VOLUME I: RESEARCH COMPONENT

CHALLENGING BEHAVIOUR ASSESSMENT IN INDIVIDUALS WITH INTELLECTUAL DISABILITIES AND AUTISM SPECTRUM DISORDER

by

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THESIS OVERVIEW

This thesis is submitted to the University of Birmingham in partial fulfilment of the requirements for the degree of Doctorate of Clinical Psychology. The thesis comprises of two volumes.

Volume I reports a systematic review and an empirical research paper. The systematic review examines the association between gastrointestinal disease and behavioural change in individuals with intellectual disabilities and autism spectrum disorder. A systematic search of literature databases identified fourteen relevant research articles. Several behaviour classes associated with gastrointestinal disorder were identified, including challenging behaviour, sleep problems, and anxiety and mood related behaviours. Multiple forms of gastrointestinal disorder were also identified as did the quality of its assessment. There was varying degrees of evidence for an association between certain behaviours and gastrointestinal disorders due to assessment methods and behavioural definitions.

The empirical paper reports the development of a challenging behaviour report form. A functional assessment tool and protocol was developed to take into account various factors when assessing challenging behaviour such as pain related behaviours, affect, and precursor behaviours. The assessment was trialled on footage of experimental functional analysis of non-verbal children with autism spectrum disorder. The development of the assessment and inter-observer agreement is reported as well as future directions in the development of the assessment. Volume I also contains a public domain briefing paper, which provides an overview of the systematic review and empirical study.

Volume II documents the clinical component and contains five Clinical Practice Reports (CPR) completed over the course of training. The reports represent clinical and empirical work carried out during placements in an adult community mental health service, an

older adult hospital service, a research centre in learning disabilities and a specialist neurorehabilitation service. CPR 1 presents a cognitive-behavioural and psychodynamic formulation of an adult male experiencing anxiety and low mood. CPR 2 a service evaluation of staff training needs in an adult community mental health service. CPR 3 presents a single-case experimental design of an experimental functional analysis of challenging behaviour in a non-verbal child with autism spectrum disorder. CPR 4 presents an assessment, formulation, and intervention with an adult male with low mood and alcoholism in a physical health setting. CPR5 is an abstract of a presentation on a neuropsychological assessment, formulation, and intervention with an adult female with multiple sclerosis and acquired brain injury.

Dedicated to my parents. Thank you for everything.

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CONTENTS

VOLUME I: RESEARCH COMPONENT

SYSTEMATIC REVIEW: GASTROINTESTINAL DISEASE AND BEHAVIOURAL CHANGE: IS THERE AN ASSOCIATION IN INDIVIDUALS WITH INTELLECTUAL DISABILITIES AND AUTISM SPECTRUM DISORDER?

THE DEVELOPMENT OF A CHALLENGING BEHAVIOUR REPORT FORM

PUBLIC DOMAIN BRIEFING PAPER

SYSTEMATIC REVIEW

EMPIRICAL PAPER:

LIST OF ILLUSTRATIONS

SYSTEMATIC REVIEW: GASTROINTESTINAL DISEASE AND BEHAVIOURAL CHANGE: IS THERE AN ASSOCIATION IN INDIVIDUALS WITH INTELLECTUAL DISABILITIES AND AUTISM SPECTRUM DISORDER?

CHALLENGING BEHAVIOUR ASSESSMENT IN INDIVIDUALS WITH INTELLECTUAL DISABILITIES AND AUTISM SPECTRUM DISORDER

LIST OF TABLES

SYSTEMATIC REVIEW: GASTROINTESTINAL DISEASE AND BEHAVIOURAL CHANGE: IS THERE AN ASSOCIATION IN INDIVIDUALS WITH INTELLECTUAL DISABILITIES AND AUTISM SPECTRUM DISORDER?

THE DEVELOPMENT OF A CHALLENGING BEHAVIOUR REPORT FORM

LIST OF APPENDICES

SYSTEMATIC REVIEW: GASTROINTESTINAL DISEASE AND BEHAVIOURAL CHANGE: IS THERE AN ASSOCIATION IN INDIVIDUALS WITH INTELLECTUAL DISABILITIES AND AUTISM SPECTRUM DISORDER?

THE DEVELOPMENT OF A CHALLENGING BEHAVIOUR REPORT FORM

VOLUME II: CLINICAL COMPONENT

CLINICAL PRACTICE REPORT 1: COGNITIVE BEHAVIOURAL AND PSYCHODYNAMIC FORMULATION OF A MAN WITH SYMPTOMS OF LOW MOOD AND ANXIETY.

CLINICAL PRACTICE REPORT 4: CASE STUDY OF A 64 YEAR OLD WITH ANXIETY AND ALCOHOL MISUSE.

CLINICAL PRACTICE REPORT 5: CASE STUDY: NEUROPSYCHOLOGICAL ASSESSMENT OF A 53 YEAR OLD WOMAN IN A NEUROLOGICAL REHABILITATION SETTING

LIST OF ILLUSTRATIONS

CLINICAL PRACTICE REPORT 1: COGNITIVE BEHAVIOURAL AND PSYCHODYNAMIC FORMULATION OF A MAN WITH SYMPTOMS OF LOW MOOD AND ANXIETY

CLINICAL PRACTICE REPORT 2: SERVICE EVALUATION: STAFF TRAINING AND SUPERVISION NEEDS

CLINICAL PRACTICE REPORT 3: A SINGLE CASE EXPERIMENTAL STUDY OF A 6 YEAR OLD BOY WITH CEREBRAL PALSY, SHOWING SELF-INJURIOUS BEHAVIOUR

CLINICAL PRACTICE REPORT 4: CASE STUDY OF A 64 YEAR OLD WITH ANXIETY AND ALCOHOL MISUSE

CLINICAL PRACTICE REPORT 5: CASE STUDY: NEUROPSYCHOLOGICAL ASSESSMENT OF A 53 YEAR OLD WOMAN IN A NEUROLOGICAL REHABILITATION SETTING

No appendices

LIST OF TABLES

CLINICAL PRACTICE REPORT 1: COGNITIVE BEHAVIOURAL AND PSYCHODYNAMIC FORMULATION OF A MAN WITH SYMPTOMS OF LOW MOOD AND ANXIETY

LINICAL PRACTICE REPORT 2: SERVICE EVALUATION: STAFF TRAINING AND SUPERVISION NEEDS

CLINICAL PRACTICE REPORT 3: A SINGLE CASE EXPERIMENTAL STUDY OF A 6 YEAR OLD BOY WITH CEREBRAL PALSY, SHOWING SELF-INJURIOUS BEHAVIOUR

CLINICAL PRACTICE REPORT 4: CASE STUDY OF A 64 YEAR OLD WITH ANXIETY AND ALCOHOL MISUSE

CLINICAL PRACTICE REPORT 5: CASE STUDY: NEUROPSYCHOLOGICAL ASSESSMENT OF A 53 YEAR OLD WOMAN IN A NEUROLOGICAL REHABILITATION SETTING

No tables

LIST OF APPENDICES

CLINICAL PRACTICE REPORT 1: COGNITIVE BEHAVIOURAL AND PSYCHODYNAMIC FORMULATION OF A MAN WITH SYMPTOMS OF LOW MOOD AND ANXIETY

CLINICAL PRACTICE REPORT 2: SERVICE EVALUATION: STAFF TRAINING AND SUPERVISION NEEDS

CLINICAL PRACTICE REPORT 3: A SINGLE CASE EXPERIMENTAL STUDY OF A 6 YEAR OLD BOY WITH CEREBRAL PALSY, SHOWING SELF-INJURIOUS BEHAVIOUR

CLINICAL PRACTICE REPORT 4: CASE STUDY OF A 64 YEAR OLD WITH ANXIETY AND ALCOHOL MISUSE

CLINICAL PRACTICE REPORT 5: CASE STUDY: NEUROPSYCHOLOGICAL ASSESSMENT OF A 53 YEAR OLD WOMAN IN A NEUROLOGICAL REHABILITATION SETTING

No appendices

SYSTEMATIC REVIEW: GASTROINTESTINAL DISEASE AND BEHAVIOURAL CHANGE: IS THERE AN ASSOCIATION IN INDIVIDUALS WITH INTELLECTUAL DISABILITIES AND AUTISM SPECTRUM DISORDER?

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Abstract

Introduction

Individuals with intellectual disorders (ID) and autism spectrum disorder (ASD) experience heightened rates of physical health conditions and are also more likely to display challenging behaviour. This systematic review aims to evaluate if the literature supports the hypothesis that pain as a result of gastrointestinal disorder (GI) is associated with changes in behaviour, specifically challenging behaviour and sleep problems, in these individuals.

Method

A systematic search for studies containing behavioural, developmental disability, and gastrointestinal disorder terms was conducted.

Results

A total of fourteen papers were included in the review. The review focussed on two main areas; the methodological issues regarding research into this area and evidence for the association between GI disorder and challenging behaviour, disordered sleep, and behaviours related to anxiety and depression. A quality framework was applied (Downs & Black, 1998).

Conclusions

The majority of papers used medical notes and assessments by medically trained professionals while the remaining used parental report. There is inconsistency in how behaviour topography of challenging behaviour and other behaviours were reported across the studies. There was equivocal evidence for and against an association between self-injurious

behaviour and GI disorder. There appears to be an association between some behaviour, such as aggression and stereotyped/hyperactive behaviour, and GI disorder, and there is some evidence that GI disorder may be associated with depression and/or anxiety in this population. Finally, there is limited evidence for the association of sleep problems and GI disease from the papers in this review.

Future research should use operational definitions of potential behavioural indicators of GI disorder to increase replicability and to increase the clinical implications of having a potential way to screen for GI disorder in non-verbal individuals.

Introduction

Intellectual disabilities affect approximately 1 in 100 individuals (Maulik, Mascarenhas, Mathers, Dua, & Saxena, 2011), with a wide range of prenatal, antenatal and postnatal causes implicated. Intellectual disability (ID) is defined by deficits in intellectual functioning (reasoning, planning) and adaptive behaviour (independent living) (American Psychiatric Association, 2013). Importantly, individuals with ID experience heightened rates of physical health conditions and are also more likely to display challenging behaviour (Cervantes & Matson, 2015; Kohane et al., 2012; Emerson, 2001). Challenging behaviour is also highly frequent in individuals with autism spectrum disorder (ASD; Richards, Oliver, Nelson, & Moss, 2012), a pervasive developmental disorder characterised by difficulties in social communication and the presence of repetitive behaviours (American Psychiatric Association, 2013). Challenging behaviour has been defined as 'culturally abnormal behaviour(s) of such an intensity, frequency, or duration that the physical safety of the person or others is likely to be placed in serious jeopardy, or behaviour which is likely to seriously limit use of, or result in the person being denied access to, ordinary community facilities (Emerson, 1995). This includes behaviours such as self-injurious behaviour or aggression; the presence of which has been associated with increased risk of psychiatric hospitalisation (Mandell, 2008), use of physical interventions (Allen, Lowe, Brophy, & Moore, 2009) and a lower quality of life for those who engage in it (Beadle-Brown, Murphy, & DiTerlizzi, 2009). Parents of children who engage in challenging behaviour and staff experience higher levels of stress and are at higher risk of burnout respectively. (Hastings, 2003; McIntyre, Blacher, & Baker, 2002)

In addition to challenging behaviour, there are a number of clinically important problems that are associated with individuals with ID/ASD, among the most commonly

identified areas are sleep disorders, anxiety and depression. Disordered sleep is more common in individuals with ID than in the typically developing population (Doran, Harvey, Horner, & Scotti, 2006; van de Wouw, Evenhuis, & Echteld, 2012). Additionally, disordered sleep is associated with challenging behaviour (van de Wouw et al., 2012) and, like challenging behaviour, disordered sleep can increase stress in the families of children with ID (Richdale, Francis, Gav, 2000). Higher prevalence of anxiety has been reported in samples of individuals with ASD (Mannion, Leader, & Healy, 2013) as has depression, with prevalence rates of up to 10% reported (Leyfer et al., 2006). Emerson (2003) also reported higher prevalence rates of anxiety and depression in individuals with ID. Challenging behaviour has been used as a "depressive equivalent" for depression, although a recent review concluded that there is insufficient evidence to support this and it is possible that other variables, such as pain, may account for the association (Davies and Oliver, 2014). This has clear clinical significance as undiagnosed and untreated mental health problems in individuals with ID can impact on daily functioning (Hassiotis & Turk, 2012). Given the heightened prevalence of challenging behaviour, sleep disorders, anxiety, and depression in individuals with ID and ASD and the deleterious consequences of these behaviours for individuals, parents and carers, it is essential that putative causes for these co-morbidities are identified and treated.

Recent research suggests that individuals with ASD and/or ID are at greater risk of experiencing painful physical health problems (Cervantes & Matson, 2015; Sigafoos, Arthur & O'Reily, 2003); one such condition is gastrointestinal disease (GI). GI disease refers to diseases and disorders involving the gastrointestinal tract, including the oesophagus, stomach, and small and large intestines. A common GI disease is Gastroesophageal Reflux Disease (GORD), which is a chronic condition with mucosal damage caused by stomach acid coming up from the stomach into the oesophagus (Bredenoord, Pandolfino, & Smout, 2013). One

study reported that the prevalence of GORD in institutionalised individuals with IQ<50 was 50% and that risk factors included having an IQ of less than 35 (Böhmer, Klinkenberg-Knol, Niezen-de Boer, & Meuwissen (2000). Similarly, Charlot et al., (2011) reported that 60% of individuals with ID admitted to inpatient psychiatric care had constipation, while 38% experienced GORD. Other research has reported prevalence rates of medication for GORD as 36% (Lee et al., 2011) in a sample with ID. Similarly, Van Der Heide, Van Der Putten, Van Den Berg, Taxis, & Vlaskamp (2009) found that in a sample of individuals with IQ less than 25, 68% were prescribed laxatives and 44% were prescribed medication for GORD. Within the literature on ASD, a review by Horvath & Perman (2002) reported that 40% of children with ASD suffer from abdominal pain, which may be related to GORD. Thus, the data from individuals with ID and/or ASD indicate a similar pattern of heightened GI disease in these populations, which is likely related to pain and discomfort for individuals.

Pain is reported to be common in children with ID and is a critical determinant of quality of life (Massaro, Pastore, Ventura, & Barbi, 2013). Caregivers, using validated pain measures, reported pain in 20% of residents in inpatient units for individuals with ID (Boerlage et al., 2013). However, pain ratings by caregivers may, at times, under report pain in individuals with ID (Boerlage et al., 2013). Self-report is the ‘‘gold standard’’ in pain assessment for both children and adults (McIntosh, 1997) and thus individuals with ID are more likely to experience unrecognised and untreated pain as self-report may be compromised (Findlay, Williams, & Scior, 2014, McGuire, Daly, & Smyth, 2010). When assessing pain in individuals with ID and communication difficulties, validated and effective tools are available and should be used. Hunt, Mastroyannopoulou, Goldman, & Seers (2003) suggest that three forms of knowledge are needed for the optimal assessment of pain in children: knowing the child, familiarity with children with the same or similar conditions, and

knowing the science. However, there seems to be poor awareness among paediatricians on pain assessment in non-verbal children (Massaro et al., 2013). This is reflected in the Death by Indifference (Mencap, 2007) report which stated that people with ID are seen as low priority within services and that many healthcare professionals have little understanding of ID, often attributing behaviour to the underlying developmental disability (Horvath & Perman, 2002). Similarly, the white paper 'Our health, our care, our say' (Health, 2006) states that people with ID face particular inequalities and that historically the NHS has not served them well. The Mencap report (2007) also identifies several cases where treatment was withdrawn for individuals with ID as the individual could not tolerate/understand the procedures. As individuals with ID and ASD are more likely to experience painful physical health difficulties and are less likely to have them correctly assessed, more effective ways of recognising pain are required that might aid diagnosis. Of these physical health difficulties, GI disease is common in this population and pain related behaviours may offer a way to conduct a cursory assessment of the disease.

The operant model, through applied behaviour analysis, provides one explanation for the association between pain and changes in an individuals' behaviour. Operant learning theory can account for challenging behaviour as a learned response to environmental stimuli (Oliver, 1995), that is maintained and modified via positive / negative reinforcement, positive/negative punishment (Lovaas & Simmons, 1969). Demanding tasks and low levels of attention are examples of antecedents which may evoke challenging behaviour (Carr & Durand, 1985). Pain is formulated as a motivating operation, a condition which increases or decreases the effectiveness of a reinforcer or a punisher (Laraway, Snycerski, Michael, & Poling, 2003). Motivating operations can be further separated into establishing operations, which increase the effectiveness of a consequence as a reinforcer or as a punisher and

abolishing operations which reduce the effectiveness of a consequence. While operant learning theory has led to successful interventions for challenging behaviour, not all interventions are successful, even when a maintaining function has been identified. This suggests that there may be other processes underlying the behaviour.

There is some evidence that undiagnosed health conditions that cause pain in non-verbal individuals with ID may precipitate challenging behaviour (Carr & Owen-DeSchryver, 2007). Children with ASD are twice as likely to engage in self-injury when health problems are present (Richards et al., 2012). Research on the temporal sequence of these behaviours suggests that the occurrence of hypothesized pain behaviours prior to self-injurious behaviour underlie pain and discomfort, whereas the occurrence of these behaviours following self-injurious behaviour is thought to be related to pain caused by the self-injurious behaviour itself (Courtemanche, Schroeder, Sheldon, Sherman, & Fowler, 2012). In studies such as these, the presence of pain is generally inferred from the presence of observable behavioural indicators of pain based on direct observation or informant-report. However, the presence of an underlying physical health condition or biological indicators of pain are not usually verified during these studies. As a result, it has been noted that the hypothesized that behavioural indicators of pain may be observable signs of general distress (Courtemanche et al., 2012). Furthermore, the evaluation of the effects of treatment of a painful health condition on self-injurious behaviour, hypothesized behavioural and physiological indicators of pain and underlying physiology may also prove useful (Carr & Owen-DeSchryver, 2007). For example, symptoms such as unexplained feeding difficulties, refusing to feed, gagging or choking, distressed behaviour, faltering growth, chronic cough, hoarseness or a single episode of pneumonia can be associated with GORD (NICE, 2015). Behavioural indicators may also take the form of other non-medical behavioural changes, such as the emergence or

changes in self-injurious behaviour and aggression. Other behavioural changes, such as disordered sleeping, may indicate GI disorder. Chronic acid reflux is often associated with frequent arousals during sleep and it is also considered to be a risk factor for obstructive sleep apnoea because of the associated mucosal oedema (Owens & Witmans, 2004). A systematic review of gastro oesophageal reflux and sleep in typically developing individuals suggested that disordered sleep could occur from slow clearance of acid reflux that occurs during arousals or awakenings from sleep (Dent, Holloway, & Eastwood, 2013). Regarding depression, it's possible that pain could act as a setting event for it (Breau & Camfield, 2011). A recent study by Greenlee, Mosley, Veenstra-VanderWeele, and Gotham (2016) reported higher incidence of depression in children and adolescents with ASD and gastrointestinal problems although the direction of causality is unclear and warrants more research in the area.

In summary, it can be challenging to assess GI disease and pain in individuals with ID and/or ASD. Individuals with communication difficulties may find it difficult to communicate pain and the presence of physical conditions to others which may mean that diagnoses are missed and care is not provided. This difficulty in communicative ability amongst this population is compounded by wider, more systemic issues. Additionally, untreated GI disease may lead to challenging behaviour, sleep disturbance and more broad behavioural change. Thus, the aim of this systematic review is to evaluate if the literature supports the hypothesis that pain as a result of GI disease is associated with changes in behaviour, specifically challenging

behaviour and sleep problems, in individuals with ID and or ASD[1]. To accomplish this, the review will evaluate the broad behavioural changes associated with GI. It will also review the types of GI disorders that are most commonly researched and how GI disease is assessed in individuals with ID.

Method

Electronic searches and choice of search terms

Three databases were searched: Ovid Medline (R; 1946-November week 4 2015), Ovid PsychINFO (1967-November week 4 2015), and Ovid Embase (1974-November week 4 2015). Search terms for developmental disabilities and GI disease were sourced using a previous systematic review (De Winter et al., 2011). This systematic review (De Winter et al., 2011) aimed to determine the physical conditions associated with challenging behaviour, included papers up until 2008, and included a wide range of medical conditions. It concluded that there was a low level of evidence for an association between GI disease and challenging behaviour. Search terms for areas of associated behavioural change were selected as broadly as possible based on known literature and clinical practice. Terms were explored within each database and added to the final search to be as inclusive as possible. Sleep problems search terms was sourced using a previous review of sleep problems (van de Wouw et al., 2012). This review included papers that explored physical conditions; however, these

[1] A previous systematic review reviewed the physical conditions, including gastro intestinal issues, associated with challenging behaviour in individuals with ID (de Winter, Jansen, & Evenhuis, 2011). This review covered a wide range of physical illnesses and in papers up to 2008. It was felt that a systematic review focussing solely on gastrointestinal disorders was warranted and included relevant literature up to 2015. Also, studies that included participants with ASD were also included as the literature supports the increased prevalence of GI disorders and challenging behaviours in this population.

did not include GI disorders. Terms were searched for in all fields (title, abstract, keywords) and medical subject heading (MeSH) terms were used where indicated.

Eligibility criteria

Relevant peer reviewed empirical/observational studies published up to and including 30th October 2015 were included. All articles were written in English. Papers included had to have a sample that comprised participants with a diagnosis of ID or ASD. The papers included also had to have a measure of GI or participants had to have a diagnosis/suspect diagnosis of GI disease. Finally, papers needed to report a measure of behaviour change or reported behavioural change.

Search method

In the first search, search terms for developmental disabilities and gastrointestinal disease were used. In the second search, challenging behaviour search terms were combined with the first search. Terms for intellectual disability/developmental disabilities, gastrointestinal disease in table, and challenging behaviour in are displayed in Table 1.

Table 1: Subject Headings and Keyword Search Terms

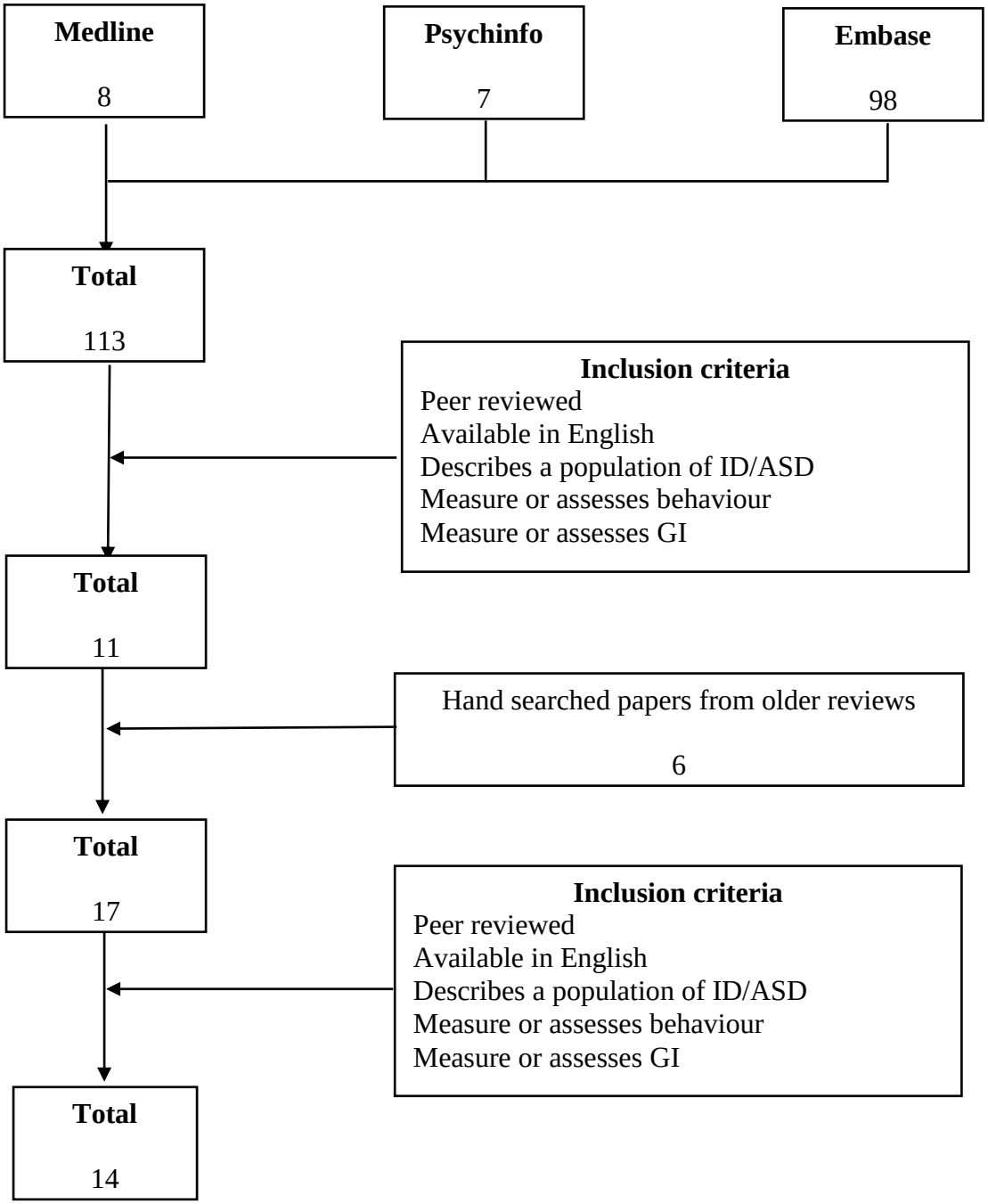
Search terms		
Intellectual disability/developmental disabilities	Gastrointestinal disease	Challenging behaviour and sleep disorder
Developmental Disabilities (MeSH)	Digestive System Diseases (MeSH)	Self-injurious behaviour (MeSH)
Intellectual Disability (MeSH)	Gastrointestinal Diseases (MeSH)	Aggression (MeSH)
Learning Disorders (MeSH)	Gastrointestinal Diseases Digestive System (MeSH)	Challenging behavio*
Developmental disab*	Gastrointestinal Diseases	Sleep Deprivation
Intellectual disab*	Gastrointestinal Disorder	Sleep Disorders, Intrinsic
Learning disab*	Gastrointestinal Tract (MeSH)	Sleep Arousal Disorders
Mental retard*	Gastrointestinal Disorders (MeSH)	Stereotyped Behaviour (MeSH)
Intellectual Development Disorder (MeSH)	Gastrointestinal System (MeSH)	Destructive Behavio*
Learning Disabilities (MeSH)	Digestive System(MeSH)	Aggress*
Pervasive Developmental Disorders	Gastrointestinal Disease (MeSH)	Sleep Disorders
Developmental disab*		Sleep Initiation and Maintenance Disorders
Intellectual Impairment		Sleep disorder*
Learning Disorder (MeSH)		
Learning Disabilities (MeSH)		
Autism (MeSH)		

*Note: MeSH=Medical Subject Heading; *=truncated search term*

Data collection and analysis

A search using the above terms was run across the three databases. One hundred and thirteen studies were identified, screened, and assessed for eligibility. The abstracts of the papers were read and once the exclusion criteria were applied, a total of 11 suitable papers were identified. A hand search of references in a key review that provided search terms (de Winter, Jansen, & Evenhuis, 2011) was also conducted and eight papers that reached criteria were included. Two of the papers from this review had already been identified in the database search. The six remaining papers were added to the present review. Overall, 17 papers were identified and read by the author. However, on reading the papers in full, three papers did not meet full inclusion criteria. As a result, the total number of papers included in this systematic review is 14. The results of the search and search process are summarised in Figure 1.

Figure 1. Search Process of the Literature Review



Data extraction and management

The author then analysed the study characteristics and the methodological quality of the of the research studies. Data were extracted on the following study characteristics: study design, sample size, demographic information (gender, age) diagnosis of ID/ASD, method of diagnosis, and proportion of the sample with ID/ASD diagnosis of GI, diagnostic method of GI, behaviour topography, behavioural measures utilised, percentage of sample with GI, percentage of sample with GI and behaviour change, percentage of the sample that were treated, percentage of the sample that saw a reduction in behaviour as a result of treatment, post measures (how was behaviour measured post treatment).

Assessment of the level of evidence

A quality checklist for randomised and non-randomised trials was adapted to evaluate the methodological quality of each study (Downs & Black, 1998). The Quality Index used had high internal consistency (KR-20: 0.89) as did the subscales apart from external validity (KR-20: 0.54). Test-retest (r 0.88) and inter-rater (r 0.75) reliability of the Quality Index were good. Reliability of the subscales varied from good (bias) to poor (external validity). Test-retest (r 0.88) and inter-rater (r 0.75) reliability were good. A copy of the quality checklist is included in Appendix 1. The checklist was designed for intervention studies and not all of the papers included in this review were of that nature. This checklist was used in an attempt to evaluate those studies that did include interventions. The studies that did not include interventions were not marked on intervention criteria and the items were omitted.

Results

Participants

The papers reviewed included participants with various developmental diagnoses. The majority of papers (n=11) focussed on individuals with ID, ranging from mild to severe. One of these papers expanded on the diagnosis and provided specific diagnoses and mechanisms of intellectual disability. Two of the papers described participants with ASD. Finally, one paper had participants with Cornelia de Lange syndrome. A summary of the papers can be seen in Table 2.

Table 2: Summary of Papers

Author, Year, Country	Sample	Diagnostic method and diagnosis of GI and behaviour	Percentage with GI and behaviour change and quality of evidence
Bosch et al., USA (1997)	**Size:** 25 inpatients, 3 cases presented. **Gender:** 2 female, 1 male. **Diagnosis ID/PDD:** Severe-Profound ID.	**Diagnosis GI**: Ulceration and inflammation of oesophagus, stomach and duodenum, h. pylori, constipation, delayed gastric emptying hiatal hernia, gastro reflux. **Diagnostic method GI:** Medical notes, OT, radiography, Gastroenterologist. **Behaviour topography:** Hand mouthing, self-injurious behaviour, kicking, hitting, hitting self, pushing into the wall, head banging, body slamming, whining, hair pulling, aggression, hand-biting. **Behavioural measures:** Chart review. MDT assessment, functional analysis	**Percentage with GI:** 24% **Percentage with GI and Behaviour change:** 100%, Quality of evidence: Lower (33/52)
Breau & Camfield, Canada (2011)	**Size:** N=123 No-pain group (n=86), Treated pain group (n=21) Untreated pain group (n=16). **Gender:** 56 Female, 67 Male. **Diagnosis ID/PDD:** ID.	**Diagnosis GI**: Caregiver report. **Diagnostic method GI:** Method unclear. **Behaviour topography:** Sleep behaviours, Childhood Sleep Habits Questionnaire. **Behavioural measures:** Childhood Sleep Habits Questionnaire, Vineland Adaptive Scale, Children's Deviation Intelligence Quotient.	**Percentage with GI:** Overall n=11 (9%). Untreated group=19%, Treated group=38%. **Percentage with GI and Behaviour change:** Not clear by disorder. Differences in sleep score between pain and no pain. Quality of evidence: Lower (31/52)

Clarke et al., UK (2006)	**Size:** N=36 **Gender:** NA **Diagnosis ID/PDD:** ID mild-severe.	**Diagnosis GI**: H. pylori. **Diagnostic method GI:** Antibodies from blood samples. **Behaviour topography:** Behaviour disorder given as a diagnosis, but no information on topography. **Behavioural measures:** None.	**Percentage with GI:** 36% **Percentage with GI and Behaviour change:** 36% Quality of evidence: Lower (36/52)
Hall et al., UK (2008)	**Size:** 54 syndrome, 44 comparison. **Gender:** Male=46%, Female=54% **Diagnosis ID/PDD:** CdLS.	**Diagnosis GI**: No diagnosis. **Diagnostic method GI:** Parent report. **Behaviour topography:** Sleep problems, self-injurious behaviour. **Behavioural measures:** Infant sleep questionnaire, Health questionnaire Challenging behaviour questionnaire.	**Percentage with GI:** 44% **Percentage with GI and Behaviour change:** No link. Quality of evidence: Higher (32/34)
Nikolov et al., USA (2008)	**Size:** 172 **Gender:** Male=145, Female=27 **Diagnosis ID/PDD:** 152=ASD, 6=Asperger's, 14=PDD-NOS.	**Diagnosis GI**: Constipation, diarrhoea, reflux, vomiting, pyloric stenosis, bowel malrotation, enterocolitis, lactose intolerance, colon polyps, and stomach cramps. **Diagnostic method GI:** Medical history, physical exam by nurse practitioner, child psychiatrist or paediatrician. **Behaviour topography:** Communication, stereotypy, hyperactivity, inappropriate speech, compulsive behaviours, irritability, social withdrawal, anxiety. **Behavioural measures:** The Vineland Adaptive Behavior Scale, The Aberrant Behavior Checklist, CYBOCS-PDD, CASI,	**Percentage with GI:** 39 (22.7%) **Percentage with GI and Behaviour change:** Participants without GI were twice as likely to respond to medication. GI higher irritability, social withdrawal, anxiety then no GI. Quality of evidence: Higher (50/52)

Gossler et al., Austria (2007)	**Size:** N=19. **Gender:** Male=10, Female=9. **Diagnosis ID/PDD:** Neurological impairment.	**Diagnosis GI**: GORD. **Diagnostic method GI:** 24 hr pH monitoring using 3-point pH catheters for all 19, barium swallows and gastric emptying in 18 of 19. **Behaviour topography:** Agitation: increased movements, decrease in cooperation and sleep, moaning, crying, and difficulties to pacify. Autoaggressive: scratching, biting, or hitting. **Behavioural measures:** Parental/caregiver report.	**Percentage with GI:** 100% **Percentage with GI and Behaviour change:** 100% Quality of evidence: Higher (39/52)
Bohmer et al., Netherlands (1999)	**Size:** N=186 **Gender:** Male=108, Female=78 **Diagnosis ID/PDD:** ID moderate-profound	**Diagnosis GI**: Esophagitis, hiatal hernia, gastritis or H. pylori found. Barrett's oesophagus was found in 18 (14.0%) and peptic strictures in five (3.9%) cases. **Diagnostic method GI:** pH-metry catheter, endoscopy to confirm. Biopsy for h. pylori and gastritis. **Behaviour topography:** vomiting, hematemesis, rumination, regurgitation, food refusal, automutilation, aggression, fear, episodes of screaming, depression, restlessness. **Behavioural measures:** Arbitrary definition. Present ten times in the last month, after consultation with the physician.	**Percentage with GI:** 15.90% **Percentage with GI and Behaviour change:** Vomiting, hematemesis, rumination, depression significantly more in those with abnormal pH. Quality of evidence: Higher (34/34)

Swender et al., USA (2009)	**Size:** 60 (30 engage in HM, 30 do not. **Gender:** Male=14, Female=16 in both conditions**.** **Diagnosis ID/PDD:** 2 severe ID, 58 profound ID.	**Diagnosis GI**: GORD. **Diagnostic method GI:** Medical records**.** **Behaviour topography:** HM. **Behavioural measures:** Questions about Behavioural Function.	**Percentage with GI: 60%** **Percentage with GI and Behaviour change:** If engage in HM, then 36.7% more likely to have GORD. SHM had higher scores on non-social. Quality of evidence: Higher (33/34)
Williams et al., Ireland (2014)	**Size:** 109 **Gender:** Male=80, Female=25 **Diagnosis ID/PDD:** 42% had ID. 25% had anxiety	**Diagnosis GI**: Abdominal Pain, constipation, diarrhoea, nausea, bloating. **Diagnostic method GI:** Gastrointestinal Symptom Inventory (parental report (GSI). **Behaviour topography:** Symptoms of anxiety (not specified), Sleep (bedtime resistance, onset delay, duration, anxiety, wakings, parasomnias, disordered breathing, daytime sleepiness. self-injurious behaviour, aggression, destruction. **Behavioural measures:** Child behaviour checklist, Children's sleep habits questionnaire, Behaviour Problems Inventory-Short.	**Percentage with GI:** 80% **Percentage with GI and Behaviour change:** Anxiety and GSI r (109) =.21, p<.05) small effect size. Anxiety and nausea r (109) =.19, p<.05) small effect. Anxiety and constipation r (109) =.20, p<.05) small effect. GI not emerge as a predictor of anxiety Quality of evidence: Lower (28/34).

Wallace et al., Australia (2002)	**Size:** 168. **Gender:** Male=94, Female=74. **Diagnosis ID/PDD:** ID.	**Diagnosis GI**: H. pylori. **Diagnostic method GI:** Medical, gastro exam and history. **Behaviour topography:** Trustworthiness, Stereotyped/hyperactive, social engagement, disturbing interpersonal behaviour **Behavioural measures:** Adaptive behaviour scale part 1 and part 2.	**Percentage with GI:** 74% as ever infected, 67% currently. **Percentage with GI and Behaviour change:** Currently infected: Higher levels of disability (ABS) Higher rates of maladaptive behaviour (see topography). Also higher level of ID. Quality of evidence: Higher (31/34)
Maenner et al., USA (2012)	**Size:** N=487. **Gender:** Male=88.6%**,** Female=11.4% **Diagnosis ID/PDD:** ASD, PDD, Asperger's. DSM criteria. ID, C. Palsy, Seizure like criteria.	**Diagnosis GI**: Constipation, encopresis, GORD. **Diagnostic method GI:** ADDM verbatim descriptions provided by doctor as per inclusion criteria. **Behaviour topography:** Sleep disturbance, stereotypic/repetitive, self-injurious behaviour, abnormal eating, aggression, mood, oppositional, tantrums, oblivious to other children, Lack imaginative play, lack of fear, insistence on sameness, delayed motor, abnormal cognitive development. **Behavioural measures:** Paediatric consensus report.	**Percentage with GI:** 35 (7.2%). **Percentage with GI and Behaviour change:** Sig. Sleep disturbance 3.1%, Eating 2.7%, Oppositional 2.5%. However, these behaviours were frequent in both with and without GI so has limited utility in screening. Quality of evidence: Higher (31/34)
Bohmer et	**Size:** N=1580.	**Diagnosis GI**: Vomiting, hematemesis, anaemia, rumination,	**Percentage with GI:** 6%

al., Netherlands (1997b)	**Gender:** IQ<35 (Male= 668, Female=410), IQ 35-50 (Male=368, Female=241). **Diagnosis ID/PDD:** ID moderate-profound.	regurgitation, behaviour problems. **Diagnostic method GI:** Records, research physician. Endoscopy in records. **Behaviour topography:** screaming, aggression, fear, restlessness. **Behavioural measures:** Coded from records. No measures.	(n=107) **Percentage with GI and Behaviour change:** All of diagnosis GI box except regurgitation were sig. more likely in patients than controls Quality of evidence: Higher (33/34)
Bohmer et al., Netherlands (1997a)	**Size:** 338. **Gender:** Male=167, Female=171. **Diagnosis ID/PDD:** ID.	**Diagnosis GI**: H. pylori. **Diagnostic method GI:** EIA-g test (antibodies) **Behaviour topography:** Looked at nonambulancy, rumination, faecal soiling, drooling only rumination. **Behavioural measures:** Medical notes	**Percentage with GI:** 85.3% **Percentage with GI and Behaviour change:** Association: Male p=0.04, IQ<50 p=0.04, rumination p=0.04. Non ambulant, drooling, and faecal soiling not significant. Quality of evidence: Higher (39/52)
Rogers et al. USA	**Size:** 23.	**Diagnosis GI**: Abnormalities. Regurgitation.	**Percentage with GI:** 10.4%

(1992)	**Gender:** Male=10, 11=female**.** **Diagnosis ID/PDD:** Profound ID.	**Diagnostic method GI:** Developmental paediatrician, radiographer (host of tests). **Behaviour topography:** Emesis (83%), Rechewing and swallow (17%), Self-stimulation (70), hands in mouth (43%), self-injurious behaviour (39%), PICA (26%) Aggression (26%). **Behavioural measures:** None stated, provided by Psychologists. Observations at mealtime.	**Percentage with GI and Behaviour change:** 10 patients (43%) hand mouthing which has led to diagnosis of rumination and regurgitation in the past. Quality of evidence: Lower (28/34)
Note: Abbreviations: OT=occupational therapy, MDT=multi-disciplinary team, CDLS=Cornelia de Lange syndrome, GI=gastro-intestinal, ASD=autism spectrum disorder, PDD-NOS=pervasive developmental disorder-not otherwise specified, CYBOCS-PDD= Children's Yale-Brown Obsessive Compulsive Scales modified for pervasive developmental disorders, CASI= Child and Adolescent Symptom Inventory Anxiety Scale, GORD= Gastroesophageal reflux disease, HM= handmouthing, ADDM=Autism and Developmental Disabilities Monitoring			

Methodological issues

Quality framework. A quality framework (Appendix 1) based on a checklist for the assessment of the methodological quality both of randomised and non-randomised studies of health care interventions (Downs & Black, 1998) was used to evaluate the methodological strengths and weaknesses of the papers in this review. The quality of the papers is presented in a 'traffic light' display; red meaning poor quality or absent, orange meaning some features were present, and green meeting the quality criteria outlined. Where there are blank spaces, the criterion was not applied due to being not applicable.

A scoring system was devised to give an overall indicator of the quality of a study. Items that were green were given a value of two, orange items a value of one, and red items a value of zero. The quality framework did not provide score cut-off points. In this case, the average score of the papers was calculated and scores falling above were judged as higher quality while scores falling below were judged as lower quality. It is important to note that the papers were compared against each other and that this scoring system presents a basic method for judging included paper's strength. The scores are provided in Table 2.

Overall, the papers were of good quality. Using the above quality scoring scheme, nine papers were found to be of higher quality (Böhmer et al., 1999; Böhmer, Niezen-de Boer, Klinkenberg-Knol, Nadorp, & Meuwissen, Oct 1997b; Böhmer et al., Oct 1997b; Gössler, Schalamon, Huber-Zeyringer, & Höllwarth, 2007; Hall, Arron, Sloneem, & Oliver, 2008; Maenner et al., 2012; Nikolov et al., 2008; Swender, Matson, Mayville, Gonzalez, & McDowell, 2006; Wallace, Webb, & Schluter, 2002). Five papers were found to be of lower quality (Bosch, Van Dyke, Milligan Smith, & Poulton,1997; Breau & Camfield, 2011; Clarke, Vemuri, Gunatilake, & Tewari, 2008; Rogers, Stratton, Victor, Kennedy, & Andres, 1992; Williams, Leader, Mannion, & Chen, 2015). There were some weaknesses that were

consistent across the majority of papers. Of the 14 papers, seven reported using interventions and only four of these reported them to the standard of the framework. Five papers reported the distribution of potential confounding variables. Five papers considered the adverse effects of the research on the participants. This is important as several of the studies used potentially invasive medical assessments. One paper in the review reported that the researchers were blind to the conditions. Also, only one of these studies reported intervention compliance.

Finally, the majority of studies were not randomised control trials, so only some of the quality frame work is appropriate. The results of the quality framework are summarised in Appendix 1.

Assessment of gastrointestinal disease. There are multiple ways in which GI disease was defined and diagnosed in these studies. Participants varied in their presentations and how they were diagnosed. A number of studies used direct assessments, conducted either by the researchers or by medically trained individuals, to assess GI disease. Three studies were identified that directly assessed Gastro-Oesophageal Reflux disease (GORD). Gössler, Schalamon, Huber-Zeyringer, & Höllwarth (2007) assessed reflux in participants using 24 hr pH monitoring using a combination of pH catheters, barium swallows, and gastric emptying. Similarly, Böhmer et al. (1999) used pH-metry catheters to assess the possibility of reflux. Where it was indicated, an endoscopy was performed to confirm the diagnosis of reflux. Nikolov et al. (2008) assessed past, current, and chronic GI problems through medical records and a non-specified physical examination by a medical professional. Within a sample of 172 children, 39 (22.7%) were said to have GI problems. These conditions included, in order of most common to least common, constipation, diarrhoea, reflux, vomiting, pyloric stenosis, bowel malrotation, enterocolitis, lactose intolerance, colon polyps and stomach cramps. (Rogers et al., 1992) used a series of tests conducted by a developmental paediatrician and

radiographer to assess regurgitation, which is the expulsion of material from the oesophagus. However, there was limited information presented about the specifics of the tests.

Of the studies that directly assessed GI disorder, four papers assessed the presence of *Helicobacter pylori (h. pylori)*. H. pylori has been noted as the most important cause of chronic active gastritis (Kuipers et al., 1995). Wallace, Webb, & Schluter (2002) assessed for h. pylori using medical histories, physical exam, and samples were taken of faeces and blood. Böhmer et al. (1997) used EIA-g antibody test to assess the presence of h. pylori. Similarly, (Clarke et al., 2008) tested 36 participants for h. pylori using antibodies from blood samples. Finally, Böhmer et al. (1999) tested the presence of h. pylori and gastritis by performing a biopsy. It would appear from this selection of the literature that the assessment of the presence of h. pylori was conducted using medical testing of blood, faeces, and tissue samples.

While direct assessments were not carried out, a further three papers in the review used medical notes to assess the presence of GI disorder. Swender, Matson, Mayville, Gonzalez, & McDowell (2006) used medical records to ascertain a diagnosis of GORD. In order to be included in the study, the participant's record must have had reference to the medical testing involved in the diagnosis (e.g. pH testing and/or diagnosis). However, assessment details were not reported. On the other hand, Bosch, Van Dyke, Milligan Smith, & Poulton (1997) described three cases in detail. All information in this study was taken from medical records. Patient A was diagnosed with ulceration and inflammation of the oesophagus, stomach, and duodenum and h. pylori was present. These were diagnosed from previous medical notes and assessments from occupational therapy, radiography, and a gastroenterologist. Patient B presented with a hiatal hernia, gastro-oesophageal reflux, delayed gastric emptying, and a duodenal ulcer. Similarly, these were diagnosed using

medical notes, a barium swallow, radiography, and gastroenterology. Finally, Patient C was diagnosed with constipation from medical notes which reported the results from a barium swallow and a small bowel study. Böhmer, Niezen-de Boer, Klinkenberg-Knol, Nadorp, & Meuwissen, (1997) reported presenting problems such as vomiting, haematemisis, anaemia, rumination, regurgitation. A research physician collected data from medical records.

The remaining four papers used caregiver report. One paper used a measure that had been used in previous research (Williams et al., 2015), one paper used a bespoke method of using descriptions from a database (Maenner et al., 2012) and the other two papers asked caregivers to report any issues/diagnosis (Breau & Camfield, 2011; Hall et al., 2008). Williams et al. (2015) used the Gastrointestinal Symptom Inventory, a parental report, in their study. The most commonly reported difficulties, in order of most to least, were abdominal pain, constipation, diarrhoea, nausea, and bloating. Maenner et al. (2012) found the participants presented with, in order of most common to least common, constipation, encopresis, and GORD. This was identified using data that contained verbatim descriptions in the Autism and Developmental Disabilities Monitoring Network. Breau & Camfield (2011) used a caregiver report to assess GI problems. However, the content of the report was not included in the study nor was how or when the participants were originally given diagnoses. Hall et al. (2008) used topographies of health problems provided by caregiver report, but the paper does not report how the participants were originally given diagnoses.

As can be seen, there is considerable variability in assessment methodology and type of GI disease assessed. All illnesses fall under the umbrella of GI disease but vary in the organ affected; from the upper tract to bowel. How diagnosis was reached is more uniform, in that five studies used medical notes and eight reported assessments carried out by medically trained professionals. Of these studies, three reported using both methods. Ten

papers in this review assessed and diagnosed medical issues either by direct medical assessment or from medical notes. The other four papers utilised parental report for symptoms. Direct medical assessment could be seen as the most efficacious as they give a current diagnosis using biological markers. Some of the papers only noted that medical notes were used and did not specify what tests had been done in the past and by what professional. Finally, considerable variation in caregiver report was noted. Some caregivers reported diagnoses given by professionals but once again, what diagnoses and how they were reached were not reported. Other studies used symptom checklists.

Assessment of behaviour topography. In terms of defining and reporting on behaviour, the behavioural topography reported in these studies varies from precise descriptions of the behaviour, such as headbanging, to broad diagnoses, such as behavioural disorder.

Four papers reported specific self-injurious behaviour topographies. Bosch et al. (1997) reported hand mouthing, kicking, hitting self, pushing into the wall, head banging, body slamming, whining, hair pulling, aggression, scratching chest. One of the participants was noted to engage in self-injurious behaviour but the topography is not elaborated on. These behavioural topographies were collected using patient chart reviews, MDT assessment, and functional analyses of behaviour. Gössler et al. (2007) used parental and caregiver reports to document agitation, which was defined as increased movements, decreased cooperation, decreased sleep, moaning, crying, difficulties in pacifying. Auto aggressive behaviour was defined as scratching, biting, and hitting. Specific topographies of behaviour were not reported in the results section. No validated measures were used. Böhmer et al. (1999) reported behaviours such as hematemesis, rumination, regurgitation, food refusal, auto mutilation, aggression, fear, screaming, depression, and restlessness. The authors defined

rumination as the deliberate regurgitation of food into the mouth with some being ejected and the reset swallowed. These symptoms were arbitrarily defined by a physician and they must have been present four times in the past month. Some symptoms were defined, such as rumination, but others were not, such as depression. Swender et al. (2006) assessed hand-mouthing through medical records. The Questions about Behavioural Function (QABF) assessment was used to indicate the possible function behind the hand-mouthing. These were the only four papers that reported the specific topography of the self-injurious behaviour.

In addition to self-injurious behaviour another study reported on sleep problems in individuals with CdLS (Hall et al., 2008). The Infant Sleep Questionnaire and the Challenging Behaviour Questionnaire were used but the paper did not report the specific types of self-injury and sleep problems. Williams et al. (2015) utilised the Child Behaviour Checklist, the Children's Sleep Habits Questionnaire and the Behaviour Problems Inventory-Short form to assess anxiety, sleep problems, self-injurious behaviour, aggression, and property destruction. However, symptoms for anxiety were not specified and neither were the specific topographies from the Behaviour Problems Inventory. Rogers et al. (1992) reported the presence of emesis, rechewing and swallowing, self-stimulation, hand mouthing, self-injurious behaviour, PICA, and aggression using observations provided by psychologists at mealtime. Clarke et al., (2008) reported that some participants had diagnoses of behaviour disorder, but no details were provided on the topography of these behaviours. How the diagnosis was reached was not reported either. Böhmer et al. (1997) coded challenging behaviour from patients' medical records. Screaming, aggression, fear, and restlessness were identified. However, no measures were noted from the medical records. Böhmer et al. (1997b) assessed non-ambulancy, IQ<50 rumination, faecal soiling, and drooling and found that rumination and IQ<50 were significantly associated with h. pylori. Nikolov et al. (2008)

used several scales (the Vineland Adaptive Behavior Scale, the Aberrant Behavior Checklist, the Children's Yale-Brown Obsessive Compulsive Scale for Pervasive Developmental Disorder (CYBOCS-PDD), and the Child and Adolescent Symptom Inventory (CASI) Anxiety Scale to measure communication, stereotypy, social development, hyperactivity, inappropriate speech, compulsive behaviours, irritability, social withdrawal, and anxiety. Wallace et al. (2002) used the Adaptive Behaviour Scale parts 1 and 2 and reported the item scores in full within the intellectual functioning and maladaptive behaviours domains. Maenner et al. (2012) used a list of behavioural features identified in a paediatric consensus report such as abnormalities in sleeping, stereotyped and repetitive motor mannerisms, self-injurious behaviours, abnormal eating habits, abnormalities in mood or affect, argumentative, oppositional, defiant, or destructive behaviours, aggression and temper tantrums. These papers reported on overarching behavioural classes and did not report on specific topographies.

Three studies measured sleep problems in this population. Breau & Camfield (2011) assessed sleep behaviours using the Childhood Sleep Habits Questionnaire (CSHQ) which uses subscales such as bedtime resistance, sleep onset, delay, sleep duration, sleep anxiety, night wakings, parasomnias, sleep disordered breathing, and daytime sleepiness. Williams et al. (2015) reported sleep problems subscales including sleep anxiety, sleep duration, and night wakings. Hall et al. (2008) assessed sleep problems in individuals with CdLS but specific sleep problems were not reported.

Several papers use parental and caregiver report to identify challenging behaviours (Gössler et al., 2007; Hall et al., 2008; Nikolov et al., 2008; Swender et al., 2006; Wallace et al., 2002; Williams et al., 2015) and sleep problems (Breau & Camfield, 2011; Hall et al., 2008; Williams et al., 2015) while there were a number of papers that provided behaviour

information using medical history and direct observation by staff (Böhmer et al.,1997a, 1997b; Böhmer et al., 1999; Bosch et al., 1997; Maenner et al., 2012; Rogers et al., 1992) and one paper did not give information on how behaviour was defined or collected (Clarke et al., 2008). Various topographies of behaviour were reported in the above studies. However, only four reported exact definitions (Böhmer et al.,1997a; Bosch, J. et al., 1997; Gössler et al., 2007; Swender et al., 2006) while 10 reported the overarching behavioural class (Böhmer et al., 1997a; Böhmer et al., 1999; Breau & Camfield, 2011; Clarke et al., 2008; Hall et al., 2008; Maenner et al., 2012; Nikolov et al., 2008; Rogers et al., 1992; Wallace et al., 2002; Williams et al., 2015). Thus, the studies that did not operationally define the target behaviours may lack internal validity.

Behavioural correlates with gastrointestinal disease

Self-injurious behaviour. Five studies reported that the topography of self-injurious behaviour was associated with GI disease. A study assessing at the role of medical conditions in self-injurious behaviour reported that seven individuals within an inpatient service for individuals with self-injurious behaviour were treated for unidentified medical conditions (Bosch, Van Dyke, Milligan Smith, & Poulton, 1997). Six of these patients were diagnosed with, and treated for, a GI disease (24% of all referrals) and five of these saw subsequent reductions in self-injurious behaviour. While this was a positive outcome, the measures of challenging behaviour reduction were variable, ranging from baseline and follow-up functional analysis to anecdotal. A study examining GORD in neurologically impaired children reported that all participants with auto-aggression and agitation had increased inflammation of the oesophagus compared to individuals without behavioural abnormalities (Gössler et al., 2007). Children with behavioural problems also had a significantly higher reflux index, which was defined as oesophageal acid exposure of more than 4% of total reflux

time when reflux time is a distal oesophageal pH of less than four for at least 15 seconds measured 3 to 5 cm from the oesophagogastric junction, which indicates that pathological reflux needs to be considered. The research concluded that self-injurious behaviour and agitation were associated with GORD and could be used as valid indicators of reflux in this population. However, the scale used to measure agitation was not provided. The scale appeared to be based on subjective reports from caregivers, and was created for the purpose of the study. Wallace, Webb, & Schluter (2002) reported higher levels of maladaptive behaviour, including self-injury, in individuals with higher levels of h. pylori. Swender, Matson, Mayville, Gonzalez, & McDowell (2006) demonstrated that hand mouthing, a form of self-injurious behaviour, had a greater prevalence in individuals with a diagnosis of GORD than in individuals without a diagnosis. A criticism of this is that, through behavioural checklist, hand mouthing was shown to be maintained by non-social reinforcement rather than pain. That said, the authors suggest that this is not unexpected as all the participants were receiving treatment for GI disease and hand-mouthing may have acquired a secondary function. Finally, Rogers, Stratton, Victor, Kennedy, & Andres (1992) noted that 10 patients (43%) in their sample engaged in hand mouthing which had led to diagnoses of rumination and regurgitation in the past.

There appears to be reasonable evidence from these studies supporting an association between self-injurious behaviour and GI disorder. Self-injurious behaviour in general, and specifically hand mouthing, were cited as indicators of GI disorder, namely reflux, rumination, and regurgitation and there is some limited evidence that treating the GI disorder leads to a reduction of self-injurious behaviour.

There are four studies in the review that found no association between self-injurious behaviour and GI disease. Hall, Arron, Sloneem, & Oliver (2008) assessed sleep and health

problems in individuals with CdLS. GI symptoms were reported in 44% of the sample but no association was found between GI symptoms and the presence of self-injurious behaviour. This is in contrast to 65% prevalence reported by Luzzani, Macchini, Valadè, Milani, & Selicorni (2003). However, GI was not diagnosed using medical methods and symptoms were obtained by parent report. No link between self-injurious behaviour and GI disorders was found in a study on an inpatient population (Böhmer et al., 1997). Maenner et al. (2012) looked at several behavioural topographies, including self-injurious behaviour, and their possible relationship with GI disease. They too found no association between self-injurious behaviour and GI disease. Nikolov et al. (2008) did report a link between the presence of GI disease and higher scores on an irritability subscale, but when taking into account that their sample was over represented with individuals with self-injurious behaviour and when the diagnosis of GI disorder was made they concluded that the association was not supported. Three papers provided good evidence not supporting the association between GI disorder and self-injurious behaviour while one paper offers reasonable evidence for no association as information on GI disorder was provided by parental report, not medical notes. Finally, Williams, Leader, Mannion, & Chen, (2015) measured self-injurious behaviour and GI disorder but did not test the association between the variables.

There appears to be evidence for and against an association between self-injurious behaviour and GI disorder. Five papers report an association, with various degrees of evidence. Three of the studies used medical examination or medical notes to diagnose GI disorder (Rogers et al., 1992; Swender et al., 2006; Wallace et al., 2002). Two of these papers (Swender et al., 2006; Wallace et al., 2002) also used validated measures of behaviour, such as the QABF and Adaptive Behavior Scale to measure behaviour while Rogers et al. (1992) used observations made by psychologists trained to observe and record behaviour. The other

two papers, Bosch et al. (1997) and Gössler et al. (2007), used inconsistent outcome evaluations and subjective behaviour recording respectively. Four papers tested the association but did not find enough evidence to support it (Böhmer et al.; Hall et al., 2008; Maenner et al., 2012; Nikolov et al., 2008) while the final paper measured both self-injurious behaviour and GI but did not attempt to test the association between them. Given the equivocal associations reported, it is difficult to reach a conclusion on the strength of the association between self-injurious behaviour and GI disorder given the studies included in this review.

Other challenging behaviours. Challenging behaviours other than self-injurious behaviour were also reported. Clarke, Vemuri, Gunatilake, & Tewari (2008) researched the association between h. pylori and behaviour disorder and found that 36% of a sample with behaviour disorder and ID tested positive for the strain. However, the study did not describe the topography of the behaviour and while all patients were treated for h. pylori, there was no post-intervention measure of behaviour change. Behaviour disorder was not defined and could include other forms of challenging behaviour, such as self-injurious behaviour, aggression, and property destruction. Wallace et al. (2002) found that individuals currently infected with h. pylori displayed higher levels of disability on the Adaptive Behaviour Scale in the areas of trustworthiness, stereotyped/hyperactive behaviour, social engagement difficulties, and disturbing interpersonal behaviour.

As well as concluding that self-injurious behaviour can be used as indicator for GORD, Gössler et al. (2007) noted that agitated behaviour was also correlated with the severity of GORD where agitation was defined as increased movements, decrease in co-operation and sleep, moaning, crying, and difficult to pacify. Maenner et al., (2012) found significant associations between eating difficulties, and oppositional behaviours and GI

disorder. However, as these behaviours occurred in individuals with and without GI problems these behaviours have limited specificity to GI disease. This is important as identifying behavioural indicators of GI disorder may make screening and diagnosis easier in non-verbal populations. In a sample of individuals in an institution, 6% were found to have GI disorder (Böhmer et al. 1997). Individuals with GI disorder were more likely to have behaviour problems such as screaming, aggression, fearfulness, and restlessness than individuals without GI disorder.

However, Nikolov et al. (2008) found no differences in ASD symptomatology between those with GI disorder and those without GI disorder. The authors also concluded that irritability was not associated with GI disorder despite several individuals with current GI disorder scoring higher on irritability than those without GI disorder.

In summary, five papers reported an association between other behaviours and GI disorder. These included behaviour disorder in general, stereotyped/hyperactive behaviour, social engagement difficulties, disturbing interpersonal behaviour, agitation, eating difficulties, oppositional behaviours, screaming, aggression, fearfulness, and restfulness. Only one paper found no association between communication, social behaviour, repetitive behaviour and irritability. This suggests that there is an association between some maladaptive behaviours and GI disorder, in that these behaviours are more likely to occur in individuals with GI disorder.

Sleep disturbance. Four papers reported on both sleep problems and GI disorder. However, not all evaluated associations between the two variables. Maenner et al., (2012) found a significant association between sleep disturbance and GI disorder. However, sleep disturbance occurred in individuals with and without GI disorder so sleep problems may have

a limited specificity to GI disorder. A limitation of this study is that a validated measure of GI disorder was not used. Breau & Camfield (2011) reported increased sleep problems in individuals experiencing pain due to GI disorder compared to individuals not experiencing pain.

Two papers measured sleep and GI disease but did not test the association between the two variables (Hall *et al.*, 2008; Williams et al., 2015).

There is limited evidence from the studies included in this review for the association of sleep problems and GI disorder. Two papers concluded that GI disorder may cause increased sleep disturbance. However, one of these papers noted that there is limited utility in using sleep problems as a screen for GI disorder, due to a lack of specificity. The other two papers included in this review did not test the association between sleep and GI disorder.

Anxiety/Depression. The relationship between GI disorder and anxiety and/or depression was also explored in several papers. Neither anxiety nor depression were included in the search terms for the review. However, several papers that met inclusion criteria made reference to them. Given the potential for a relationship between these and GI disorder, and the possibility that such a relationship could indicate the presence of GI disorder, they would be included in the review. Where depression was not directly measured as a construct, behaviours that could be construed as symptoms were, such as crying and withdrawal.

Similarly, anxiety and symptoms of it such as agitation and restlessness were measured. Böhmer et al., (1999) reported that depression is significantly more common in those with abnormal pH levels. However, clinical judgement was used and without a set of pre-specified criteria, it is not possible to evaluate or replicate these findings. Williams et al. (2015) showed a significant association with a small effect size between anxiety and GI

disorder. However, anxiety did not emerge as a predictor of GI disorder. Nikolov et al. (2008) found that those with GI disorder scored higher on irritability, social withdrawal, and anxiety than those without GI disorder. Agitation, defined as increased movements, decrease in cooperation and sleep, moaning, crying, and difficulties to pacify, was found to be a marker for GORD (Gössler et al., 2007).

Four papers assessed depression and/or anxiety in relation to GI disorder. However, where an association was made the results are potentially confounded by the symptoms also being associated with pain (Davies and Oliver, 2014) in that pain from medical conditions could act as a setting event for it (Breau & Camfield, 2011). For example, the definition of agitation used by Gössler et al. (2007) could be construed as symptoms of anxiety or as symptoms of pain and Greenlee et al. (2016) reported an association between depression and gastrointestinal problems in a sample of individuals with ASD.

Other behavioural correlates. The papers included in this review measured other variables that may be associated with or impacted by GI disorder. Nikolov et al. (2008) noted that those with GI disorder were less responsive to risperidone than those without GI disorder. Böhmer et al., (1999) found that vomiting, hematemesis (vomiting of blood), and rumination occurred significantly more in those with abnormal pH. Similarly, Böhmer et al. (1997) found that rumination was more likely in individuals with h. pylori.

It should be noted that all four papers that assessed the presence of h. pylori reported associations between h. pylori and the presence of maladaptive behaviour (Wallace, Webb, & Schluter,2002; Clarke, Vemuri, Gunatilake, & Tewari, 2008) rumination (Böhmer et al. 1997b; Böhmer et al. 1999) and depression (Böhmer et al. 1999).

Discussion

This review has evaluated the association between GI disorders and behaviour such as challenging behaviour, sleep problems, anxiety, and affect related behaviours. The review focussed on two main areas; the methodological issues regarding research into this area and evidence for the association between GI disorders and the behaviours listed above.

Regarding how diagnosis of GI disorder was reached, medical notes and assessments by medically trained professionals were carried out in the majority of papers while the rest used parental report for symptoms. There were multiple forms of GI disorder reported, such as GORD, vomiting, diarrhoea, rumination, regurgitation. H. pylori, a leading cause of gastritis (Kuipers et al., 1995), was reported in four of the studies reported. Regarding reporting types of challenging behaviour, there is inconsistency in how behaviour topography of challenging behaviour and other behaviours were reported across the studies. Some studies reported general behavioural categories, such as behaviour disorder while other studies used specific behavioural topographies, such as hand mouthing and body banging.

There was equivocal evidence for an association between self-injurious behaviour and GI disorder. Five papers reported an association but a further four papers did not find enough evidence to support an association. Regarding other behaviours besides self-injurious behaviour, there appears to be an association between some behaviour, such as aggression and stereotyped/hyperactive behaviour, and GI disorder, in that these behaviours are more likely to occur in individuals experiencing GI disorder. However, there is limited evidence for the association of sleep problems and GI disease from the papers in this review. Finally, there is some evidence that GI disorder may be associated with depression and/or anxiety in this

population but the behavioural markers used to establish a diagnosis may be potentially confounded by the symptoms also being associated with pain.

There are several limitations of the studies included in the review. There are multiple ways in which GI disorder is assessed and multiple different GI disorders reported. Also, the quality of definitions of the various topographies of behaviour varied in the studies included. The majority of the papers included did not operationally define the target behaviours. Instead, broad categories of behaviours were frequently used, such as 'behaviour disorder' and aggression. This makes it difficult to evaluate and replicate research into specific topographies that may serve as behavioural markers for GI disorder.

There are several strengths and weaknesses in this review. Both a potential strength and weakness was a potentially over-inclusive search strategy. Research evaluating challenging behaviour in general were included along with other behaviours and GI disorders in general that impacted on the lives of those who engage in them and also in those who care and work with them. While this broadened the scope of the review it perhaps prevented a focus on a specific behaviour topography or GI disorder. Also, GI search terms were not truncated. While this made the number of search results more manageable, it may have reduced the number of articles identified, potentially excluding relevant papers. Furthermore, three databases were used to manage the amount of results. They were chosen on the basis of their relevance to the research question e.g. Medline was chosen as it contains most papers on GI disorders. Other databases may also have reported relevant papers but it was felt that the databases used was comprehensive. Another limitation is that there were not many papers that reached inclusion criteria. This may be due to the search strategy but it seems more likely that this is still an emerging research area. Finally, the quality criteria used (Downs & Black, 1998) was designed to be used with intervention studies and over half of the papers

included were not intervention studies. However, many of the items on the checklist were applicable and using the same criteria for all studies allowed for a more direct comparison between these criteria.

Direct medical assessment is seen as the 'gold standard' (NICE, 2015) and was frequently employed in the studies included in the review. Medical notes/histories were also used. Interestingly however, research has suggested that data extracted from medical notes tend to under report GI disorder compared to medical exams and parental report, which were also used (Wang, Tancredi, & Thomas, 2011). This could mean that some of the studies that used medical notes could have under reported the presence of GI disorder. Also, the studies included in the review provided limited evidence for an association between disordered sleep and GI disorder. However, there is evidence in the literature using typically developing populations that sleep is affected by GI disorder (Dent et al., 2013; Johnson, 2005).

There are several implications for clinical psychology that can be drawn from the findings of this review. When assessing challenging behaviour, physical illnesses such as GI disease should be assessed and ruled out early in the process. While there is evidence for and against the association between GI disease and challenging behaviour, the literature supporting the association and the literature surrounding the difficulties for non-verbal individuals in communicating physical distress suggests that physical examination should be high on a clinician's agenda. Many papers did not operationally define the target behaviours, which is important in assessing and intervening (Cooper, Heron, & Heward, 2006).

In conclusion, it is recommended that more intervention studies are conducted on this population. The ultimate goal should be to have reliable behavioural markers as to the presence of GI disorders. Research should use operational definitions of potential behavioural

indicators of GI disorder to increase replicability and to increase the clinical implications of having a potential way to screen for GI disorder in non-verbal individuals. Similarly, how GI disorder is diagnosed/assessed in these studies should be more uniform and if possible, rigorous (NICE, 2015). For example, GORD should be assessed using pre- and post-treatment pH-level monitoring after initial endoscopic examination (Eryılmaz et al., 2012). However, these procedures are invasive and are not always possible in the clinical setting.

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THE DEVELOPMENT OF A CHALLENGING BEHAVIOUR REPORT FORM

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Abstract

Introduction

Challenging behaviour is shown by between 10% and 15% of people with intellectual disabilities (ID). The high prevalence of challenging behaviour and its significant impact warrant early, effective intervention. A valid and reliable assessment is required to understand the underlying causes of challenging behaviour effectively intervene.
The aim of this paper is to develop such an assessment, called the Challenging Behaviour Report Form (CBRF-R). It will include variables to assess behavioural function, behavioural indicators of pain, behavioural indicators of affect, precursor behaviours, and severity. The reliability of the form will be assessed using experimental functional analyses footage of challenging behaviour in non-verbal children with Autism Spectrum Disorder (ASD).

Method

The CBRF-R was developed from an existing assessment through a process of discussion with other clinicians and researchers. Once completed, the reliability of the CBRF-R was assessed on experimental functional analysis footage of children and adolescents with ASD and challenging behaviour.

Results

A total of 125 CBRF-R forms were completed by Observer 1 from footage of 21 participants who engaged in challenging behaviour. Observer 2 completed 31 (25%) CBRF-R forms for the purpose of reliability. The majority of variables included in the form had fair to strong inter observer agreement while ten had poor reliability.

Conclusion

The CBRF-R provides an efficient and simple method for recording incidents of challenging behaviour. It has been shown to reliably measure many factors that occur in incidents of challenging behaviour. Future development of the form is CBRF-R is possible by applying it to natural observations and assessing its validity by comparing its results to more established methods such as experimental functional analyses.

Introduction

Challenging behaviour is shown by between 10% and 15% of people with intellectual disabilities (ID; Emerson, 2001) and the term includes behaviours such as self-injury (SIB) aggression and destruction of the environment. These behaviours lead to decreased quality of life (Beadle-Brown et al., 2009)**,** increased risk of psychiatric hospitalisation (Mandell, 2008) and greater likelihood of reactive physical intervention (Allen et al., 2009). Challenging behaviour also affects carers, with parents experiencing higher levels of stress (Hastings, 2003; McIntyre, Blacher, & Baker, 2002) and staff being at higher risk of burnout (Mills & Rose, 2011).

The high prevalence of challenging behaviour in this population and its significant, negative consequences warrant early, effective intervention. To do this, a valid and reliable assessment is needed to understand the underlying causes of challenging behaviour. Prevailing interventions for challenging behaviour includes Positive Behaviour Support (PBS), an approach to the delivery of behavioural services that has been substantially informed by the Applied Behaviour Analysis (ABA) literature (Johnston, Foxx, Jacobson, Green, & Mulick, 2006). The goals of this approach are to apply 'behavioural principles in order to reduce problem behaviours and build appropriate behaviours that result in durable change and a rich lifestyle' (Carr, 1999). A feature of this approach, which is rooted in the ABA literature, is an emphasis on the assessment of function to tailor interventions to bring changes in behaviour and quality of life to the individual (Carr, 1999).

Evidence from ABA studies demonstrates that operant learning theory can account for behaviours such as SIB as a learned response to environmental stimuli (Iwata, Pace, et al., 1994). Operant learning theory proposes that challenging behaviour is learned and

maintained and modified using positive / negative reinforcement, positive/negative punishment (Iwata, Pace, et al., 1994). Demanding tasks and low levels of attention are examples of antecedents which may elicit SIB (Beavers, Iwata, & Lerman, 2013; Iwata, Pace, et al., 1994).

It is argued that in order to effectively reduce challenging behaviour the targeted behaviour should be assessed in a valid and reliable way (Beavers et al., 2013). There are multiple methods of assessing the function of behaviour including naturalistic observations (ABC analysis) and informant based measures (Beavers et al., 2013). A highly effective method is using experimental functional analysis to uncover the function of challenging behaviour which allows for a targeted intervention to be put in place (British Psychological Society, 2007; Iwata, Pace, et al., 1994; NICE, 2015). One of the first studies using experimental functional analysis observed participants repeatedly across multiple analogue conditions (Iwata, Dorsey, Slifer, Bauman, & Richman, 1982). This demonstrated that it is possible to identify variables that affect self-injury and had significant influence on the treatment of challenging behaviour as it allowed for targeted interventions depending on the function identified, whereas before interventions were not targeted (Carr & Durand, 1985; Iwata, Dorsey, Suifer, Bauman, & Richman, 1994). However, Matson & Minshawi (2007) argue that there is still much work to be done before experimental functional analysis can be described as a valid technology due to a lack of group comparison studies comparing it to checklists or functional assessment.

There are ethical and practical issues that surround experimental functional analysis and it seems to be rarely used in applied settings, instead being reserved for academic research (Matson & Minshawi, 2007). Hastings and Noone (2005) highlighted that experimental functional analysis may not be suitable for instances where severe challenging

behaviour is present. It was also noted that the results from the assessment may not always be clear as multiple functions may be identified. There is a growing call for a less labour intensive and more ethical assessment for challenging behaviours while still assessing the function in a reliable and valid way (Matson & Minshawi, 2007).

A less invasive method of assessing function is the use of informant based methods (Floyd, Phaneuf, & Wilczynski, 2005) such as the Questions about Behavioral Function (Paclawskyj, Matson, Rush, Smalls, & Vollmer, 2000) and the Motivational Assessment Scale (Durand & Crimmins, 1988). Indirect assessment instruments conducted with knowledgeable informants may yield relevant information that aids in the identification of functional relations such as motivating operations affecting problem behaviours that may not be apparent if functional analysis is used exclusively. However, the benefits of indirect methods are offset by the fact that reports stem from recollections of the problem behaviours and personal judgments about behaviour environment interactions and not their direct measurement and erroneous hypotheses may be developed (Floyd et al., 2005) and may be less reliable than direct observation (Beavers et al., 2013).

Naturalistic methods may also be used. A common method is the use of ABC charts, in which descriptions of the antecedent, behaviour, and consequence are recorded by an observer. However, these charts provide little in the way of structure and guidance to the observer, resulting in data that are often not reliable (Toogood & Timlin, 1996). A variation of these open ended charts are structured ABC charts. These take the form of checklists that prompt the observer and aid interpretation (O'Neill, Storey, Horner, & Sparague, 2014). However, these charts often contain information that relates to behavioural function without reference to other salient factors.

In summary, there are several main issues regarding the methods used in the assessment of function. Naturalistic observation methods do not provide much structure and guidance to the observer and may provide unreliable data (Toogood & Timlin, 1996), informant based measures may exclude the assessment of other salient factors (Floyd et al., 2005), and experimental functional analysis has ethical and practical issues and there is demand for less labour intensive assessments for challenging behaviour (Matson & Minshawi, 2007). There is scope to develop such direct observational methods to include more contextual information about an episode of challenging behaviour.

While operant learning theory has led to successful interventions for challenging behaviour, not all interventions are successful even when a maintaining function has been identified (Matson & LoVullo, 2008). This suggests that there may be other processes underlying the behaviour and that the assessment of antecedents may aid in the assessment of challenging behaviour (Oliver, 1995). Several other contributing factors have been identified in the literature as direct causes and setting events of challenging behaviour. These include pain, affect, and precursor behaviours and their assessment may aid the assessment of challenging behaviour.

Pain has received increased attention in recent times (Carr & Owen-DeSchryver, 2007). Pain could be formulated as a motivating operation which accounts for conditions which increase or decrease the effectiveness of a reinforcer or a punisher (Laraway et al., 2003). There is some evidence that undiagnosed health conditions that cause pain in non-verbal individuals with ID may precipitate challenging behaviour (Carr & Owen-DeSchryver, 2007; Carr, Smith, Giacin, Whelan, & Pancari, 2003; O'Reilly, 1997). For example, children with Autism Spectrum Disorder (ASD) are twice as likely to engage in SIB when health problems are present (Richards et al., 2012). Research on the temporal sequence of these

behaviours suggests that the occurrence of hypothesized non-verbal indicators of pain prior to SIB underlie pain and discomfort, whereas the occurrence of these behaviours following SIB is thought to be related to pain caused by the SIB itself (Eden, 2013). In studies such as these, the presence of pain is generally inferred from the presence of observable behavioural indicators of pain based on direct observation or informant-report. However, the presence of an underlying physical health condition or biological indicators of pain are not usually verified during these studies. As a result, it has been noted that the hypothesized behavioural indicators of pain may actually be observable signs of general distress (Courtemanche et al., 2012). Observational assessments such as the The Face, Legs, Activity, Cry, Consolability (FLACC) has been shown to be a valid measure of pain (Eden, 2013).

Along with behavioural indicators of pain, affect has been highlighted as a potential setting event. Lowry (1998) has suggested that when a person feels depressed they may find environmental events more aversive and be more likely to engage in challenging behaviour to escape. A study was conducted to assess whether or not there is a correlation between mood ratings and occurrences of SIB, the results of which demonstrated that mood ratings had predictive validity for problem behaviour (Carr, Magito McLaughlin, Giacobbe-Grieco, & Smith, 2003). Therefore, it appears important to include affect in any assessment of challenging behaviour.

Precursor behaviours should also be considered when assessing challenging behaviour. A precursor behaviour is a behaviour that reliably precedes an incident of challenging behaviour and have been used in adapted experimental functional analysis designs in cases where challenging behaviour is too severe to be elicited (Najdowski, Wallace, Ellsworth, MacAleese, & Cleveland, 2008). Najdowski et al. (2008) operationalised precursor behaviours as occurring immediately before the challenging behaviour. These

behaviours can also be identified for infrequent but severe challenging behaviour (Dracobly & Smith, 2012). Successful interventions using functional communication training (FCT) have also been implemented based on the variables identified that maintain the precursor behaviours (Najdowski et al., 2008). Other studies using descriptive assessment of precursor behaviours allowed for reactive interventions once the behaviour is displayed, pre-emptively avoiding the occurrence of SIB (Dracobly & Smith, 2012; Herscovitch, Roscoe, Libby, Bourret, & Ahearn, 2009). Results such as these suggest that the analysis of precursor behaviours to challenging behaviour may provide an alternative and indirect method to the assessment of function.

In addition to this, Beavers et al. (2013) suggested that clinically useful information such as severity and the duration of an episode of challenging behaviour should be recorded as this information can impact on decision making. Assessments should also be relatively quick to complete in the clinical setting and need as few measures as possible to create a summary statement of the behaviour in question (Newcomer & Lewis, 2004).

The "Challenging Behaviour Report Form" (CBRF) was developed to efficiently record details of incidents of challenging behaviour (Appendix 1; Snape, 2010). This item checklist provides a structure for recording incidents of challenging behaviour that provides a more detail than would typically be recorded from analogue conditions. It contains factors that occur during incidents of challenging behaviour alongside tick-boxes, in addition to a short personal details section.

The form was developed using video footage of functional analyses of challenging behaviour in 60 young children with rare genetic syndromes (Angelman Syndrome (AS), Cornelia de Lange syndrome (CDLS) & Cri du Chat syndrome (CDC). Once incidents of

challenging behaviour were identified from the video footage, as much information as possible about the incident and the 10 seconds preceding it was recorded. This included the topographies of challenging behaviour, behaviours performed by the participant immediately prior to challenging behaviour, behaviours performed by adults immediately prior to challenging behaviour, affect during the challenging behaviour, and behaviours accompanying the challenging behaviour were also recorded.

In conclusion, a more comprehensive assessment of challenging behaviour can be developed that combines environmental variables and internal events such as pain, into an efficient, simple and clinically useful assessment tool that is valid. Some researchers have proposed the use of functional assessments checklists that may be more acceptable in applied settings. The aim of this research is to further develop the CBRF to meet this need. The developed form will include variables to assess behavioural function, behavioural indicators of pain to assess its presence, behavioural indicators of affect to assess potential setting events, precursor behaviours that reliably predict challenging behaviour, and severity to help clinical decision making. It will also include the behaviours of those present after the occurrence of the challenging behaviour to assess the consequences for those behaviours. The form will be developed to be as efficient and user-friendly as possible for the observer and provide clinically relevant data. Once developed, the reliability of the form will be assessed using experimental functional analyses footage of challenging behaviour in non-verbal children with ASD. Functional analyses footage will be used as it provides discrete conditions in which challenging behaviour is expected to occur, increasing the feasibility of assessing reliability as there will be more instances of challenging behaviour.

Method

Setting

This study was part of a larger research project that aimed to describe and assess the causes of SIB in a cohort of 30 children with a severe ID and ASD and evaluate the effects of treatment of a health condition on suspected pain-related SIB. Data on self-injury, sleep disorder, self-restraint, health, pain and other related behavioural characteristics were collected. The present study used the functional assessment component of this data set only to further develop the CBRF.

Participants

Participants were referred to the research project by NHS professionals (community paediatricians, school nurses, a CAMHS LD team, special school staff from the West Midlands, the research centre participant database, and self-referral from the research centre's website. To be eligible for the study, all participants were non-verbal or had limited speech, and behavioural characteristics of ASD and SIB were present. They also had to be aged between 2 years, 0 months and 14 years, 11 months at the time of recruitment.

In total, 64 potential participants were referred. All potential participants were contacted with information about the research and asked for an expression of interest. Interested participants were screened either by telephone or by post/email. The screen took approximately 15 minutes to complete. Twenty-nine participants were recruited to the project. Of these participants, 21 (18 male, 3 female, mean age 8 years 10 months, SD=3.3) engaged in some form of challenging behaviour during the experimental analyses of their self-injury. The footage of these experimental functional analyses were analysed using the modified CBRF form, henceforth referred to as the CBRF-R. Challenging behaviour was

defined as engaging in behaviour that was self-injurious, aggressive to others or caused destruction to the environment.

Measures

Screening measures. Prior to inclusion in the study, participants completed several measures by phone, by post, or by email to assess eligibility for the study. Table 1 contains the measures used. A more comprehensive description is presented in Appendix 2.

Table 1: Screening for Eligibility in the Study

Screening measures	Purpose
Background Questionnaire	Demographic information
The Wessex	Measures ability on the Social and Incapacity and the Speech, Self-help and Literacy sub-scales
The Social Communication Questionnaire (SCQ)	Screening measure for ASD
Modified Checklist for Autism in Toddlers (MCHAT)	Screening measure for ASD in children aged under 30 months
The Challenging Behaviour Questionnaire (CBQ)	Assess topography, frequency, and severity of challenging behaviour

Table 2: Structure and contents of the CBRF-R

Section title	Purpose	Number of items	Items
1. Child's behaviour prior to challenging behaviour	Assess potential functional, precursor, and pain related behaviours of the child	39	1. Attempts/ accepts / resists eye contact initiates / accepts / resists verbal interaction 2. Initiates / accepts / resists physical contact engaged with / attempting to avoid or escape task /situation/sensory stimulus (e.g. light/noise) 3. Already in possession of/ attempts to access items / activities 4. Initiates/accepts/ resists transition (staff/materials/task/location)

				5. Waiting 6. Intelligible / unintelligible verbal / non-verbal communication 7. Movement that appears non-purposeful 8. Consistently holding particular part of body (own or other) / clothing / items 9. Pain related behaviours (leg tremors, gasping, shivering, grimacing, squirming, gagging, bruxism, guarding or rubbing body part, groan) 10. Other behaviours.
2.	Child's affect prior to challenging behaviour	Assess behaviours related to affect	5	1. Happy 2. Fearful 3. Sad 4. Angry 5. Neutral (mood)
3.	Child's vocalisation prior to challenging behaviour	Assess child's vocalisations	4	1. Laughing 2. Crying 3. Whining 4. Neutral (vocalisations)
4.	Other's behaviour prior to challenging behaviour	Assess potential functional behaviours of the other	24	1. Attempts/ terminates eye contact with child 2. Provides / terminates verbal interaction with child; provides / terminates physical contact with child 3. Looks at / talks to person other than target child 4. Provides / removes demands (verbal / physical) 5. Prevents / provides access to items/activity 6. Announces (verbally / non-verbally) onset of a transition (staff/activity/location) 7. Unsure of meaning of child's communication 8. Complies with/can't comply with child's requests 9. Provides/terminates physical restraint
5.	Challenging behaviour	Assess challenging behaviour topography and directional orientation of the child	10	1. Self-injury 2. Aggression 3. Destruction of environment 4. Facing/looking towards the other 5. Facing away from the other 6. Facing a third party 7. Topography 1

				8. Topography 2 9. Topography 3 10. Topography 4
6.	Child's affect during challenging behaviour	Assess behaviours related to affect	5	1. Happy 2. Fearful 3. Sad 4. Angry 5. Neutral (mood)
7.	Child's vocalisation during the challenging behaviour	Assess child's vocalisations	4	1. Laughing 2. Crying 3. Whining 4. Neutral (vocalisations)
8.	Child's behaviour after the challenging behaviour	Assess potential functional, precursor, and pain related behaviours of the child	39	1. Attempts/ accepts / resists eye contact initiates / accepts / resists verbal interaction 2. Initiates / accepts / resists physical contact engaged with / attempting to avoid or escape task /situation/sensory stimulus (e.g. light/noise) 3. Already in possession of/ attempts to access items / activities 4. Initiates/accepts/ resists transition (staff/materials/task/location) 5. Waiting 6. Intelligible / unintelligible verbal / non-verbal communication 7. Movement that appears non-purposeful 8. Consistently holding particular part of body (own or other) / clothing / items 9. Pain related behaviours (leg tremors, gasping, shivering, grimacing, squirming, gagging, bruxism, guarding or rubbing body part, groan) 10. Other behaviours.
9.	Child's affect after challenging behaviour	Assess behaviours related to affect	5	1. Happy 2. Fearful 3. Sad 4. Angry 5. Neutral (mood)
10.	Child's vocalisations after	Assess child's vocalisations	4	1. Laughing 2. Crying 3. Whining

challenging behaviour			4. Neutral (vocalisations)
11. Other's behaviour after challenging behaviour	Assess potential functional behaviours of the other	24	1. Attempts/ terminates eye contact with child 2. Provides / terminates verbal interaction with child; provides / terminates physical contact with child 3. Looks at / talks to person other than target child 4. Provides / removes demands (verbal / physical) 5. Prevents / provides access to items/activity 6. Announces (verbally / non-verbally) onset of a transition (staff/activity/location) 7. Unsure of meaning of child's communication 8. Complies with/can't comply with child's requests 9. Provides/terminates physical restraint
12. Severity of the challenging behaviour	Duration, effect on health, and carer concern	11	1. Duration <1 min 2. Duration <5 min 3. Duration ≥5 min 4. Effect on health none 5. Effect on health mild 6. Effect on health moderate 7. Effect on health severe 8. Carer concern none 9. Carer concern mild 10. Carer concern moderate 11. Carer concern severe

Table 3: Reliability Values

Kappa statistic	Strength of agreement	R	Strength of agreement
< 0.00	Poor	0.00-0.19	Very weak
0.00-0.20	Slight	0.20-0.39	Weak
0.21-0.40	Fair	0.40-0.59	Moderate
0.41-0.60	Moderate	0.60-0.79	Strong
0.61-0.80	Substantial	0.80-1.00	Very strong
0.81-1.00	Almost perfect		

Note: R=correlation coefficient

Eligibility. Participants were included in the study if they met the following criteria: severely limited speech or non-verbal based on parent/ carer responses to the background questionnaire and Wessex, score above 15 on the Social Communication Questionnaire for children older than four years old or two fails on critical items on the Modified Checklist for Autism in Toddlers for children younger than four years old, and reported the presence of any SIB over the last month in the Challenging Behaviour Questionnaire.

Functional Analysis

Behavioural measure. Participants who met criteria and accepted an invitation to take part in the study completed the Questions About Behavioral Function Questionnaire (Appendix 3; QABF; Paclawskyj et al., 2000), which is a 25 item questionnaire that is intended to give an indication as to the function of a particular target behaviour. There are five subscales of the QABF: attention, non-social, escape, physical and tangible. There are five items for each subscale. A 40-item ASD-specific version of the QABF was used (Richards & Oliver, 2012, unpublished). This version adds subscales such as social escape and routines and rituals that are associated with ASD.

Experimental functional analysis. The participants then progressed to the research day where the experimental functional analysis was conducted. Parents/carers chose the location of the experimental functional analysis which ranged between a laboratory setting at the university, the participant's home, and the participant's school.

Design

The CBRF-R was developed in the context of a larger study using experimental functional analyses. Single-case experimental designs were used. Participants experienced high attention, low attention, task demand, and access to tangibles in an ABACAD alternating treatment design. Additional analogue conditions were conducted where information from the informant based measures indicated their applicability.

In the standard functional analysis conditions, high attention, low attention and demand-escape were included. A tangible condition was included if SIB was identified in the pre-EFA assessments for access to tangibles.

The *high attention condition* was the control condition and provided a baseline for the other conditions where the participant had access to play materials, free access to verbal and physical attention and no demands were placed on them. There were no planned consequences for the behaviour. In the *low attention condition* social attention was provided contingent on the occurrence of challenging behaviour. In the *demand-escape condition* the requirement to complete a task was removed on the occurrence of challenging behaviour. In the *tangible condition* the participant had access to a toy/preferred item for two minutes prior to the condition beginning. The item was then removed and returned to the participant contingent upon the occurrence of challenging behaviour. Three sets of alternating treatments (ABACAD) were run for each participant. Each condition was 2.5 minutes long.

In addition to these standard experimental functional analysis conditions, four ASD specific conditions were used with some participants. They were used if certain 'high risk' conditions were identified at screening. In the *no interaction condition* the participant has access to items and was free to move around. The researcher was present but stayed away from the participant and did not offer any social contact and there were no planned consequences for challenging behaviour. During the *social-escape condition* the researcher provided high levels of verbal attention and stayed close to the participant. On the occurrence of challenging behaviour the researcher withdraws social attention and decreases physical proximity from the participant. In the *sensory-escape condition* the participant was exposed to a sensory stimulus (e.g. sound) that has been reported to elicit SIB through the pre-experimental functional analysis assessments. On the occurrence of SIB, the sensory stimulus was removed. In the *rituals and routines condition* the participant had access to a known ritual or routine. This was identified in the pre-experimental functional analysis assessment. The researcher then prevented access to the ritual. The participant was able to access it contingent on the occurrence of SIB. The ASD specific conditions were alternated three times using pairwise comparisons e.g. EFEFEF.

Finally, idiosyncratic conditions that were unique to each participant were identified through the QABF and screening measures. These conditions were designed based on information obtained prior to the experimental functional analysis.

Developing and evaluating the Challenging Behaviour Report Form-R

Further content development. The 'Challenging Behaviour Report Form' (CBRF; Snape, 2010) was developed to detail incidents of challenging behaviour along and events in the environment before and during the challenging behaviour with the aim of ascertaining function. It records the challenging behaviours observed, such as aggression,

self-injury, and destruction and the behaviours of the participant and carers immediately preceding the incident (e.g. child trying to leave the room, carer makes physical contact with the participant) and the affect and other behaviours accompanying challenging behaviour during the episode of challenging behaviour (e.g. vocalisations)

There is scope to develop the CBRF by including more information about the incidents of challenging behaviour. To accomplish this, initial discussions with clinicians and researchers with years of experience in conducting functional assessment of challenging behaviour were held and research literature on the underlying causes of challenging behaviour were taken into consideration. Through this process more items for the form were generated and refined. It was decided that items pertaining to behavioural function, indicators of pain, indicators of affect, precursor behaviours, and measures of severity were to be included. A section recording the behaviour of the individuals involved after the occurrence of challenging behaviour was also included. Following these discussions, a draft form was created (see Appendix 4). A workshop was organised for researchers and clinicians to trial the use of the form. The workshop consisted of a practical session in which two post-doctoral researchers, the author, and three research assistants watched functional analysis footage while completing the form. From this workshop the final version of the form was developed which was edited to be more intuitive (see Appendix 5). An associated protocol was developed that describes the use of the form in detail and defines the items on the form (see Appendix 6).

The final modified CBRF-R comprised of 12 sections which are summarised in Table 2. The 'child' is defined as the individual that is being observed. The 'other' is defined as the individual who is interacting/ in proximity to the child.

Full descriptions of each of these items can be found in the protocol in Appendix 8.

Additional content. Clinically relevant variables such as challenging behaviour severity, and additional precursor behaviours were introduced to the form. The severity items were adapted from the Challenging Behaviour Interview (Chris Oliver et al., 2003), which was developed to assess the severity of challenging behaviour. In addition to this, items related to the behavioural markers of pain were also included from the Face, Legs, Activity, Cry, Consolability (FLACC; Appendix 7; Malviya, Voepel-Lewis, Burke, Merkel, & Tait, 2006). Affect was altered from the original version. A section on the participant's and adult's behaviour after the challenging behaviour has occurred was also added.

Organisation of items. Many of the items have multiple components contained within. These items are related to each other and provide more information. For example, "attempts to get items/in possession" refers to a situation where the child is trying to access an item or is already in possession of one. To mark this item, an observer would tick/number the box to the left of the description and circle the corresponding description. In this example, if the child was trying to access an item the observer would tick the item's box and circle the words "to get items." Having multiple items like this helps to condense the form, allowing it to be completed with greater ease. An example of this can be seen in Figure 1. Some items were coloured in an attempt to help observers differentiate between items more readily. In total, there are 174 variables on the CBRF-R. The 'prior' and 'after' sections are identical, as this allows easier completion of the form.

Figure 1. Example of CBRF-R items with Multiple Components

☐ Attempts/ accepts / resists eye contact

☐ Initiates / accepts / resists verbal interaction

☐ Initiates / accepts / resists physical contact

Instructions of use. Each CBRF-R records one episode of challenging behaviour. An episode of challenging behaviour is defined as beginning 15 seconds before the target behaviour occurs and ends once there has been no occurrence of the target behaviour for 10 seconds. The time period of 15 seconds prior was based on the original CBRF protocol which stated 10 seconds. The time was increased to 15 seconds on discussion with researchers and clinicians to maximise the amount of information recorded while still maintaining feasibility. The time of the end of an episode, which is 10 seconds after the last occurrence of challenging behaviour, was chosen as that is the period of time that reinforcement is delivered during the experimental functional analysis conditions.

Observer training. Following the modification of the CBRF-R, the author completed the form using experimental functional analysis footage from a previous research project on challenging behaviour with individuals with Cri du Chat, Angelman syndrome, and Cornelia de Lange syndrome for training purposes. Observer 1 was a doctoral student who has some experience coding incidents of challenging behaviour and several years' experience working with individuals who engage in challenging behaviour. Observer 2 was a post graduate student who has some experience of coding challenging behaviour via observing videos of analogue conditions. Footage from 10 participants was used. This footage was chosen as the nature of the experimental functional analysis and the population were similar to that of the current study. Observer 2 was provided with a copy of the CBRF-R and the CBRF-R

protocol. A practice session was held in which both observer 1 and 2 studied the CBRF-R and practiced it on some of footage that was not already coded by the author. Observer 2 then completed 33 per cent of the same clips that observer 1 had completed. Inter-rater reliability for the training session was found to be 60% agreement between observers. This was deemed acceptable and items that were not agreed upon were evaluated and changes were made to the form and the protocol accordingly. After these changes, a new reliability criterion of 70% agreement was set for observer 2 on a further ten forms. Following this further training, observer 2 reached the reliability criterion and coding of the newly collected experimental analysis footage commenced.

Coding. The form was designed to be of use in a clinical setting and to provide clinically useful information. The time taken to complete each form was recorded to evaluate how long it would take someone to complete the CBRF-R as it was required to be clinically useable.

Assessment of inter-observer agreement

Observer 1 and 2 were separately shown video clips of incidents of challenging behaviour from the experimental functional analysis and completed a copy of the CBRF-R for each clip. There was a section on the top of the form for identification purposes so that each form could be matched for reliability testing. Both observers were instructed to tick any box that described what was happening in the footage and circle any corresponding descriptions. Observer 1 completed the CBRF-R for each participant, up to a maximum of 10 episodes for each participant. Observer 2 then completed the CBRF-R for 25% of these clips. The clips were chosen by allocating a number to each clip and using a random number generator. The corresponding clips were given to observer 2 to watch and code.

Inter-observer agreement was established using Cohen's Kappa and Kendall's Tau b. Both observers were compared for each clip of footage on: items that both observers report as having happened, items that observer 1 but not observer 2 reported as having occurred in the clip, items that observer 2 but not observer 1 reported as having occurred in the clip, and items that neither observer report as having occurred. From this information a Kappa value of between 0.00 and 1.00 was calculated for each item to measure inter-observer agreement. According to Landis & Koch (1977), the labels in Table 3 may be assigned to the corresponding ranges of Kappa. There were several variables that had cumulative scores greater than 1. This was because when the variables on the form were collapsed to reduce the number of variables in the reliability analysis, multiple variables within a category were sometimes scored. As such, Kendall's Tau b was seen as a more appropriate statistical test for these variables to assess inter-observer agreement. A correlation coefficient (r) between -1.00 and 1.00 was provided. Evans (1996) suggests that the labels in Table 3 can be given to the r values.

Scoring protocol

In order to reduce the number of variables to conduct inter-observer reliability on, the 174 variables on the CBRF-R were collapsed into 69 categories. These categories were decided on after the form had been finalised through discussion between researchers and clinicians. For example, the "attention prior" category, which refers to items that may suggest the child is engaging in attention maintained behaviours, contains the items: attempts eye contact, accepts eye contact, initiates verbal interaction, accepts verbal interaction, initiates physical contact, accepts physical contact, initiates transition staff, accepts transition staff, terminates eye contact, terminates verbal interaction, terminates physical contact, removes demands (verbal/physical). Each category had a subscale score range. For example, the

"attention prior" category had a range of scores from 0-7. The full list of the categories used and the CBRF-R items that they contain are presented in Appendix G.

Scoring considerations. The decision was made to give some variables/categories priority over others. The process of deciding to give precedent to some items over others was theoretical rather than methodological and was done via discussion with researchers and clinicians skilled in the area. This was done to minimise the impact of one observer being over inclusive in the items they recorded. For example, items in the "task escape other" category were given priority over "attention other." This was due to an issue during the observer training phase where variables that were in the "attention other" were recorded as well as the "task escape other." While all items were correctly selected, such as "removes demands (physical) and "terminates physical contact with the child," if one of the observers selected both items, and one recorded "removes demands (physical), it reduced inter-observer agreement despite both observers recording the same behaviours.

Results

Number of forms completed and time taken

Footage from a total of 29 functional analyses was watched. From this footage, 21 participants were identified as engaging in challenging behaviour. A total of 125 CBRF-R forms were completed for the 21 participants who engaged in challenging behaviour. The average number of forms completed per participant was 6.3 (min=1, max=10). It took approximately 3 minutes to complete each form. Observer 2 completed 31 (25%) CBRF-R forms.

Reliability

In this section, the variables are presented in categories that summarise related data together. Variables presented as percentage agreement are presented this way due to it not being possible to calculate Kappa or Kendall's Tau b. This was because these variables were scored entirely as being present or not present by at least one observer, and neither Kappa nor Kendall's Tau b could be calculated. As such, they are presented as percentage agreement. Table 4 presents the reliability scores for causal variables which consist of items associated with behavioural function and pain. Table 5 contains the reliability scores for communication, precursor behaviours, and affect. Table 6 presents the reliability of the items related to the severity of the challenging behaviour. Table 7 contains the reliability scores for the challenging behaviour topography. Finally, table 8 contains the reliability scores for the restraint related behaviours. The range of possible scores is also presented in Tables 4, 5, 6, 7, and 8.

Table 4: Causal Variables

Category title	Before/after challenging behaviour	Kappa/R	Range	Interpretation
Behavioural function				
Child: Behavioural indicators of attention maintained behaviour	Before	0.609[b]	0-7	Strong
	After	0.619[b]	0-7	Strong
Child: Behavioural indicators of task escape maintained	Before	0.611[b]	0-3	Strong
	After	0.674[b]	0-3	Strong
Child: Behavioural indicators of sensory escape maintained behaviour	Before	0.667[a]	0-1	Substantial
	After	0.911[a]	0-1	Almost perfect
Child: Behavioural indicators of access to sensory stimulation maintained behaviour	Before	-0.053[a], *ns*	0-1	Poor
	After	0[a]	0-1	Poor
Child: Behavioural indicators of escape from social contact maintained behaviour	Before	0.353[b]	0-4	Weak
	After	0.554[a]	0-4	Moderate
Child: Behavioural indicators of access to self-stimulation maintained behaviour	Before	0.444[a]	0-1	Moderate
	After	0.474[a]	0-1	Moderate
Child: Behavioural indicators of access tangible items maintained behaviour	Before	0.713[b]	0-5	Strong
	After	0.429[b]	0-5	Moderate
Other: Behavioural indicators of social escape maintained behaviours	Before	0.66[b]	0-4	Strong
	After	0.867[b]	0-4	Very strong
Other: Behavioural indicators of attention maintained behaviours	Before	0.865[b]	0-4	Very strong
	After	0.845[b]	0-4	Very strong
Other: Behavioural indicators of task escape maintained behaviours	Before	0.763[b]	0-2	Strong
	After	0.896[b]	0-2	Very strong
Other: Behavioural indicators of access to tangible items maintained behaviours	Before	0.87[a]	0-1	Almost perfect
	After	1[a]	0-1	Almost perfect
Other: Difficulty in communication	Before	1[a]	0-2	Almost perfect
	After	100%	0-2	
Other: Miscellaneous behaviours	Before	1[a]	0-2	Almost perfect
	After	0.634[a]	0-2	Substantial

Pain	Before	0.902[a]	0-9	Almost perfect
	After	0.714[a]	0-9	Substantial

Note: [a]=Kappa, [b]=Tau b. All reliability scores are significant at the p<.05 level unless noted ns=not statistically significant.

Table 5: Communication, Precursor Behaviours, Affect

Category title	Before/during/after challenging behaviour	Kappa/R	Range	Interpretation
Communication				
Child: Communicative behaviours before occurrence of challenging behaviour	Before	0.57[b]	0-3	Moderate
	After	0.72[b]	0-3	Strong
Precursor behaviours				
Child: Other precursor behaviours	Before	0.634[a]	0-2	Substantial
	After	0.609[a]	0-2	Moderate
Affect				
Child: Behavioural indicators of positive affect	Before	0.318[a]	0-2	Fair
	During	0[a]	0-2	Poor
	After	0.783[a]	0-2	Substantial
Child: Behavioural indicators of negative affect (sad)	Before	0.835[b]	0-2	Very strong
	During	0.575[b]	0-2	Moderate
	After	0.838[b]	0-2	Very strong
Child: Behavioural indicators of negative affect (fear)	Before	0.25[a]	0-2	Fair
	During	0.254[a]	0-2	Fair
	After	0.063[a], *ns*	0-2	Poor
Child: Behavioural indicators of negative affect (anger)	Before	0.242[a]	0-1	Fair
	During	0.34[a]	0-1	Fair
	After	0.634[a]	0-1	Substantial
Child: Behavioural indicators of neutral affect	Before	0.772[b]	0-2	Strong
	During	0.6[a]	0-2	Strong
	After	0.762[b]	0-2	Strong

Note: [a]=Kappa, [b]=Tau b. All reliability scores are significant at the p<.05 level unless noted ns=not statistically significant.

Table 6: Clinical Severity

Category title		Percentage agreement	Range
Duration	Duration of challenging behaviour less than a minute	100%	0-1
	Duration of challenging behaviour less than 5 minutes, greater than a minute	100%	0-1
	Duration of challenging behaviour equal to or greater than 5 minutes	100%	0-1
Severity	None	93.8%	0-1
	Mild	6.3%	0-1
	Moderate	100%	0-1
	Severe	100%	0-1
Carer concern	None	93.8%	0-1
	Mild	6.3%	0-1
	Moderate	100%	0-1
	Severe	100%	0-1

Note: [a]=Kappa, [b]=Tau b. All reliability scores are significant at the p<.05 level unless noted ns=not statistically significant.

Table 7: Challenging Behaviour Topography

Category title	Percentage/ Kappa	Range	Interpretation
Self-injury	1	0-1	Almost perfect
Aggression	0	0-1	Poor
Destruction	100%	0-1	
Child facing towards the other	0.412	0-1	Moderate
Child facing away from the other	0.545	0-1	Moderate
Child facing 3rd party	0	0-1	Poor
Topography 1 of challenging behaviour	1	0-1	Almost perfect
Topography 2 of challenging behaviour	0.783	0-1	Substantial
Topography 3 of challenging behaviour	1	0-1	Almost perfect
Topography 4 of challenging behaviour	1	0-1	Almost perfect

Note: All reliability scores are significant at the p<.05 level unless noted ns=not statistically significant.

Table 8: Restraint

Category	Before/after challenging behaviour	Kappa/ percentage of agreement	Range	Interpretation
Child: Self restraint	Before	0.173[a], *ns*	0-1	Poor, *ns*
	After	0	0-1	Poor
Other: applies restraint	Before	0	0-1	Poor
	After	100%	0-1	
Other: removes restraint	Before	0	0-1	Poor
	After	0.034[a], *ns*	0-1	Poor

Note: [a]=Kappa. All reliability scores are significant at the p<.05 level unless noted ns=not statistically significant.

Regarding causal variables, the variables related to behavioural function had reliability ranging from poor to perfect. Two variables fared poorly, five had moderate reliability, and the rest had strong or better reliability. The pain related variables had almost perfect reliability before the challenging behaviour and substantial reliability after. The communication variables had moderate and strong reliability. Precursor behaviours had substantial agreement before and after challenging behaviour. Regarding the child's affect, there was strong or higher reliability for seven variables, moderate agreement for one, and fair agreement for five variables. Two had poor agreement between observers. The clinical severity variables are expressed as percentage agreement as at least one observer had constant scores for a variable, which did not make the use of Cohen's Kappa or Kendall's Tau b possible. Overall, very strong agreement was recorded. Regarding challenging behaviour, the majority of variables scored moderate or high, with only two variables scoring poorly. Finally, variables related to restraint had overall poor reliability.

Discussion

The purpose of this study was to develop a close-ended report form with which incidents of challenging behaviour in individuals with intellectual disabilities could be recorded in detail. This version was developed to include behaviours after the incident of challenging behaviour and other clinically relevant variables such as challenging behaviour intensity, severity, duration, emotional impact on others and additional precursor behaviours were introduced to the form. In addition to this, items related to the behavioural markers of pain were also included and affect was altered from the original version. The original version of the CBRF (Snape, 2010) provides a more structured method of recording incidents of challenging behaviour than traditional ABC recording and a richer source of information than experimental functional analysis (Carr & Durand, 1985; Iwata, Dorsey, et al., 1994). The version developed in this study goes further again and adds more variables in an easy to use format that gives clinicians a fast way of getting a sense of what is happening during an incident of challenging behaviour, as the form takes on average 3 minutes to complete. This may be an effective and efficient way for clinicians to obtain a comprehensive overview of an incident of challenging behaviour, especially in the natural environment, although it has not been tested on naturalistic observations yet.

While the majority of items had fair to strong inter observer agreement, there were ten categories that had poor reliability. They were "Child: Self-restraint" before and after, "Other: applies restraint" before, "Other: removes restraint" before and after, "Aggression", "Child: Behavioural indicators of positive affect" during, "Child: Behavioural indicators of negative affect (fear)" after and "Child: Behavioural indicators of access to sensory stimulation maintained behaviour" before and after.

There are several possibilities for these results. Observers may have deviated from the provided definitions. This is known as "observer drift" (O'Leary & Kent, 1973). Attempts were made to minimise this by observer training, a protocol containing operational definitions, and combining items into overarching categories. However, some of the items that had poor reliability were in categories on their own or had two items and may have been easily missed. Also, some of the behaviours were infrequent within the footage. For example, there were few instances of restraint, either application or removal, within the footage. It is possible that "events occurring at a low frequency will be less likely to be detected as vigilance will be lower if motor patterns are infrequent" (Caro, Roper, Young, & Dank, 1979). Finally, observer fatigue (Caro et al., 1979) may have played a role, where long periods of observation are tiring and may have affect vigilance over time. While an attempt was made to minimise this by suggesting frequent breaks while coding, fatigue will vary between observers. However, this may not have been a major factor as the majority of items had fair to strong inter-observer agreement.

There are several limitations of this study. The CBRF-R was used entirely on experimental functional analysis footage that included participants that were selected based solely on self-injurious behaviour. This means that self-injurious behaviour was perhaps over represented and other forms of challenging behaviour underrepresented. This may have affected reliability for items such as "Aggression." As such, the CBRF-R may not be as reliable if used on other analogue footage. Also, the CBRF-R was not tested on naturally occurring incidents of challenging behaviour; instead it was used on incidents of challenging behaviour that occurred during analogue conditions.

Regarding future research, these items with poor reliability need to be revisited on the form, in the protocol, and in observer training to improve the accuracy and reliability of the

overall form. This could be achieved by creating a more comprehensive training package where visual footage containing an example of each item is watched by potential observers. This could potentially aid in the identification of less frequent behaviours. Also, reliability testing could be carried out on a range of challenging behaviours from experimental functional analysis footage and subsequently on non-structured naturalistic observations.

A scoring algorithm could be developed from the CBRF-R results to ascertain the possible function of challenging behaviour (based on 10 completed forms) and compared to other forms of functional assessment, or indeed the results of the experimental functional analysis itself to assess construct validity.

The CBRF-R described here is the second version of this report form and was an attempt to include more variables while keeping the form usable. The items included in it should not be considered to be a definitive list of every behaviour that may coincide with challenging behaviour.

A limitation of using closed-ended forms is that it is possible that the user may want to record a behaviour that is not contained within the form. However, the benefit of using a closed-ended report form is that it guides the users to use correct, defined choices free from presupposition in a structured manner, making it easier for the reader to ascertain what was going on during the incident. As the form goes through further development new behaviours may be added and refined. It will be important to find a balance between the form being as inclusive and comprehensive as possible and being concise enough to be quickly and easily completed.

Despite these limitations, there are several clinical and ethical implications that arise from this. Experimental functional analysis requires excessive amounts of resources, but in

completing the assessment, staff training, and data analysis (Hastings & Noone, 2005). Hastings and Noone (2005) also noted issues with the clarity and validity of results. A less labour intensive (Matson & Minshawi, 2007) and swifter (Hastings & Noone, 2005) assessment, such as the CBRF-R, could potentially deliver reliable and valid data on the function of challenging behaviour while circumventing the ethical dilemma of placing and individual in a condition where they are expected to engage in challenging behaviour Hastings & Noone, 2005). This could potentially mean that resources that would normally be used on experimental functional analysis could be used elsewhere and increase its social validity.

In conclusion, the CBRF-R is a close-ended report form which provides an efficient and simple method for recording incidents of challenging behaviour. It has been shown to reliably measure many factors that occur in incidents of challenging behaviour. Eventually, it is hoped that it will reliably determine the function of challenging behaviour in individuals with intellectual disability and ASD and provide clinically useful information for the clinician. The CBRF-R may offer a less demanding and more time efficient method of ascertaining function of challenging behaviour than the use of traditional experimental analysis.

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PUBLIC DOMAIN BRIEFING PAPER

CHALLENGING BEHAVIOUR ASSESSMENT IN INDIVIDUALS WITH INTELLECTUAL DISABILITIES AND AUTISM SPECTRUM DISORDER

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SYSTEMATIC REVIEW

GASTROINTESTINAL DISEASE AND BEHAVIOURAL CHANGE: IS THERE AN ASSOCIATION IN INDIVIDUALS WITH INTELLECTUAL DISABILITIES AND AUTISM SPECTRUM DISORDER

Introduction

Individuals with intellectual disorders (ID) and autism spectrum disorder (ASD) experience heightened rates of physical health conditions and are also more likely to display challenging behaviour (Cervantes & Matson, 2015; Kohane et al., 2012, Richards, Oliver, Nelson, & Moss, 2012). In addition to challenging behaviour, there are a number of clinically relevant difficulties that are associated with individuals with ID/ASD, among the most commonly identified areas are sleep disorders, anxiety and depression (van de Wouw et al., 2012; Mannion, Leader, & Healy, 2013; Leyfer et al., 2006). The aim of this systematic review is to evaluate if the literature supports the hypothesis that pain as a result of gastrointestinal disorder (GI) is associated with changes in behaviour, specifically challenging behaviour and sleep problems, in individuals with ID and or ASD.

Method

A systematic search for studies containing behavioural, developmental disability, and gastrointestinal disorder terms was conducted using several databases. These were Ovid Medline, PsychINFO, and Ovid Embase. Several included papers were also handpicked from an earlier review.

Results

From the search strategy, a total of fourteen papers were included in the review. The review focussed on two main areas; the methodological issues regarding research into this area and evidence for the association between GI disorder and challenging behaviour, disordered sleep, and behaviours related to anxiety and depression. Various behavioural topographies, gastrointestinal disorders, and assessment methodologies were reported. Quality was assessed using an adapted quality index for randomised and non-randomised trials (Downs & Black, 1998).

Conclusions

The majority of papers used medical notes and assessments by medically trained professionals while the remaining used parental report. There is inconsistency in how behaviour topography of challenging behaviour and other behaviours were reported across the studies.

Regarding the association between GI disorder and behaviour, there was equivocal evidence for and against an association between self-injurious behaviour and GI disorder. There appears to be an association between some behaviour, such as aggression and stereotyped/hyperactive behaviour, and GI disorder, in that these behaviours are more likely to occur in individuals experiencing GI disorder. There is some evidence that GI disorder may be associated with depression and/or anxiety in this population but the behavioural markers used to establish a diagnosis may be potentially confounded by the symptoms also being associated with pain. Finally, there is limited evidence for the association of sleep problems and GI disease from the papers in this review.

It is recommended that more intervention studies are conducted on this population. Research should use operational definitions of potential behavioural indicators of GI disorder to increase replicability and to increase the clinical implications of having a potential way to screen for GI disorder in non-verbal individuals. Similarly, how GI disorder is diagnosed/assessed in these studies should be more uniform and if possible, rigorous.

EMPIRICAL PAPER:

THE DEVELOPMENT OF A CHALLENGING BEHAVIOUR REPORT FORM

Introduction

Challenging behaviour is shown by between 10% and 15% of people with intellectual disabilities (ID; Emerson, 2001). The term includes behaviours such as self-injury (SIB) aggression and destruction of the environment. The high prevalence of challenging behaviour in this population and its significant, negative consequences warrant early, effective intervention. A valid and reliable assessment is required to understand the underlying causes of challenging behaviour effectively intervene.

There is evidence that undiagnosed health conditions that cause pain in non-verbal individuals with ID may precipitate challenging behaviour (Carr & Owen-DeSchryver, 2007; Carr, Smith, et al., 2003; O'Reilly, 1997). Research suggests that the occurrence of hypothesized non-verbal indicators of pain prior to SIB underlie pain and discomfort, whereas the occurrence of these behaviours following SIB is thought to be related to pain caused by the SIB itself (Eden, 2013). Along with behavioural indicators of pain, affect has been highlighted as a potential setting event and their assessment may provide an alternative and indirect method to the assessment of function. Beavers et al. (2013) suggested that clinically useful information such as severity and the duration of an episode of challenging behaviour should be recorded as this information can impact on decision making.

Aim

To develop the Challenging Behaviour Report Form (CBRF-R) to include variables to assess behavioural function, behavioural indicators of pain to assess its presence, behavioural indicators of affect to assess potential setting events, precursor behaviours that reliably predict

challenging behaviour, and severity to help clinical decision making. It also includes the behaviours of those present after the occurrence of the challenging behaviour to assess the consequences for those behaviours. To assess the reliability of the form using experimental functional analyses footage of challenging behaviour in non-verbal children with Autism Spectrum Disorder (ASD).

Method

The CBRF-R was developed from an existing assessment. Through a process of discussion with other clinicians and researchers an assessment form was developed to include pain related behaviours, affect, precursor behaviours, and severity of the challenging behaviour. Once completed, the reliability of the CBRF-R was assessed on experimental functional analysis footage of children and adolescents with ASD and challenging behaviour.

Results

Footage from a total of 29 experimental functional analyses identified 21 participants who engaged in challenging behaviour. A total of 125 CBRF-R forms by Observer 1. Observer 2 completed 31 (25%) CBRF-R forms for the purpose of reliability. The majority of variables included in the form had fair to strong inter observer agreement while ten had poor reliability.

Conclusion

The CBRF-R provides an efficient and simple method for recording incidents of challenging behaviour. It has been shown to reliably measure many factors that occur in incidents of challenging behaviour. Future development of the form is CBRF-R is possible by applying it to natural observations and assessing its validity by comparing its results to more established methods such as experimental functional analyses.

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APPENDICES