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Physical conditions and challenging behaviour in people with intellectual disability: a systematic review

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Abstract

Background Challenging behaviour is a major problem among people with intellectual disabilities. Physical factors may be an important cause. The aim of the present systematic review was to determine the physical conditions associated with challenging behaviour.

Methods A literature search was conducted in PubMed and the Cochrane systematic review database for empirical studies published between 1990 and 2008. The quality of all the studies that met the inclusion criteria was assessed using the SIGN-50 methodology checklists.

Results The search identified 45 studies, which looked at general medical conditions, motor impairment, epilepsy, sensory impairment, gastrointestinal disease, sleep disorders, dementia and others. There were four high-quality observational studies, seven well-conducted observational studies, 21 observational studies of low methodological quality and 13 non-analytical studies. There were significant and independent associations between challenging behaviours and urinary incontinence, pain related to cerebral palsy and chronic sleep problems, and between self-injurious behaviour and visual impairment. No association was found with hearing impairment, bowel incontinence, mobility impairment or epilepsy. Many other physical conditions were not addressed at all.

Conclusion Medical conditions can play a role in challenging behaviour, and this should be evaluated in the clinical setting. So far, the level of evidence is generally low, and longitudinal studies are completely lacking. We recommend a systematic approach to research examining the role of physical conditions in challenging behaviour, the ultimate aim being to establish a basis for the development of clinical guidelines.

Keywords aggression, challenging behaviour, intellectual disability, medical conditions, self-injurious behaviour, systematic review

Introduction

Challenging behaviour is a major problem in people with intellectual disabilities (ID). The estimated prevalence of these severe behaviour problems in large population studies is between 10% and 15% (Harris 1993; Sigafoos *et al.* 1994; Emerson *et al.* 2001a; Holden & Gitlesen 2006; Jones *et al.* 2008).

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The most common forms of challenging behaviour are aggression (7%), destructive behaviour (4–5%) and self-injury (4%; Emerson *et al.* 2001a). Furthermore, the most demanding forms of these behaviours tend to persist throughout life (Emerson *et al.* 2001b; Murphy *et al.* 2005; Totsika *et al.* 2008). Challenging behaviours can have serious disadvantages for the children and adults in question, such as interference with development, quality of life, social participation (Holden & Gitlesen 2006), physical injury and even death (Noel & Clarke 1982; Jan *et al.* 1994; Carlock *et al.* 1997; Nissen & Haveman 1997). People displaying aberrant behaviours also tend to be overmedicated (Matson *et al.* 2000; Stolker *et al.* 2008). Family and staff can find it very stressful and emotionally difficult to deal with people with challenging behaviour (Hastings & Brown 2002).

The causal and maintaining mechanisms underlying challenging behaviour are multifactorial (Applegate *et al.* 1999; Schroeder *et al.* 2001; McClintock *et al.* 2003; Matson & Boisjoli 2007; Tenneij & Koot 2008). Although functional analysis and treatment have focused mainly on behavioural aspects (Hassiotis & Hall 2008), physical factors also play an important role in different forms of challenging behaviour (self-injury, aggression, stereotypes, pica and rumination) (Applegate *et al.* 1999; Matson & Boisjoli 2007). Bosch *et al.* (1997) performed an exploratory study in people with self-injurious behaviour (SIB) and ID. In 28% of the patients, they found previously undiagnosed medical conditions that could be expected to cause pain or discomfort. Conditions that may cause pain (and may go unnoticed) are inflammatory or infectious diseases, motor impairment, pulmonary or cardiac disease, gastrointestinal conditions, malignancies, lacerations or fractures, headache, ear, nose, throat or eye diseases (Bosch *et al.* 1997; Ryan & Sunada 1997; van Schrojenstein Lantman-de Valk & Walsh 2008). People with ID and SIB have intact pain sensation (Symons *et al.* 2009) and SIB can target the site of the pain (Breau *et al.* 2003). Pain may very well be a cause of SIB (Symons *et al.* 2009; Oliver & Richards 2010), and one can imagine that this is also the case for other types of challenging behaviour, especially if people are unable to express their complaints otherwise (Kastner *et al.* 2001).

It has been proposed that challenging behaviours should be assessed on multidisciplinary lines and that medical conditions should be taken into account (Loschen & Osman 1992; Kwok & Cheung 2007; The National Association for the Dually Diagnosed 2007). The aim of this systematic review is to determine which physical conditions are associated with challenging behaviour.

Methods

Selection criteria for studies covered by this review

Types of studies

We considered relevant empirical/observational studies published between January 1990 and July 2008 with a minimum sample size of five participants. The papers were written in English, Dutch or German.

Types of participants

Participants were children and adults with ID. All levels of ID were included.

Types of exposure

All physical medical conditions were included. Medication side effects and substance abuse were excluded. We excluded studies of specific syndrome phenotypes, although syndromes may include both physical conditions and specific behavioural problems. Behavioural problems are widely considered to be related to the syndrome (and psychiatric profile) rather than to physical co-morbidity, and possible causal relationships are not studied. We do not intend to describe behavioural phenotypes.

Types of outcome

We defined challenging behaviour using the description of problem behaviours in 'Diagnostic criteria for psychiatric disorders for use with adults with learning disabilities (DC-LD)', axis III, level D: general diagnostic criteria for problem behaviours, verbally aggressive behaviour, physically aggressive behaviour, destructive behaviour, self-injurious behaviour, sexually inappropriate behaviour, oppositional behaviour, demanding behaviour, wandering

behaviour, mixed problem behaviour, other problem behaviours and mixed other problem behaviours. We also included problematic feeding disorders: food rumination/regurgitation disorder and pica, DC-LD axis III, level B (Royal College of Psychiatrists 2001).

Search methods for identification of studies

Electronic searches

We searched Medline/PubMed and the *Cochrane Database of Systematic Reviews*. Research terms were selected to search very broadly on ID, all forms of problem behaviours and all possible medical conditions that might be associated with behavioural problems (based on known literature and clinical practice). Terms were searched for as MeSH terms when indicated, and otherwise they were searched for in all fields (title, abstract, keywords).

In a first search, research terms for intellectual disability were combined with terms for problem behaviour. In a second search, terms for intellectual disability were combined with terms for medical conditions and 'behavior'.

Terms for intellectual disability were: 'Mental Retardation [MeSH]' OR 'Intellectual Disability' OR 'Developmental Disability' OR 'Mental Handicap' OR 'Multiple Handicap' OR 'Intellectual Impairment' OR 'Communicative Impairment' OR 'Learning Disability' OR 'Neurodevelopmental Disability' OR 'Cognitive Impairment'.

Terms for problem behaviour were: 'Aggression [MeSH]' OR 'Self-Injurious Behavior [MeSH]' OR 'Feeding Behavior [MeSH]' OR 'Sexual Behavior [MeSH]' OR 'Stereotyped Behavior [MeSH])' OR 'Challenging Behavior' OR 'Rumination' OR 'Pica'.

Terms for physical conditions were: 'Pain [MeSH]' OR 'Epilepsy [MeSH]' OR 'Menopause [MeSH]' OR 'Menstrual Cycle [MeSH]' OR 'Vision Disorders [MeSH]' OR 'Hearing Disorders [MeSH]' OR 'Otitis Media [MeSH]' OR 'Bacterial Infections and Mycoses [MeSH]' OR 'Parasitic Diseases [MeSH]' OR 'Virus Diseases [MeSH]' OR 'Dementia [MeSH]' OR 'Cardiovascular Diseases [MeSH]' OR 'Digestive System Diseases [MeSH]' OR 'Disorders of Environmental Origin [MeSH]' OR 'Female Urogenital Diseases [MeSH]' OR 'Hemic and Lymphatic Diseases [MeSH]' OR 'Immune System Diseases [MeSH]' OR 'Male Urogenital Diseases [MeSH]' OR 'Musculoskeletal Diseases [MeSH]' OR 'Neoplasms [MeSH]' OR 'Nervous System Diseases [MeSH]' OR 'Nutritional and Metabolic Diseases [MeSH]' OR 'Eye Diseases [MeSH]' OR 'Otorhinolaryngologic Diseases [MeSH]' OR 'Pathological Conditions, Signs and Symptoms [MeSH]' OR 'Respiratory Tract Diseases [MeSH]' OR 'Skin and Connective Tissue Diseases [MeSH]' OR 'Stomatognathic Diseases-[MeSH]'.

Searching other resources

We scrutinised the bibliographies of articles that seemed relevant.

Data collection and analysis

Selection of studies

One reviewer (C. F. dW.) screened the titles and abstracts from the search. The full texts of papers that appeared relevant were retrieved. Four articles were available as epub ahead of print at the time of the search. The actual publication dates were outside our inclusion period, but they were included in this review. A total of 45 articles fulfilled our inclusion criteria and they were included in the present review.

Data extraction and management

Two reviewers (C. F. dW. and H. M. E.) then analysed the study characteristics and methodological quality/level of evidence of the articles independently. Where there were gaps in the available information, we attempted to contact the authors. When the reviewers judged the articles differently, they discussed the results in order to reach agreement. A third reviewer was on hand to make a final decision if agreement could not be reached. This was not necessary as full agreement was reached about the study characteristics and the level of evidence of all articles.

A note was made about the following study characteristics for all articles:

- Study design.
- Participants: number of participants, age of the participants, sex, level of ID and study population

Table 1 Reliability of method of exposure and outcome assessment

*	No evidence: method of assessment not described, or unstructured staff reports or file data only
**	Checklist or specific assessment used, but moderate validity
***	Evidence from other sources is used to demonstrate that the method of assessment is reliable and valid

(e.g. representative for the general ID population or referred patients).

• Exposure: the medical conditions discussed, evaluation method and reliability (Table 1).

• Outcome: kind of problem behaviour, and assessment of behaviour and reliability (Table 1).

• Results of the study and relevant conclusion for the present review.

Assessment of the level of evidence

At present, there is no consensus about a tool for the assessment of quality in observational studies (Sanderson *et al.* 2007). For case–control studies and for cohort studies (SIGN 2008), we used the SIGN-50 methodology checklists as they are designed for observational studies and comprise most of the items indicated by the STROBE statement for the publication of observational studies (von Elm *et al.* 2007). Although SIGN-50 was originally designed as a basis for guideline development, we used it as a quality tool for the present systematic review. We used the SIGN methodology checklist for cohort studies and adapted this for cross-sectional studies. The SIGN-50 methodology checklists include items covering internal validity (study question, selection of participants, assessment, confounding and statistical analysis), the overall assessment of the study and the description of the study. We checked whether all items indicated by the STROBE guidelines for reporting on observational studies (von Elm *et al.* 2007) were adequately addressed by the SIGN checklists. We also looked at whether a power analysis had been conducted, because the STROBE guidelines require reports of observational studies to explain how study size is determined (von Elm *et al.* 2007).

Accordingly, the studies were rated for level of evidence (LE) using the SIGN criteria (SIGN 2008; Table 2). The SIGN criteria do not mention cross-sectional studies and we therefore considered cross-sectional studies without any statistical analysis to be non-analytical studies (LE 3). We allocated cross-sectional studies with statistical analysis to the same category as case–control and cohort studies (LE 2).

Table 2 Levels of evidence (LE) according to the SIGN criteria (SIGN 2008)

1++	High-quality meta-analyses, systematic reviews of randomised controlled trials (RCTs) or RCTs with a very low risk of bias
1+	Well-conducted meta-analyses, systematic reviews or RCTs with a low risk of bias
1–	Meta-analyses, systematic reviews or RCTs with a high risk of bias
2++	High-quality systematic reviews of case–control or cohort studies High-quality case–control or cohort studies with a very low risk of confounding or bias and a high probability that the relationship is causal
2+	Well-conducted case–control or cohort studies with a low risk of confounding or bias and a moderate probability that the relationship is causal
2–	Case–control or cohort studies with a high risk of confounding or bias and a significant risk that the relationship is not causal
3	Non-analytical studies, e.g. case reports, case series
4	Expert opinion

Results

The 45 articles identified in the search were allocated to eight different categories according to the type of exposure: physical conditions in general, motor disorders, sensory impairment, epilepsy, gastrointestinal disease, sleep disorders, dementia and others. There were many categories about which no articles were found, examples being menopause, cardiac and pulmonary disease, infectious disease and malignancies. Most articles addressed our review area as a secondary issue. In these cases, our quality assessment was confined to the questions related to the review area. The results of the review of study characteristics, study results and level of evidence have been described by category and they can be found in Tables 3–10. All studies included both males and females, with the exception of one

male-only (Day 1994) and one where female-only study (Taylor *et al.* 1993). Many of the study groups were very heterogeneous, including participants with both known and unknown aetiology. Furthermore, many studies had a cross-sectional design but they were also retrospective, being based on file data. This area is addressed in Tables 3–10 under the headings Exposure and Outcome measures.

Physical conditions in general

Our search identified 11 cross-sectional studies looking at general medical conditions and challenging behaviour (Table 3). The quality of the studies varied: there were five non-analytical studies, three analytical studies with a high risk of bias and three high-quality studies (Table 3).

The three high-quality studies all looked at the same study population. A study of problem behaviour in general found a significant correlation with visual impairment, urinary incontinence and not having a severe physical disability. There was no association with hearing impairment, bowel incontinence, impaired mobility and epilepsy (Jones *et al.* 2008). In the same study population, there was a significant correlation between SIB and visual impairment. There was no association with hearing impairment, bowel and urinary incontinence, impaired mobility and epilepsy (Cooper *et al.* 2009a). Urinary incontinence was significantly and independently associated with aggressive behaviour in adults, whereas no significant correlation was found for vision impairment, hearing impairment, bowel incontinence, impaired mobility and epilepsy (Cooper *et al.* 2009b). In studies with a higher risk of bias, Collacott *et al.* (1998) found an association between SIB and hearing impairment and impaired mobility, but not with epilepsy and visual impairment. Deb *et al.* (2001) found the same prevalence of behavioural disorders in people with or without physical disabilities or illness.

Low-quality studies suggest that previously undiagnosed and acute medical conditions are associated with severe problem behaviours. The disease categories are gastrointestinal (lactose intolerance, constipation, colon carcinoma), neurological (epilepsy, Parkinson's, sleep apnea, migraine, stroke, encephalitis, hypothalamic hamartoma), urogenital (urinary tract infection, renal insufficiency, pelvic inflammatory disease, gynaecological malignancies, prostatitis), ear–nose–throat (otitis media, sinus problems, dental disease), cardiological (valve disease, heart failure), trauma (lacerations, broken bones, head trauma), eye diseases (dry eyes, cataracts), endocrine (diabetes mellitus, thyroid disease), pulmonary (chronic obstructive pulmonary disease, lung cancer), dermatological (eczema, cellulites), muscolo-skeletal (arthritis, scoliosis), haematological (anaemia) and others (dehydration; Peine *et al.* 1995; Bosch *et al.* 1997; Ryan & Sunada 1997; Kastner *et al.* 2001). Davidson *et al.* (1994) found no differences in medical determinants between people with aggressive behaviour compared to people with other behavioural problems.

In conclusion, urinary incontinence is associated with challenging behaviour (high level of evidence). Visual impairment is associated with SIB, but not with aggressive behaviour (high level of evidence). People with severe physical disabilities have a lower risk of problem behaviours (moderate level of evidence). Previously undiagnosed medical conditions and acute medical conditions are frequently noted in people exhibiting these behaviours. Some of these conditions cause acute or chronic pain (very low level of evidence).

Motor disorders

Two cross-sectional studies looking at cerebral palsy and one at non-ambulatory persons were identified. One of them was a high-quality study, one was an analytical study with a high risk of bias and the third was a non-analytical study (Table 4).

In the high-quality study, which examined children with cerebral palsy, almost 25% had significant behaviour problems. Children with more severe functional limitations had fewer behaviour problems, while children with severe pain had more problems. Sensory impairments and seizures were not significantly correlated with behaviour in children with cerebral palsy (Parkes *et al.* 2008). Blacher & McIntyre's (2006) study suggested that young adults with cerebral palsy and ID had fewer internalising and externalising behaviour problems and were less aggressive than young adults with ID only and equal SIB and sexual problems, but this remains insufficiently proven. Kobe *et al.* (1994) noted many problem behaviours in non-ambulatory

Table 3 Characteristics and level of evidence of studies on general medical conditions

Study	Design	*n*	Age	Level ID	Study population	Exposure measure	Outcome measure	Analysis & results	Level of evidence
Cooper *et al.* (2009a)	Cross-sectional	1023	Adults	All	Representative	***21st Century Health Check	***SIB by DC-LD	Multivariate logistic regression: OR = 1.939, $P = 0.041$ for visual impairment and SIB	2++
Cooper *et al.* (2009b)	Cross-sectional	1023	Adults	All	Representative	***21st Century Health Check	***Aggression by DC-LD	Multivariate logistic regression: OR = 1.995, $P = 0.007$ for urinary incontinence and aggression	2++
Jones *et al.* (2008)	Cross-sectional	1023	Adults	All	Representative	***21st Century Health Check	***Problem behaviour by DC-LD	Multivariate logistic regression: visual impairment OR = 1.460, $P = 0.033$, urinary incontinence OR = 2.053, $P = 0.000$, severe physical disabilities OR = 0.179, $P = 0.000$	2++
Deb *et al.* (2001)	Cross-sectional	101	Adults	All	Representative	**Purpose-designed questionnaire (physical disabilities or illness)	***Psychiatric interview and Disability Assessment Scale	No significant difference in behaviour disorders in people with and without physical impairment or illness	2–
Kastner *et al.* (2001)	Cross-sectional	209	Adults	All	Referred	*Medical files (retrospectively)	**Behaviour rating scale: SIB, aggressive, disruptive, inappropriate habits, others	12% undiagnosed condition†	3
Collacott *et al.* (1998)	Cross-sectional	2101	Adults	All	Representative	*Not described	***SIB by interview and Disability Assessment Schedule	Backward stepwise logistic regression analysis: hearing impairment and immobility $P = 0.0001$	2–
Bosch *et al.* (1997)	Case series	25	Children & adults	Moderate to profound	Referred	*Medical files (retrospectively)	*SIB	28% undiagnosed condition that may have caused pain or discomfort†	3

Table 3 *Continued*

Study	Design	*n*	Age	Level ID	Study population	Exposure measure	Outcome measure	Analysis & results	Level of evidence
Ryan & Sunada (1997)	Cross-sectional	1135	Adults	Moderate to severe	Referred	***Medical examination and additional test protocols (medical files, retrospectively)	*Not described	75% previously undiagnosed or undertreated medical condition†	3
Peine *et al.* (1995)	Case series	10	≥45 years	All	Residence	*Medical files	*Staff records on SIB, aggression, agitation, noncompliance, self-stimulation	Challenging behaviour may coincide with acute medical conditions†	3
Davidson *et al.* (1994)	Cross-sectional	199	Children & adults	All	Referred	*Medical files	*Files and intake chart	χ^2: not significant for epilepsy and cerebral palsy in aggression vs. other challenging behaviour	2–
Hyman *et al.* (1990)	Cross-sectional	97	Children	Not described	Referred	*Medical files (retrospectively): cerebral palsy, sensory impairments, otitis media, seizures	*SIB	Comparison to published data. Co-occurrence of SIB and visual impairment and a history of infantile spasms†	3

Exposure and Outcome measure *, **, ***: see Table 1.
Level of evidence 2++, 2−, 3: see Table 2.
All = mild to profound ID.
Representative = study population is representative for ID population.
Referred = referred patients to a multidisciplinary team for people with ID and/or challenging behaviour.
Residence = study population is derived from a residence for people with ID.
ID, intellectual disabilities; SIB, self-injurious behaviour; DC-LD, diagnostic criteria for psychiatric disorders for use with adults with learning disabilities.
† No statistical analysis.

Table 4 Characteristics and level of evidence of studies on motor disorders

Study	Design	*n*	Age	Level ID	Study population	Exposure measure	Outcome measure	Analysis & results	Level of evidence
Parkes *et al.* (2008)	Cross-sectional	818	Children	All	Representative	***Cerebral palsy by Gross Motor Function Classification System (GMFCS), visual acquity and Bimanual Fine Motor Function, hearing loss (decibels), seizures, pain (Child Health Questionnaire)	***Strenghts and Difficulties Questionnaire: conduct, hyperactivity, emotion and peer problems	Multivariate logistic regression: GMFCS score IV (severe limitations) OR 0.4 (95% CI 0.2–0.8), GMFCS score V (total assistance) OR 0.2 (95% CI 0.1–0.3) severe pain OR 2.7 (95% CI 1.5–4.6)	2++
Blacher & McIntyre (2006)	Cross-sectional	282	Young adults	Moderate to profound	Representative, but cultural-specific	*Cerebral palsy, diagnosed by state agencies	***Scales of Independent Behavior – Revised (SIB-R), Problem Behavior Scale: internalising and externalising, and Reiss Screen for Maladaptive Behavior: aggression, SIB, sexual problems	ANOVA (cerebral palsy group compared to ID only, all less problems in CP): SIB-R: general index $P < 0.05$, externalised index $P < 0.05$, internalised index $P < 0.05$, asocial index not significantly different. Reiss: aggression $P < 0.05$, SIB not significantly different, sexual problems not significantly different	2–
Kobe *et al.* (1994)	Cross-sectional	203	Children & adults	Profound	Residence	*Medical files: non-ambulatory	***Aberrant Behavior Checklist: SIB, stereotypic, aggression	High prevalence of SIB, stereotypy, aggression†	3

Exposure and Outcome measure *, **, ***: see Table 1.
Level of evidence 2++, 2–, 3: see Table 2.
All = mild to profound ID.
Representative = study population is representative for ID population.
Referred = referred patients to a specialised centre/team.
Residence = study population is derived from a residence for people with ID.
ID, intellectual disabilities; SIB, self-injurious behaviour.
† No statistical analysis.

people with ID, but a possible causal relationship was not investigated.

The severe functional limitations associated with motor disorders seem to prevent challenging behaviour, while the pain caused by the conditions is associated with increased challenging behaviour (Blacher & McIntyre 2006; high level of evidence).

Sensory impairments

We found three cross-sectional studies of sensory impairments: a well-conducted study, one with a high risk of bias and a non-analytical study (Table 5).

The well-conducted study from Sjoukes *et al.* (2009) found that visual impairment is not significantly related to challenging behaviour in adults who are visually impaired or blind compared to people without visual impairment. The second, lower-quality study indicates that persons with SIB were more often diagnosed with vision and hearing impairments (Wieseler *et al.* 1995). A likely bias is that more diagnostic examinations may have been performed in people with this behaviour. A high prevalence of visual impairment was noted in eye-poking children. Many of the eye-poking children also exhibited other types of SIB (Jan *et al.* 1994).

It has therefore not been proven that hearing impairment leads to more challenging behaviour in people with ID (low level of evidence). Visual impairment is not significantly associated with challenging behaviour (moderate level of evidence), but specific behaviours (such as SIB/eye-poking) may be related to visual impairment (low level of evidence).

Epilepsy

Our search traced eight studies of epilepsy. There were four well-conducted analytical studies and four analytical studies with a high risk of bias (Table 6).

There is no increased prevalence of physical aggression and other behavioural problems in adults with epilepsy compared to people without epilepsy (Matson *et al.* 1999; Espie *et al.* 2003; Tyrer *et al.* 2006; Matthews *et al.* 2008). Specific subgroups (people with additional visual impairment, motor handicaps, more severe and more frequent seizures and medication side effects, people with generalised EEG activity) may be more at risk for behavioural problems (Deb & Hunter 1991; Espie *et al.* 2003).

In a scientifically unsatisfactory study, Mendez *et al.* (1993) found that interictal violence was correlated with ID and psychopathology, but not with seizure variables.

Two low-quality studies looked at children. Children with epilepsy and ID had more behaviour problems than children with higher IQs, but this relationship was not significant after adjusting for seizure severity (Buelow *et al.* 2003). Lewis *et al.* (2000) found that children with epilepsy did not have more problem behaviour than those without epilepsy.

The group of epileptic patients as a whole does not therefore seem to exhibit more challenging behaviour, with the exception of some specific subgroups (moderate level of evidence).

Gastrointestinal disease

We identified eight studies of gastrointestinal disease. Seven were analytical studies with a high risk of bias or confounding, and one was a non-analytical study. Only one study had a prospective character, but it was uncontrolled. Six looked at gastro-oesophageal reflux disease (GORD) and two at infections with *Helicobacter pylori* (Table 7).

The study of Gössler *et al.* (2007) had a high risk of bias, but the results are nevertheless clinically relevant. Children with more gastro-oesophageal reflux showed significantly more agitation or SIB (as reported by parents). This was also true when there was recurrent reflux after treatment. Children with behavioural abnormalities had significantly higher levels of oesophageal inflammation.

Böhmer *et al.* (1999) found that rumination was more common in institutional residents with GORD. Behaviour problems (rumination, SIB, aggression, fear, screaming, restlessness) do not predict, but may co-occur with, GORD (Böhmer *et al.* 1997b). A significant association was found between reflux oesophagitis and problem behaviours or changed behaviour (rumination, aggression, fear, screaming, restlessness; Böhmer *et al.* 1997c). Rogers *et al.* (1992) found indications that rumination and meal-time challenging behaviours may be caused by dysphagia, GORD or aspiration. Adults

Table 5 Characteristics and level of evidence of studies on sensory impairments

Study	Design	*n*	Age	Level ID	Study population	Exposure measure	Outcome measure	Analysis & results	Level of evidence
Sjoukes *et al.* (2009)	Cross-sectional	269	Adults	Moderate to profound	Representative	***Visual functioning according to IASSID guidelines. Assessment of visual acuity, visual fields, contrast sensitivity, auto-refraction, skiascopy, strabismus	***Development Behaviour Checklist: disruptive/ antisocial, self-absorbed, communication disturbance, anxiety, social relating	Multivariate linear regression: not significant	2+
Wieseler *et al.* (1995)	Cross-sectional	209	Children & adults	All	Residence	*Medical files (retrospectively)	*SIB	Wilcoxon matched-pairs signed-ranks test: $P < 0.001$	2−
Jan *et al.* (1994)	Case series	21	Children	Profound	Referred (visually impaired program)	*Medical examination	*Eye-poking	Frequent visual impairment in eye-poking and other SIB†	3

Exposure and Outcome measure *, **, ***: see Table 1.

Level of evidence 2+, 2−, 3: see Table 2.

All = mild to profound ID.

Representative = study population is representative for ID population.

Referred = referred patients to a specialised centre/team.

Residence = study population is derived from a residence for people with ID.

ID, intellectual disabilities; IASSID, International Association for the Scientific Study of Intellectual Disabilities; SIB, self-injurious behaviour.

† No statistical analysis.

Table 6 Characteristics and level of evidence of studies on epilepsy

Study	Design	*n*	Age	Level ID	Study population	Exposure measure	Outcome measure	Analysis & results	Level of evidence
Matthews *et al.* (2008)	Cross-sectional	318	Adults	Not described	Representative	**Epilepsy interview by trained nurse	***Aberrant Behavior Checklist	Comparison between epileptic and non-epileptic group (not different on level of functioning and psychiatric diagnoses). Mann–Whitney $U = 1254.0$, $P = 0.122$	2+
Tyrer *et al.* (2006)	Cross-sectional	3065	Adults	All	Representative	*Epilepsy by report of carer	***Disability Assessment Schedule: physical aggression towards others	Multivariate logistic regression epilepsy present: OR 0.95 (95% CI 0.74–1.21), $P = 0.67$	2+
Buelow *et al.* (2003)	Cross-sectional	164	Children	Borderline to mild (compared to normal and high IQ)	Representative	***Diagnosis of epilepsy, seizure severity scale for adults	***Child Behaviour Checklist: internalising, externalising problems	No comparison between presence and absence of epilepsy. Comparison between groups of different levels of ID: univariate analysis, total problem score higher in group with low IQ: $P = 0.0055$. After adjusting for seizure severity: $P = 0.094$	2–
Espie *et al.* (2003)	Cross-sectional	186	Adults	All	Referred (hospital-based epilepsy clinics, community ID teams, specialist teams for people with ID and epilepsy)	***Neurologist diagnosis, seizure diaries and Epilepsy and Learning Disabilities Quality of Life Scale	***Aberrant Behavior Checklist	No comparison between presence and absence of epilepsy. Stepwise linear regression analysis: For irritability predictors: ambulant $P = 0.029$. For stereotypic behaviours: visual impairment $P = 0.038$.	2+
Lewis *et al.* (2000)	Cross-sectional	392	Children	All	Representative	*History of seizures	***Developmental behaviour Checklist	Multivariate analysis (MANCOVA): total behaviour problem score in epileptic group compared to non-epileptic group $P > 0.05$	2–
Matson *et al.* (1999)	Cross-sectional	706	Adults	All	Residence	***Diagnosis of epilepsy by neurologist	***Aberrant Behavior Checklist	Univariate analysis (ANOVA): more aberrant behaviour in non-epileptic Group $F(1.705) = 19.1$, $P < 0.001$	2–

Table 6 *Continued*

Study	Design	*n*	Age	Level ID	Study population	Exposure measure	Outcome measure	Analysis & results	Level of evidence
Mendez *et al.* (1993)	Case–control	44–88	Adults	Mild to moderate	Referred (university-affiliated neurology clinic)	*File study, measure not described	***Overt Aggression Scale: violence/aggression (verbal/physical/destructive/SIB)	No comparison between presence and absence of epilepsy. Comparison between violent group with epilepsy and non-violent group with epilepsy. Univariate analysis: more ID in the violent group, McNemar χ^2: $P < 0.01$	2–
Deb & Hunter (1991)	Case–control	150–150	Adults	Mild to severe	Representative	***Epilepsy clearly defined, classification according to the International Classification of Epileptic Seizures	***Profile of Abilities and Adjustment (PAA) schedule (maladaptive behaviour section)	Groups comparable. Univariate analysis between epileptic (EP) and non-epileptic (NEP) groups: total PAA score and severe problem score not significantly different. EP less cooperative, $Z = -2.21$, $P = 0.027$. EP more echolalia, $Z = -2.36$, $P = 0.018$. Subgroup analysis: severe ID EP less aggressive than NEP ($Z = -1.97$, $P = 0.049$). Single-type seizure EP less aggressive than NEP ($Z = -2.29$, $P = 0.022$). Only slow background EEG activity EP less aggressive than NEP ($Z = -2.53$, $P = 0.011$) and EP less overactive than NEP ($Z = -2.01$, $P = 0.044$). Only generalised epileptiform EEG activity EP more temper tantrum than NEP ($Z = -2.47$, $P = 0.013$) and EP more irritable than NEP ($Z = -2.42$, $P = 0.016$)	2+

Exposure and Outcome measure *, **, ***: see Table 1.

Level of evidence 2+, 2–: see Table 2.

All = mild to profound ID.

Representative = study population is representative for ID population.

Referred = referred patients to a specialised centre/team.

Residence = study population is derived from a residence for people with ID.

ID, intellectual disabilities; SIB, self-injurious behaviour.

Table 7 Characteristics and level of evidence of studies on gastrointestinal disease

Study	Design	*n*	Age	Level ID	Study population	Exposure measure	Outcome measure	Analysis & results	Level of evidence
Gössler *et al.* (2007)	Uncontrolled prospective study	19	Children	Not described	Referred (paediatric department of university hospital)	***Yes: 24-h pH monitoring and endoscopy	*Auto-aggression, agitation reported by caregivers	Univariate analysis. Children with behaviour abnormalities more GOR than without $P < 0.0004$. SIB more GOR than agitation $P < 0.01$. Children with behaviour abnormalities higher degree of inflammation than without $P < 0.05$. Degree of inflammation not different between SIB and agitation $P > 0.05$	2–
Swender *et al.* (2006)	Case–control	60	Adults	Severe-profound	Residence	*Medical records GORD (by pH testing or endoscopy)	*Hand mouthing	Higher of frequency of GORD diagnosis in people with than without hand mouthing. $\chi^2 = 8.30$, $P < 0.01$	2–
Wallace *et al.* (2002)	Cross-sectional	168	Adults	Not described	Representative	***HP in blood & faecal samples	***Adaptive Behaviour Scale	Univariate analysis: more maladaptive behaviour in HP-positive than HP-negative group: stereotyped/ hyperactive $P = 0.04$, SIB $P = 0.05$. No difference: sexual behaviour $P = 0.06$, disturbing interpersonal behaviour $P = 0.37$	2–
Böhmer *et al.* (1999)	Cross-sectional	435	Children & adults	Moderate to profound	Residence	***24-h pH-metry and endoscopy	*Physician report: rumination, SIB, aggression, fear, screaming, restlessness	Multivariate stepwise logistic regression GORD: rumination $P = 0.001$, other behaviours not significant	2–

Table 7 *Continued*

Study	Design	*n*	Age	Level ID	Study population	Exposure measure	Outcome measure	Analysis & results	Level of evidence
Böhmer *et al.* (1997a)	Cross-sectional	338	Children & adults	Moderate to profound	Residence	***Serum test HP	*Staff report: rumination	Univariate analysis: rumination in HP-positive patients, compared to HP-negative patients: OR 2.0 (95% CI 1.07–3.64) $P = 0.04$	2–
Böhmer *et al.* (1997b)	Cross-sectional	110	Children & adults	Moderate to profound	Residence	***24-h pH-metry and endoscopy	*Staff report: rumination, SIB, aggression, fear, screaming, restlessness	Univariate analysis: comparison of frequency of behaviour problems in patients with GORD and without: all not significant	2–
Böhmer *et al.* (1997c)	Cross-sectional	1687	Children & adults	Moderate to profound	Residence	***24-h pH-metry and endoscopy (retrospective file study)	*Physician report: rumination, SIB, aggression, fear, screaming, restlessness	Univariate analysis: comparison of frequency of behaviour problems in patients with reflux oesophagitis and without: rumination $P < 0.0001$, changed behaviour $P = 0.01$	2–
Rogers *et al.* (1992)	Case series	23	Adults	Profound	Residence	***Modified barium swallows and esophagrams	*Staff supervisors: regurgitation Psychologist reports: other behaviour problems	High prevalence of dysphagia and gastro-oesophageal abnormalities in rumination†	3

Exposure and Outcome measure *, **, ***: see Table 1.
Level of evidence 2–, 3: see Table 2.
All = mild to profound ID.
Representative = study population is representative for ID population.
Referred = referred patients to a specialised centre/team.
Residence = study population is derived from a residence for people with ID.
ID, intellectual disabilities; SIB, self-injurious behaviour; GORD, gastro-oesophageal reflux disease; HP, *Helicobacter pylori*.
† No statistical analysis.

who engaged in hand mouthing were diagnosed with GORD more often (Swender *et al.* 2006).

Current *H. pylori* infection was associated with more problem behaviour (stereotyped, hyperactive and self-abusive). *H. pylori* infection was also associated with a lower level of ID. This was not taken into account in the analysis of the correlation with problem behaviour (Böhmer *et al.* 1997a; Wallace *et al.* 2002).

Gastro-oesophageal reflux disease may therefore contribute to behaviour difficulties such as rumination, SIB and agitation, but this is insufficiently substantiated (low level of evidence).

Sleep disorders

Seven studies of sleep disorders were found: one well-conducted, five analytical with a high risk of bias or confounding, and one non-analytical study (Table 8).

In the well-conducted study, children with sleep problems had more daytime problem behaviours (irritability, stereotypy, hyperactivity, aggression, screaming, temper tantrum, non-compliance and impulsivity) than children without sleep problems. When factors for specific sleep problems were analysed, after correction for age and level of ID, only irritability was associated with severe sleep problems. The cross-sectional design implies that it is not possible to determine whether the problem behaviours or the sleep problems are the cause (Didden *et al.* 2002).

In studies with a high risk of bias, looking at both children and adults, the subjects with problem behaviours (SIB) had more sleep disturbance than those without problem behaviours (Piazza *et al.* 1996; Symons *et al.* 2000), and people with sleep problems displayed more, and more severe, problem behaviour (stereotypes, irritability, SIB, aggression, temper tantrums, screaming) than those without sleep problems. It is unclear whether sleep problems cause the behavioural problems or vice versa, or if both have a shared neurobiological base (Quine 1991; Chaney *et al.* 1994; Wiggs & Stores 1996; Brylewski & Wiggs 1999).

Sleep problems are therefore associated with daytime challenging behaviour, but the nature of the relationship remains unestablished (moderate level of evidence).

Dementia

We found two non-analytical studies of dementia (which did not relate specifically to Down syndrome only; Table 9).

In people with dementia, behaviour problems (aggression, temper tantrums, pica, SIB, screaming, wandering, repetitive behaviours) are seen, and may arise early in the disease process (Duggan *et al.* 1996; Cooper 1997; very low level of evidence).

Others

We found three articles about conditions not mentioned previously (Table 10).

In a well-conducted study of pain in children with ID, Breau *et al.* (2003) showed that children with SIB did not express pain differently. Children with chronic pain did not have more SIB than children without chronic pain, but the locations were different (near the site of pain instead of mostly to the head and hand).

A non-analytical study of male sex offenders with ID found that many had a distinctive physical disability that might have contributed to the behaviour (Day 1994).

In a non-analytical study of the menstrual cycle in women with SIB, Taylor *et al.* found that SIB might be exacerbated during certain phases of the menstrual cycle (Taylor *et al.* 1993; very low level of evidence).

Discussion

This systematic review of physical conditions associated with challenging behaviour in children and adults with ID identified 11 well-conducted studies that found significant and independent associations with urinary incontinence, pain related to cerebral palsy and chronic sleep problems. Visual impairment is significantly associated with SIB. Because of the cross-sectional design of all the studies, no firm conclusions can be drawn about the causative character of the physical conditions discussed. Longitudinal studies are the only way to complete the evidence. No association was found with hearing impairment, bowel incontinence, mobility impairment or epilepsy. Twenty-one analytical studies of unsatisfactory quality and 13 non-analytical studies

Table 8 Characteristics and level of evidence of studies on sleep disorders

Study	Design	*n*	Age	Level ID	Study population	Exposure measure	Outcome measure	Analysis & results	Level of evidence
Didden *et al.* (2002)	Cross-sectional	286	Children	All	Representative	***Simonds & Parraga Sleep Questionnaire	***Aberrant Behavior Checklist	Mann–Whitney *U*, sleep problem compared to no sleep problem: irritability $Z = -3.93$ $P < 0.001$, stereotypy $Z = -3.60$ $P < 0.001$, hyperactivity $Z = -3.73$ $P < 0.001$, SIB $Z = -1.11$ $P = 0.266$, aggression $Z = -3.15$ $P < 0.01$, screaming $Z = -2.90$ $P < 0.01$, temper tantrum $Z = -3.68$ $P < 0.001$, non-compliance $Z = -4.31$ $P < 0.001$, impulsivity $Z = -2.09$ $P < 0.05$. Multivariate stepwise logistic regression analysis for specific sleep problems: only irritability significant correlation with severe sleep problems: Wald $\chi^2 = 10.23$ $P < 0.01$	2+
Symons *et al.* (2000)	Case–control	60	Adults	Profound	Residence	**Sleep observations by staff	*SIB (as reported by staff)	SIB less sleep than people without SIB: Wilcoxon signed-rank test: $Z = 2.31$ $P < 0.02$, and more variability in sleep pattern: χ^2 $P < 0.01$	2–
Brylewski & Wiggs (1999)	Cross-sectional	205	Adults	All	Representative	***Simonds & Parraga sleep questionnaire	***Aberrant Behavior Checklist	*t*-test for differences between groups with and without sleep problems: irritability $t = -3.76$ $P < 0.001$, stereotypies $t = -3.07$ $P < 0.01$, hyperactivity $t = -2.62$ $P = 0.01$, aggression/temper tantrums $t = -3.12$ $P < 0.01$, non-compliance $t = -1.81$ $P = 0.073$, SIB $t = -2.83$ $P < 0.01$, screaming $t = -3.26$ $P = 0.001$	2–
Wiggs & Stores (1996)	Cross-sectional	486	Children	Severe	Representative	***Simonds & Parraga sleep questionnaire	***Aberrant Behavior Checklist	*t*-test for differences between groups with and without sleep problems: irritability $t = 4.43$ $P < 0.001$, stereotypies $t = 3.69$ $P < 0.001$, hyperactivity $t = 4.18$ $P = 0.001$, SIB $t = 2.76$ $P < 0.01$, aggression $t = 5.22$ $P < 0.001$, screaming $t = 5.54$ $P < 0.001$, temper tantrums $t = 3.96$ $P < 0.001$, non-compliance $t = 3.60$ $P < 0.01$ impulsivity 2.90 $P < 0.01$	2–

Table 8 *Continued*

Study	Design	*n*	Age	Level ID	Study population	Exposure measure	Outcome measure	Analysis & results	Level of evidence
Piazza *et al.* (1996)	Cross-sectional	51	Children & young adults	All	Referred (inpatient unit for people with severe behaviour problems)	**Sleep observations	*Behaviour reports by psychologist	Less sleep in children with ID and behaviour problems than peers of the same age†	3
Chaney *et al.* (1994)	Cross-sectional	40	Adults	Moderate to profound	Residence	**Sleep observations	***Client Development Evaluation report: aggression, SIB, destructiveness, reaction to frustration, stereotypy, hyperactivity, temper tantrums, inacceptable social behaviour	Fisher's exact probability test patients with sleep disturbance compared to those without: more stereotypic behaviour: $P = 0.01$, SIB: $P = 0.056$	2–
Quine (1991)	Cross-sectional	166	Children	Severe	Representative	***Sleep index and settling and waking problems from the Behaviour Screening Questionnaire	***Behaviour Screening Questionnaire and Disability Assessment Schedule	χ^2 between children with and without sleep problems: management problems $P < 0.001$, activity $P < 0.001$, concentration $P < 0.001$, attention seeking $P < 0.01$, sexual problems $P < 0.001$, runs away $P < 0.001$, interferes $P < 0.001$, destructive $P < 0.01$, pica $P < 0.01$, swears $P < 0.01$, disruptive $P < 0.01$, temper tantrums $P < 0.05$, problems with peers $P < 0.05$, habits $P < 0.001$, repetitive activities $P < 0.05$, echolalia $P < 0.05$	2–

Exposure and Outcome measure *, **, ***: see Table 1.
Level of evidence 2+, 2–, 3: see Table 2.
All = mild to profound ID.
Representative = study population is representative for ID population.
Referred = referred patients to a specialised centre/team.
Residence = study population is derived from a residence for people with ID.
ID, intellectual disabilities; SIB, self-injurious behaviour.
† No statistical analysis.

Table 9 Characteristics and level of evidence of studies on dementia

Study	Design	*n*	Age (years)	Level ID	Study population	Exposure measure	Outcome measure	Analysis & results	Level of evidence
Cooper (1997)	Cross-sectional	134	≥65	Mild to severe	Representative	***Present Psychiatric State-Learning Disabilities: dementia	***Present Psychiatric State-Learning Disabilities: maladaptive behaviours	Aggression, temper tantrums and other behaviour problems may arise in dementia†	3
Duggan *et al.* (1996)	Case series	12	≥48	Mild to severe	Referred (psychiatrists for people with ID)	**If available dementia diagnosis by psychiatrist in medical files and interview with carers	**Past Behavioural History Inventory	Behavioural problems (pica, aggression, SIB, screaming, wandering, repetitive behaviours) can occur early in dementia†	3

Exposure and Outcome measure *, **, ***: see Table 1.

Level of evidence 3: see Table 2.

All = mild to profound ID.

Representative = study population is representative for ID population.

Referred = patients identified by specialised consultants.

ID, intellectual disabilities; SIB, self-injurious behaviour.

† No statistical analysis.

Table 10 Characteristics and level of evidence of other conditions

Study	Design	*n*	Age	Level ID	Study population	Exposure measure	Outcome measure	Analysis & results	Level of evidence
Breau *et al.* (2003)	Cross-sectional	101	Children	All	Referred (tertiary paediatric centre)	***Pain: Non-communicating Children's Checklist – Revised	***SIB: Behavior Problem Inventory	MANOVA: not significant for pain expression between children with and without SIB. *t*-test: children with chronic pain compared to those without: less body surface $P = 0.01$, fewer body sites $P = 0.04$	2+
Day (1994)	Case series	47	Adolescents & adults	Borderline to moderate	Referred (hospital department of psychiatry)	*Medical files (retrospectively)	**Sexual offences/ incidents	51% had a distinctive physical disability (speech deficits, dysmorphias, sensory impairments, epilepsy)†	3
Taylor *et al.* (1993)	Case series	9	Adolescents & adults	Severe to profound	Residence	*Menstrual cycle	***Client Development Evaluation Report: SIB	Exacerbation of SIB during early and late follicular phases of the menstrual cycle†	3

Exposure and Outcome measure *, **, ***: see Table 1.
Level of evidence 2+, 3: see Table 2.
All = mild to profound ID.
Referred = referred patients to a specialised hospital/tertiary centre.
Residence = study population is derived from a residence for people with ID.
ID, intellectual disabilities; SIB, self-injurious behaviour.
† No statistical analysis.

or case series, usually based on file data, suggest associations with GORD, dysphagia, dementia, menstrual cycle phases and specific sub-types of epilepsy. However, so far, these are insufficiently substantiated.

Reports listing undiagnosed or untreated conditions found during the diagnostic work-up of persons with challenging behaviours (Ryan & Sunada 1997; Kastner *et al.* 2001) have only limited value, as it is well known that even severe undiagnosed conditions are found in any group of persons with ID (van Schrojenstein Lantman-de Valk & Walsh 2008). This results from the ineffective communication of subjective symptoms by these persons and by a lack of training in medical issues among caring staff.

Four medical conditions have been proven to be associated with challenging behaviour. Urinary incontinence is significantly correlated with aggressive behaviour. As the authors in question point out, it remains unclear whether there is a shared underlying mechanism (e.g. autonomic sympathetic discharge), whether the incontinence contributes to aggression (because people are ashamed or experience discomfort) or whether the aggression contributes to the incontinence (Cooper *et al.* 2009b). Second, visual impairment is significantly correlated with SIB only. The cross-sectional character of the studies means that the direction of this relationship has not been established. Visual impairment may contribute to the behaviour (people express communication difficulties with problem behaviour, or engage in eye-poking in order to get visual stimulation); alternatively, visual impairment may be caused by the tissue damage resulting from SIB. Third, increased behavioural difficulties are seen only if cerebral palsy is complicated by severe pain (Parkes *et al.* 2008). Finally, the shared conclusion in studies of sleep disorders is that people with behavioural problems have more sleep disturbance (Quine 1991; Chaney *et al.* 1994; Piazza *et al.* 1996; Wiggs & Stores 1996; Brylewski & Wiggs 1999; Symons *et al.* 2000). It remains unclear whether the association is because of a common underlying neurochemical mechanism accounting for both factors, and whether sleep disturbance contributes to behaviour problems, or vice versa (Symons *et al.* 2000). These findings are supported by single-case experimental studies, in which problem behaviours are exacerbated in people who are naturally deprived of sleep (Kennedy & Meyer 1996; O'Reilly & Lancioni 2000).

A few conditions that appear clinically relevant have been insufficiently investigated. Almost all studies of the behavioural effects of gastrointestinal disease are side measurements of other studies. Dysphagia, GORD and *H. pylori* infections are frequent in rumination cases (Rogers *et al.* 1992; Böhmer *et al.* 1997a), which indicates that there should be diagnostic testing for these conditions in people with rumination. Changes in behaviour may be caused by oesophageal pain, colic and discomfort resulting from GORD (Gössler *et al.* 2007) and reflux oesophagitis (Böhmer *et al.* 1997c).

Dementia can result in behavioural changes, even at an early stage (Duggan *et al.* 1996; Cooper 1997). One study indicates that distinctive physical features may cause aberrant behaviour, possibly resulting from low self-esteem (Day 1994). The menstrual cycle may affect SIB, either through hormonal changes or pain and discomfort (Taylor *et al.* 1993). The role of pain requires further study, because there is no proof that chronic pain causes more SIB (Breau *et al.* 2003), even though there is a proven link between severe pain and behaviour in children with cerebral palsy (Parkes *et al.* 2008). Acute and chronic pain may very well be expressed by challenging behaviour (Dubois *et al.* 2010).

In certain physical conditions, it has not been possible to prove a causal relationship. Motor disorders are not correlated with more problem behaviours (Blacher & McIntyre 2006; Jones *et al.* 2008; Parkes *et al.* 2008). This may be because people with impaired mobility may receive higher levels of support from carers (to meet their disability needs). Furthermore, the physical disabilities may preclude problem behaviour in these people (Jones *et al.* 2008). Nor has hearing impairment been proven to cause more problem behaviours (Wieseler *et al.* 1995; Jones *et al.* 2008; Cooper *et al.* 2009a,b). However, many studies used file data and it is known that hearing impairment frequently goes unnoticed (Evenhuis *et al.* 2001; Meuwese-Jongejeugd *et al.* 2008), so this association should be investigated further.

Conclusions in studies of epilepsy are unambiguous. Generally, the population with epilepsy does not have more behaviour problems than those

without epilepsy (Deb & Hunter 1991; Matson *et al.* 1999; Lewis *et al.* 2000; Buelow *et al.* 2003; Espie *et al.* 2003; Tyrer *et al.* 2006; Jones *et al.* 2008; Matthews *et al.* 2008; Cooper *et al.* 2009a,b). However, it has been suggested that specific subgroups (e.g. people with generalised epileptic activity, more severe or frequent seizures, medication side effects and co-morbidity) may be prone to more behavioural difficulties (Deb & Hunter 1991; Espie *et al.* 2003).

A possible explanation of why associations are difficult to detect is that, in larger studies, the groups of participants were very heterogeneous. This may have reduced the effect sizes.

There are many medical conditions (e.g. infectious disease, migraine, menopause, allergy, and cardiac and pulmonary disease) where it is conceivable that pain or discomfort could lead to challenging behaviour. Particularly, people with more severe forms of ID, with less capacity to communicate their discomfort, may express this through altered behaviour. However, this review failed to find studies of many of these conditions. This does not mean that there is no correlation between these conditions and challenging behaviour; it means that further investigation is required. Areas warranting urgent attention in future research are gastrointestinal disease (GORD, but also constipation), infectious disease (e.g. ear infections, which can cause pain), hormonal influences (menstrual cycle and thyroid function), dental disease and cardiopulmonary disease. The fact that the life expectancy of the population with ID is rapidly increasing means that research is needed into the behavioural effects of physical deterioration and menopause in older age.

Only a few studies directly address the question of which physical conditions may cause problem behaviour and, with a few exceptions, these were generally low-quality studies. More often, behaviour was measured in conjunction with a single physical condition, or behaviour was described, but as part of a larger study of the condition. Moreover, the design of almost all the studies was cross-sectional. The implication is that the nature of the associations found in these studies cannot be established. This makes firm conclusions about causal relationships difficult but some suggestions for clinical practice can be made.

When people with ID and challenging behaviours are examined, urinary incontinence, visual impairment, sleep problems and pain should be considered. In people who engage in rumination, hand mouthing or mealtime challenging behaviour, diagnostic testing should be performed looking at dysphagia, GORD and *H. pylori* infections. Other conditions that might be considered are early dementia, discomfort in certain menstrual cycle phases and specific sub-types of epilepsy (people with more severe and more frequent seizures, medication side effects and generalised EEG activity).

The present review highlights the role that medical conditions can play in challenging behaviour and the need for evaluating those conditions in clinical practice. It also reveals major gaps in the evidence, because many studies are of low quality and many physical illnesses have not been investigated.

There is a strong need for comprehensive high-quality longitudinal research, with clearly defined measures of both physical conditions and behavioural disorders, to establish firm evidence as a basis for clinical guidelines for the prevention, differential diagnosis and treatment of challenging behaviour.

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