

The relationship between challenging behaviour and anxiety in adults with intellectual disabilities: a literature review

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Abstract

Background Anxiety and challenging behaviour (CB) often occur simultaneously in people with intellectual disabilities (ID). Understanding the associations between anxiety and CB may contribute to more accurate diagnoses and management of both anxiety and CB in this population.

Aims To examine the relationship between anxiety and CB.

Methods A literature review covering the period from January 2000 to January 2012.

Results Seven studies about the relationship between psychiatric disorders, including anxiety, and CB were identified. These studies confirm the relationship between anxiety and CB in people with ID, although the precise nature of this relationship remains unclear.

Conclusions The study points toward the existence of a moderate association between anxiety and CB.

Further research is needed to clarify the complex nature of the association between anxiety and CB.

Keywords anxiety, challenging behaviour, intellectual disabilities

Introduction

People with intellectual disabilities (ID) are exposed to a variety of biological, developmental, psychological and social stressors (Deb *et al.* 2001; Allen 2008). As a result, they exhibit challenging behaviours (CB)¹ more frequently and severely than the general population (Hastings *et al.* 2004; Allen 2008).

Rates of CB vary considerably across studies, ranging from 2% to 60% in the ID population (Borthwick-Duffy 1994; Emerson *et al.* 2001; Crocker *et al.* 2006). Methodological variations and the heterogeneity of the study populations (e.g.

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¹ In literature, 'challenging behaviours' (CB) and 'problem behaviours' are used interchangeably. For ease of readability, we use CB throughout the text.

level of ID, age and residential setting) account for these differences. Also, the definition of CB varies across the studies, which further limits the comparability of the studies.

Understanding CB in people with ID is important to control or prevent such behaviours. A number of contributing factors to CB stand out in this population: pain, physical diseases, psychiatric disorders, and attachment problems (Smith & Howie 2009). Anxiety is also considered to be one of these influencing factors (Cooper *et al.* 2007). According to Deb *et al.* (2001) and Ramirez & Lukenbill (2007), people with ID experience more mood and anxiety disturbances than the general population. Emerson & Hatton (2007) reported a three to four times higher risk of emotional problems and related CB (disruptive/antisocial behaviour, self-absorbed behaviour, communication problems, anxiety) in people with ID compared with those without ID.

In research and clinical practice, however, anxiety in relation to CB is often a neglected aspect. Understanding the associations between anxiety and CB may contribute to more accurate diagnoses and management of both psychiatric (anxiety) disorders and CB in the ID population.

When considering anxiety as an influencing factor, it is important to realise that anxiety is influenced by a number of other variables (Stavrakaki & Lunskey 2009). There are several aetiological factors that are generally attributed to anxiety symptoms or anxiety disorders in people with ID, such as hereditary factors, genetic syndromes (e.g. fragile X syndrome, Williams syndrome), brain damage, cognitive deficits (particularly in temporal memory), poor adaptive coping skills, trauma/abuse, attachment problems or exposure to stress (Stavrakaki & Lunskey 2009).

The purpose of this literature review was to examine the relationship between anxiety and CB in persons with ID. The key question was: What is known about the relationship between anxiety and CB in persons with ID?

Method

A literature search was undertaken using the PsycINFO and Medline databases, combining the

following MeSH/Thesaurus terms describing both the target group and relevant variables. Applied key terms referring to the target group were: *mental retardation; intellectual disability/ies; learning disability/ies*. Key terms used to describe outcome variables were *anxiety (disorder); problem behaviour; behavioural problems; aggression and challenging behaviour*. The search for these terms was in both singular and plural forms, in US and British terminology. We included English-language articles on empirical studies published in peer-reviewed journals between January 2000 and January 2012.

To be included in the review, articles had to focus on the relationship between anxiety symptoms/anxiety disorders, on the one hand, and any form of CB, on the other. Articles that did not refer to adults with ID and articles strictly referring to specific genetic syndromes were excluded. Figure 1 shows a flow chart of the systematic review process that was utilised.

The first step was to screen the available articles for relevance by reading the titles and abstracts. This step was carried out by two independent researchers (A. P. & W. N.). In cases of lack of consensus, the papers were discussed until consensus was reached. Twelve articles remained after this first round. The full texts of these papers were read and analysed by the first author, and the bibliographies from these 12 articles were examined for relevant cross-references. Following the full text analysis, three articles were added. Eight articles did not provide relevant empirical research data or were opinion or review articles without research data. In the end, seven articles remained.

The level of evidence of the findings was assessed by making use of the Cochrane criteria for the methodological quality of individual studies (CBO Kwaliteitsinstituut voor de Gezondheidszorg 2007).

Results

The results of this literature review are summarised in Table 1, which lists the number of participants, level of ID, study design, measurement instruments and findings for each of the studies.

In a matched case control study (Table 1, #1), Myrbakk & Tetzchner (2008) investigated the relationship between psychiatric disorders and CB in

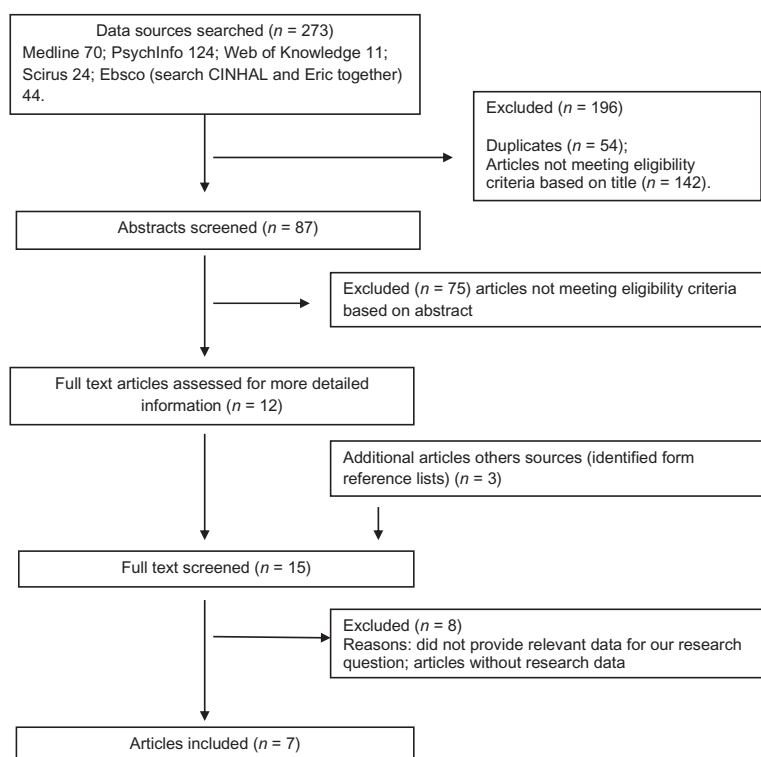


Figure 1 Flow chart for systematic review.

persons with ID. The Aberrant Behaviour Checklist (ABC) was used to assess CB. They found that temper tantrums/outbursts (42%) and aggression toward other people (38%) were the most frequent CB. The researchers used four different instruments to screen for psychiatric symptoms: the Reiss Screen, the Mini Psychiatric Assessment Schedule for Adults with Developmental Disorders (Mini PAS-ADD), the Diagnostic Assessment of the Severely Handicapped (DASH-II), and the Assessment of Dual Diagnosis (ADD). In all checklists, persons with CB scored statistically significantly higher than the comparison group without CB on both general psychopathology measures and the diagnosis-specific symptom scales. The majority (69%) of the participants with CB showed symptoms of the main psychiatric disorders, including anxiety disorders. The Mini PAS-ADD revealed scores for anxiety symptoms in 21% of persons with CB, but none in the comparison group (Fisher Exact test, $P = 0.000$). Measurements with DASH-II revealed anxiety disorders in 32% ($n = 43$) of the persons with CB and 13% of the persons in

the control group (Fisher, $P = 0.046$). The prevalence of aggression, tantrums, screaming and self-injury were positively correlated with anxiety severity. The correlation between anxiety and tantrums was moderate ($r = 0.54$; $P < 0.01$) according to the standards by Floyd *et al.* (2006), whereas the correlations between anxiety and aggression, screaming and self-injury were weak to very weak ($r = 0.36, 0.34, 0.18$; ns, respectively). The conclusion of the study was that the majority of the participants with CB showed symptoms of various psychiatric disorders, suggesting that many CB problems may be (unconventional) symptoms of these psychiatric disorders. According to the authors, however, CB could also be related to difficult life situations caused by psychiatric disorders. Another possible explanation was that difficult life situations contribute to both psychiatric disorders and CB in individuals with ID.

In a matched case control design (Table 1, #2), Holden & Gitlesen (2003) used the PAS-ADD in a sample of 165 adults showing all levels of ID. This study also revealed a correlation between CB and

Table 1 Results literature review: the relationship between challenging behaviour (CB) and anxiety

Authors	Level	Design	Sample	Topic	Measurement instruments	Level of evidence	Findings
1. Myrbakk & Tetzchner (2008)	Mild to profound	Matched case control	142	Psychiatric symptoms/disorders	ABC Reiss Screen, Mini PAS-ADD DASH-II and ADD	B	Group with CBs scored significantly higher on symptoms of psychiatric disorder than the comparison group. PAS-ADD: 21% anxiety symptoms in the group with CB and none in the control group DASH-II: 32% anxiety symptoms in the group with CB and 13% in the control group.
2. Holden & Gitlesen (2003)	Mild to profound	Matched case control	165	Psychiatric symptoms/disorders	PAS-ADD Rating CB	B	CB are associated with an increased prevalence of psychiatric symptoms including anxiety. PAS-ADD: 27% anxiety symptoms in the group with CB and 12% in the control group.
3. Felce <i>et al.</i> (2009)	Mild to profound	Survey	312	Psychiatric symptoms/disorders	ABS ABC DAS PIMRA	C	CB scored significantly higher among participants with psychiatric symptoms; this was more pronounced for people with severe ID. 46% persons met at least one threshold level indicator of mental illness, 24% showed symptoms of anxiety.
4. Tsiouris <i>et al.</i> (2003)	Moderate to profound	Prospective open clinical study	26	Psychiatric symptoms/disorders	DSM III-R/DSM IV criteria Repeated observations Use of protective devices BPI OAS	C	At the start: all 26 persons had SIB, only 7 had a psychiatric diagnosis. After a multidimensional diagnostic approach, all 26 persons were found to have a psychiatric diagnosis: 5 persons were diagnosed with anxiety disorders. Beside the first diagnosis there were 15 persons with co-morbid diagnoses, including anxiety. Treatment of the psychiatric disorders led to a decrease in SIB.

Table 1 Continued

Authors	Level	Design	Sample	Topic	Measurement instruments	Level of evidence	Findings
5. Moss <i>et al.</i> (2000)	Not mentioned	Control study	320	Psychiatric symptoms/disorders	PAS-ADD Rating CB on a four-point scale	B	Increasing severity of CB was associated with an increased prevalence of psychiatric symptoms. This was strongest for depression; the association was present for anxiety (and psychosis) but did not reach statistical significance.
6. Rojahn <i>et al.</i> (2004)	Severe and profound	Survey	180	Psychiatric symptoms/disorders	DASH-II BPI	C	The presence of CB increases the likelihood of psychopathology by up to three times. CB, particularly SIB and aggressive/destructive behaviour, are not general atypical manifestations (of mood and) anxiety disorders. On the other hand, there is a high risk of serious CB contributing to a high score on the anxiety sub-scale of the DASH-II.
7. Tsiouris <i>et al.</i> (2011)	Mild to profound	Survey	4069	Psychiatric symptoms/disorders	IBR-MOAS	C	Anxiety was most strongly associated with verbal aggression and physical aggression against self and obsessive compulsive disorder with physical aggression against objects.

ABC, Aberrant Behavior Checklist/community; Reiss Screen, Reiss Screen for Maladaptive Behavior; PAS-ADD-Checklist, (Mini)Psychiatric Assessment for Adults with Developmental Disabilities Checklist; DASH-II, Diagnostic Assessment of the Severely Handicapped; ADD, Assessment of Dual Diagnosis; ABS, Adaptive Behavior Scale; DAS, Disability Assessment Scale; PIMRA, Psychopathology Instrument for Mentally Retarded Adults; ID, intellectual disabilities; BPI, Behavior Problems Inventory; OAS, Overt Aggression Scale; IBR-MOAS, Institute for Basic Research Modified Overt Aggression Scale.

increased prevalences of psychiatric symptoms, but focused specifically on anxiety and psychosis. CB were measured by rating the extent to which the persons showed aggression to others, destruction of property, self-injurious behaviour (SIB) or CB not specified. Anxiety was present in almost 12% of persons in the control group with minimal or no CB ($n = 59$), while 27% of persons in the CB group ($n = 96$) showed symptoms of anxiety (χ^2 , $P \leq 0.05$).

Felce *et al.* (2009) carried out a survey among 312 adults with ID (Table 1, #3). This study confirmed the existence of a relationship between psychiatric status and the occurrence of CB. Measured with the ABC, CB appeared to be more prevalent among participants meeting threshold levels on the psychiatric screening of the Psychopathology Instrument for Mentally Retarded Adults (PIMRA) (31.7%) than those who were categorised as below the threshold for mental illness (15.0%, $U = 6617$, $P < 0.001$). This relationship was more pronounced in people with severe ID than in people with moderate or mild ID. The percentage of individuals meeting at least one threshold level indicator of possible mental illness in this study was 46%. Of these 46%, 24% of the persons had an anxiety disorder, 14% an adjustment disorder, 12% an affective disorder, 8% a somatoform disorder, 5% suffered from schizophrenia, and 5% had a personality disorder.

In another study among 320 persons with ID (Table 1, #5), Moss *et al.* (2000) found an association between symptoms of anxiety and the occurrence of SIB. They also concluded that the severity of CB (none, less demanding, more demanding) was positively associated with the prevalence of psychiatric symptomatology as measured with the PAS-ADD. From the individuals meeting defined diagnostic, in relation to severity of CB, the association was notably strong for depression (15%, χ^2 test, $P < 0.001$), but less so for anxiety (9%) and psychosis (9%), where the positive association was present but did not reach statistical significance.

Tsiouris *et al.* (2003) employed a different methodological approach by using an intervention study with a pre-post-test design (Table 1, #4). The intervention consisted of treating previously undiagnosed psychiatric disorders in individuals with ID in order to decrease or eliminate SIB. Data were collected from 26 individuals with ID who showed

SIB. At the start of the study, most of the subjects were treated with psychotropic drugs, protective devices and/or behaviour modification. Only seven of them had a psychiatric diagnosis at the start of the study and none of them was diagnosed with an anxiety disorder. A diagnosis of ID with CB or SIB was the carrying diagnosis in the other 19 persons. Using a multidimensional diagnostic approach, all 26 subjects were ultimately found to have a psychiatric diagnosis. Major depression (12), impulse control disorder (12), anxiety disorder (5), and bipolar disorder (6) were the most common psychiatric diagnoses (including double counts) in this group. In fifteen cases, the individuals also had co-morbid psychiatric diagnoses, such as an impulse-control disorder, an anxiety disorder, major depression or a bipolar disorder. Treatment of these psychiatric disorders with appropriate psychotropic medication led to positive impacts, i.e. statistically significant ($P < 0.001$) decreases in the frequency and severity of SIB (from a moderate, severe or extreme degree to mild degree). SIB was completely eliminated in 12 cases.

Rojahn *et al.* (2004) (Table 1, #6) carried out a risk ratio analysis to explore the association between CB and psychiatric disorders. They reported that persons with severe and profound ID exhibiting self-injurious, stereotypical, aggressive or destructive behaviours all had statistically significant higher scores for psychopathology on the DASH-II than persons without CB. The presence of CB increased the likelihood of psychopathology by up to three times. Factor analysis was used to determine whether specific types of CB were clustered with certain psychiatric conditions. The analysis confirmed that CB tended to be associated with psychiatric conditions traditionally known to be associated with such behaviours, e.g. impulse control and conduct disorders, and to a lesser extent, with conditions such as mood and anxiety disorders. The moderate strengths of the associations reveal that CB (and in particular SIB and aggressive/destructive behaviours) are not typical manifestations of mood and anxiety disorders.

Risk ratios were computed to explore to what extent the probability of having a certain psychiatric condition (DASH-II scores) was related to CB (BPI scores). These results revealed a different view than the outcomes of the factor analysis. Risk ratios

ranged from 0.7 to 3.0, with a mean risk ratio of 1.5 (SD = 0.4). The Anxiety sub-scale (DASH-II) showed a risk ratio of 2.3, which means that having a serious aggression/destructive problem more than doubled the risk of scoring in the 90th percentile of the DASH-II Anxiety sub-scale. The items and sub-scales of the DASH-II were developed to reflect major DSM (Diagnostic and Statistical Manual of Mental Disorder) categories most pertinent to this population.

The relationships between psychiatric conditions and CB showed to be complex and manifested considerable individual variation. The authors concluded as follows: 'The associations may suggest that CB are generally not atypical manifestations of psychiatric disorders based on the same biochemical substrate, but that both are related to genetic pre-dispositions combined with diminished resilience and other mechanisms that assist individuals in fending off psychosocial problems' (p. 31).

In a large survey of 4069 participants (Table 1, #7), Tsiouris *et al.* (2011) examined the association between certain psychiatric disorders and aggressive behaviour, controlling for sex, age, autism, and level of ID. The researchers used a modification of the Modified Overt Aggression Scale (MOAS) with the following five domains of aggression: verbal aggression toward others (VAOTH); physical aggression against other people (PAOTH); physical aggression against objects (PAOBJ); physical aggression against self (PASLF), and verbal aggression toward self (VASLF). The results indicated that anxiety, disturbances in impulse control and bipolar disorders, as well as young age and having severe ID or autism, were positively associated with physical aggression against self. Anxiety (clinical diagnoses made by psychiatrists) was most strongly associated with both verbal aggression (21% more than those without the disorder; regression coefficient = 0.189, $P < 0.001$) and physical aggression against self (34%; 0.295, $P < 0.001$); and obsessive compulsive disorder with physical aggression against objects (21%; 0.209, $P < 0.001$). Anxiety, impulse control disorders, and bipolar disorders showed to be common co-morbid psychiatric disorders in persons with autism spectrum disorders and were found to contribute to the initiation or exacerbation of aggression against self and objects in this study group.

Most of the studies presented above confirm the relationship between anxiety disorders/anxiety symptoms and CB. However, the associations between anxiety and CB must be interpreted within the context of associations found between other psychiatric disorders and CB. In three of the studies, anxiety had the strongest association with CB; in the other four studies, depression, bipolar disorder, psychosis or impulse control disorder had stronger associations with CB than anxiety.

Two of the seven studies did not identify any statistically significant relationships between anxiety and CB, but they did notice signs of such relationships.

Discussion

The principal finding of the present literature review is that most of the studies we included show statistically significant associations (from weak to strong) between psychiatric disorders, including anxiety, and CB in persons with ID. It should be noted that the specific relationship between anxiety and CB has not much been researched to date. For this reason, we also gave attention to studies that examined the relationship between CB and psychiatric disorders in a broader sense. These studies were analysed in particular for that part of the data that explored the relationship between anxiety and CB. Noticeably, although the focus in those studies was not specifically on the relationship between anxiety and CB, they did include many anxiety-related issues.

We may conclude that the association between anxiety and CB is rather complex in nature. We found different patterns of associations which are open to a number of interpretations. For example, we do not know whether an increase in psychopathology, including anxiety, leads to more CB, or whether the opposite is true: more CB leads to higher levels of anxiety, or more generally, to more psychopathology. Thus, the direction which the found relationship takes remains unclear and may go either way.

The complexity is increased even further when other influencing factors associated with both anxiety disorders and CB are considered. Examples of these influencing factors are coping and life

events (MacHale & Carey 2002; Sequeira *et al.* 2003; Owen *et al.* 2004; Hartley & MacLean 2009). Poor coping skills in persons with ID, especially a lack of ability to control aggression, influence their capacity to deal with stressful social interactions, which may lead to more symptoms of anxiety and distress (Hartley & MacLean 2009). Other studies show that adults with ID who have experienced negative life events display more frequent CB, and are at increased risk for anxiety disorder (MacHale & Carey 2002; Sequeira *et al.* 2003; Owen *et al.* 2004).

Moreover, the precise nature of anxiety cannot always be identified. Questions arise such as: Are the symptoms really anxiety symptoms? Does this specific person really have an anxiety disorder according to DSM-criteria? Are the anxiety symptoms part of another psychiatric disorder?

The same types of question arise in connection with the issue of how we should understand CB. Is this type of behaviour a direct result of the anxiety experienced? Or conversely, does CB create feelings of anxiety, e.g. because of negative responses from the person's environment? Another hypothesis is that the presence of CB may increase the likelihood of risk factors for anxiety, such as environmental precursors. Myrbakk & Tetzchner (2008) state 'that CB may reflect underlying psychopathology or a difficult life situation related to the presence of a psychiatric condition, or a difficult life situation may contribute to both psychiatric disorders and CB in people with ID' (p. 329). People with ID are particularly vulnerable to stressful events and stressful social interactions (Hartley & MacLean 2005). They have difficulties in coping with stress, with an increased risk of rather high levels of psychological distress in their everyday life.

Another hypothesis is that anxiety and CB are not directly linked but coincide because of underlying biological pathology. Rojahn *et al.* (2004) and Tsiouris *et al.* (2011) refer to the etiological factors of both anxiety and aggression: there is a similar phenotype predisposition, combined with diminished resilience for fending off psychosocial adversities.

We noted that different types of CB were addressed in the different studies. This makes it difficult to interpret the outcomes. Also, various types of instruments were used in the studies included,

both for measuring CB and for determining psychopathology. Not all these instruments have been specifically developed for and tested in people with ID, which challenges their validity and reliability for this group. Generally, an assessment of psychopathology and other psychological variables in this population poses various problems. Cognitive and communication deficits make assessment through self-report difficult to achieve, and in most cases even impossible. Then, there is the problem of diagnostic overshadowing, i.e. attributing symptoms of psychopathology to a person's intellectual disability. Moreover, the usefulness of current diagnostic classification systems [e.g. ICD-10 (International Classification of Disease) and DSM-IV] for our target group is restricted, as those systems have been validated only for the general population (Fletcher *et al.* 2007). All studies included struggle with the above mentioned problems and for this reason the findings should be interpreted with caution. An important prerequisite for improving research in persons with ID is the availability of validated instruments for the population of people with ID.

Despite the different approaches and the methodological limitations, all studies show an association between anxiety and CB. As stated before, this relationship is complex in nature, probably bidirectional, and therefore no causal relationships can be inferred. It is difficult to draw unambiguous conclusions on the basis of the various types of studies we included in this review. Nevertheless, we studied the papers thoroughly and conscientiously, discussed results and were cautious when drawing conclusions.

For clinical practice, we recommend multidimensional diagnostic procedures, focusing on the biological, psychological and social dimension of existence, to fully account for the complexity of the relationship between psychiatric disorders, including anxiety, and CB. Different diagnostic data gathering methods should be used, including diagnostic interviews, use of (validated) rating scales, and observational methods, to obtain an optimum understanding of the CB in people with ID and the possible role of biological, psychological and social factors in the origin of this behaviour. This puts the presence of a psychiatric disorder, including anxiety, in a broader perspective of influencing factors that contribute to the actual manifestation of CB.

Finally, the present review makes clear that more research is needed to clarify the relationship between anxiety and CB, and the possible role of other influencing factors in this relationship.

Conclusion

In conclusion, despite a number of methodological limitations, this review points toward the existence of a moderate association between anxiety and CB. The complex nature of this association requires further clarification.

Implications for practice: (1) professionals should be aware of the complex relationship between psychopathology/anxiety and CB in patients with ID; and (2) to analyse those complex relations in individual persons with ID, professionals should perform thorough diagnostic procedures, taking into account different dimensions of functioning, i.e. biological/physiological, psychological, social and environmental dimensions. Therefore, a multidisciplinary and multidimensional approach in these diagnostic procedures is needed to explore the dynamics between psychopathology/anxiety and CB.

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