wide; eyes frowning	0	1	2	3
Turning down of mouth, not smiling	0	1	2	3
Lips puckering up, tight, pouting or quivering	0	1	2	3
Clenching or grinding teeth, chewing or thrusting tongue out	0	1	2	3
Not moving, less active, quiet	0	1	2	3
Jumping around, agitated, fidgety	0	1	2	3
Floppy	0	1	2	3
Stiff, spastic, tense, rigid	0	1	2	3
Gesturing to or touching a part of the body that hurt	0	1	2	3
Protecting, favouring or guarding a part of the body that hurt	0	1	2	3
Flinching or moving the body part away, being sensitive to touch	0	1	2	3
Moving the body in specific way to show pain (e.g. head back, arms down, curls up etc.)	0	1	2	3
Shivering	0	1	2	3
Change in colour, pallor	0	1	2	3
Sweating, perspiring	0	1	2	3
Tears	0	1	2	3
Sharp intake of breathe, gasping	0	1	2	3
Breath holding	0	1	2	3

Please indicate whether any of the following occurred anytime today:

	Not at all	Just a little	Fairly often	Very often
Eating less, not interested in food	0	1	2	3
Increase in sleep	0	1	2	3
Decrease in sleep	0	1	2	3

SECTION 3

Please complete this section even if Patty had NO pain today.

Did Patty show these behaviours today? Please circle "yes" or "no".

Turn eyes and head toward sound.	Yes	No
Smile in response to presence of a caregiver.	Yes	No
Willingly allow caregiver to wipe nose.	Yes	No
Look at face of caregiver.	Yes	No
Respond to voice of caregiver or another person.	Yes	No
Express two or more recognisable emotions such as pleasure, sadness, fear, or distress.	Yes	No
Hold head erect for at least 15 seconds without assistance.	Yes	No
Sit supported for at least one minute.	Yes	No

Is there anything else that Patty usually does that she did not do today? (E.g. Usually brushes her own hair and did not today).

Is there anything Patty did today that she has never done before? (E.g. Has never co-operated in bath before, but was co-operative today).

APPENDIX D: MEASURES USED IN VALIDATION OF THE NON-COMMUNICATING CHILDREN'S PAIN CHECKLIST-POSTOPERATIVE VERSION

Consent to Participate in Study of Postoperative Pain

STUDY TITLE: POST-OPERATIVE PAIN IN COGNITIVELY IMPAIRED CHILDREN

INVESTIGATORS:

Lynn Breau, Clinical Psychology PhD. Student,
Dalhousie University and Pediatric Pain Research Lab, IWK Grace
Dr. Patrick McGrath, Psychologist,
IWK Grace Pediatric Pain Service, Co-Director IWK Grace Pediatric Pain Research Lab
Dr. Carol Camfield, Pediatrician,
IWK Grace Division of Neurology
Dr. G. Allen Finley, Anesthetist,
IWK Grace Dept. Of Anesthesiology and Pediatric Pain Servic,e Co-Director IWK Grace Pediatric Research Pain Lab

INTRODUCTION: You and your child are being asked to take part in a study to describe the way children with mental and verbal deficits show that they are in pain after surgery.

PURPOSE OF THIS STUDY: This study may let us know if the pain following surgery can be measured by the facial expression and behaviour of children with mental and speech problems.

STUDY DESIGN: No treatments are involved. No pain will be inflicted. We will learn from watching your child; this is called an observational study.

WHO CAN PARTICIPATE IN THIS STUDY:

You and your child are able to be a part of this study if:

Your child is a patient of the Neurology Clinic.
Your child is between the ages of 3-18 years.
Your child has cognitive impairments and speech deficits.
You have been the primary caregiver of your child for at least 6 months.
You speak English.
Your child is currently enrolled in the Longitudinal Study of the Pain Expression of Cognitively Impaired Children.

PROCEDURES OF THE STUDY:

If you agree to take part in this study, you will be asked to:

1) Provide a researcher with the time and location of your child's scheduled surgery and allow us to contact his/her physician to make arrangements to conduct the study.

2) On the morning before surgery: watch your child for 10 minutes, and then complete a checklist of behaviours you may have seen. You will also be asked to rate how strong your child's pain was during those 10 minutes. You will also be asked to let a researcher watch your child, fill out the checklist, and rate his/her pain, and to let your child's nurse rate his/her pain.

3) Allow a researcher to videotape your child's face while they are in the recovery room. This will be from the time the nurse says he/she is awake until they leave the recovery room (maximum 60 minutes).

4) Repeat the 10 minute observation, checklist, and pain rating later that day after surgery has been completed. This will be done once, just before your child is given pain medication, and a second time after the medication has begun to work. You will also be asked to let the researcher and nurse repeat watching your child and completing their ratings at these times.

5)Repeat the 10 minute observation, checklist, and pain rating once on the second day your child is in the hospital, and once on the third day your child is in the hospita (if they are still in the hospital). This will be done just before pain medication is given. Allow the researcher and nurse to complete their observations and ratings on the second and third day your child is in the hospital.

6) Allow the researcher to get information from his/her hospital chart concerning your child's surgery and stay in the hospital, such as the type of surgery, the medications they received, other painful procedures your child had during the hospital stay.

RISKS AND DISCOMFORTS: There are no risks or discomforts for you or your child if you agree to take part in this study.

POSSIBLE BENEFITS: There are no direct benefits for your child if they take part in this study. However, the information gained from this study may help caregivers and health professionals provide better pain management for cognitively impaired children in the future.

COMPENSATION: There will be no costs to your family for being in this study.

CONFIDENTIALITY: Neither you nor your child will be named in any reports or publications based on this research. All videotapes and information collected from you will be stored in a locked cabinet at the Pediatric Pain Laboratory, Psychology Department, Dalhousie University. Only staff immediately involved in the research will have access to the videotapes and information you give us.

QUESTIONS OR PROBLEMS: If you have any questions about this study, please feel free to contact Lynn Breau at 494-1938. You may also contact the IWK Grace Research Services Office at 428-8765 if you would like additional information from an outside source.

OTHER PERTINENT INFORMATION: You are free to withdraw from this study at any point without it affecting the care your child receives. At completion of this project, a summary of the results will be sent to everyone who took part.

I, _____ agree to take part in the study entitled: **POST-OPERATIVE PAIN IN COGNITIVELY IMPAIRED CHILDREN** being conducted by: Lynn Breau, Dr. Patrick McGrath, Dr. Carol Camfield, and Dr. G. Allen Finley. I also give consent for my child _____ to take part in this study. I have been informed of the purpose and requirements of the study. I understand that I may withdraw at any point without it affecting my child's care.

Signature: _____ Date: _____

Consent Obtained by: _____ Date: _____

Note. Original printed on IWK Health Centre letterhead.

Consent to Participate in Study of Postoperative Pain: Updated October 1999

STUDY TITLE: POST-OPERATIVE PAIN IN COGNITIVELY IMPAIRED CHILDREN

INVESTIGATORS:

Lynn Breau, Clinical Psychology PhD. Student, Dalhousie University and Pediatric Pain Research Lab, IWK Grace
Dr. Patrick McGrath, Psychologist, IWK Grace Pediatric Pain Service, Co-Director IWK Grace Pediatric Pain Research Lab
Dr. Carol Camfield, Pediatrician, IWK Grace Division of Neurology
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You speak English.
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PROCEDURES OF THE STUDY:

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1) Provide a researcher with the time and location of your child's scheduled surgery and allow us to contact his/her physician to make arrangements to conduct the study.

2) On the morning before surgery: watch your child for 10 minutes, and then complete a checklist of behaviours you may have seen. You will also be asked to rate how strong your child's pain was during those 10 minutes. You will also be asked to let a researcher watch your child, fill out the checklist, and rate his/her pain, and to let your child's nurse rate his/her pain.

2) Allow a researcher to videotape your child's face while they are in the recovery room. This will be from the time the nurse says he/she is awake until they leave the recovery room (maximum 60 minutes). They will do this with a hand-held camera, standing about 2 meters away from your child. They will film your child's face only.

4) Repeat the 10 minute observation, checklist, and pain rating later that day after surgery has been completed. This will be done once, just before your child is given pain medication, and a second time after the medication has begun to work. You will also be asked to let the researcher and nurse repeat watching your child and completing their ratings at these times.

5)Repeat the 10 minute observation, checklist, and pain rating once on the second day your child is in the hospital, and once on the third day your child is in the hospital (if they are still in the hospital). This will be done just before pain medication is given. Allow the researcher and nurse to complete their observations and ratings on the second and third day your child is in the hospital.

6) Allow the researcher to get information from his/her hospital chart concerning your child's surgery and stay in the hospital, such as the type of surgery, the medications they received, other painful procedures your child had during the hospital stay.

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POSSIBLE BENEFITS: There are no direct benefits for your child if they take part in this study. However, the information gained from this study may help caregivers and health professionals provide better pain management for cognitively impaired children in the future.

COMPENSATION: There will be no costs to your family for being in this study.

CONFIDENTIALITY: Neither you nor your child will be named in any reports or publications based on this research. All videotapes and information collected from you will be stored in a locked cabinet at the Pediatric Pain Laboratory, Psychology

Department, Dalhousie University. Only staff immediately involved in the research will have access to the videotapes and information you give us.

QUESTIONS OR PROBLEMS: If you have any questions about this study, please feel free to contact Lynn Breau at 428-2702 (Collect if necessary) 0r 494-1938. You may also contact the IWK Grace Research Services Office at 428-8765 if you would like additional information from an outside source.

OTHER PERTINENT INFORMATION: You are free to withdraw from this study at any point without it affecting the care your child receives. At completion of this project, a summary of the results will be sent to everyone who took part.

Also:
If you agree to participate, you are free to stop any activity at any time if you believe your child is not willing to continue or is uncomfortable. If a researcher feels your child is unwilling to participate in any activity, they will also stop. In particular, you are free to ask the researcher to stop filming or watching your child, or to leave the room if at any time you feel your child is made uncomfortable by their presence. The researcher will also stop or leave if they or your child's nurse/doctor believe your child is unwilling to continue or is made uncomfortable by them being there or filming. You are also free to not answer any questions or provide information if you would rather not.

I, _____ agree to take part in the study entitled: POST-OPERATIVE PAIN IN COGNITIVELY IMPAIRED CHILDREN being conducted by: Lynn Breau, Dr. Patrick McGrath, Dr. Carol Camfield, and Dr. G. Allen Finley. I believe my child_____ is willing to take part in this study and give my consent for him/her to participate. I also consent to having a researcher videotape my child's face during their time in the recovery room. I have been informed of the purpose and requirements of the study. I understand that I may withdraw at any point without it affecting my child's care or that I may stop any activity at any point if I wish.

Signature: _____ Date: _____

Consent Obtained by: _____ Date: _____

Note. Original printed on IWK Health Centre letterhead.

Observation Record Sheet - Satellite Study 2: Post-Operative Pain

Name of Child:_____ Observer: _____

Date: Day_____ Month _____ Year _____

Day of Hospitalization: ☐ Day of Surgery ☐ Post-operative Day 1
☐ Post-operative Day 2

Time of observation: Start_____ End_____

How often has this child shown these behaviours in the last 10 minutes? Please circle a number for each behaviour.

	Not at all	Just a little	Fairly often	Very often
Moaning , whining, whimpering (Fairly soft)	0	1	2	3
Crying (Moderately loud)	0	1	2	3
Screaming/yelling (Very loud)	0	1	2	3
A specific sound or word for pain (for example: a word, cry, or type of laugh)	0	1	2	3
Not co-operating, cranky, irritable, unhappy	0	1	2	3
Less interaction with others, withdrawn	0	1	2	3
Seeking comfort or physical closeness	0	1	2	3
Being difficult to distract, not able to satisfy or pacify	0	1	2	3
A furrowed brow	0	1	2	3
A change in eyes, including: squinching of eyes, eyes opened wide; eyes frowning	0	1	2	3
Turning down of mouth, not smiling	0	1	2	3
Lips puckering up, tight, pouting or quivering	0	1	2	3

	Not at all	Just a little	Fairly often	Very often
Clenching or grinding teeth, chewing or thrusting tongue out	0	1	2	3
Not moving, less active, quiet	0	1	2	3
Jumping around, agitated, fidgety	0	1	2	3
Floppy	0	1	2	3
Stiff, spastic, tense, rigid	0	1	2	3
Gesturing to or touching part of the body that hurt	0	1	2	3
Protecting, favouring or guarding part of the body that hurts	0	1	2	3
Flinching or moving the body part away, being sensitive to touch	0	1	2	3
Moving the body in specific way to show pain (e.g. head back, arms down, curls up etc.)	0	1	2	3
Shivering	0	1	2	3
Change in colour, pallor	0	1	2	3
Sweating, perspiring	0	1	2	3
Tears	0	1	2	3
Sharp intake of breathe, gasping	0	1	2	3
Breath holding	0	1	2	3

How strong would you rate the pain experienced by this child during this 10 minute period? Please indicate your rating by putting a mark on the line below:

No Pain At all — Worst Pain Ever

Nurse's Pain Rating Sheet - Satellite Study 2: Post-Operative Pain

Name of Child:_____ Observer: _____

Date: Day_____ Month _____ Year _____

Day of Hospitalization: ❑ Day of Surgery ❑ Post-operative Day 1
❑ Post-operative Day 2

Time of observation: Start_____ End_____

How strong would you rate the pain experienced by this child during this 10 minute period? Please indicate your rating by putting a mark on the line below:

No Pain At all

Worst Pain Ever

Surgical Information Sheet - Satellite Study 2: Post-Operative Pain

Child's Name _____ Completed by:_____

Type of procedure:

Length of procedure:

Length of time in recovery room (minutes):

General anesthesia during surgery? Yes No

Intravenous Opiods during surgery? Yes No

Were there any unusual occurrences related to this surgery?

Hospitalization Information Sheet - Satellite Study 2: Postoperative Pain

Name of Child:

Date admitted:

Reason for admission:

Illness Status: ☐ chronic ☐ acute

Date released:

Date of Surgery(dd/mm/yy):

Type of surgery:

Post-Operative Analgesics

Day of Surgery:

Analgesic type:	Prescribed	Administered
Opiod		
Non-opiod		

Post-Operative Day 1:

Analgesic type:	Prescribed	Administered
Opiod		
Non-opiod		

Post-Operative Day 2:

Analgesic type:	Prescribed	Administered
Opiod		
Non-opiod		

Invasive Procedures Performed

Type	Number of each procedure performed:		
	Day of Surgery	Post-op Day 1	Post-op Day 2
Finger prick			
IV insertion			
Venous bloodwork			
Nasogastric tube insertion			
Intramuscular injection			
Subcutaneous injection			
Urinary catheter			
Lumbar puncture			
Bone marrow aspiration			
Other:			

Unusual Occurrences During Hospitalization:

APPENDIX E: MEASURES USED IN THE STUDY USING FACIAL ACTION TO DETECT POSTOPERATIVE PAIN IN CHILDREN WITH SEVERE COGNITIVE IMPAIRMENTS

Consent to Participate in Study of Postoperative Pain

STUDY TITLE:
POST-OPERATIVE PAIN IN
COGNITIVELY IMPAIRED CHILDREN

INVESTIGATORS:

Lynn Breau, Clinical Psychology PhD. Student, Dalhousie University and Pediatric Pain Research Lab, IWK Grace
Dr. Patrick McGrath, Psychologist, IWK Grace Pediatric Pain Service, Co-Director IWK Grace Pediatric Pain Research Lab
Dr. Carol Camfield, Pediatrician, IWK Grace Division of Neurology
Dr. G. Allen Finley, Anesthetist, IWK Grace Dept. Of Anesthesiology and Pediatric Pain Servic,e Co-Director IWK Grace Pediatric Research Pain Lab

INTRODUCTION: You and your child are being asked to take part in a study to describe the way children with mental and verbal deficits show that they are in pain after surgery.

PURPOSE OF THIS STUDY: This study may let us know if the pain following surgery can be measured by the facial expression and behaviour of children with mental and speech problems.

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Your child is a patient of the Neurology Clinic.
Your child is between the ages of 3-18 years.
Your child has cognitive impairments and speech deficits.
You have been the primary caregiver of your child for at least 6 months.
You speak English.
Your child is currently enrolled in the Longitudinal Study of the Pain Expression of Cognitively Impaired Children.

PROCEDURES OF THE STUDY:

If you agree to take part in this study, you will be asked to:

1) Provide a researcher with the time and location of your child's scheduled surgery and allow us to contact his/her physician to make arrangements to conduct the study.

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3) Allow a researcher to videotape your child's face while they are in the recovery room. This will be from the time the nurse says he/she is awake until they leave the recovery room (maximum 60 minutes).

4) Repeat the 10 minute observation, checklist, and pain rating later that day after surgery has been completed. This will be done once, just before your child is given pain medication, and a second time after the medication has begun to work. You will also be asked to let the researcher and nurse repeat watching your child and completing their ratings at these times.

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POSSIBLE BENEFITS: There are no direct benefits for your child if they take part in this study. However, the information gained from this study may help caregivers and health professionals provide better pain management for cognitively impaired children in the future.

COMPENSATION: There will be no costs to your family for being in this study.

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Department, Dalhousie University. Only staff immediately involved in the research will have access to the videotapes and information you give us.

QUESTIONS OR PROBLEMS: If you have any questions about this study, please feel free to contact Lynn Breau at 494-1938. You may also contact the IWK Grace Research Services Office at 428-8765 if you would like additional information from an outside source.

OTHER PERTINENT INFORMATION: You are free to withdraw from this study at any point without it affecting the care your child receives. At completion of this project, a summary of the results will be sent to everyone who took part.

I, _____ agree to take part in the study entitled: **POST-OPERATIVE PAIN IN COGNITIVELY IMPAIRED CHILDREN** being conducted by: Lynn Breau, Dr. Patrick McGrath, Dr. Carol Camfield, and Dr. G. Allen Finley. I also give consent for my child _____ to take part in this study. I have been informed of the purpose and requirements of the study. I understand that I may withdraw at any point without it affecting my child's care.

Signature: _____ Date: _____

Consent Obtained by: _____ Date: _____

Note. Original printed on IWK Health Centre letterhead.

Consent to Participate in Study of Postoperative Pain: Updated October 1999

STUDY TITLE: POST-OPERATIVE PAIN IN COGNITIVELY IMPAIRED CHILDREN

INVESTIGATORS:

Lynn Breau, Clinical Psychology PhD. Student,
Dalhousie University and Pediatric Pain Research Lab, IWK Grace
Dr. Patrick McGrath, Psychologist,
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Signature: _____ Date: _____

Consent Obtained by: _____ Date: _____

Note: Original printed on IWK Health Centre letterhead.

Facial Actions of the Child Facial Coding System[a]

Facial Actions	Possible Score
Brow Lower	0-2
Squint	0-2
Eye Squeeze	0-2
Nose Wrinkle	0-2
Nasolabial Furrow	0-2
Cheek Raiser	0-2
Upper Lip raise	0-2
Lip Corner Pull	0-2
Vertical Mouth Stretch	0-2
Horizontal Mouth Stretch	0-2
Blink	0-1
Flared Nostril	0-1
Open Lips	0-1

Note. [a](Chambers et al., 1996)

Videotape Time Segment Sheet

Tape # _____ Participant # _____

Time:

a) Time in Recovery Room _____

b) Time Filming Started _____

c) Time Elapsed before filming (b - a) _____

KEY
Coding Segment = a 10 second period to be coded
Phase = whenever counter starts at a new number.
Phase start = First second of phase
Second Start = First second to be coded
Second Stop = last second to be coded

SEGMENTS TO BE CODED:

Coding Segment	Phase	Phase Start	Second Start	Second End
1				
2				
3				
4				
5				
6				

Visual Analogue Coding of Pain Sheet

Date Completed: Day_____ Month _____ Year _____
Participant #_____
Tape #_____

Time Start Segment 1____:____:____

No Pain At all — Worst Pain Ever

Time Start Segment 2____:____:____

No Pain At all — Worst Pain Ever

Time Start Segment 3____:____:____

No Pain At all — Worst Pain Ever

Time Start Segment 4____:____:____

No Pain At all

Worst Pain Ever

Time Start Segment 5____:____:____

No Pain At all

Worst Pain Ever

Time Start Segment 6 ____:____:____

No Pain At all

Worst Pain Ever

Visual Analogue Coding of Sedation Sheet

Date Completed: Day ____ Month _____ Year _____
Participant #_____
Tape #_____

Time Start Segment 1____:____:____

No Sedation At all — Worst Sedation Ever

Time Start Segment 2____:____:____

No Sedation At all — Worst Sedation Ever

Time Start Segment 3____:____:____

No Sedation At all — Worst Sedation Ever

Time Start Segment 4____:____:____

No Sedation At all

Worst Sedation Ever

Time Start Segment 5____:____:____

No Sedation At all

Worst Sedation Ever

Time Start Segment 6 ____:____:____

No Sedation At all

Worst Sedation Ever

Surgical Information Sheet

Child's Name _____ Completed by:_____

Type of procedure:

Length of procedure:

Length of time in recovery room (minutes):

General anesthesia during surgery? Yes No

Intravenous Opiods during surgery? Yes No

Were there any unusual occurrences related to this surgery?

Hospitalization Information Sheet

Name of Child:

Date admitted:

Reason for admission:

Illness Status: ☐ chronic ☐ acute

Date released:

Date of Surgery(dd/mm/yy):

Type of surgery:

Post-Operative Analgesics

Day of Surgery:

Analgesic type:	Prescribed	Administered
Opiod		
Non-opiod		

Post-Operative Day 1:

Analgesic type:	Prescribed	Administered
Opiod		
Non-opiod		

Post-Operative Day 2:

Analgesic type:	Prescribed	Administered
Opiod		
Non-opiod		

Invasive Procedures Performed

Type	Number of each procedure performed:		
	Day of Surgery	Post-op Day 1	Post-op Day 2
Finger prick			
IV insertion			
Venous bloodwork			
Nasogastric tube insertion			
Intramuscular injection			
Subcutaneous injection			
Urinary catheter			
Lumbar puncture			
Bone marrow aspiration			
Other:			

CFCS Coding Sheet

Surgery Study of Nonverbal Children

Participant Number_____ Coder _____ Date _____

Segment 1

Time	BRLOi	SQTi	ESQi	BNK	FNST	NWRi	NLFi	CHRi	OL	ULRi	LCPi	VMSTi	HMSTi

Segment 2

Time	BRLOi	SQTi	ESQi	BNK	FNST	NWRi	NLFi	CHRi	OL	ULRi	LCPi	VMSTi	HMSTi

Segment 3

Time	BRLOi	SQTi	ESQi	BNK	FNST	NWRi	NLFi	CHRi	OL	ULRi	LCPi	VMSTi	HMSTi

Segment 4

Time	BRLOi	SQTi	ESQi	BNK	FNST	NWRi	NLFi	CHRi	OL	ULRi	LCPi	VMSTi	HMSTi

Segment 5

Time	BRLOi	SQTi	ESQi	BNK	FNST	NWRi	NLFi	CHRi	OL	ULRi	LCPi	VMSTi	HMSTi

PLEASE NOTE ANYTHING UNUSUAL ABOUT THIS TAPE OR THE CODING OF IT

ON THE REVERSE OF THIS PAGE.

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