VOLUME I

RESEARCH COMPONENT

THE ASSOCIATION BETWEEN PAIN, LOW MOOD AND CHALLENGING BEHAVIOUR IN INTELLECTUAL DISABILITY

BY

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Overview

Both volume I and II of this thesis are submitted in partial fulfilment of the requirements for the degree of Doctorate of Clinical Psychology (ClinPsyD) at the University of Birmingham. Volume I includes the research component, comprising of three papers; a literature review, empirical paper and public domain briefing document. Both the literature review and empirical paper were prepared for submission to the *American Journal on Intellectual and Developmental Disabilities,* although contrary to journal requirements (see Appendix A for author's guidelines), tables and figures have been integrated into the text.

The literature review examined the concept of challenging behaviour as a "depressive equivalent" using a systematic review to investigate the association between depression and challenging behaviour (specifically aggression and self-injury) in individuals with an intellectual disability. Fifteen papers were identified which contained data related to the association between depression and aggression and/or self-injury. Conclusions drawn from these studies indicated that the association between depression and both aggression and self-injury is equally ambiguous and based on these studies, it would appear that there is currently insufficient evidence to support the use of challenging behaviour as a depressive equivalent.

The aims of the empirical paper were to examine the validity of the pain subscale of the Questions About Behavioral Function (QABF) and the relationship between pain related challenging behaviour (as measured by the QABF) and other operant functions. Findings gained provide support for the use of the QABF pain subscale in clinical practice and the potential role of pain as a setting event for challenging behaviour. More broadly the results

highlight the need to address the health needs and related pain of individuals with intellectual disabilities.

The public domain briefing document provides an accessible summary of the literature review and empirical paper.

Volume II includes the clinical component, comprising of five papers; a formulation of one client from two psychological perspectives, a service evaluation, a single case experimental design and two case studies. Client anonymity is ensured throughout through the use of pseudonyms which have been applied to all individuals included within each report.

This thesis is dedicated to

Lindsey Anne Davies

my beautiful mum and friend,

forever in our hearts

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Table of Contents

Chapter Three: Public Domain Briefing Document

List of Tables

Chapter One

Chapter Two

List of Appendices

Chapter One

A Systematic Review of the Association between Depression, Aggression and Self-Injury in Intellectual Disability

Abstract

The prevalence of depression in individuals with an intellectual disability is estimated to lie between 3% and 6%. It has been suggested that symptoms of depression in this population might be atypical and include unusual features such as challenging behaviour. However, there is significant disagreement regarding the use of challenging behaviour as a 'depressive equivalent'. The aim of this review is to evaluate published research reporting on the association between challenging behaviour, specifically aggression and self-injury, and depression in people with an intellectual disability as a first step toward evaluating whether challenging behaviours might be considered as depressive equivalents. The results of the studies identified indicated that the association between depression and aggression, and depression and self-injury are equivocal and the interpretations of the results limited by threats to validity. Based on this analysis, there is insufficient evidence to support the use of challenging behaviour as a depressive equivalent. Potentially confounding variables which could account for the association between challenging behaviour and depression, such as pain, are proposed based on the emerging literature on unidentified health problems in people with intellectual disability and their association with challenging behaviour. Further research to examine potentially confounding variables and the association between challenging behaviour and depression using methodologically robust designs and measures is clearly warranted.

Introduction

For many years, it was assumed that individuals with an intellectual disability were comparatively immune to the development of psychiatric disorder (Matson, Barrett, & Helsel, 1988; Sovner & Hurley, 1983). However, contemporary research has indicated that a substantial proportion of individuals with an intellectual disability, between 10% and 40%, experience mental health problems (Bakken et al., 2010; Cooper, Smiley, Morrison, Williamson, & Allan, 2007; Grey, Pollard, McClean, MacAuley, & Hastings, 2010). This increased risk of psychiatric disorder associated with the presence of an intellectual disability, supports the value of the use of dual diagnoses within this population (Bernal & Hollins, 1995; Einfeld, Ellis, & Emerson, 2011). The experience of compromised mental health is likely to further disadvantage individuals with pre-existing cognitive deficits (Reynolds & Baker, 1988) and consequently this is an important area for research. Policy initiatives acknowledge this as evidenced in the Health of the Nation Document (DoH, 1995) which called for improved identification and treatment of psychiatric disorder in people with an intellectual disability.

The presentation of depression in people with an intellectual disability has received particular attention within the literature due to the relatively high prevalence which is reported to be between 3% and 6% (Hurley, 2008; Cooper, 1997; Cooper et al., 2007). Prevalence rates of depression are underestimated within the general population (Paykel & Priest, 1992) and underestimation in people with intellectual disability is potentially exacerbated by pre-existing associated cognitive and behavioural impairments. The validity of self report, typically central to the identification of the symptoms of depression, is compromised for individuals with an intellectual disability due to more constrained expressive communication

(Levitas, Hurley, & Pary, 2001). As a result, carer report is often relied upon more heavily as a source of information for diagnosis, despite the poor reliability of this method for disorders such as depression (Burt, 1999). It has also been proposed that individuals with an intellectual disability might show symptoms of psychiatric illness that are different to the typically developing population, a phenomenon termed psychosocial masking (Reiss, 1994). Consequently, symptoms of depression, for example, might include a number of unusual features, not classified as core symptoms for diagnosis (Reiss, 1993; Sturmey, 1995). As a result a number of additional symptoms for the diagnosis of depression have been suggested, including increased somatic complaints, reduction in speech and onset of challenging behaviour (Smiley & Cooper, 2003).

Published prevalence rates of challenging behaviour in people with an intellectual disability are as high as 45%, although they vary widely (e.g. Emerson et al., 2001; Grey et al., 2010; Lowe et al., 2007). These high prevalence rates have been attributed to the presence of additional psychiatric disorders in individuals with an intellectual disability by a small number of researchers (Myrbakk & von Tetzchner, 2008) and there is evidence that indicates an association between the two (e.g. Grey et al., 2010; Laud & Matson, 2006, Moss et al., 2000; Tyrer et al., 2006). These studies have demonstrated a significant positive association between the presence of challenging behaviour and psychiatric disorder and significant group differences, so that participants with particular psychiatric symptoms demonstrate significantly more challenging behaviour and vice versa.

The concept of challenging behaviour as a "behavioural equivalent" for psychiatric disorder is already widely used (Hurley, 2006), particularly for depression as illustrated in the Diagnostic

Criteria for Psychiatric Disorders for Use with Adults with Learning Disabilities/Mental Retardation (DC-LD; Royal College of Psychiatrists, 2001), in which aggression is cited as a symptom of depression. Research assessing the knowledge of professionals, including direct care staff, regarding depression, indicates that they are aware of the inclusion of challenging behaviour as a symptom of depression (Munden & Perry, 2002). In combination, these observations strongly suggest that the concept of challenging behaviour as a depressive equivalent is becoming established in clinical practice.

The results of a number of studies have been used as evidence for challenging behaviour as a "depressive equivalent", although conceptual and methodological limitations of the studies call into question the validity of the conclusions that are drawn. To illustrate, several studies have used only two or three participants (e.g. Durand & Mapstone, 1998; Lowry & Sovner, 1992), limiting the generalisability of the findings. Challenging behaviour has also been identified as a depressive equivalent based on the results of medication trials, whereby symptoms of depression, including challenging behaviour, have reduced following the administration of anti-depressants (Clarke & Gomez, 1999; Jawed, Krishnan, Prasher, & Corbett, 1993). However, these results might be explained by the treatment of a common underlying mechanism (Aman, Arnold, & Armstrong, 1999; Ellis, Singh, & Ruane, 1999). Thus, studies based on the introduction of anti-depressant medication are not considered robust evidence of challenging behaviour as a depressive equivalent. Additionally, some studies have used broad definitions of challenging behaviour that include phenomena such as lethargy which are also used to diagnose depression (Paclawskyj, Matson, Bamburg, & Baglio, 1997), thus potentially inflating the association between challenging behaviour and depression. Although other studies have provided interesting data on the association between

specific types of challenging behaviour and particular psychiatric diagnoses, these do not employ statistical analyses and thus the significance of these findings is unclear (Grey et al., 2010; Marston, Perry, & Roy, 1997).

Due to these methodological limitations, there is an emerging consensus that the conclusion drawn from studies such as these, that challenging behaviour is a depressive equivalent, should be considered more critically. In publishing the unequivocally entitled paper "Challenging behaviours should not be considered as depressive equivalents in individuals with intellectual disability," Tsiouris, Mann, Patti and Sturmey (2003) openly challenged the use of challenging behaviour as a diagnostic criterion for depression, based on the current evidence. In their review of the assessment of mood in adults with an intellectual disability, Ross and Oliver (2003) stated that challenging behaviour is increasingly being labelled as a depressive equivalent with an apparent lack of explicit or robust rationale. McBrien (2003) argued that challenging behaviour was being used incorrectly to diagnose depression to avoid missing cases which did not fulfil current diagnostic criteria as a result of impairments associated with an intellectual disability, such as an inability to express worthlessness or suicidal ideation. Similarly, Holden and Gitlesen (2004) argued against the use of challenging behaviour as a depressive equivalent because of the risk of falsely identifying depression in this population.

To summarise, although the results of numerous studies indicate a possible association between challenging behaviour and depression, support for the use of challenging behaviour as a depressive equivalent is often derived from studies which are not methodologically robust. Consequently, the use of challenging behaviour as a depressive equivalent has become

a contentious issue both within the research and clinical communities that needs to be resolved to assess and more effectively treat both challenging behaviour and depression in individuals with an intellectual disability. Without clarification, the potential danger is that depression is misdiagnosed due to the presence of challenging behaviour and neither will be treated appropriately.

The aim of this paper is to review published studies of the association between challenging behaviour, specifically aggression and self-injury, and depression in people with an intellectual disability, as a first step toward evaluating the validity of the use of challenging behaviour as a depressive equivalent. These specific forms of challenging behaviour are reviewed due to their clinical significance and generally well defined nature. It is also important to more specifically define the challenging behaviours of interest as different types might or might not be significantly associated with depression. Inclusion criteria for this review are made explicit and only studies deemed to meet a defined level of methodological quality will be included. If there is a case to be made that challenging behaviours should be considered as depressive equivalents, then it is reasonable to expect that there should be evidence of an association, albeit potentially weak given the number of possible diagnostic criteria, between the diagnosis of depression and challenging behaviour across studies.

Method

Search criteria

All peer reviewed, published articles examining the association between depression and challenging behaviour between 1967 and June 2011 were identified by a systematic literature

search using the search engine PsycINFO®. Table 1 lists the search terms (including both English and American spellings) that were employed. All possible combinations of the main search terms and variations of these were used to identify relevant papers.

Table 1: Terms employed in the literature search for studies reporting the association between depression and aggression and/or self-injury

Search term	Variations
Challenging behavio*	Problem behavio*, aberrant behavio*, behavio* disorder, aggress*, self injur*, self destruct*
Intellectua* disab*	Learning disab*, mental retard*, mental handica*, develop* disab*
Depress*	Mood, affect*

Although the association between depression and general challenging behaviour (no specifically defined types) is not examined in this study, search terms related to challenging behaviour were included to ensure no data regarding aggression or self-injury included as a subclass of challenging behaviour were overlooked. 'Intellectual disability' and variations of this term were included in order to limit the data reviewed to this population. The reference lists of all identified papers were also inspected to check for any omissions.

The inclusion criteria were that studies were written in English and contained data on the association between depression and aggression and/or self-injury that was analysed statistically. Depression was defined within this study as the presence of core features of depression (low mood and/or reduced interest and pleasure; see Ross & Oliver, 2002) or a diagnosis of depression. Aggression and self-injury were defined as potentially causing

physical harm to others and/or self (Oliver et al., 2003). Studies were excluded if the sample was atypical by being limited in inclusion criteria and homogeneous (e.g. including participants with a specific syndrome only) or only general challenging behaviour, as opposed to aggression or self-injury specifically, was examined. Papers including an examination of the effect of medication on depression and aggression and/or self-injury were excluded because, as noted in the introduction, these were not deemed to demonstrate robust evidence of an association between depression and the two topographies of challenging behaviour under investigation (see Appendix B for flowchart outlining papers excluded). The reliability and validity of the measures used to assess both aggression and self-injury as well as severity of intellectual disability in each study were examined (where reported) in order to appraise the quality of each, but not to exclude, given the paucity of papers meeting the remaining criteria.

Identified papers

Fifteen studies, thirteen regarding aggression and twelve self-injury (ten of the fifteen studies provided data regarding both), meeting criteria were identified and included in this review. The methodology employed and results reported in each study are described in Tables 2 and 3.

Table 2: Methodology of thirteen studies reporting the association between aggression and depression in people with intellectual disability

Authors	Sample		Measures			Results
	Participant Characteristics	Recruitment	Aggression	Depression	Intellectual Disability	
Laman & Reiss (1987)	45 adults with mild ID	Clients from a sheltered workshop and patients from a mental health clinic for people with ID	Social Performance Survey Schedule ("threatens others verbally or physically")	Psychopathology Instrument for Mentally Retarded Adults Depression Subscale and Illinois-Chicago Informant Rating Scale for Depression	Vocabulary subset of the WAIS-R	"High" depressed group (12 highest scorers) scored significantly higher on "threatens others verbally and physically" than the "low" depressed group (12 lowest scorers)
Reiss & Rojahn (1993)	528 children and adults with mild to moderate (60.6%) and severe to profound (39.4%) ID	Residents from community based agencies from three American states	Reiss Screen for Maladaptive Behavior: Child (Conduct Disorder Scale) and Adult (Aggressive Behavior Scale)	Reiss Screen for Maladaptive Behavior: Child (Depression Scale) and Adult (Depression Behavioral Signs and Depression Physical Signs Scales)	No measure described	Presence of depression (scoring at or above cut-off on child or either of adult depression scales) associated with significantly increased risk in probability of an aggressive behaviour problem (at or above cut-off on appropriate aggression scale). The depressed group scored significantly higher on measures of aggression than the non-depressed group.
Meins (1995)	178 adults aged between 20 and 76 years (mean age = 39.1 years) with mild (71%) to severe (29%) ID	Individuals from residential facilities or two psychiatric hospitals in Germany	Abbreviated version of the Disability Assessment Schedule (Aggressive Behaviour: Persons)	Psychiatric examination (mostly in line with DSM-III-R), including mild and brief, occasionally supplemented by concurrent atypical symptoms	Disability Assessment Schedule: Adaptive Behaviour Score	No significant difference in mean score of aggression between participants with a diagnosis of depression and those without (the control group)

Authors	Sample		Measures			Results
	Participant Characteristics	Recruitment	Aggression	Depression	Intellectual Disability	
Bihm, Poindexter, & Warren (1998)	170 children and adults aged between 8 and 56 years (mean age = 32.41 years) with severe (21%) and profound (79%) ID	Individuals from a residential facility	Reiss Screen for Maladaptive Behavior (Aggression Subscale), BPI (Aggression/ Destructive Behavior), Behavior Incident Report (BIR), Physical Aggression Inventory (PAI)	Reiss Screen for Maladaptive Behavior (Depression Behavioural Signs and Depression Physical Signs)	No measure described	Aggression as measured by the BIR, PAI and BPI had significant positive associations with depression. Aggression as measured by the PAI (but not BIR or BPI) significantly predicted by depression (behavioural signs only).
Ross & Oliver (2002)	24 adults with severe and profound ID, 39 years mean age	Randomly selected community sample	Challenging Behaviour Interview: physical aggression	Mood, Interest and Pleasure Questionnaire	Wessex Scale	No significant difference in occurrence of aggression between "low mood" (12 participants with lowest MIPQ scores) and comparison group (12 participants with highest MIPQ scores)
Tsiouris, Mann, Patti, & Sturmey (2003)	92 adults (42.6 years mean age) with mild (24%), moderate (30.4%), severe (26%) and profound (10.9%) ID (8.7% unspecified)	Clients referred to a regional clinic for assessment of challenging behaviour and psychiatric diagnoses	CBCPID: Aggression item scored as present or absent during the last two weeks	CBCPID and diagnoses based on an algorithm of DSM IV. Diagnoses of depression include major depression, bipolar disorder (depressed phase) and schizoaffective disorder depressed phase.	No measure described	No significant difference in endorsement of aggression item between "depressed" and "non-depressed" (other psychiatric diagnoses e.g. psychotic disorder) groups. Aggression did not load on factor analysis of depression or show a significant association with core features of depression.

Authors	Sample		Measures			Results
	Participant Characteristics	Recruitment	Aggression	Depression	Intellectual Disability	
Rojahn, Matson, Naglieri, & Mayville (2004)	180 adults aged between 20 and 91 years (mean age = 50.6 years) with mild (2.2%), moderate (5.6%), severe (15%) and profound (66.7%) ID (10.5% unspecified)	Residents from a developmental centre	BPI (Aggression/Destructive Behavior)	Diagnostic Assessment for the Severely Handicapped II	No measure described	Depression showed a significant, positive association with aggression, although aggression formed a separate factor across all factor analyses to depression. Participants demonstrating aggression had a significantly higher depression score than those not showing aggression, but only when this was defined liberally. Relative risk of aggression given depression < 2.
Tsiouris, Mann, Patti, & Sturmey (2004)	92 adults (42.6 years mean age) with mild (24%), moderate (30.4%), severe (26%) and profound (10.9%) ID (8.7% unspecified)	Clients referred to a regional clinic for assessment of challenging behaviour and psychiatric diagnoses	CBCPID: Aggression item scored as present or absent during the last two weeks	Independent diagnosis by psychiatrist and CBCPID	No measure described	Small likelihood of depression given the presence of aggression (result below arbitrary cut off for indicating variable as a useful predictor, as measured by Bayes' Formula)
Kishore, Nizamie, & Nizamie (2005)	60 children and adults (21 years mean age) with mild (36.7%), moderate (43.3%) and severe/profound (20%) ID	Individuals with ID and behavioural problems from a psychiatric institute	Reiss Screen Test Manual, Reiss Screen for Maladaptive Behavior and the AAMD Adaptive Behaviour Scale II	Semi-structured psychiatric diagnostic interview	Stanford Binet Intelligence Scale, Vineland Social Maturity Scale	"Affective" (bipolar, mania and depression) group scored significantly higher on measures of depression than the "behaviour" (unspecified behaviour problems) and "other" disorder group. On measures of aggression, the "affective" scored higher than the "behaviour" group only.

Authors	Sample		Measures			Results
	Participant Characteristics	Recruitment	Aggression	Depression	Intellectual Disability	
Hemmings, Gravestock, Pickard, & Bouras (2006)	214 adults (72% of sample aged over 35 years) with mild to moderate (64%) and severe (36%) ID	From a local register for people with ID	Disability Assessment Schedule (Behaviour Problems Section)	Psychiatric Assessment Schedule for Adults with a Developmental Disability Checklist	Clinical level of ID following ICD-10 diagnostic criteria from clinical notes and/or assessment	Symptoms of depression ("suicidal", "weight change", "early waking" and "irritable mood", but not "low mood") were significantly associated with aggression. Aggression significantly predicted by "early waking" "loss of energy" and "irritable mood" but not "low mood".
Hurley (2008)	300 adults with mild (58%), moderate (27.5%), severe (7.5%) and profound (7%) ID	Clients seen in a speciality clinic between 1993 and 2003	In case notes, no measure described	Psychiatric diagnostic interview and record review	In case notes, no measure described	Depressed group reported as being significantly more aggressive than control group (participants who did not receive a psychiatric diagnosis)
Langlois & Martin (2008)	1302 older adults (57.8% aged over 50 years) with mild to moderate (42.4%) and severe (57.8%) ID	Clients from community agencies and residents from institutional settings	InterRAI ID Aggression Rating Scale, verbal or physical abuse, socially inappropriate or disruptive behaviour and resisting care. Also physical abuse.	InterRAI ID Depression Rating Scale, psychiatric interview and record review	No measure described	Participants with a diagnosis of depression were significantly more likely to demonstrate aggression than those without a diagnosis

Authors	Sample		Measures			Results
	Participant Characteristics	Recruitment	Aggression	Depression	Intellectual Disability	
Myrbakk & von Tetzchner (2008)	142 adolescents and adults aged between 14 and 72 years (40.7 years mean age) with mild (5.6%), moderate (31%), severe (46.5%) and profound (16.9%) ID	Individuals referred to a specialist challenging behaviour service in Norway and from the community receiving services from their municipality	Aberrant Behavior Checklist: one aggression item	Reiss Screen for Maladaptive Behavior, Mini Psychiatric Assessment Schedule for Adults with Developmental Disability, Assessment of Dual Diagnosis and the Diagnostic Assessment of the Severely Handicapped II	Leiter International Performance Scale Revised, Wechsler Intelligence Scale for Children Revised III, Wechsler Adult Intelligence Scale, Vineland Adaptive Behaviour Scale and clinical judgement	Aggression showed significant, positive association with depression (except when measured using the Assessment of Dual Diagnosis)

BPI = Behaviour Problem Inventory, CBCPID = Clinical Behaviour Checklist for Persons with Intellectual Disability, DSM = Diagnostic and Statistical Manual, ID = Intellectual Disability, WAIS-R = Wechsler Adult Intelligence Scale – Revised.

Table 3: Methodology of twelve studies reporting the association between self-injury and depression in people with intellectual disability

Authors	Sample		Measures			Results
	Participant Characteristic	Recruitment	Self-Injury	Depression	Intellectual Disability	
Meins (1995)	178 adults aged between 20 and 76 years (mean age = 39.1 years) with mild (71%) to severe (29%) ID	Individuals from residential facilities or two psychiatric hospitals in Germany	Abbreviated version of the Disability Assessment Schedule (Aggressive Behaviour: Persons)	Psychiatric examination (mostly in line with DSM-III-R), including mild and brief, occasionally supplemented by concurrent atypical symptoms	Disability Assessment Schedule: Adaptive Behaviour Score	Participants without a diagnosis of depression scored significantly higher on measures of SIB than those with a diagnosis of depression
Ross &Oliver (2002)	24 adults (39 years mean age) with severe and profound ID	Randomly selected community sample	Challenging Behaviour Interview	Mood, Interest and Pleasure Questionnaire	Wessex Scale	No significant difference in occurrence of SIB between "low mood" (12 participants with lowest MIPQ scores) and comparison group (12 participants with highest MIPQ scores).
Holden & Gitlesen (2003)	165 adults aged between 18 and 46+ years with mild (14%), moderate (27%), severe (35%) and profound (23%) ID	All participants lived in or received respite care from residential facilities. Participants with challenging behaviour were referred to a habilitation service due to problematic behaviour.	Informant rated	Psychiatric Assessment Schedule for Adults with a Developmental Disability Checklist. Depression measured using the item "depressed mood and/or suicidal thoughts/actions"	Staff asked to classify using DSM definitions of ID	Participants with aggression/ unacceptable behaviour without SIB scored significantly higher on depression measure than those with aggression/unacceptable behaviour and SIB

Authors	Sample		Measures			Results
	Participant Characteristic	Recruitment	Self-Injury	Depression	Intellectual Disability	
Tsiouris, Mann, Patti, & Sturmey (2003)	92 adults (42.6 years mean age) with mild (24%), moderate (30.4%), severe (26%) and profound (10.9%) ID (8.7% unspecified)	Clients referred to a regional clinic for assessment of challenging behaviour and psychiatric diagnoses	CBCPID: SIB item scored as present or absent during the last two weeks	CBCPID and diagnoses based on an algorithm of DSM IV. Diagnoses of depression include major depression, bipolar disorder (depressed phase) and schizoaffective disorder depressed phase.	No measure described	No significant difference in endorsement of SIB item between "depressed" and "non-depressed" (other psychiatric diagnoses e.g. psychotic disorder) groups. SIB did not load on factor analysis of depression or show a significant association with core features of depression.
Rojahn, Matson, Naglieri, & Mayville (2004)	180 adults aged between 20 and 91 years (mean age = 50.6 years) with mild (2.2%), moderate (5.6%), severe (15%) and profound (66.7%) ID (10.5% unspecified)	Residents from a developmental centre	BPI (behaviours that can damage the body and occur repeatedly in unvarying presentation)	Diagnostic Assessment for the Severely Handicapped II	No measure described	Depression showed a positive, significant association with SIB. SIB formed a separate factor across all factor analyses to depression. Participants demonstrating SIB scored significantly higher on depression than those not showing SIB, but only when this was defined liberally. Relative risk of SIB given depression ≤ 2.
Tsiouris, Mann, Patti, & Sturmey (2004)	92 adults (42.6 years mean age) with mild (24%), moderate (30.4%), severe (26%) and profound (10.9%) ID (8.7% unspecified)	Clients referred to a regional clinic for assessment of challenging behaviour and psychiatric diagnoses	CBCPID: SIB item scored as present or absent during the last two weeks	Independent diagnosis by psychiatrist and CBCPID	No measure described	Moderate likelihood of depression (as measured by Bayes' Formula) given the presence or absence of SIB

Authors	Sample Participant Characteristic	Recruitment	Measures Self-Injury	Depression	Intellectual Disability	Results
Kishore, Nizamie, & Nizamie (2005)	60 children and adults (21 years mean age) with mild (36.7%), moderate (43.3%) and severe/profound (20%) ID	Individuals with ID and behavioural problems from a psychiatric institute	Reiss Screen Test Manual, Reiss Screen for Maladaptive Behaviors and the AAMD Adaptive Behavior Scale II	Semi-structured psychiatric diagnostic interview	Stanford Binet Intelligence Scale, Vineland Social Maturity Scale.	Affective" (bipolar, mania and depression) group scored significantly higher on measures of depression than the "behaviour" (unspecified behaviour problems) and "other" disorder group. There was no significant difference in SIB scores across these groups.
Hemmings, Gravestock, Pickard, & Bouras (2006)	214 adults (72% of sample aged over 35 years) with mild to moderate (64%) and severe (36%) ID	From a local register for people with ID	Disability Assessment Schedule (Behavior Problems Section)	Psychiatric Assessment Schedule for Adults with a Developmental Disability Checklist	Clinical level of ID following ICD-10 diagnostic criteria from clinical notes and/or assessment	Symptoms of depression ("sad/down", "suicidal", "loss of appetite", "weight change", "loss of confidence", "early waking" and irritable mood") significantly associated with SIB. SIB significantly predicted by "irritable mood" and "suicidal ideas" but not "sad/down".
Hurley (2008)	300 adults with mild (58%), moderate (27.5%), severe (7.5%) and profound (7%) ID	Clients seen in a speciality clinic between 1993 and 2003	In case notes, no measure described	Psychiatric diagnostic interview and record review	In case notes, no measure described	"Depressed group" reported as showing significantly more SIB than the control group (participants who did not receive a psychiatric diagnosis)
Langlois & Martin (2008)	1302 older adults (57.8% aged over 50 years) with mild to moderate (42.4%) and severe (57.8%) ID	Clients from community agencies and residents from institutional settings	InterRAI ID self-injury scale	InterRAI ID Depression Rating Scale, psychiatric interview and record review	No measure described	Participants with a diagnosis of depression were significantly more likely to show SIB than those without a diagnosis

Authors	**Sample** Participant Characteristic	Recruitment	Self-Injury	**Measures** Depression	Intellectual Disability	**Results**
Myrbakk & von Tetzchner (2008)	142 adolescents and adults aged between 14 and 72 years (40.7 years mean age) with mild (5.6%), moderate (31%), severe (46.5%) and profound (16.9%) ID	Individuals referred to a specialist challenging behaviour service in Norway and from the community receiving services from their municipality	Aberrant Behavior Checklist: three SIB items	Reiss Screen for Maladaptive Behavior, Mini Psychiatric Assessment Schedule for Adults with Developmental Disability, Assessment of Dual Diagnosis and the Diagnostic Assessment of the Severely Handicapped II	Leiter International Performance Scale Revised, Wechsler Intelligence Scale for Children Revised III, Wechsler Adult Intelligence Scale, Vineland Adaptive Behaviour Scale and clinical judgement	SIB showed a significant, positive association with depression (except when measured using the Reiss Screen)
Sturmey, Laud, Cooper, Matson, & Fodstad (2010)	693 adults (48 years mean age) with mild (2.6%), moderate (5.5%), severe (12.8%) and profound (79.1%) ID	Residents from a care facility	Diagnostic Assessment for the Severely Handicapped II: SIB Subscale.	Diagnostic Assessment for the Severely Handicapped II: Depression Subscale. DSM criteria imposed by a psychiatrist and psychologist, mental status exam, record review, staff interview and behavioural observations	Previously determined by a psychologist using DSM-IV-TR. Standardised measures (e.g. Stanford Binet-IV or Leiter) behavioural observations, the Vineland Adaptive Behavior Scales and the Matson Evaluation of Social Skills for the Severely Retarded	Out of 29 correlations between behavioural items (SIB being one of them) and depression, all but 4 were significant at .01 or higher (range = -.01 to .37)

BPI = Behaviour Problem Inventory, CBCPID = Clinical Behaviour Checklist for Persons with Intellectual Disability, SIB = self-injurious behaviour, ID = intellectual disability

Results

Methods employed by the studies identified

Of the fifteen studies identified, the majority (twelve) included participants with a range of intellectual disability from mild/moderate to severe/profound, one included participants with a mild level of disability only and another two included participants with a severe/profound disability only. Six studies recruited participants from community settings, four from inpatient or clinic settings and five from both. In order to examine challenging behaviour and depression, most studies used standardised assessments, including questionnaires and structured interviews. Only one study used entirely non-standardised measures whilst the remaining six studies utilised both standardised and bespoke measures (e.g. diagnostic interview). The majority of studies (eight) employed a group comparison design, four used correlational methodology and the remaining three used mixed methods. Thus, within the studies identified, the methods employed varied widely in terms of sample size, characteristics and origin, and the measures and design used.

The association between depression and aggression and/or self-injury

Data indicating an association between depression and aggression and/or self-injury

Of the fifteen studies identified with data regarding the association between depression and aggression and/or self-injury, six provided data in support of an association between depression and these forms of challenging behaviour (Holden & Gitlesen, 2003; Hurley, 2008; Laman & Reiss, 1987; Langlois & Martin, 2008; Reiss & Rojahn, 1993; Tsiouris et al., 2004; both Hurley and Langlois & Martin provided data regarding an association between

depression and both types of challenging behaviour). Sample size varied largely across studies, ranging from 45 to 1302. Five of these included participants with the whole range of intellectual disabilities, with one employing participants with only a mild intellectual disability; five studies included adults only and one both children and adults. Only one study recruited participants purely from the community, the rest recruited from institutional or speciality clinics (two) or a combination of the two (three). In terms of measures of challenging behaviour, four studies used various types of screening tools, the remaining two used case note and informant report. Two studies did not give a definition of aggression, the other two defined it broadly, including both verbal and physical aggression. No definitions of self-injury were given. One study relied on psychiatric interview for the diagnosis of depression, three used a variety of rating scales and another two studies used a combination of the two. Two studies disclosed how intellectual disability was measured (using a standardised measure and informant rating against diagnostic criteria), the remaining four did not. Group designs were utilised in five studies, whereby a group of participants classified as having depression were compared on measures of aggression and/or self-injury to those labelled as not experiencing depression, or experiencing depression to a lesser extent, one of these also employed a correlational method. The remaining study relied on a correlational method alone. Thus, less than half of the studies identified with data regarding the association between depression and aggression and/or self-injury provided data in support of an association. Two of these reported data regarding an association between depression and both types of challenging behaviour.

Data indicating the absence of an association between depression and aggression and/or self-injury

Five studies did not demonstrate a significant association between depression and aggression and/or self-injury (Kishore et al., 2005; Meins, 1995; Ross & Oliver, 2002; Tsiouris et al., 2003; 2004); two of these were based on the same sample and three of these contained data indicating the absence of an association with depression for both aggression and self-injury. Sample size ranged from 24 to 178 participants. All of these studies employed adult participants only, except one which recruited both adults and children, four used participants with the whole range of intellectual disabilities, and another included participants with a severe/profound intellectual disability only. One study recruited participants solely from the community, three from speciality clinics and another from both settings. Each study used a rating scale to measure challenging behaviour, although operational descriptions were not supplied. The measurement of depression differed across studies so that two studies used psychiatric examination, another employed a rating scale and the final two studies used both. These two studies did not describe the measurement of intellectual disability, the other three studies described the use of standardised measures. Three studies employed a group design, another used correlational statistics and another, a mixed methodology. Groups compared comprised of participants diagnosed as having depression, rated as low mood or scoring higher on measures of depression and participants without a diagnosis of depression, without low mood or scoring lower on measures of depression respectively. Thus, a third of the studies identified as reporting on the association between depression and aggression and/or self-injury did not provide data in support of an association. Two of these were based on the same sample and three of these contained data indicating the absence of an association with depression for both aggression and self-injury.

Inconclusive data regarding the association between depression and aggression and/or self-injury

According to the data of six of the studies identified, the association between aggression and depression was unclear (Bihm et al., 1998; Hemmings et al., 2006; Kishore et al., 2005; Myrrbakk & Tetzchner, 2008; Rojahn et al., 2004; Sturmey et al., 2010; three of these studies reported data on the association between depression and both aggression and self-injury). The results produced by these studies were deemed inconclusive because, as in the case of three studies, the results within the study depended on how challenging behaviour and/or depression was measured, so that both significant and non-significant findings were produced depending on the measures used. In another study utilising a group design, the affective group only scored significantly higher on measures of aggression than one of two comparison groups, both scoring lower on measures of depression than the affective group. The statistics and design used also affected the results produced in the fifth study, so that correlational statistics produced a significant finding, as did the group design, but factor analysis and relative risk did not. The remaining study conducted a range of correlational analyses but was unclear as to whether those relating to self-injury were significant.

Sample sizes within these studies varied from 60 to 693. Four of these included child and adult participants, two recruited adults only. The majority of studies (five) included participants with a range of intellectual disabilities, just one study included participants with severe/profound intellectual disability only. Four studies recruited participants from the community, another from a psychiatric institute and another recruited participants from both settings. Each of the studies utilised rating scales to measure challenging behaviour, two of these gave some description as to how this was operationally defined and both were based on

the presentation of aggression and destruction as a measure of aggression. With regard to the measurement of depression, four studies used checklists, one study utilised a semi-structured diagnostic interview and another used both. Two studies did not disclose how level of intellectual disability was measured, a further two used a variety of standardised scales, another used diagnostic criteria based on case notes and the remaining study used diagnosis and standardised measures. Two of the studies used a group design; one based on the comparison of participants with a diagnosis of affective disorder and those with other psychiatric diagnoses, who scored significantly lower on measures of depression, the other based groups on the presence or absence of self-injury. This study also conducted correlational analyses, as did the other four studies. Thus, from the data of six of the studies identified, the association between aggression and depression was unclear. Three of these studies reported data on the association between depression and both aggression and self-injury

Summary of results

Only four of the studies identified as providing data on the association between aggression and depression demonstrated the presence of an association. Another four studies did not support these findings and another five studies provided data which was equivocal as to the nature of this association. Similarly, four of the studies identified as providing data on the association between self-injury and depression demonstrated the presence of an association. A further four studies did not support these findings and another four studies provided data which was equivocal as to the nature of this association.

Discussion

The aim of this paper was to evaluate peer reviewed, published studies reporting on the association between challenging behaviour, specifically aggression and self-injury, and depression in people with an intellectual disability. Whilst studies were not excluded on the basis of employing measures of challenging behaviour or intellectual disability with poor or no reported reliability or validity, the methodologies employed by each study were reported and appraised critically to examine the quality of the results produced.

The results of this review indicated that associations between depression and both aggression and self-injury are, at best, equivocal. In both cases, four studies identified an association, four did not and the results of a further five in the case of aggression and four for self-injury were inconclusive. Based on this evaluation there is currently insufficient evidence to support the use of challenging behaviour as a depressive equivalent. It is reasonable to suppose that if self-injury or aggression were symptoms of depression then they would reliably be associated with this diagnosis, even if the strength of the association was moderate. These results must be considered in light of a few methodological limitations. Whilst there were no substantial differences in the methodologies employed by studies which did and did not find an association between depression and aggression and/or self-injury, the sample sizes employed by studies identifying an association tended to be larger which might indicate a lack of power in studies which did not find an association. It must also be noted that many of the studies (six) which examined the association between both aggression and self-injury found the same result for both (either an association, no association or equivocal results), only three studies with data regarding both types of challenging behaviour found different results for aggression

and self-injury, indicating that the discrepant methodologies employed by the studies may have had an effect on the results.

Methodological limitations of this review must also be considered. Firstly, papers were included based on the provision of data regarding the association between depression and challenging behaviour, whereby depression was defined as the presence of low mood and/or reduced interest and pleasure. This was deemed a useful, basic definition given the varied conceptualisation of depression within the literature and the problems of identifying depressive symptomatology in this population. However, had a more restricted definition of depression been applied, such as diagnosis by a psychiatrist, many studies included within this review would not have met inclusion criteria. Similarly, aggression and self-injury were broadly defined within this review as including physical harm to others or self respectively, so that several of the studies included used definitions which were not tightly defined. Studies were also included which claimed to measure aggression and/or self-injury, although the exact definition of these were not provided, so that they might not have met inclusion criteria had more information regarding these variables been provided. Finally, although it was deemed useful to segregate challenging behaviour into aggression and self-injury to examine their unique relationship with depression, it is likely that a great deal of overlap between these behaviours remained, so that data pertaining to each type of behaviour was not distinct and many participants used to provide data on each individual type of challenging behaviour actually demonstrated both (e.g. Davies & Oliver, in preparation).

There are a number of methodological limitations of the studies identified, as with much of the wider literature, which are likely to confound results, as highlighted in previous reviews

(McBrien, 2003; Ross & Oliver, 2003). Firstly, due to concerns regarding the under detection of depression, a number of studies have been conducted specifically to provide evidence in support of challenging behaviour as a depressive equivalent. Thus, participants have been recruited who are already known to both demonstrate challenging behaviour and experience psychiatric disorder (as in Kishore et al., 2005 and Tsiouris, et al., 2004), so that the resultant association between these two disorders is unsurprising. Several researchers have also included challenging behaviours within checklists of depression used to diagnose the disorder, leading to a circular argument whereby participants are diagnosed with depression as a result of demonstration of challenging behaviour, and these findings are taken as evidence that depression exists within this population and challenging behaviour is a reliable symptom. To illustrate, Tsiouris et al. (2004) used a measure depression which included self-injury as a clinical indicator of depression and reported an association between self-injury and depression, thus potentially inflating this association.

Measures of depression administered to people with an intellectual disability, including both rating scales and psychiatric interview, have also been criticised for being unsuitable, with poor or unevaluated psychometric properties. Despite being adapted to best suit the characteristics of individuals with severe and profound intellectual disabilities, measures administered continue to include items such as suicidal ideation, which are difficult to assess in this population. It is also often unclear as to how to score such items, as not applicable or as not occurring, which will affect the resultant aggregate score. For example, suicidality and self reproach have both been used as symptoms of depression to assess the association between challenging behaviour and depression in samples with a large proportion of participants with severe intellectual disability (Hemmings et al., 2006; Tsiouris et al., 2003;

2004). As is generally the case for psychological measures used with individuals with an intellectual disability, measures of depression adapted to suit this population are often less robust in terms of psychometric properties or the properties have not been established. Bihm et al. (1998) for example, used a primary measure of aggression without documented reliability or validity. When items have been used from standardised measures, it is not always clear as to whether reliability and validity data have been provided at this level (as in the study conducted by Hemmings et al., 2006).

Other equivalents to depressive symptoms have been proposed within the literature, including irritable mood and psychomotor agitation or retardation (e.g. Charlot, Doucette, & Mezzacappa, 1993; Matson et al., 1999; Tsiouris, 2001). However, these, like challenging behaviour, have not been applied consistently across studies so that the assessment of depression varies widely. As a result, the diagnosis of depression, according to Ross and Oliver (2002), is often inappropriately and inconsistently applied. As relatively little is understood about the expression of depression in individuals with an intellectual disability, degree of depression has not been investigated, so that only milder forms of depression, for example, might require equivalents in order to be diagnosed; more severe forms which are, arguably, easier to identify in this population might not require change to the diagnostic criteria. Both aggression and self-injury have also been conceptualised differently across studies. To illustrate, aggression is on occasion measured in conjunction with destruction of property, with no delineation between verbal and physical aggression or, more broadly, alongside behaviours more commonly conceptualised as conduct disorder type behaviours (e.g. Langlois & Martin, 2008; Rojahn et al., 2004).

Small samples are often recruited by relevant studies (e.g. Laman & Reiss, 1987; Ross & Oliver, 2002), reducing their power. Many of the participants recruited are on medication at the time of the study, but rarely are the potential confounding effects of this considered in the interpretation of resultant data (e.g. Tsiouris et al., 2003; 2004). Although group designs are common within the literature, the matching of groups on relevant variables, such as other psychopathology (e.g. autism spectrum disorder), age or sex is less common (e.g. Kishore et al., 2005; Sturmey et al., 2010). Despite agreement within the research community that depression is expressed by individuals with a mild intellectual disability in a similar manner to that of the general population (Pawlarcyzk, & Beckwith, 1987; Sovner, 1986), and that diagnostic criteria need only be modified for people with a severe or profound intellectual disability, few studies in this area (with the exception of Ross & Oliver, 2002 and Bihm et al., 1998) have recruited only participants with a severe or profound intellectual disability. Across studies, participants have been recruited from a range of settings, from institutions to community residences; random selection from the community is rare (except in the case of Ross & Oliver, 2002) so that samples are often biased. Within some studies, this problem has been further exacerbated so that participants recruited from different settings, or indeed experiencing different levels of intellectual disability, are subject to different assessment procedures (e.g. Meins, 1995). Contrast or control groups also vary in their composition across studies, including participants with fewer symptoms or no symptoms of depression, participants with other psychiatric symptoms or none, rendering interpretation of resultant data difficult.

Studies in this area often investigate the association between depression and challenging behaviour using correlational designs, as is evident in seven of the studies reviewed.

Alongside the often small samples and differences between these in terms of key variables which could confound the results, it has so far been impossible to infer causation in this area. Retrospective, as opposed to the more methodologically robust prospective design, is also commonly employed (as in Hurley, 2008 and Meins, 1995). This is significant since contemporaneous presentation of challenging behaviour and depression has been proposed as a criteria for a depressive equivalent (Meins, 1995), which is often more difficult to identify in retrospective studies, although cannot be guaranteed in prospective designs (as in Ross & Oliver, 2002, where there was a time delay between measurement of challenging behaviour and depression). The informants used to complete measures of challenging behaviour and depression are often selected for convenience with disregard to their qualification as accurate informants (e.g. Holden & Gitlesen, 2003; Laman & Reiss, 1987), with different informants completing different measures (e.g. Meins, 1995).

Despite these and other methodological limitations, it has been concluded by a number of researchers that challenging behaviour is a depressive equivalent. Significant positive associations between challenging behaviour and depression have perhaps been too readily conceptualised as part of the same disorder. Indeed, some researchers have suggested that such an association could merely indicate the presence of two co-existing disorders (Paclawskyj, Matson, Bamburg, & Baglio 1997), since both are relatively common within this population. It has also been proposed that distress might underlie both challenging behaviour and psychiatric disorder, so that they both arise from a common pathway (Charlot et al., 2007). As demonstrated by Laman and Reiss (1987) the association between depression and challenging behaviour might be due to the social skills deficits experienced by people with depression, limiting their behavioural repertoire so that social interaction is less appropriate.

Lowry (1994) proposed a more comprehensive account for the association between depression and challenging behaviour without reducing it to a behavioural equivalent. According to Lowry, the experience of symptoms of depression causes an individual to perceive environmental events as more aversive, provoking challenging behaviour such as aggression or self-injury. Thus, depressive symptomatology acts as a setting event for challenging behaviour and both challenging behaviour and depression are conceptualised as being distinct from each other. This model has since been corroborated by the findings of a few studies which, using mood induction, have demonstrated that lowering mood results in an increased rate of challenging behaviour (Carr et al., 2003; Durand & Mapstone, 1998). Whilst Lowry's model is certainly more sophisticated than the arguably reductionist depressive equivalent approach, it does not account for other variables which could be acting as setting events for both depression and challenging behaviour, such as pain.

Research has indicated a clear association between pain and both challenging behaviour and low mood (Breau & Camfield, 2011; Carr & Owen-DeSchryver, 2007; Symons & Danov, 2005; Tervo, Symons, Stout, & Novacheck, 2006). Thus, it is feasible, given the prevalence of health problems and related pain in this population (van Schrojenstein Lantman-De Valk, Metsemaker, Haveman, & Crebolder, 2000; Stallard, Williams, Lenton, & Velleman, 2001), that pain is the underlying variable for both challenging behaviour and pervasive low mood and loss of interest and potentially acting as a setting event. Thus, although a possible direct association between challenging behaviour and depression cannot be ignored at this stage, given the strong association between self-injury and pain within the literature, it might be beneficial for clinicians to assume that low mood in the context of challenging behaviour is the result of pain in the first instance.

Further research in this area is important to provide clarification of the status of challenging behaviour as a depressive equivalent. Currently, the conceptualisation of depression experienced by individuals with an intellectual disability varies dramatically across researchers and practitioners, so that little diagnostic standardisation exists. As a result, individuals with an intellectual disability are potentially at risk of being incorrectly diagnosed with depression due to the demonstration of challenging behaviour, so that neither disorder is formulated and thus treated appropriately. Other individuals experiencing symptoms of depression requiring treatment might also go undiagnosed due to the absence of challenging behaviours. Based on the current evidence it is unwise to continue to diagnose depression using challenging behaviour as a depressive equivalent. Future research should also investigate other putative third variables involved in the association between depression and challenging behaviour (as proposed by Ross & Oliver, 2003), including pain. Such research could help to clarify if depressive symptomatology, such as low mood and loss of interest, and challenging behaviour co-exist due to the presence of a common third variable. It might also be useful to focus research efforts onto individuals known to be at high risk of challenging behaviour, those with a severe intellectual disability and autism (McClintock, Hall, & Oliver, 2003), to see what role, if any, is played by pain and depression.

To provide this clarification, more robust measures of depression and tightly operationalised definitions of challenging behaviour are required. Participants must also be selected randomly so that data are generalisable to the population of people with an intellectual disability and to avoid inflating the association between depression and challenging behaviour. In order to establish causality, more robust methodologies must also be employed, including natural

observations and functional analytic techniques so that variables underlying the demonstration of challenging behaviour and depressive symptoms might be detected.

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Chapter Two

Pain, Aggression and Self-Injury in Children with Intellectual Disability

Abstract

The growing evidence for an association between pain and challenging behaviour in people with intellectual disability highlights the need for clinical assessment of pain related challenging behaviour and its relationship to well documented operant functions. The aims of this study are to examine 1) the validity of the pain subscale of the Questions About Behavioral Function (QABF) and 2) the relationship between pain related challenging behaviour and other operant functions. QABF data and measures of pain related behaviour were collected on 46 children aged between 4 and 15 years with a range of syndromes associated with intellectual disability. Children who were identified by the QABF pain subscale as showing pain related challenging behaviour scored significantly higher on pain indices than a contrast group (participants classified as having non pain related challenging behaviour). Of those identified as having pain related challenging behaviour, the vast majority (95.6%) also demonstrated challenging behaviour with a function additional to pain and significantly more functions of challenging behaviour as compared to the non pain related challenging behaviour group. These findings provide support for the use of the pain subscale of the QABF in clinical practice and the potential role of pain as a setting event for challenging behaviour. More broadly the results highlight the need to address the health needs and related pain of individuals with intellectual disabilities.

Introduction

Prevalence rates of challenging behaviour in people with an intellectual disability vary between approximately 5% and 45% (Borthwick-Duffy, 1994; Emerson & Bromley, 1995; Emerson et al., 2001; Grey, Pollard, McClean, MacAuley, & Hastings, 2010; Lowe et al., 2007; Qureshi & Alborz, 1992). A growing body of research indicates that challenging behaviour has a detrimental effect on quality of life and leads to social exclusion and more limited service provision (Emerson, 2001; Kiernan & Qureshi, 1993; Murphy, 2009). Challenging behaviour is also a major source of stress for families as well as staff and the impact of challenging behaviour is evident in the NHS, with a high cost of service provision (Campbell, 2011; Gallagher et al., 2008; Knapp, Comas-Herrera, Astin, Beecham, & Pendaries, 2005; Unwin & Deb, 2011;).

The causes of challenging behaviour and demonstrations of intervention efficacy have dominated research in this area, with operant theory arguably the most well supported account. Evidence for the operant paradigm is derived from the results of both experimental and descriptive functional analyses and an extensive applied behaviour analytic intervention literature (Iwata et al., 1994; Scotti, Evans, Meyer, & Walker, 1991). Theoretical models of the development of self-injury, such as Guess and Carr's (1991) stage model, impute operant processes as maintaining and driving the emergence of more severe behaviour and there is evidence for this model from longitudinal studies of early self-injury (Oliver, Hall, & Murphy, 2005). Given the weight of evidence, it is highly likely that operant processes are influential in the development and maintenance of challenging behaviour for most people at some point. However, other causes of challenging behaviour cannot be ruled out as operant theory alone cannot, for example, explain the association between challenging behaviour and

pain related health conditions (de Winter, Jansen, & Evenhuis, 2011). This observation warrants further study as it might help account for the introduction of challenging behaviour into the repertoire prior to social reinforcement becoming influential, variability in the course of challenging behaviour and challenging behaviour that appears unrelated to environmental contingencies.

People with an intellectual disability are more likely to experience health problems and consequently pain and discomfort. van Schrojenstein Lantman-De Valk, Metsemaker, Haveman and Crebolder (2000) demonstrated a two-fold increase in the prevalence of health conditions in people with an intellectual disability, as compared to the general population. Health conditions common to individuals with an intellectual disability are numerous, including epilepsy and osteoporosis as well as disorders of the skin and gastrointestinal, respiratory and cardiovascular systems (Böhmer et al., 1999; Emerson, Baines, Allerton, & Welch, 2011; Jansen, Krol, Groothoff, & Post, 2004; Kerr, Fraser, & Felce, 1996; Srikanth, Cassidy, Joiner, & Teeluckdharry, 2011). As a result, individuals with an intellectual disability are more likely to experience a high degree of pain and discomfort (Breau, Camfield, McGrath, & Finley, 2003; Stallard, Williams, Lenton, & Velleman, 2001).

There is emerging evidence of an association between painful health conditions and challenging behaviour. Carr and Owen-DeSchryver (2007) reported higher frequency and intensity of self-injury and aggression on ‘sick’ than ‘well’ days across twelve participants. Using a prospective, correlational design, Symons and Danov (2005) showed that maternal ratings of a child’s pain were significantly higher when self-injury was occurring. An association between pain and challenging behaviour has also been reported in children with

particular genetic syndromes associated with intellectual disability, such as Cornelia de Lange syndrome (CdLS), in which gastro-oesophageal reflux and self-injury co-occur (Luzzani, Macchini, Valade, Milani, & Selicorni, 2003). More generally, Walsh, Morrison and McGuire (2011) report a significantly higher prevalence of challenging behaviour in participants experiencing chronic pain than those who are not, in a large, community sample. There is also evidence to suggest that treatment of specific health conditions results in the reduction of challenging behaviour in children with autism (Horvarth & Perman, 2002).

The observation of higher rates of challenging behaviour when poor health or pain is evident is complemented by a literature on health and pain in those who show challenging behaviour. Individuals with intellectual disabilities who self-injure demonstrate significantly more non-verbal behavioural signs of pain than those who do not (Symons, Harper, McGrath, Breau, & Bodfish, 2009) and participants experiencing chronic pain have been observed to self-injure near to the site of pain (Breau et al., 2003). Additionally, it has been suggested that individuals with intellectual disabilities who self-injure might show amplified pain expression (Defrin, Pick, Peretz, & Carmeli, 2004). These observations might be accounted for by interpreting the self-injury as an attempt to physically remove a source of pain or by pain gating. Melzack and Wall's (1965) gate control theory would explain self-injury as functioning to reduce the pain experienced at another body site through the activation of non-nociceptive fibres which do not respond to pain stimuli. Such activation works to 'close the gate' on the transmission of pain signals to the brain, and thus reduce the perception of pain. These explanations suggest a direct relationship between pain and self-injury that is not mediated by the environment, so that other functions of the self-injury are unlikely to be operative.

In order to explore an alternative explanation for the association between pain and challenging behaviour, researchers have used functional analysis to explore the role of pain as a setting event (Kennedy & O'Reilly, 2006). Setting events are variables which influence behavioural responses by altering their relationships with antecedent stimuli and are thus closely related to establishing operations (Bijou & Baer, 1961; Michael, 1982). The relationship between an antecedent stimulus and a behavioural response can be made more or less probable by the presence or absence of a setting event (Carr & Smith, 1995). O'Reilly (1997), for example, reported that self-injury in a 2 year old girl with William's Syndrome was only demonstrated in a loud noise condition during episodes of otitis media. Further support for the role of pain as a setting event for challenging behaviour using a larger sample has been provided by Carr, Smith, Giacin, Whelan and Pancari (2003). Using natural observations and experimental functional analysis across three participants, significantly more self-injury was demonstrated in the presence of both menses and task demands than in menses alone, so that menstrual pain acted as a setting event for self-injury. Thus, when acting as a setting event, pain is identified as a function of challenging behaviour alongside well established operant functions.

Evidence is accumulating to support an association between pain and challenging behaviour imputing both a direct and mediated cause. However, this has predominantly been investigated for self-injury, with less evidence regarding other forms of challenging behaviour. The use of single case experimental designs has also limited the external validity of the findings obtained. Further research is required to investigate the relationship between pain and multiple forms of challenging behaviour in samples of individuals with intellectual disability.

The identification of pain in individuals with an intellectual disability is difficult as assessment of pain relies heavily on self report which may be compromised for this population. In order to examine a causal relationship between pain and challenging behaviour empirically, temporal associations must first be observed and then manipulations of independent variables undertaken. The latter is both impractical and clearly unethical. However, a questionnaire, completed by carers, can overcome these problems. The Questions About Behavioral Function (QABF; Matson & Vollmer, 1995) has been commonly used for the purpose of functional analysis. The QABF has good test-retest reliability and, in comparison to other measures such as the Motivation Assessment Scale (Durand & Crimmins, 1988), good inter-rater reliability, high internal consistency for both individual subscales and the scale as a whole and good discriminant and convergent validity with the results of experimental functional analysis and other checklists (Nicholson, Konstantinidi, & Furniss, 2006; Paclawskyj, Matson, Rush, Smalls, & Vollmer, 2000, 2001; Zaja, Moore, Ingen, & Rojahn, 2011). Clearly this measure could be useful for identifying pain related challenging behaviour if the pain subscale of the QABF has robust psychometric properties. However, to date, the validity of this subscale specifically has not been examined and thus the first aim of this study is to examine, using a range of pain behaviour measures, the comparative levels of pain behaviour shown by children demonstrating pain and non pain related challenging behaviour, as identified by the QABF.

The results of several studies have indicated an association between low mood and pain in individuals with an intellectual disability. Tervo, Symons, Stout and Novacheck (2006) observed that pain is expressed by children with limited verbal communication through deviation from typical mood. Breau and Camfield (2011) also demonstrated that pain was

scored higher in children with a clinically significant low mood score. The association between pain and mood is so widely accepted within the literature that low mood has often been used as an indicator of pain (Symons, Shinde, & Gilles, 2008). Observational measures of pain such as the FLACC (Merkel, Voepel-Lewis, Shayevitz, & Malviya, 1997) have also used activity levels as a pain marker, as these variables have been shown to co-occur within the literature. In a sample of children with an intellectual disability, activity was shown to decrease during incidents of pain (Breau, Camfield, McGrath, Rosmus, & Finley, 2001). Conversely, Luzzani et al. (2003) observed an increase in activity in the presence of pain. Thus, although an association between pain and activity has been identified within the literature, the exact nature of this association is unclear. A second aim of this study is to examine mood and activity levels in children demonstrating pain and non pain related challenging behaviour.

As noted above, pain is hypothesised to act as a setting event or a direct cause for challenging behaviour. However, data relevant to these possibilities have not been reported for a large sample of individuals with an intellectual disability. Identification of a pain function, alongside other functions, would support a setting event interpretation. Conversely, pain as a direct cause of challenging behaviour would be more likely to be identified in the absence of additional functions. However, clearly a combination of these possibilities is feasible given that pain might lead directly to self-injury, for example, at times but also act as a setting event at other times. The final aim of this study is to examine the relationship between pain related challenging behaviour and social and stimulatory functions of challenging behaviour in a large group of children with intellectual disability using the QABF, to examine, how

frequently pain might operate as a setting event and appears to be an unmoderated cause of challenging behaviour.

To summarise, the aims of this study are to:

1) examine the validity of the identification of pain related challenging behaviour by the QABF by comparing children showing pain related challenging behaviour (as identified by the QABF) to children showing challenging behaviour unrelated to pain, on indices of pain behaviour
2) examine differences in mood and activity in children identified as having pain and non pain related challenging behaviour
3) describe the proportion of children with an intellectual disability showing challenging behaviour identified as showing pain related challenging behaviour by the QABF
4) examine the relationship between pain related challenging behaviour and social and stimulatory functions of challenging behaviour

Method

Participants

46 children aged between 4 and 15 years (mean = 11 years, 0 months) participated. Participants were diagnosed with a range of syndromes, including autism spectrum disorder (n = 8), Rubinstein-Taybi (n = 8), Angelman (n = 6), Tuberous Sclerosis Complex (n = 6), 1p36 deletion (n = 5), Fragile X (n = 4), Cri du Chat (n = 3), Cornelia de Lange (n = 2), Prader Willi (n = 2), Down (n = 1) and 8p23 deletion (n = 1) syndromes. Twenty four

participants (52.2%) were female. The majority of the sample was verbal (69.6%) and ambulant (80.4%), with normal vision (84.8%) and hearing (69.6%). Almost half of the sample (44%) was classed as "not able" according to the Wessex. Forty two (91.3%) participants' adaptive behaviour standard score was classified as low, the rest were classified as moderately low. SCQ scores indicated that 84.8% of the sample scored above the cut-off for ASD and 52.5% for autism, with a mean total score of 39. According to the CBI, 76.1% of the sample showed self-injury, 87% aggression and 63% both self-injury and aggression.

Measures

The Activity Questionnaire (TAQ; Burbidge et al., 2010)

Using 18 items scored on a five point Likert scale ("0 - never/almost never" to "4 - always or most of the time"), the TAQ measures total activity (scored between 0 and 72), impulsivity (scored between 0 and 36) and overactivity (scored between 0 and 36). The internal consistency, internal and inter-rater reliability of the TAQ have been established (Burbidge et al., 2010).

The Challenging Behaviour Interview (CBI; Oliver et al., 2003)

The CBI provides a detailed description of the severity and impact of challenging behaviour and is conducted in two parts. Firstly, the respondent is asked whether the participant has shown specific types of challenging behaviour (e.g. aggression, self-injury) within the last month using operational definitions and examples of each. The second part of the interview assesses the severity of each form of behaviour identified in part one through the aggregate of

fourteen items scored on a four or five point Likert scale. The authors reported good inter-rater, test-retest reliability and content validity for the interview.

The Health Questionnaire (HQ; Hall, Arron, Sloneem, & Oliver, 2008)

The HQ requires informants to rate the presence and severity of 15 health conditions over the previous month on a four point Likert scale ranging from 0 (never) to 3 (severe). Total severity scores range from 0 to 48. Hall et al. (2008) reported good internal reliability for the scale, although the validity of the questionnaire is yet to be determined.

The Mood, Interest and Pleasure Questionnaire – Short Form (MIPQ-S; Ross & Oliver, 2003; Arron, Oliver, Moss, Berg, & Burbidge, 2011).

The MIPQ-S consists of 12 items which informants rate on a five point Likert scale, based on observations made over the last two weeks. The data derived from these items can be used to calculate total affect (scored between 0 and 24) and subscale scores: mood and interest/pleasure (each scored between 0 and 12). Good levels of internal consistency, test-retest and inter-rater reliability for this version of the scale have been reported (Arron, Oliver, Moss, Berg, & Burbidge, 2011).

The Non-Communicating Child Pain Checklist-Revised (NCCPC-R; Breau, McGrath, Camfield, & Finley, 2002)

The NCCPC-R requires informants to rate the frequency of 30 observable pain related behaviours on a five point Likert scale. The data derived from these items can be used to calculate total (scored between 0 and 90) and subscale scores: vocal (scored between 0 and 12), social (scored between 0 and 12), facial (scored between 0 and 15), activity (scored

between 0 and 6), body/limbs (scored between 0 and 18), physiological (scored between 0 and 18) and eating/sleeping (scored between 0 and 9). The version utilised within this study was adapted to measure observable pain related behaviours over the previous week (as opposed to the previous two hours in the original version) in order to identify chronic or intermittent as opposed to acute health problems and related pain. Adapting this measure to a week-long observation period also brought this in line with other measures used within this study. Examination of the psychometric properties of this adapted version indicates good levels of inter-rater and test-retest reliability (Eden, 2012).

Questions About Behavioral Function (QABF; Matson & Vollmer, 1995)

The QABF measures the function of behaviour using 25 items, each requiring the informant to rate on a four point Likert scale from 0 (never) to 3 (often) the frequency with which challenging behaviour was demonstrated in a particular situation. Item scores are used to derive five subscales; attention, task escape, self-stimulation, pain and tangibles, each comprising five items and scored on a range of 0 to 15. Validity, internal consistency and inter-rater reliability of the QABF are robust (Matson, Bamburg, Cherry, & Paclawskyj, 1999; Nicholson, Konstantinidi, & Frederick, 2006).

The Social Communication Questionnaire: Current Version (SCQ; Rutter, Bailey, Berument, Lord, & Pickles, 2003)

The SCQ was used as a measure of autism spectrum disorder (ASD). Consisting of 40 items related to behaviours and characteristics associated with ASD demonstrated during the previous three months, the informant rated questionnaire provides scores for three subscales; social interaction, communication and repetitive behaviour. All items require a yes/no

response and are scored as 0 or 1 respectively. Total scores range from 0 to 40. A cut off of 15 and 22 for ASD and autism respectively has been proposed (Rutter *et al*., 2003). Good concurrent validity has been found with various other measures of ASD (Howlin & Karpf, 2004).

The Vineland Adaptive Behavior Scale Survey Edition (VABS; Sparrow, Balla, & Cicchetti, 1984)

The VABS comprises four subscales; communication, daily living, socialisation and motor skills. Subscales are derived from 383 items, scored between 0 (never) and 2 (usually). The adaptive behaviour standard score was used as a measure of adaptive behaviour. The authors report good validity, internal consistency, test-retest and inter-interviewer reliability. The VABS adaptive behaviour standard score was used as the primary measure of adaptive behaviour within the study.

The Wessex Questionnaire (Kushlick, Blunden, & Cox, 1973)

The Wessex is an informant based questionnaire that measures adaptive behaviour, communication, physical disability and sensory impairment using 16 items, scored on a three or four point Likert scale. Total scores range from 16 to 49. Examination of the psychometric properties of the Wessex indicates good validity and reliability at a subscale level (Kushlick et al., 1973) as well as inter-rater reliability across different populations and settings (Palmer & Jenkins, 1982). The Wessex was utilised within the study to provide information as to the physical characteristics of participants (such as sight and hearing) and a basic adaptive behaviour score.

Recruitment

Participants were recruited from a database held at the Cerebra Centre for Neurodevelopmental Disorders at the University of Birmingham. This database holds contact information and data for almost 1600 people with various syndromes associated with an intellectual disability. Participants were originally recruited to the database via syndrome support groups and took part in a continuing questionnaire study, investigating syndrome specific characteristics.

All participants were recruited as part of a larger study investigating pain in children with an intellectual disability, for which the primary inclusion criterion was that all participants were between 4 and 15 years of age with a syndrome associated with intellectual disability. For this study, an additional inclusion criterion was aggression and/or self-injury within the previous month, according to parents/carers responses to the CBI. These types of challenging behaviour were chosen due to their clinical importance.

Procedure

Parents/carers of children whose data are held on the database were contacted by telephone if they had given consent to be contacted regarding future research and the child they cared for was aged between 4 and 15 years. Information regarding the study (see Appendix C) and a questionnaire pack including the Wessex, SCQ, NCCPC-R, HQ, MIPQ and TAQ (see measures and Appendix D other questionnaires were also included in the pack but were not utilised within this study) were sent to each parent/carer who had given verbal consent to take part in the study over the telephone. Upon receipt of a consent form (Appendix E) and completed questionnaire pack, parents/carers were contacted to conduct the telephone

interview, involving completion of the QABF, VABS and the CBI, taking approximately one hour (see Appendix F). The QABF was conducted up to four times per participant, with regard to up to two topographies of aggression and self-injury identified during the CBI. When children demonstrated more than two topographies of aggression and/or self-injury, parents/carers identified the most frequent and/or severe (causing most concern to the parent/carer because of risk of injury to individual and/or others) topography of aggression and/or self-injury for the basis of QABF completion.

Ethical approval to conduct this study, as part of a larger project, was obtained from Birmingham, East, North and Solihull Research Ethics Committee (see Appendix G).

Data analysis

The authors of the QABF (Matson & Vollmer, 1995) suggest that an endorsement (scoring 1 or above) of at least four or five items on one QABF subscale, with no other subscales including 'significant' endorsements, is indicative of 'function'. Endorsement of four items on one subscale of one QABF, irrespective of other subscale endorsements, was employed as a definition of function for that subscale within this study, so that multiple functions could be identified. The pain related challenging behaviour group consisted of participants with four or more QABF pain item endorsements on at least one QABF, whilst the non pain related challenging behaviour group included participants who received three or less QABF pain item endorsements on all QABFs. A total 'severity' score was also derived from the QABF for each subscale. This was calculated by summing all QABF items of each subscale across all QABFs for each participant.

Statistical analysis of the dataset using multiple Sharipo-Wilk tests indicated that much of the data was not normally distributed and skewed in various ways. As a result, the remaining data analysis involved only non parametric tests, including Spearman's Rho, Mann Whitney U and Fisher's Exact Test. All analyses conducted were two-tailed, unless a significant difference between groups was hypothesised. Relative risk analyses with 95% confidence intervals were employed.

Results

Participant total and subscale scores on all measures are shown in Table 1.

Table 1: Total and subscale scores for the whole sample on all measures

Measure	Subscale	Median Score	IQR
NCCPC	Vocal	3	4
	Social	4	3
	Facial	4	5
	Activity	2	2.25
	Body and Limbs	2	5
	Physiological	1	3
	Eating/Sleeping	0	2
	Total	**17.5**	**14.75**
HQ	**Total**	**3**	**4**
MIPQ	Mood	20	3.5
	Interest/Pleasure	17	4
	Total	**38**	**5.5**
TAQ	Impulsivity	20	12
	Overactivity	16	17.5
	Total	**35**	**26**
QABF	Total Pain Severity	8	24.75
	Total Attention Severity	13	17.25
	Total Task Escape Severity	16	17
	Total Self Stimulation Severity	9.5	17
	Total Tangible Severity	21	26

HQ score = severity of health problems (see Measures)

Pain and non pain related challenging behaviour groups

In order to evaluate the validity of the QABF, pain and non-pain related challenging behaviour groups were formed (see Method). Twenty three participants comprised each group. Table 2 shows demographic information for these groups.

Table 2: Demographic information for the pain and non pain related challenging behaviour groups (median and inter-quartile ranges)

	Pain Function Group (n = 23)	Non Pain Function Group (n = 23)
Age	12 (4)	12 (5)
Male	8 (34.8%)	14 (60.9%)
VABS adaptive behaviour standard score	43 (18)	58 (25)
SCQ total score	20 (14.8)	23 (6.9)

To examine the comparability of the groups, demographic data were evaluated. Statistical analyses indicated that the groups were significantly different with regard to adaptive behaviour, as measured by the VABS adaptive behaviour standard score (U = 152, p = .01), with the pain function group scoring significantly lower than the non pain function group. There was no significant difference between groups for age (U = 217.5, p = .30), gender (χ^2 (1, N = 46) =, p = .14) or SCQ total score (U = 206, p = .29).

Given the significant difference between the groups on the VABS adaptive behaviour standard score, it is possible that this could confound analyses of group differences on pain related indices. Consequently, in order to examine the relationship between adaptive behaviour and pain related indices, Spearman's rho correlations were derived between all pain and health related measures (QABF pain severity; NCCPC-R total and subscales; HQ total severity score), variables purported to be associated with pain (MIPQ total and subscales; TAQ total and subscales) and adaptive behaviour as measured by the total VABS adaptive

behaviour standard score, within each group. Results indicated that for the vast majority of analyses (32 correlations, see Appendix H for correlation matrix) VABS adaptive behaviour standard score was not significantly correlated with pain indices except for a significant correlation with NCCPC facial (r = -.39, p = .04) for the pain related challenging behaviour group and TAQ overactivity (r = -.53, p = .01) for the non pain related challenging behaviour group. These results indicate that adaptive behaviour, as measured by the VABS adaptive behaviour standard score, is unlikely to be a confounding variable in group comparison analyses as it was significantly correlated with so few pain related measures.

Validity of the identification of pain related challenging behaviour

In order to examine the validity of the identification of pain related challenging behaviour by the QABF, the pain and non pain related challenging behaviour groups were compared on indices of pain behaviour. The results of a series of Mann Whitney U tests, as shown in Table 3, indicated that the pain function group scored significantly higher on all measures of pain except NCCPC-R activity.

Table 3: Median, inter-quartile range and Mann Whitney U analyses for total and subscale scores for measures of pain

Measure	Variable	Pain Function Group (n = 23)	Non Pain Function Group (n = 23)	U (1 tailed)	*P*	Z
QABF Pain	Severity	24 (12)	0 (4)	8	<.001	-5.69
	Total	22 (20)	12 (12)	111	<.001	-3.38
NCCPC	Vocal	4 (3)	2 (2)	147.5	.005	-2.59
	Social	6 (4)	3 (2)	127	.001	-3.05
	Facial	5 (5)	3 (4)	136.5	.003	-2.84
	Activity	2 (2)	1 (1)	202	.078	-1.42
	Body/Limbs	3 (4)	0 (3)	175.5	.023	-2
	Physiological	2 (3)	0 (1)	117	<.001	-3.34
	Eat/Sleep	1 (1)	0 (1)	152	.004	-2.69
HQ	Total	5 (6)	3 (4)	151	.017	-2.13

HQ score = severity of health problems

All of these results were in the hypothesised direction with a large difference across groups for some pain indices. The significant group difference for health conditions across groups as measured by the HQ also indicates that adaptive behaviour, as measured by the VABS adaptive behaviour standard score, is unlikely to be a confounding variable as the HQ is not a measure of behaviour but physical condition.

Additional variables associated with pain

In order to examine differences in mood and activity in children identified as having pain and non pain related challenging behaviour, further statistical analyses were conducted. The

results of Mann Whitney U analyses, as illustrated in Table 4, demonstrated that no significant differences between groups were identified for the total MIPQ score, interest/pleasure, TAQ total, impulsivity or overactivity, although the difference between the groups with regard to mood and overactivity approached significance.

Table 4: Median, inter-quartile range and Mann Whitney U analyses for total and subscale scores for variables associated with pain

Measure	Variable	Pain Function Group (n = 23)	Non Pain Function Group (n = 23)	U (1 tailed)	*P*
MIPQ	**Mood**	19.5 (5.25)	21 (3)	167.5	.05
	Interest/ Pleasure	17 (6.25)	18 (5)	239	.75
	Total	**37 (7)**	**38 (5)**	**197.5**	**.21**
TAQ	**Impulsivity**	20 (12.5)	20 (11)	230	.60
	Overactivity	21 (18.5)	12 (7)	175	.08
	Total	**39 (23.25)**	**33 (31)**	**218.5**	**.43**

Functions of challenging behaviour

To describe the proportion of children with intellectual disability showing challenging behaviour identified as showing pain related challenging behaviour by the QABF, the functions of challenging behaviour for the whole sample and participants with pain related challenging behaviour only were examined. Of the total sample, 32 participants (69.6%) demonstrated challenging behaviour as a function of access to tangibles, 32 (69.6%) task escape, 27 (58.7%) attention, 23 (50%) pain and 20 (43.5%) self stimulation.

Of those identified as having pain related challenging behaviour, the majority also demonstrated challenging behaviour with a function additional to pain, so that only one participant (4.4%) demonstrated pain related challenging behaviour without another function. Another participant (4.4%) demonstrated one other function, five participants (21.7%) two functions, another five participants (21.7%) three functions and eleven participants (47.8%) showed all four other functions.

The relationship between pain related challenging behaviour and social and stimulatory functions of challenging behaviour

Two statistical methods were utilised in order to investigate the social and stimulatory functions of challenging behaviour demonstrated by the pain and non pain related challenging behaviour groups. First, the difference in total severity of the relevant QABF subscales was compared using Mann Whitney U analyses. The results of this analysis, as reported in Table 5, demonstrated a significant difference between groups for attention, task escape and self stimulation, with the pain related challenging behaviour group scoring significantly higher on total severity for each of these subscales than the non pain related challenging behaviour group. No significant difference across groups was found with regard to severity on the QABF tangible subscale.

Table 5: Median, inter-quartile range and Mann Whitney U analyses for QABF functions

QABF Subscale Severity	Pain Function Group (n = 23)	Non Pain Function Group (n = 23)	U (1 tailed)	p
Attention	15 (13)	4 (17)	175	.049
Task Escape	22 (16)	9 (21)	148.5	.011
Self Stimulation	15 (5)	3 (14)	155	.016
Tangible	23 (23)	10 (24)	200	.156

Fisher's Exact test was also used in order to identify significant differences in the number of children identified as being functional for each of the remaining QABF subscales (defined as scoring one or above on four or more items for a particular subscale on at least one QABF) across pain function groups. The results of this analysis indicated that significantly more children in the pain function group were also functional for attention (p = .02) and task escape (p = .01), but not self stimulation (p = .14) or tangible (p = .34). The relative risks for attention and task escape function given pain related challenging behaviour were 2.53 (CI = 1.14-5.63) and 4.59 (CI = 1.24-16.98) respectively.

To summarise, the pain related challenging behaviour group scored significantly higher for severity of three other functions (attention, task escape and self stimulation) and demonstrated significantly more additional functions (attention and task escape) than the non pain related challenging behaviour group.

Discussion

In order to further validate the QABF, participants showing pain related challenging behaviour (as identified by the QABF) were compared to participants whose challenging behaviour was unrelated to pain, on indices of pain behaviour. Group differences with regard to mood and activity were also examined in order to investigate the association between these variables and challenging behaviour in the context of pain. A description of the proportion of children with an intellectual disability showing challenging behaviour identified as showing pain related challenging behaviour by the QABF was also provided. The final aim of this study was to examine the relationship between pain related challenging behaviour and social and stimulatory functions of challenging behaviour. The number and specificity of the pain measures used to validate the QABF and relatively large sample employed are strengths of this study.

The pain function group scored significantly higher on all measures of pain, except NCCPC-R activity, than the non pain related challenging behaviour group. These results are supportive of the validity of the QABF pain function subscale. Despite a significant difference between pain and non pain related challenging behaviour groups on the VABS adaptive behaviour standard score, adaptive behaviour was not considered a confounding variable within this or the remaining analyses since it was not significantly correlated with the majority of pain indices and variables associated with pain. Additionally, the pain related challenging behaviour group experienced a higher number of health conditions and this, in combination with the lack of within group correlations between the VABS and pain indices, strongly suggests that the differences between groups are more likely to be attributable to pain and

discomfort as a result of health conditions than the potentially confounding variable of adaptive behaviour.

Further statistical analysis indicated that the differences in mood and overactivity between the pain and non pain related challenging behaviour group approached significance, although no significant differences between groups were identified for total MIPQ score, mood, interest/pleasure, TAQ total score, overactivity or impulsivity. Thus, the results of this study indicate that mood and activity warrant further investigation in this context. At this stage it appears that they are too broad as measures of possible pain or that they are associated with specific types of pain and discomfort.

Access to tangibles and task escape were the most common functions of challenging behaviour identified. The challenging behaviour of children demonstrating pain related challenging behaviour appeared to be multifunctional, so that only one participant was not reported to have an additional function. Further statistical analyses indicated that the pain related challenging behaviour group scored higher on the attention, task escape and self stimulation QABF subscales, although no significant group differences were identified for the tangible subscale. Significantly more participants in the pain than non pain challenging behaviour group were also classified as functional for attention and task escape, but not self stimulation or tangible reinforcement. The strongest association identified (relative risk ratio of over four) between pain and task escape has also been highlighted in a recent systematic review of the literature (Langthorne, unpublished), which proposes a specific relationship between health problems and related pain and escape maintained challenging behaviour.

The validation of one aspect of the QABF demonstrated within this study further supports the utility of this measure when attempting to identify the function of challenging behaviour in individuals with an intellectual disability (Matson, Tureck, & Rieske, 2012). Due to the time consuming nature of, and ethical concerns regarding, experimental functional analyses, the QABF is a much needed resource, due to its ease of completion and demonstrable psychometric properties. However, the reliability of the QABF is compromised for low rate behaviours (Matson & Wilkins, 2009) and those with multiple functions, which were demonstrated by the vast majority of children in this study and is common within this population (Matson & Boisjoli, 2007). This is to be expected as challenging behaviour is commonly influenced by the environment and thus would be demonstrated at different times within various contexts in which it receives reinforcement (Matson et al., 1999). Had a more stringent definition of function been adopted within this study (i.e. five or more endorsements or absence of significant endorsements within other subscales), such multiple functions would probably not have been identified, thus indicating the need to use the recommended definitions of function with caution (Matson & Vollmer, 1995).

Although a relationship between both mood and challenging behaviour (Carr, McLaughlin, Giacobbe-Grieco, & Smith, 2003; Durand & Mapstone, 1998; Lindauer, DeLeon, & Fisher, 1999) and activity and challenging behaviour (Arron et al., 2011; Davies & Oliver, in preparation) has been demonstrated in the literature, neither mood nor activity, according to the results of this study, appeared to be strongly associated with pain related challenging behaviour. These non-significant findings might be a result of the way in which these variables were measured, so that more detailed, observational methodologies might have identified subtle differences in the nature of activity levels between these groups. Difficulties

in measuring mood due to the communication deficits experienced by individuals with intellectual disabilities might also mean that low mood in this population goes undetected. Further research is required in order to refine the concept of low mood in this population and how it might be more accurately detected and assessed.

The identification of social and stimulatory functions of challenging behaviour in all but one participant with pain related challenging behaviour indicates that, in line with previous research (Carr et al., 2005; O'Reilly, 1997), pain potentially acts as a setting event for challenging behaviour. Thus, individuals with an intellectual disability experiencing pain which impacts on the challenging behaviour they demonstrate, might experience particular environmental stimuli as less tolerable. Such situations, according to the results of this study, include low levels of attention and task escape, but not lack of access to tangibles. Support for attention (e.g. Sloneem, Oliver, Udwin, & Woodcock, 2011; Tiger, Fisher, Toussaint, & Kodak, 2009) and task escape (e.g. Lalli, Kates, & Casey, 1999; McComas, Hoch, Paone, & El-Roy, 2000) as functions for challenging behaviour is apparent within the literature, although empirical evidence for the potential interaction between pain and these functions of challenging behaviour has not previously been available. An interaction with self stimulation appears to be less clear.

Limitations of the study

Despite the utility of the QABF as a measure of pain related challenging behaviour, the addition of experimental functional analysis or observational data would have strengthened the methodology of this study and made identification of challenging behaviour function more accurate (Matson et al., 2012). Furthermore, although the use of multiple pain indices

comprised a major strength of this study, measures of pain in individuals with an intellectual disability remain inferior to those utilised with typically developing individuals and thus conclusions drawn from this study must be considered in light of this. An observational measure such as the FLACC might have increased the validity of this study. The use of the NCCPC-R with verbal participants might be perceived as a limitation of this study, but was deemed necessary in order to measure pain in a standardised manner across all participants, a third of whom were not verbal. Finally, although pain scores were demonstrably higher in the pain related challenging behaviour group, whether the pain was directly related to challenging behaviour was not examined.

A less stringent definition of function was utilised within the study in order to enable identification of multiple functions of challenging behaviour. As a result, it is possible that errors of commission have been made, in that functions of challenging behaviour were wrongly identified. However, given the robust association between pain and challenging behaviour, it is, arguably, more beneficial to this population that this type of error is made, rather than functions of challenging behaviour being undetected. In this study, function of challenging behaviour was assumed across all types of challenging behaviour demonstrated by each participant if only identified in aggression or self-injury, although this might not have been the case. Again, whilst this prevented an error of omission, it also restricted investigation of different functions of different types of challenging behaviour, which might have proved insightful given the documented association between specific types of challenging behaviour and functions (Dawson, Matson, & Cherry, 1998).

Because of the different types of non-normal distributions present across data, transformation to a normal distribution was not practical and thus, more powerful analyses, such as ANCOVA which could have taken differences in adaptive behaviour across groups into account, could not be applied. A larger sample would have also allowed for separate analyses of self-injury and aggression, which as noted above, might have indicated differences with regard to function across different types of challenging behaviour. Sample size also limited the power of the statistical analysis employed, which might have been responsible for the non significant Fisher's exact test related to self stimulation.

Implications and further research

The results of this study clearly highlight the need to address the health needs and related pain of individuals with intellectual disabilities. Confirmatory evidence for the role of pain as a setting event for challenging behaviour has been obtained and although this finding must be replicated within this population, it is clear that pain is associated with challenging behaviour and when related appears to contribute to particular environmental stimuli as experienced as more aversive. As a result, levels of challenging behaviour may be more varied across the same environmental contexts and thus, clinical interventions aimed at reducing the frequency of such behaviour will need to address health problems and related pain. The young sample with whom these results were identified also indicates the importance of this work within an early intervention context.

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Chapter Three

Public Domain Briefing Document

Overview

Challenging behaviour, such as aggression and self-injury, is a significant issue for individuals with an intellectual disability, demonstrated by up to 45% of this population according to the literature (e.g. Grey, Pollard, McClean, MacAuley, & Hastings, 2010). Adverse consequences of challenging behaviour include social exclusion, more limited service provision, stress experienced by families and staff and a financial strain on the NHS (Campbell, 2011; Emerson, 2001; Gallagher et al., 2008; Knapp, Comas-Herrera, Astin, Beecham, & Pendaries, 2005; Murphy, 2009). Although much research has already been conducted in this area, further research is required to increase our understanding regarding the causes and function (purpose) of challenging behaviour in this population.

Literature Review

Background

Within the literature, it has been suggested that challenging behaviour is a symptom of depression in people with an intellectual disability, and is thus a "depressive equivalent" (e.g. Marston, Perry, & Roy, 1997; Smiley & Cooper, 2003). There is certainly evidence to indicate an association between depression and challenging behaviour (e.g. Clarke & Gomez, 1999; Lowry & Sovner, 1992), although methodological limitations inherent within studies

producing this data weaken conclusions drawn from them, so that several researchers have questioned the use of challenging behaviour as diagnostic criteria for depression (McBrien, 2003; Ross & Oliver, 2003).

Aim

A systematic review of studies containing data regarding the association between depression and aggression and/or self-injury was conducted.

Method

All peer reviewed, published articles examining the association between depression and challenging behaviour between 1967 and June 2011 were identified by a systematic literature search. The inclusion and exclusion criteria for studies were set so that included papers; were written in English, contained data on the association between depression and aggression and/or self-injury specifically which had been statistically analysed, employed a sample that was generally representative of the population of people with an intellectual disability and did not rely on the examination of the effect of medication on depression and aggression and/or self-injury, as this was not deemed robust evidence of an association between these variables. Fifteen studies, thirteen regarding aggression and twelve regarding self-injury (ten of the fifteen studies provided data regarding both aggression and self-injury), meeting these criteria were identified and included in this review.

Results and conclusions

The results of the studies identified indicated that the association between depression and both aggression and self-injury is equally ambiguous. In both cases, four studies identified an

association, four did not and the results of a further five in the case of aggression and four for self-injury were equivocal. Based on these studies, it would appear that there is currently insufficient evidence to support the use of challenging behaviour as a depressive equivalent. These results must be considered in light of a few methodological limitations however, both within this study and the studies forming the basis of this review. For example, despite the development of clear inclusion and exclusion criteria, it could be argued that these were not specific enough, so that more strict criteria could have been employed, although this would have further limited the number of papers included. Methodological limitations within the studies identified also existed, so that for example, the samples employed by some studies were small and recruited from institutions, thus potentially inflating the association between depression and challenging behaviour and reducing the validity of these findings.

An alternative account to challenging behaviour as a depressive equivalent was proposed whereby pain is a third variable and is thus responsible for the association between challenging behaviour and depression as both are associated with pain. This was deemed feasible given the given the prevalence of health problems and related pain in this population (e.g. Stallard, Williams, Lenton, & Velleman, 2001) and thus it was suggested that clinicians should assume that depression in the context of challenging behaviour is the result of pain in the first instance, before assuming a direct relationship between depression and challenging behaviour. Future research is required to clarify the status of challenging behaviour as a depressive equivalent. Such research should focus on third variables like pain involved in the association between depression and challenging behaviour and must be more methodologically robust than those conducted previously.

Empirical Paper

Background

Due to the high prevalence of health conditions and associated pain in individuals with an intellectual disability (Breau, Camfield, McGrath, & Finley, 2003) the role of pain as a function of challenging behaviour (e.g. self-injury as a method of pain reduction) has received much attention within the research literature. Using a variety of methodologies, researchers have demonstrated an association between pain and challenging behaviour (e.g. Carr & Owen-DeSchryver, 2007; Symons & Danov, 2005), although studies have typically employed small samples, limiting the validity of these findings, and focused largely on self-injury. The exact nature of the association between pain and challenging behaviour is also yet to be determined. The Gate Control Theory of pain (Melzack & Wall, 1965) proposes a direct association between pain and challenging behaviour, so that self-injury directly reduces the experience of pain and is thus reinforced, so that the individual is more likely to demonstrate self-injury in the future. A setting events model (Carr & Smith, 1995) has also been applied in this context, so that researchers (including O'Reilly, 1997 and Carr, Smith, Giacin, Whelan, & Pancari, 2003) have proposed a less direct relationship between pain and challenging behaviour, whereby pain acts as a setting event for challenging behaviour, making environmental stimuli more aversive and thus challenging behaviour more probable.

Aims

The aims of this study were to examine the relationship between pain related challenging behaviour and other operant functions (e.g. task escape, attention) in a large sample of individuals with intellectual disability, as well as the validity of the pain subscale of the

Questions About Behavioral Function (QABF), used to measure pain related challenging behaviour.

Method

QABF data and measures of pain related behaviour were collected on 46 children aged between 4 and 15 years with a range of syndromes associated with intellectual disability. Participants were recruited from a database held at the Cerebra Centre for Neurodevelopmental Disorders at the University of Birmingham. Parents/carers of children whose data are held on the database were contacted by telephone if they had given consent to be contacted regarding future research and the child they cared for was aged between 4 and 15 years. Information regarding the study and a questionnaire pack were sent to each parent/carer who had given verbal consent to take part in the study over the telephone. Upon receipt of a consent form and completed questionnaire pack, parents/carers were contacted to conduct a telephone interview.

Results and conclusions

Statistical analysis indicated that the pain and non pain related challenging behaviour groups differed significantly on pain indices, and thus demonstrated the validity of the QABF as a measure of pain related challenging behaviour. Of those identified as having pain related challenging behaviour, the vast majority also demonstrated challenging behaviour with a function additional to pain and significantly more functions of challenging behaviour as compared to the non pain related challenging behaviour group. These findings provide support for the use of the pain subscale of the QABF in clinical practice and the potential role

of pain as a setting event for challenging behaviour. More broadly the results highlight the need to address the health needs and related pain of individuals with intellectual disabilities.

The number and specificity of the measures used to validate the QABF and relatively large and representative sample employed are strengths of this study. However, limitations within this study must also be acknowledged. Although the use of multiple pain indices was advantageous, measures of pain in individuals with an intellectual disability remain inferior to those utilised with typically developing individuals. The skewed distribution of the data also limited the type of statistical analysis which could be conducted whilst the sample size limited the power of the statistical analysis to detect significant differences. Additionally, a less stringent definition of function was utilised within the study than that recommended by the authors of the QABF, in order to enable identification of multiple functions of challenging behaviour. As a result, it is possible that errors of commission have been made, in that functions of challenging behaviour were wrongly identified. However, given the robust association between pain and challenging behaviour, it is arguably more beneficial to this population that this type of error is made, rather than functions of challenging behaviour being undetected.

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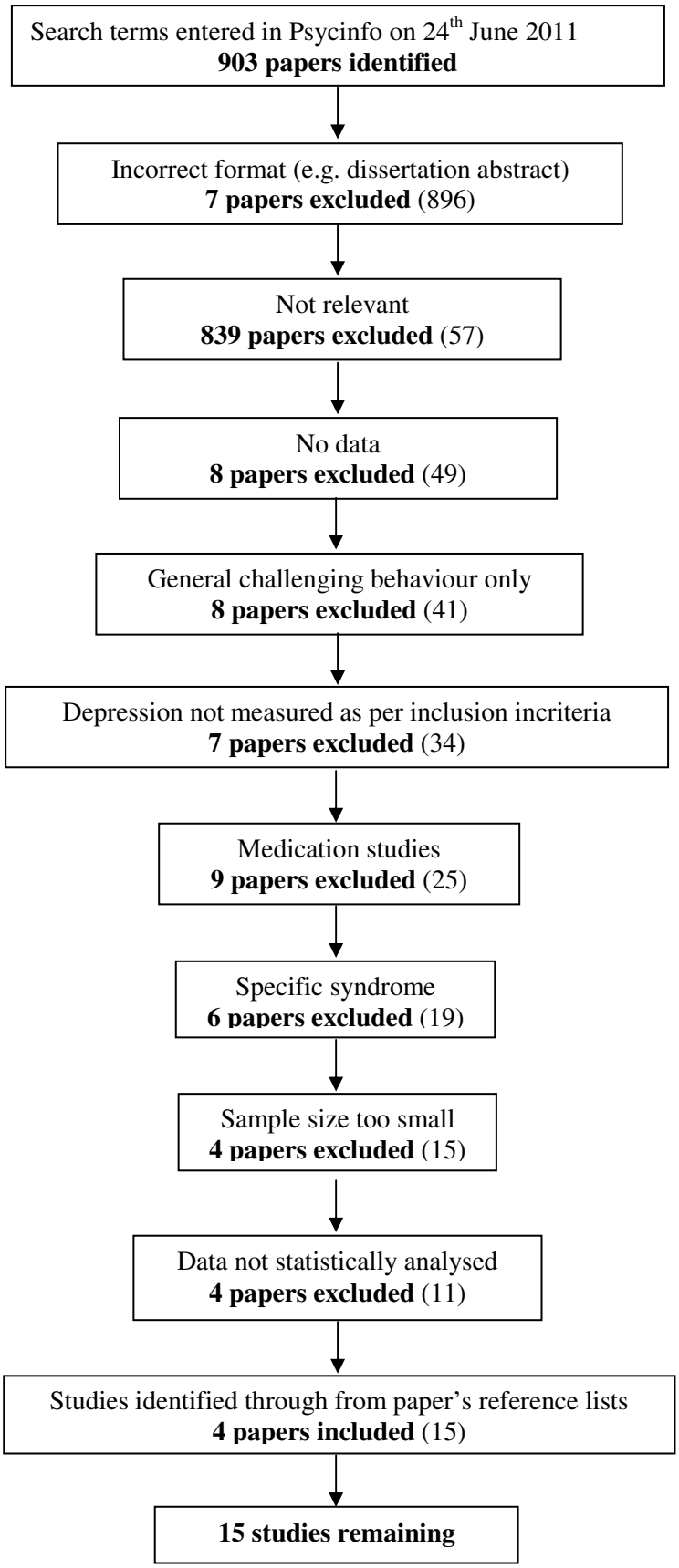
Appendix B: Flow chart to demonstrate paper selection



Appendix D: Questionnaire Pack

WESSEX QUESTIONNAIRE

These items refer to the person you care for. For each question (A, B, C, D etc …), please enter the appropriate code in each box.

A) Wetting (nights) **1** = frequently **2** = occasionally **3** = never ☐

B) Soiling (nights) **1** = frequently **2** = occasionally **3** = never ☐

C) Wetting (days) **1** = frequently **2** = occasionally **3** = never ☐

D) Soiling (days) **1** = frequently **2** = occasionally **3** = never ☐

E) Walk with help* **1** = not at all **2** = not up stairs **3** = up stairs and elsewhere ☐

*(note: if this person walks *by himself/herself* upstairs and elsewhere, please also code '3' for 'walk with help')

F) Walk by himself **1** = not at all **2** = not up stairs **3** = up stairs and elsewhere ☐

G) Feed himself **1** = not at all **2** = with help **3** = without help ☐

H) Wash himself **1** = not at all **2** = with help **3** = without help ☐

I) Dress himself **1** = not at all **2** = with help **3** = without help ☐

J) Vision **1** = blind or almost **2** = poor **3** = normal ☐

K) Hearing **1** = deaf or almost **2** = poor **3** = normal ☐

L) Speech **1** = never a word **2** = odd words only **3** = sentences and normal **4** = can talk but doesn't ☐

If this person talks in sentences, is his/her speech:

1 = Difficult to understand even by acquaintances, impossible for strangers?

2 = Easily understood for acquaintances, difficult for strangers?

3 = Clear enough to be understood by anyone? ☐

M) Reads **1** = nothing **2** = a little **3** = newspapers and/or books ☐

N) Writes **1** = nothing **2** = a little **3** = own correspondence ☐

O) Counts **1** = nothing **2** = a little **3** = understands money values ☐

Please check your answers and go on to the next questionnaire.

SOCIAL COMMUNICATION QUESTIONNAIRE © Rutter et al 2003

Please circle 'yes' if <u>any</u> one of the following behaviours is present. Although you may be uncertain about whether some behaviours are present or not, please do answer 'yes' or 'no' to every question on the basis of what you think.

1. Is she/he now able to talk using short phrases or sentences? If no, skip to question 8.	**Yes**	**No**
2. Can you have a to and fro "conversation" with her/him that involves taking turns or building on what you have said?	**Yes**	**No**
3. Has she/he ever used odd phrases or said the same thing over and over in almost exactly the same way (either phrases that she/he has heard other people use or ones that she/he has made up?	**Yes**	**No**
4. Has she/he ever used socially inappropriate questions or statements? For example, has she/he ever regularly asked personal questions or made personal comments at awkward times?	**Yes**	**No**
5. Has she/he ever got her/his pronouns mixed up (e.g., saying you or she/he for I)?	**Yes**	**No**
6. Has she/he ever used words that she/he seemed to have invented or made up her/himself; put things in odd, indirect ways; or used metaphorical ways of saying things (e.g., saying hot rain for steam)?	**Yes**	**No**
7. Has she/he ever said the same thing over and over in exactly the same way or insisted that you say the same thing over and over again?	**Yes**	**No**
8. Has she/he ever had things that she/he seemed to have to do in a very particular way or order or rituals that she/he insisted that you go through?	**Yes**	**No**
9. Has her/his facial expression usually seemed appropriate to the particular situation, as far as you could tell?	**Yes**	**No**
10. Has she/he ever used your hand like a tool or as if it were part of her/his own body (e.g., pointing with your finger, putting your hand on a doorknob to get you to open the door)?	**Yes**	**No**
11. Has she/he ever had any interests that preoccupy her/him and might seem odd to other people (e.g., traffic lights, drainpipes, or timetables)?	**Yes**	**No**
12. Has she/he ever seemed to be more interested in parts of a toy or an object (e.g., spinning the wheels of a car), rather than using the object as it was intended?	**Yes**	**No**
13. Has she/he ever had any special interests that were unusual in their intensity but otherwise appropriate for her/his age and peer group (e.g., trains, dinosaurs)?	**Yes**	**No**
14. Has she/he ever seemed to be unusually interested in the sight, feel, sound, taste, or smell of things or people?	**Yes**	**No**
15. Has she/he ever had any mannerisms or odd ways of moving her/his hands or fingers, such as flapping or moving her/his fingers in front of her/his eyes?	**Yes**	**No**

16. Has she/he ever had any complicated movements of her/his whole body, such as spinning or repeatedly bouncing up and down?	**Yes**	**No**
17. Has she/he ever injured her/himself deliberately, such as by biting her/his arm or banging her/his head?	**Yes**	**No**
18. Has she/he ever had any objects (other than a soft toy or comfort blanket) that she/he had to carry around?	**Yes**	**No**
19. Does she/he have any particular friends or a best friend?	**Yes**	**No**
20. When she/he was 4 to 5, did she/he ever talk with you just to be friendly (rather than to get something)?	**Yes**	**No**
21. When she/he was 4 to 5, did she/he ever spontaneously copy you (or other people) or what you were doing (such as vacuuming, gardening, or mending things)?	**Yes**	**No**
22. When she/he was 4 to 5, did she/he ever spontaneously point at things around her/him just to show you things (not because she/he wanted them)?	**Yes**	**No**
23. When she/he was 4 to 5, did she/he ever use gestures, other than pointing or pulling your hand, to let you know what she/he wanted	**Yes**	**No**
24. When she/he was 4 to 5, did she/he nod her/his head to mean yes?	**Yes**	**No**
25. When she/he was 4 to 5, did she/he shake her/his head to mean no?	**Yes**	**No**
26. When she/he was 4 to 5, did she/he usually look at you directly in the face when doing things with you or talking with you?	**Yes**	**No**
27. When she/he was 4 to 5, did she/he smile back if someone smiled at her/him?	**Yes**	**No**
28. When she/he was 4 to 5, did she/he ever show you things that interested her/him to engage your attention?	**Yes**	**No**
29. When she/he was 4 to 5, did she/he ever offer to share things other than food with you?	**Yes**	**No**
30. When she/he was 4 to 5, did she/he ever seem to want you to join in her/his enjoyment of something?	**Yes**	**No**
31. When she/he was 4 to 5, did she/he ever try to comfort you if you were sad or hurt?	**Yes**	**No**
32. When she/he was 4 to 5, when she/he wanted something or wanted help, did she/he look at you and use gestures with sounds or words to get your attention?	**Yes**	**No**
33. When she/he was 4 to 5, did she/he show a normal range of facial expressions?	**Yes**	**No**
34. When she/he was 4 to 5, did she/he ever spontaneously join in and try to copy the actions in social games, such as The Mulberry Bush or London Bridge Is Falling Down?	**Yes**	**No**
35. When she/he was 4 to 5, did she/he play any pretend or make-believe games?	**Yes**	**No**
36. When she/he was 4 to 5, did she/he seem interested in other children of approximately the same age whom she/he did not know?	**Yes**	**No**

37. When she/he was 4 to 5, did she/he respond positively when another child approached her/him?	**Yes**	**No**
38. When she/he was 4 to 5, if you came into a room and started talking to her/him without calling her/his name, did she/he usually look up and pay attention to you?	**Yes**	**No**
39. When she/he was 4 to 5, did she/he ever play imaginative games with another child in such a way that you could tell that they each understood what the other was pretending?	**Yes**	**No**
40. When she/he was 4 to 5, did she/he play cooperatively in games that required joining in with a group of other children, such as hide-and-seek or ball games?	**Yes**	**No**

Please check your answers and go on to the next questionnaire.

ACTIVITY QUESTIONNAIRE

Instructions:

- **Please read each item carefully and circle the appropriate number on the scale, for the person you care for.**
- **Please ensure that you indicate a response for every item. If the particular behaviour does not apply, for example, if the person is not verbal or not mobile, please circle 0 on the**

		Never/ almost never	Some of the time	Half of the time	A lot of the time	Always/ almost all the time
1.	Does the person wriggle or squirm about when seated or lying down?	**0**	**1**	**2**	**3**	**4**
2.	Does the person fidget or play with their hands and/or feet when seated or lying down?	**0**	**1**	**2**	**3**	**4**
3.	Does the person find it difficult holding still?	**0**	**1**	**2**	**3**	**4**
4.	Does the person find it difficult to remain in their seat even when in situations where it would be expected?	**0**	**1**	**2**	**3**	**4**
5.	Does the person prefer to be moving around or become frustrated if left in one position for too long?	**0**	**1**	**2**	**3**	**4**
6.	When the person is involved in a leisure activity (e.g. watching TV, playing a game etc.) do they make a lot of noise?	**0**	**1**	**2**	**3**	**4**
7.	When the person is involved in an activity, are they boisterous and/or rough?	**0**	**1**	**2**	**3**	**4**
8.	Does the person act as if they are "driven by a motor" (i.e. often very active)?	**0**	**1**	**2**	**3**	**4**
9.	Does the person seem like they need very little rest to recharge their battery?	**0**	**1**	**2**	**3**	**4**
10.	Does the person often talk excessively?	**0**	**1**	**2**	**3**	**4**
11.	Does the person's behaviour seem difficult to manage/contain whilst out and about (e.g. in town, in supermarkets etc.)?	**0**	**1**	**2**	**3**	**4**
12.	Do you feel that you need to "keep an eye" on the person at all times?	**0**	**1**	**2**	**3**	**4**
13.	Does the person you care for seem to act/do things without stopping to think first?	**0**	**1**	**2**	**3**	**4**
14.	Does the person blurt out answers before questions have been completed?	**0**	**1**	**2**	**3**	**4**
15.	Does the person start to respond to instructions before they have been fully given or without seeming to understand them?	**0**	**1**	**2**	**3**	**4**

16.	Does the person want things immediately?	**0**	**1**	**2**	**3**	**4**
17.	Does the person find it difficult to wait?	**0**	**1**	**2**	**3**	**4**
18.	Does the person disturb others because they have difficulty waiting for things or waiting their turn?	**0**	**1**	**2**	**3**	**4**

Please check your answers and go on to the next questionnaire.

MOOD, INTEREST AND PLEASURE QUESTIONNAIRE

This questionnaire contains 12 questions – <u>you should complete all 12 questions</u>. Each question will ask for your opinion about particular behaviours, which you have observed in the <u>LAST 2 WEEKS</u>. For every question you should circle <u>the most appropriate</u> response e.g.

6) In the LAST TWO WEEKS, how interested did the person appear to be in his/her surroundings?

interested all of the time	***interested most of the time***	***interested about half of the time***	***interested some of the time***	***never interested***

1) In the last two weeks, did the person seem…

sad all of the time	sad most of the time	sad about half of the time	sad some of the time	never sad

Please comment if anything has happened in the last two weeks which you feel might explain sadness if it has been observed (e.g. a bereavement):

2) In the last two weeks, how often did you hear positive vocalizations* when the person was engaged in activities*?

all of the Time	most of the the time	about half of the time	some of the time	Never

*<u>positive vocalizations</u>: e.g. laughing, giggling, "excited sounds" etc.
*<u>engaged in activities</u>: i.e. when someone is actively involved in any activity such as a mealtime, a social interaction, a self-care task or social outing etc.

3) In the last two weeks, do you think the facial expression of the person looked "flat"*…

all of the Time	most of the the time	about half of the time	some of the time	Never

*<u>flat expression</u>: expression seems lifeless; lacks emotional expression; seems unresponsive.

4) In the last two weeks, would you say the person…

cried every Day	cried nearly every day	cried 3-4 times each week	cried once or twice each week	cried less than once each week

5) In the last two weeks, how interested did the person appear to be in his/her surroundings?

interested all of the time	interested most of the time	interested about half of the time	interested some of the time	Never Interested

6) In the last two weeks, did the person seem to have been enjoying life…

all of the Time	most of the the time	about half of the time	some of the time	Never

Please comment if there are any reasons why this person might not have been enjoying him/herself e.g. illness, being in pain, experiencing a loss etc.:

7) In the last two weeks, would you say the person smiled…

at least once every day	at least once nearly every day	3-4 times each week	once or twice each week	less than once each week

8) In the last two weeks, how disinterested did the person seem to be in his/her surroundings?

disinterested all of the time	disinterested most of the time	disinterested about half of the time	disinterested some of the time	Never Disinterested

9) In the last two weeks, when the person was engaged in activities*, to what extent did his/her facial expressions* suggest that s/he was interested in the activity?

interested all of the time	interested most of the time	interested about half of the time	interested some of the time	Never Interested

*****engaged in activities: i.e. when someone is actively involved in any activity such as a mealtime, social interaction, self-care task or social outing etc.
*****facial expressions: interest might be indicated by the degree to which the person's gaze is being directed at the person/things involved in an activity.

10) In the last two weeks, would you say that the person…

laughed every day	laughed nearly every day	laughed 3-4 times each week	laughed once or twice each week	laughed less than once each week

11) In the last two weeks, how often did you see gestures which appeared to demonstrate enjoyment* when the person was engaged in activities*?

all of the Time	most of the the time	about half of the time	some of the time	Never

*gestures which appear to demonstrate enjoyment: e.g. clapping, waving hands in excitement etc.
*****engaged in activities: i.e. when someone is actively involved in any activity such as a meal time, social interaction, self-care task or social outing etc.

12) In the last two weeks, did the person's vocalizations* sound distressed…

all of the Time	most of the the time	about half of the time	some of the time	Never

*****vocalizations: any words, noises or utterances.

Please feel free to make any additional comments about the behaviour of the person over the last two weeks:

Please check your answers and go on to the next questionnaire.

THE NCCPC-R: PAIN CHECKLIST

How often has your child shown these behaviours in the last week? Please circle a number for each item. If an item does not apply to your child (for example he/she does not eat solid food or cannot reach with his/her hands), then indicate 'not applicable' for that item.

		Not at all	Just a little	Fairly often	Very often	Not applicable
I. Vocal						
1.	Moaning, whining, whimpering (fairly softly)	0	1	2	3	NA
2.	Crying (moderately loud)	0	1	2	3	NA
3.	Screaming/yelling (very loud)	0	1	2	3	NA
4.	A specific sound or word for pain (e.g. a word, cry or type of laugh)	0	1	2	3	NA
II. Social						
5.	Not cooperating, cranky, irritable, unhappy.	0	1	2	3	NA
6.	Less interaction with others, withdrawn.	0	1	2	3	NA
7.	Seeking comfort or physical closeness	0	1	2	3	NA
8.	Being difficult to distract, not able to satisfy or pacify	0	1	2	3	NA
III. Facial						
9.	A furrowed brow	0	1	2	3	NA
10.	A change in eyes, including squinching of eyes, eyes opened wide, eyes frowning.	0	1	2	3	NA
11.	Turning of mouth, not smiling	0	1	2	3	NA
12.	Lips puckering up, tight, pouting or quivering.	0	1	2	3	NA
13.	Clenching or grinding teeth, chewing or thrusting tongue out.	0	1	2	3	NA
IV. Activity						
14.	Not moving, less active, quiet	0	1	2	3	NA
15.	Jumping around, agitated, fidgety	0	1	2	3	NA
V. Body and limbs						
16.	Floppy	0	1	2	3	NA
17.	Stiff, spastic, tense, rigid	0	1	2	3	NA
18.	Gesturing to or touching part of the body that hurts	0	1	2	3	NA
19.	Protecting, favouring or guarding part of the body that hurts	0	1	2	3	NA
20.	Flinching or moving the body part away, being sensitive to touch	0	1	2	3	NA
21.	Moving the body in a specific way to show pain (e.g. head back, arms down, curls up etc)	0	1	2	3	NA

VI. Physiological						
22.	Shivering	0	1	2	3	NA
23.	Change in colour, pallor	0	1	2	3	NA
24.	Sweating, perspiring	0	1	2	3	NA
25.	Tears	0	1	2	3	NA
26.	Sharp intake of breath, gasping	0	1	2	3	NA
27.	Breath holding	0	1	2	3	NA
VII. Eating/ Sleeping						
28.	Eating less, not interested in food.	0	1	2	3	NA
29.	Increase in sleep	0	1	2	3	NA
30.	Decrease in sleep.	0	1	2	3	NA

Please check your answers and go on to the next questionnaire.

HEALTH QUESTIONNAIRE

Instructions:

- **Have these medical problems affected the person you care for in the past MONTH**
- **Please rate as 0 – if your child has not been affected by this problem in the past month, 1 - if they have been mildly affected, 2 – if the problem has moderately affected your child and 3 - if your child has been severely affected by the problem.**

	No	Mild	Moderate	Severe
1. Eye Problems (e.g. glaucoma /blocked tear duct/s)	**0**	**1**	**2**	**3**
2. Ear Problems (e.g. infections, glue ear)	**0**	**1**	**2**	**3**
3. Dental Problems (e.g. toothache / gum problems / mouth ulcers / delayed eruption of teeth)	**0**	**1**	**2**	**3**
4. Cleft Palate.	**0**	**1**	**2**	**3**
5. Gastrointestinal Difficulties (e.g. reflux / stomach problems)	**0**	**1**	**2**	**3**
6. Bowel Problems (e.g. obstruction)	**0**	**1**	**2**	**3**
7. Heart Abnormalities or Circulatory Problems (e.g. congenital heart lesions or murmur)	**0**	**1**	**2**	**3**
8. Problems with Genitalia (e.g. prostate / testicular problems i.e. undescended testes)	**0**	**1**	**2**	**3**
9. Hernia (e.g. inguinal or hiatal)	**0**	**1**	**2**	**3**
10. Limb Abnormalities (e.g. malformed arm)	**0**	**1**	**2**	**3**
11. Epilepsy / Seizures / Neurological Referrals	**0**	**1**	**2**	**3**
12. Lung or Respiratory Problems (asthma / bronchitis)	**0**	**1**	**2**	**3**
13. Liver or Kidney Problems	**0**	**1**	**2**	**3**
14. Diabetes or Thyroid Function Problems	**0**	**1**	**2**	**3**
15. Skin Problems (e.g. tinea, eczema, psoriasis, dry skin)	**0**	**1**	**2**	**3**
16. Other (please specify problem and severity from 0-3)	**0**	**1**	**2**	**3**

THANK YOU FOR COMPLETING THESE QUESTIONNAIRES

Appendix E: Consent Form

The Cerebra Centre for Neurodevelopmental Disorders

UNIVERSITY OF BIRMINGHAM

Investigating the health related pain and challenging behaviour in children with neurodevelopmental disorders

Study Director: Professor Chris Oliver

Consent form A: Please complete this form if you are a person with a neurodevelopmental disorder.

1.	Has somebody else explained the project to you or have you read the information?	YES/NO
2.	Do you understand what the project is about?	YES/NO
3.	Have you asked all of the questions you want?	YES/NO
4.	Have you had your questions answered in a way you understand?	YES/NO
5.	Do you understand it is OK to stop taking part at any time?	YES/NO
6.	Are you happy to take part?	YES/NO

If any answers are 'no' or you don't want to take part, don't sign your name!

If you do want to take part, you can write your name below

_____

You can also choose if you want to say 'yes' to these questions:

7.	Are you happy for us to contact your family again in the future?	YES/NO

Your name: _____

Date:_____

The person who explained this project to you needs to sign too. This should be your parent/guardian.

Print name:_____ Sign:_____

Date:_____

PTO…continued overleaf

<u>Consent form B:</u> Please complete this form if you are a parent/carer/guardian of a person with a neurodevelopmental disorder.

Please initial box…

1. I confirm that I have read and understood the 'pain and challenging behaviour' information sheet. I have had the opportunity to consider the information, ask questions and have had these answered satisfactorily. ☐

2. I understand that my participation and that of my child/person I care for is voluntary and that I am free to withdraw at any time without giving any reason, without my or that of my child's/person I care for's medical care or legal rights being affected. ☐

3. I agree to take part in the above study. ☐

Optional clauses: The statement below is <u>optional</u>:

4. I agree for researchers to contact me regarding mine and my child's involvement in future aspects of this study. I understand that neither I nor my child is obligated to take part in future aspects of the study. ☐

Print Name: _____ Telephone number: _____

Address:_____

_____

Relationship to participant:_____ Signature: _____

Date: _____

Appendix F: Interview Schedule

Questions About Behavioral Function (QABF)

Rate how often the person you care for demonstrates self-injurious behaviours in situations where they might occur. Be sure to rate how often each behaviour occurs, not what you think a good answer would be.

		Does not apply	Never	Rarely	Some	Often
1	Engages in the behaviour to get attention.	X	0	1	2	3
2	Engages in the behaviour to escape work or learning situations.	X	0	1	2	3
3	Engages in the behaviour as a form of 'self-stimulation'.	X	0	1	2	3
4	Engages in the behaviour because he/she is in pain.	X	0	1	2	3
5	Engages in the behaviour to get access to items such as preferred toys, food, or beverages.	X	0	1	2	3
6	Engages in the behaviour because he/she likes to be reprimanded.	X	0	1	2	3
7	Engages in the behaviour when asked to do something (get dressed, brush teeth, work, etc.).	X	0	1	2	3
8	Engages in the behaviour even if he/she thinks no one is in the room.	X	0	1	2	3
9	Engages in the behaviour more frequently when he/she is ill.	X	0	1	2	3
10	Engages in the behaviour when you take something away from him/her.	X	0	1	2	3
11	Engages in the behaviour to draw attention to him/herself.	X	0	1	2	3
12	Engages in the behaviour when he/she does not want to do something.	X	0	1	2	3
13	Engages in the behaviour because there is nothing else to do.	X	0	1	2	3
14	Engages in the behaviour when there is something bothering him/her physically.	X	0	1	2	3
15	Engages in the behaviour when you have something he/she wants.	X	0	1	2	3
16	Engages in the behaviour to try and get a reaction from you.	X	0	1	2	3
17	Engages in the behaviour to try and get people to remove a task or demand	X	0	1	2	3
18	Engages in the behaviour in a highly repetitive manner, ignoring his/her surroundings.	X	0	1	2	3
19	Engages in the behaviour because he/she is physically uncomfortable.	X	0	1	2	3
20	Engages in the behaviour when a peer has something he/she wants.	X	0	1	2	3
21	Does he/she seem to be saying 'come see me' or 'look at me' when engaging in the behaviour?	X	0	1	2	3
22	Does he/she seem to be saying 'stop asking me to do this' when engaging in the behaviour?	X	0	1	2	3
23	Does he/she seem to enjoy the behaviour, even if no one is around?	X	0	1	2	3
24	Does the behaviour seem to indicate to you that he/she is not feeling well?	X	0	1	2	3
25	Does he/she seem to be saying 'give me that (toy item, food item) when engaging in the behaviour?	X	0	1	2	3

CHALLENGING BEHAVIOUR INTERVIEW

Name: _____ Date of interview: ____/ ____/ 19____ Male Female Date of Birth: _____/ ____/ 19____

Current Address: _____ Name of Respondent: _____

_____ Profession/job: _____

Administration

1. Identify a respondent who has known the person well for at least 3 months.

2. Let the participant see a copy of the interview to help administration.

3. For part I, ask the respondent if each category of challenging behaviour has occurred in the last month by naming the category and then giving some examples from the appendix. Check the response by ensuring the month criterion and that the behaviour fulfils the operational definition. Tick the appropriate box.

4. In part II, enter the behaviour categories in the boxes above question 1. This will help you enter the ratings later on. For challenging behaviours which are included, read each question whilst the respondent looks at the question and then ask for a rating for each of the behaviour categories which have been chosen. Check the rating by asking for an example.

CHALLENGING BEHAVIOUR INTERVIEW (PART I)

1. Which of the following behaviours have occurred in the last month? (See appendix for definitions and examples)

Challenging Behaviour Category	List behaviours from examples in appendix
❑ Self –Injury (SIB)	
❑ Physical aggression (PAG)	
❑ Verbal aggression (VAG)	
❑ Disruption and destruction of property or the environment (DST)	
❑ Anal poking (AP)	
❑ Stereotyped behaviours (STB)	
❑ Inappropriate vocalisations (IV)	
❑ Inappropriate removal of clothing (IRC)	
❑ Pica (PIC)	
❑ Inappropriate or unacceptable sexual behaviour (ISB)	
❑ Smearing (SMR)	
❑ Stealing (STL)	
❑ Self-induced vomiting and regurgitation (SIV)	

CHALLENGING BEHAVIOUR INTERVIEW (PART II)

In each box, enter the category of challenging behaviour that is being considered ⇨

1. Think about how often this behaviour occurred in the last month. If there was no change and you watched this person now, then would you *definitely* see the behaviour:

⑤	④	③	②	①
In the next 15 minutes	In the next hour	By this time tomorrow	By this time next week	By this time next month

2. In the last month, for how long did the *longest* episode or burst of this behaviour last?

①	②	③	④	⑤
Less than a minute	Less than 5 minutes	Less than 15 minutes	Less than an hour	More than an hour

3. In the last month, for how long have episodes or bursts of this behaviour *typically* lasted or lasted on average?

①	②	③	④	⑤
Less than a minute	Less than 5 minutes	Less than 15 minutes	Less than an hour	More than an hour

4. For the worst episode of behaviour in the last month, what response was necessary[1]?

⓪	①	②	③	④					
Nothing	Verbal discouragement or reminder	Informal physical intervention by one member of staff e.g. blocking, holding an arm briefly, taking objects from an individual Removal to a safe environment Removal of staff or others from immediate environments	Informal physical intervention by more than one member of staff Formal restraint procedure Protective or restrictive devices employed	Seclusion PRN medication Legal involvement or legal advice has been sought Section of MHA invoked					

[1] To score, identify any items which have occurred and take highest scoring item.

5. In the last month, what has been the worst effect of this behaviour on the individual's *physical health*?

⓪	①	②	③
No effect at all	**Minor, temporary injury,** such as reddening of the skin, but *no* bruising or tissue damage	**Moderate injury,** such as bruising , cuts or abrasions **or illness lasting less than a day,** e.g. brief stomach upset, a single episode of vomiting	**Significant injury** e.g. fractured bones, sutures required, minor or major operation required **or illness lasting more than a day**

6. In the last month, what has been the worst *direct* effect of this behaviour on the *physical health* of staff or carers?

⓪	①	②	③
No effect at all	**Minor, temporary injury,** such as reddening of the skin, but *no* bruising or tissue damage	**Moderate injury,** such as bruising , cuts or abrasions **or illness lasting less than a day,** e.g. brief stomach upset, a single episode of vomiting	**Significant injury** e.g. fractured bones, sutures required, minor or major operation required **or illness lasting more than a day**

7. In the last month, what has been the worst *direct* effect of this behaviour on the *physical health* of other service users?

⓪	①	②	③
No effect at all	**Minor, temporary injury,** such as reddening of the skin, but *no* bruising or tissue damage	**Moderate injury,** such as bruising , cuts or abrasions **or illness lasting less than a day,** e.g. brief stomach upset, a single episode of vomiting	**Significant injury** e.g. fractured bones, sutures required, minor or major operation required **or illness lasting more than a day**

8. Throughout the whole of the last month, has the behaviour had any negative effects on the well-being of other service users e.g. disruption to planned activities, service users are frightened or upset, belongings or clothing are damaged or lost, meals are spoiled etc.?

⓪	①	②	③	④
No effect at all on the well-being of other service users	Effect on the well-being of other service users **about once in the last month**	Effect on the well-being of other service users **about once a week**	Effect on the well-being of other service users **about once every 3 days**	Effect on the well-being of other service users **nearly every day**

9. In the last month, what has been the *direct* effect of this behaviour on the environment in which the individual lives?

⓪	①	②	③	④					
No damage or loss at all	**Disruption or mild damage** to property or the living areas e.g., objects thrown, furniture tipped, doors slammed, meals spoiled, paint scratched. **Item does not require repair or replacement.**	**Moderate damage** to property or living areas e.g. curtains torn, furniture partly broken. **Item requires repair but can be used.**	**Significant damage** to property and living areas. **Item requires repair and cannot be used.**	**Extreme damage** to property or living areas. **Item requires replacement and cannot be used or repaired** e.g. windows broken, furniture unusable.					

10. In the last month, as a result of this behaviour, have restrictive or protective devices (e.g. arm splints, helmet) or specially designed clothing (e.g. all-in-one suit) been worn by the individual?

⓪	①	②	③	④					
Never	**Some of the time**	**About half the time**	**Most of the time**	**Almost continuously**					

(If so was it: Arm splint(s) ❑, Helmet or headgear ❑, Gloves/mittens/other items on hands ❑, Specially designed clothing ❑, Other ❑, (please specify) _____)

11. Has the environment in which the individual currently lives been modified because of this behaviour (examples of modification are given in the box below)?

⓪	①	②	③
No modifications	**Modifications to the person's possessions but not elsewhere** e.g. padding on a wheel chair, clothing which is strengthened	**Modifications have been made to the environment but are not noticeable unless pointed out e.g. curtains on Velcro, window locks**	**Modifications have been made to the environment and are noticeable**

Examples of modification to the environment: windows are not made of glass, TV is in a protective cabinet or out of reach, furniture is secured, a cupboard door is secured, a door is secured, curtains are absent (because they will be torn down), pictures are out of reach, hard or sharp surfaces are padded, service users are always visible, a room is out of bounds, cutlery is plastic, furniture is deliberately heavy, door closers are fitted to prevent slamming, wallpaper is washable in rooms apart from kitchen and bathroom, fridge is secured, ornaments are out of reach, furniture or fittings have been removed, furniture is chosen because it has particular qualities e.g. no sharp edges etc.

12. In the last month, as a result of this behaviour, has a verbal response by staff or carers been necessary e.g. discouraging the behaviour, distraction to another activity, reminder, reprimand?

⓪	①	②	③	④
Never	**At least once a month**	**At least once a week**	**At least once a day**	**At least once an hour**

13. In the last month, as a result of this behaviour, has physical contact or prevention or restraint by staff or carers been necessary e.g. blocking, taking objects from an individual, temporary restraint of an arm?

⓪	①	②	③	④					
Never	**At least once a month**	**At least once a week**	**At least once a day**	**At least once an hour**					

(If so was it a written procedure ❑ or an informal procedure ❑, please tick.)

14. In the last month, for this behaviour, was it necessary for more than one member of staff to respond when the behaviour occurred?

⓪	①	②	③	④					
Never	**At least once a month**	**At least once a week**	**At least once a day**	**At least once an hour**					

15. In the last month, has there been any contact with any of the following regarding this behaviour?

		Name and Contact Number
❑	Clinical Psychologist or Psychology Assistant working with a Clinical Psychologist	
❑	Psychiatrist	
❑	General Practitioner	
❑	Challenging Behaviour specialist or team	
❑	Speech and language therapist	
❑	Legal advisor	
❑	Other	
❑	Other	
❑	Other	

SUMMARY OF SCORES

	Behaviours				
Qu.					
1					
2					
3					
4					
5					
6					
7					
8					
9					
10					
11					
12					
13					
14					
Total					

NOTES:

Appendix H: Matrix Demonstrating Spearman's Rho Correlations Between Scores on the VABS and Pain Related Indices in Each Function Group

Function Groups				QABFTotal PainSev	NCCPC Total	NCCPC Vocal	NCCPC Social	NCCPC Facial	NCCPC Activity	NCCPC Body	NCCPC Phys	NCCPC Eat	HQ Total	GRQ Total	MIPQ Total	MIPQ Mood	MIPQ IP	TAQ Total	TAQ Imp	TAQ Overact	VABS Adapt
Spearman's Rho	Non Pain Related CB Group	QABFTotal PainSev	Corr. Coeff.		.572**	.474*	.599**	.640**	.156	.350	.741**	.317	.344	.507**	-.234	-.546**	-.040	.224	.326	.268	-.138
			Sig. (1-tail)		.002	.011	.001	.000	.239	.051	.000	.070	.054	.008	.141	.004	.429	.152	.065	.108	.265
		NCCPC Total	Corr. Coeff.			.792**	.792**	.762**	.524**	.740**	.650**	.398*	.241	.684**	-.280	-.557**	-.082	.589**	.602**	.506**	-.175
			Sig. (1-tail)			.000	.000	.000	.005	.000	.000	.030	.134	.000	.098	.003	.355	.002	.001	.007	.213
		NCCPC Vocal	Corr. Coeff.				.544**	.501**	.374*	.440*	.491**	.271	.065	.356	-.226	-.534**	-.064	.437*	.462*	.340	-.065
			Sig. (1-tail)				.004	.007	.039	.018	.009	.106	.384	.052	.150	.004	.387	.019	.013	.056	.384
		NCCPC Social	Corr. Coeff.					.580**	.276	.760**	.588**	.331	.360*	.484*	-.171	-.338	-.075	.269	.323	.211	.077
			Sig. (1-tail)					.002	.102	.000	.002	.062	.046	.011	.218	.057	.367	.107	.066	.167	.364
		NCCPC Facial	Corr. Coeff.						.205	.418*	.644**	.345	.436*	.751**	-.336	-.589**	-.085	.379*	.415*	.324	-.350
			Sig. (1-tail)						.174	.023	.000	.053	.019	.000	.058	.002	.350	.037	.024	.066	.051
		NCCPC Activity	Corr. Coeff.							.281	.366*	.237	-.020	.398*	-.091	-.223	.028	.582**	.551**	.608**	-.270
			Sig. (1-tail)							.097	.043	.138	.463	.033	.340	.153	.449	.002	.003	.001	.107
		NCCPC Body	Corr. Coeff.								.315	.325	.358*	.540**	-.156	-.259	-.063	.354*	.351	.315	-.055
			Sig. (1-tail)								.072	.065	.047	.005	.239	.116	.388	.049	.050	.072	.402
		NCCPC Phys	Corr. Coeff.									.156	.011	.580**	-.426*	-.633**	-.232	.330	.402*	.340	.004
			Sig. (1-tail)									.239	.481	.002	.021	.001	.144	.062	.029	.056	.492
		NCCPC Eat	Corr. Coeff.										.239	.478*	-.046	-.109	-.079	.360*	.439*	.330	-.271
			Sig. (1-tail)										.136	.012	.417	.311	.360	.046	.018	.062	.105
		HQ Total	Corr. Coeff.											.356	.097	-.073	.240	.046	-.002	-.010	-.282
			Sig. (1-tail)											.052	.330	.370	.135	.417	.496	.481	.096
		MIPQ Total	Corr. Coeff.													.623**	.894**	-.286	-.337	-.355*	.280
			Sig. (1-tail)													.001	.000	.093	.058	.048	.098
		MIPQ Mood	Corr. Coeff.														.255	-.272	-.368*	-.211	.061
			Sig. (1-tail)														.120	.105	.042	.167	.391
		MIPQ IP	Corr. Coeff.															-.246	-.284	-.342	.243
			Sig. (1-tail)															.129	.094	.055	.132
		TAQ Total	Corr. Coeff.																.944**	.933**	-.336
			Sig. (1-tail)																.000	.000	.059
		TAQ Imp	Corr. Coeff.																	.878**	-.308
			Sig. (1-tail)																	.000	.076
		TAQ Overact	Corr. Coeff.																		-.525**
			Sig. (1-tail)																		.005

Pain Groups			QABFTotal PainSev	NCCPC Total	NCCPC Vocal	NCCPC Social	NCCPC Facial	NCCPC Activity	NCCPC Body	NCCPC Phys	NCCPC Eat	HQ Total	GRQ Total	MIPQ Total	MIPQ Mood	MIPQ IP	TAQ Total	TAQ Imp	TAQ Overact	VABS Adapt
Pain Related CB Group	QABFTotal PainSev	Corr. Coeff.		.369*	.392*	.300	.316	.059	.125	.044	.177	.702**	.470*	-.358	-.439*	-.219	-.171	-.068	-.196	.020
		Sig. (1-tail)		.042	.032	.082	.071	.395	.285	.422	.209	.000	.014	.051	.021	.164	.223	.382	.191	.465
	NCCPC Total	Corr. Coeff.			.807**	.790**	.643**	.299	.715**	.608**	.676**	.201	.569**	-.480*	-.765**	-.291	-.013	-.060	.034	-.232
		Sig. (1-tail)			.000	.000	.000	.083	.000	.001	.000	.191	.003	.012	.000	.095	.477	.395	.441	.144
	NCCPC Vocal	Corr. Coeff.				.678**	.532**	.424*	.486**	.422*	.361*	.050	.616**	-.252	-.664**	-.080	.031	.031	.057	-.045
		Sig. (1-tail)				.000	.004	.022	.009	.022	.045	.415	.001	.129	.000	.361	.446	.446	.401	.420
	NCCPC Social	Corr. Coeff.					.439*	.270	.393*	.324	.406*	.028	.464*	-.381*	-.681**	-.201	.180	.133	.174	-.153
		Sig. (1-tail)					.018	.106	.032	.066	.027	.452	.015	.040	.000	.185	.211	.278	.219	.243
	NCCPC Facial	Corr. Coeff.						.107	.265	.202	.462*	.079	.700**	-.198	-.309	-.132	.052	-.071	.156	-.385*
		Sig. (1-tail)						.314	.111	.178	.013	.367	.000	.188	.081	.280	.409	.377	.244	.035
	NCCPC Activity	Corr. Coeff.							.145	.002	.005	.035	.215	-.126	-.297	-.033	.456*	.606**	.352	.220
		Sig. (1-tail)							.255	.497	.490	.440	.168	.289	.090	.443	.016	.001	.054	.156
	NCCPC Body	Corr. Coeff.								.520**	.391*	.162	.252	-.361*	-.616**	-.264	-.230	-.280	-.139	-.194
		Sig. (1-tail)								.006	.032	.241	.129	.050	.001	.118	.151	.103	.268	.188
	NCCPC Phys	Corr. Coeff.									.726**	.184	.280	-.385*	-.449*	-.252	-.032	-.119	-.025	.001
		Sig. (1-tail)									.000	.212	.103	.038	.018	.129	.444	.299	.457	.498
	NCCPC Eat	Corr. Coeff.										.386*	.299	-.490*	-.393*	-.424*	-.125	-.135	-.148	-.137
		Sig. (1-tail)										.042	.089	.010	.035	.025	.290	.274	.255	.266
	HQ Total	Corr. Coeff.											.274	-.562**	-.260	-.567**	-.329	-.158	-.410*	-.063
		Sig. (1-tail)											.114	.004	.128	.004	.073	.248	.033	.393
	MIPQ Total	Corr. Coeff.													.705**	.931**	.099	.148	.082	.282
		Sig. (1-tail)													.000	.000	.330	.256	.359	.102
	MIPQ Mood	Corr. Coeff.														.464*	.054	.121	-.021	.150
		Sig. (1-tail)														.015	.406	.295	.462	.253
	MIPQ IP	Corr. Coeff.															.118	.134	.128	.275
		Sig. (1-tail)															.301	.277	.286	.108
	TAQ Total	Corr. Coeff.																.877**	.951**	.142
		Sig. (1-tail)																.000	.000	.265
	TAQ Imp	Corr. Coeff.																	.720**	.347
		Sig. (1-tail)																	.000	.057
	TAQ Overact	Corr. Coeff.																		.005
		Sig. (1-tail)																		.492

CB = Challenging behaviour

*. Correlation sig at 0.05 level (1-tailed).

**. Correlation sig. at 0.01 level (1- tailed).