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Deposited on: 26 February 2016
Statistical modelling studies examining the dimensional structure of psychopathology experienced by adults with intellectual disabilities: systematic review

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Abstract

Diagnosing mental ill-health using categorical classification systems has limited validity for clinical practice and research. Dimensions of psychopathology have greater validity than categorical diagnoses but dimensional models have not had a significant impact on our understanding of mental ill-health experienced by adults with intellectual disabilities. This paper systematically reviews the methods and findings from intellectual disabilities studies that use statistical methods to identify dimensions of psychopathology from data collected using structured assessments of psychopathology. The PRISMA framework for systematic review was used to identify studies for inclusion. Study methods were compared to best-practice guidelines on the use of exploratory factor analysis. Data from the 20 studies included suggest that it is possible to use statistical methods to model dimensions of psychopathology experienced by adults with intellectual disabilities. However, none of the studies used methods recommended for the analysis of non-continuous psychopathology data and all 20 studies used statistical methods that produce unstable results that lack reliability. Statistical modelling is a promising methodology to improve our understanding of mental ill-health experienced by adults with intellectual disabilities but future studies should use robust statistical methods to build on the existing evidence base.
1. Introduction

Classification systems for the diagnosis of mental ill-health are central to clinical practice and research. A categorical model of psychopathology is the basis for existing classification systems, such as ICD-10 and DSM5. Although some authors disagree (Lawrie et al., 2010), there has been a developing consensus that classification systems based on a categorical model of psychopathology lack sufficient validity for research to investigate the aetiology and pathophysiology of mental ill-health (Insel, 2012). Dimensional models of psychopathology, empirically derived using statistical modelling, have been examined as the main alternative to categorical models and been found to have improved reliability and validity (Markon, 2010).

Concerns about categorical models of psychopathology have centred around the limited discriminant and predictive validity. With regard to such neighbouring categorical diagnoses, most studies have focused on psychopathology in the psychoses. Early studies, examining psychopathology, failed to identify points of rarity, or a bimodal distribution, that could distinguish between schizophrenia and the affective psychoses (Kendell & Gourlay 1970; Kendell & Brockington 1980). High levels of comorbidity in epidemiological studies have also been cited as evidence for the poor discriminant validity of diagnostic categories (Mineka et al., 1998). For example, in one study of comorbid depressive and anxiety diagnoses (Brown et al., 2001) participants 57% of 1,127 participants had current co-morbid mood and anxiety disorders, and the rate of lifetime comorbidity was 81% (Brown et al., 2001).
Alongside the research on discriminant validity, studies have compared the predictive validity of categorical and dimensional models of psychopathology. Two studies reported that the dimensional models of psychopathology were more strongly associated with measures of longitudinal outcome of mental ill-health (van Os et al., 1996; van Os et al., 1999) and one concluded that they were similar and complementary (Dikeos et al., 2006).

One focus of early research on dimensional models of psychopathology was psychopathology experienced by children and young people. This initial evidence supported the concept of internalising and externalising dimensions of psychopathology (Achenbach 1966; Achenbach et al., 1987). More recent studies also support the existence of internalising and externalising dimensions in adults (Krueger 1999; Slade 2007). The internalising dimension is characterised by problems with negative emotions, and includes psychopathology cutting across the boundaries of the commonly co-morbid mood, anxiety, fear/ phobic and obsessional disorders. The externalising dimension includes problems with aggression and other problem behaviours, overactivity and inattention, and disinhibition, cutting across the commonly co-morbid conduct, personality, attention-deficit and hyperactivity and substance misuse disorders.

The findings suggesting that a dimensional approach may have advantages over categorical diagnoses led to work to examine how to incorporate the evidence from dimensional models into DSM-V (Helzer et al., 2009). Since categorical classification systems for the diagnosis of mental ill-health have good utility in clinical practice, dimensional models are best considered as complementary rather than an
alternative to categorical diagnoses (Kotov et al., 2011). Whilst recognising the ongoing questions about categorical diagnoses and the research supporting the internalising and externalising frameworks, the DSM-V working group decided that there was insufficient scientific evidence to support wholesale changes in the organisation structure of DSM-V. Instead, since dimensional and categorical models can be complementary in clinical care the working group included cross-cutting, dimensional assessments of psychopathology in the DSM-V manual.

We have argued above that dimensional modelling studies have helped our understanding of psychopathology experienced by individuals who do not have intellectual disabilities. They may also offer useful insights into psychopathology experienced by individuals with intellectual disabilities. Psychopathology, and the presentation of mental disorders experienced by adults with intellectual disabilities, can differ from that seen in the general population. For example, aggression and other challenging behaviours are the type of psychopathology most commonly experienced by adults with intellectual disabilities (Cooper et al., 2007), and have been conceptualised as equivalents of depressive symptoms (DC-LD; Royal College of Psychiatrists, 2001). This conceptualisation contrasts with the proposition above that mood symptoms are best thought of as part of an internalising dimension of psychopathology and problem behaviours in the separate externalising dimension. However, given the differences in psychopathology experienced by individuals with intellectual disabilities, it cannot be assumed that the results from statistical modelling studies using data from individuals who do not have intellectual disabilities can be generalised to understanding psychopathology experienced by individuals with intellectual disabilities. Therefore, this review examined the methods and
findings from studies using a statistical approach to model the dimensional structure of psychopathology experienced by adults with intellectual disabilities.

2. Methods

The Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) statement was used as the basis for this review (Moher et al., 2009). PRISMA is an evidence-based set of guidelines on the minimum items for reporting in systematic reviews, aiming to improve the reporting of systematic reviews. There are 27 items included in the statement, and for each item an explanation of the rationale for inclusion is available, along with supporting evidence and examples of best practice (Liberati et al., 2009)

2.1 Search strategy

Four electronic databases were systematically searched:

- MEDLINE (Ovid), 1946 to February week 1 2015, last searched 6 February 2015
- EMBASE (OVID), 1947-present, last searched 6 February 2015
- PsycINFO (EBSCOhost), 1900 to February 2015, last searched 6 February 2015
- CINAHL (EBSCOnhost), 1937 to 6 February 2015.

Appropriate search terms for intellectual disabilities were combined with terms for statistical data reduction methods used to extract dimensions from large datasets (appendix A shows the Medline search strategy). To identify additional relevant studies, reference lists of retrieved studies were hand searched along with five key intellectual disabilities journals (American Journal on Mental Retardation, Journal of Autism and Developmental Disorders, Journal of Intellectual Disability Research,

2.2 Study selection
Independently screening of the titles and abstracts of identified articles, two authors (CAM, MO) had agreement of 96% with a kappa of 0.56. For each remaining article, an inclusion checklist was independently completed on the full text article by two researchers (CAM, MO) using the criteria below. There was agreement on 37/38 (97.4%) full text records and full agreement reached at a consensus meeting to finalise which articles to retain for review.

2.3 Inclusion criteria
1. Sample includes adults with intellectual disabilities
2. Data collected using a structured instrument
3. Instrument includes a broad range of psychopathology items relevant to mental ill-health
4. Data analysed using factor analysis, or similar methods.

2.4 Coding of statistical methods
There are multiple decisions about which specific methods to use in the step-by-step process involved in factor analysis, and related statistical techniques. Best practice recommendations for factor analysis (Costello & Osborne, 2005) were used as the basis for coding the methods in studies identified in the literature search (Table 1).
One component of factor analysis that the best practice guidelines (Costello & Osborne, 2005) did not make any specific recommendation about was the method used to produce the initial correlation matrix. Factor analytic methods were originally developed for use with continuous variables. Since psychopathology data is often ordinal, or binary, in nature, it is preferable to use methods specifically developed for use with non-continuous variables, such as polychoric or tetrachoric correlations (Wirth & Edwards, 2007). Therefore, in addition to the methods in Table 1, studies were coded as to whether they used poly/tetrachoric methods appropriate for the analysis of non-continuous variables.

The best practice guidelines made no specific recommendations on the minimum sample size and the case: item ratio required for factor analysis (Costello & Osborne, 2005). This was because the evidence from Monte Carlo simulations suggests that the requirements for sample size and case: item ratio are dependent on other aspects, such as the strength of item loadings (Costello & Osborne, 2005). In order to provide a benchmark to code studies against, it was decided to use a minimum sample size of 200 and case: item ratio of 5:1 which are often cited in psychopathology research (Floyd & Widaman, 1995).

Each included paper was coded independently by two authors (CAM, MO) using a specifically designed, structured tool. Where there was disagreement a meeting with a third researcher took place to reach consensus agreement about which articles to retain for review.
3. Results

Figure 1 shows the number of articles retrieved and included at each stage of the search process. The majority of full-text articles were excluded because the structured assessment instrument collected a narrow range of psychopathology, such as problem behaviours (Aberrant Behaviour Checklist; Aman et al., 1985) and depressive symptoms (Clinical Behaviour Checklist for Persons with Intellectual Disabilities, CBCPID; Tsiouris et al., 2003).

Table 2 provides an outline of the 16 papers that met the inclusion criteria. For the purposes of this review, multiple studies reported in a single paper are coded separately in the results (Watson et al., 1984; Sturmey et al., 1996). Therefore, the methods and findings from a total of 20 studies are summarised.

The high number of studies included suggests that it is feasible to use exploratory factor analysis to model psychopathology experienced by adults with intellectual disabilities. None of the studies reported problems with the methods of analysis due to problems with the data; although five studies reported excluding items of psychopathology from the analysis because of zero (Gustafsson & Sonnander, 2005; Gustafsson & Sonnander, 2002; Sturmey et al., 2004; Sturmey et al., 2010; Gerber & Carminati, 2013) or less than one per cent (Matson et al., 1991) variance.
Only one study tested the fit of the model from the exploratory factor analysis (EFA) using confirmatory factor analysis (CFA; Hatton & Taylor, 2008). In two separate analyses, the CFA found that the EFA model was a poor fit when tested in random sub-samples of the sample used for the EFA.

3.1 Methods of analysis

Seven studies reported the statistical software package used in the factor analysis; three used the Statistical Package for the Social Sciences (SPSS: (Linaker, 1991; Sturmey et al., 2005; Gerber & Carminati, 2013) and four used the Statistical Analysis System (SAS: Watson et al., 1988; Gustafsson & Sonnander, 2002; Gustafsson & Sonnander, 2005; Janssen & Maes, 2013). None of these seven papers reported the use of the programs for EFA of categorical data in SPSS (CATPCA) and SAS (PRINQUAL).

None of the studies described the use of the recommended poly/tetra-choric methods to produce the initial correlation matrix for the analysis of non-continuous data.

The best practice guidelines recommend the use of common factor analysis (CFA) and oblique rotation. All nineteen studies that provided details of the methods of analysis (Moss et al., 1998) used principal components analysis (PCA) and all 20 studies reported the results from orthogonal factor rotation (17 studies used varimax and three quartimax rotation). One study used oblique rotation initially but changed
to using orthogonal rotation because there were no items cross-loading across factors (Hatton & Taylor, 2008).

One study did not report the method used to decide the number of factors to extract from the analysis (Moss et al., 1998). Five studies used the recommended parallel analysis (Zeilinger et al., 2010) or scree plot (Matson et al., 1991; Sturmey et al., 1996; Sturmey et al., 2005; Janssen & Maes, 2013). Twelve (70.5%) studies used a minimum eigenvalue (EV) cut-off as the single method to decide how many factors to extract from the data. Hatton & Taylor (2008) used an EV cut-off of 1.0 but also included sufficient factors to account for over 60% of the variance. Therefore, although it is identified as the least accurate method in best practice guidelines, the majority of studies used the EV cut-off method as the only method to decide the number of factors to extract.

Table 3 provides summary data for the remaining three aspects of the methods that were coded.

********* insert Table 3 about here*********

Overall, a greater number of studies met the guidance on sample size, case: item ratio and the minimum item loading than the recommendations relating to methods of factor extraction and rotation.

4. Discussion
The studies included in this review made an important research contribution to the development of structured instruments for the assessment of psychopathology experienced by adults with intellectual disabilities. All of the structured instruments are useful in the assessment and management of mental ill-health experienced by adults with intellectual disabilities. None of the studies were primarily aimed at examining de novo dimensional models of psychopathology. However, two studies did give consideration to the relevance of their findings to the underlying structure of psychopathology (Watson et al., 1988; Sturmey et al., 1996). We believe that the initial data from all 20 studies provides a useful evidence-base to inform future statistical modelling studies.

One finding from this review was that studies to date have used statistical methods that have been consistently shown to produce unreliable results. In the majority of studies the “Little Jiffy” (Kaiser, 1970) combination of PCA and varimax rotation was used which has been shown to threaten the validity of findings from statistical modelling. In fact, the combination of PCA and varimax rotation still dominates the factor analysis literature more broadly. There is some suggestion that over time there has been a reduction in the number of studies using PCA and varimax (Conway & Huffcutt, 2003) but they were used in over half of the 1700 exploratory factor analysis studies identified by a search of a two-year period of Psycinfo (Costello & Osborne, 2005) and studies published between 1997 and 2008 in key developmental disabilities journals (Norris & Lecavalier, 2010).

PCA has been reported to be less accurate than common factor analysis in Monte Carlo simulation studies (Snook & Gorsuch, 1989; Widaman, 1993; Fabrigar et al.,
1999; Preacher & MacCallum, 2002) and the methods used to produce the initial correlation matrix have been shown generate unstable factor solutions when used with non-continuous data (Olsson, 1979; Bernstein & Teng, 1989; Mislevy, 1986). Since there is clear evidence to suggest that common factor analysis is preferable to PCA, future studies should consider using common factor analysis, and tetrachoric correlations, to model the dimensional structure of psychopathology experienced by adults with intellectual disabilities (Ford et al., 1986; Fabrigar et al., 1999; Preacher & MacCallum, 2002; Costello & Osborne, 2005; Henson & Roberts, 2006).

One useful finding from psychopathology research using statistical modelling has been the suggestion that the poor discriminant validity of categorical diagnoses can be partly explained by higher order, internalising and externalising dimensions of psychopathology (Krueger & Markon, 2006). Higher order dimensions of psychopathology have also been shown to have greater validity than categorical diagnoses for the investigation of the aetiology of mental ill-health (Buckholtz & Meyer-Lindentonberg, 2012). The insights into understanding psychopathology that statistical modelling studies describing higher order dimensions have provided require examination of the correlations between dimensions. However, all the studies included in this review used orthogonal factor rotation which does not allow dimensions to correlate. Therefore, future studies should use oblique rotation to examine whether higher order dimensions exist and are useful for understanding psychopathology experienced by individuals with intellectual disabilities.

Studies investigating the higher order internalising and externalising dimensions have provided novel insights into the relationship between challenging behaviours,
such as aggression, and other forms of psychopathology in children (Loth et al., 2014). There have been many studies investigating the relationship between problem behaviours and other forms of psychopathology in individuals with intellectual disabilities (Thakker et al., 2012). None of these intellectual disabilities studies investigated the relationship of problem behaviours to other types of psychopathology within a broad dimensional model.

Bearing in mind the methodological limitations described above, three of the five studies that explicitly identified challenging behaviour dimensions (Linaker, 1991; Sturmey et al., 2010; Sturmey et al., 1996; Sturmey et al., 2004; Kellett et al., 2005) were made up of a mixture of problem behaviour and affective psychopathology items (Sturmey et al., 1996; Kellett et al., 2005; Sturmey et al., 2004). The irritability/depression dimension extracted in a separate study (Tsiouris et al., 2003) also includes a mixture of items usually considered as representing problem behaviour and affective psychopathology. The consistency with which studies have identified dimensions including both problem behaviour and affective items of psychopathology is interesting and merits further study.

There has been a persistent thread of research examining whether problem behaviours are equivalents of depressive symptoms in adults with intellectual disabilities (depressive equivalents). Two previous studies used statistical modelling to examine the relationship between problem behaviours and depression in adults with intellectual disabilities (Tsiouris et al., 2003; Sturmey et al., 2010). These studies were not included in this review because the CBCPID (Marston et al., 1997) includes only ratings of problem behaviours and depressive symptoms. Similar to
the studies included in this review, the findings are limited by the use in both studies of PCA with varimax rotation (Tsiouris et al., 2003; Sturmey et al., 2010). However, both these studies concluded that problem behaviours should not be considered as equivalents to depressive symptoms.

Despite the methodological limitations, these initial studies looking at challenging behaviours within dimensional models of psychopathology provide some encouragement that statistical approaches may provide new insights into the relationship between problem behaviours and other psychopathology. Since problem behaviours are associated with significant negative impacts (Beadle-Brown et al., 2009) and costs (Felce et al., 2008) future research should use robust statistical methods to examine whether a dimensional approach to psychopathology can improve our understanding and therefore management of mental ill-health experienced by adults with intellectual disabilities.

5. Conclusions

Dimensional models of psychopathology can provide useful insights into mental ill-health. It is not yet clear whether studies of psychopathology data will further our understanding of psychopathology experienced by adults with intellectual disabilities and contribute to clinical practice. Future statistical modelling studies using psychopathology data from adults with intellectual disabilities should use robust methods recommended in best practice guidelines for EFA and CFA.

6. Acknowledgements
This study was funded by a research grant from the Chief Scientist Office, Scottish Government (Reference: CZG/2/556)
7. References


Figure 1. Flow chart of study selection process.

Records identified through database searching (n = 2338) → Records identified through other sources (n = 25) → Records after duplicates removed (n = 847) → Records screened (n = 847) → Full-text articles assessed for eligibility (n = 38) → Articles included for data extraction (n = 16) → Full-text articles excluded (n = 22)
Table 1: Best practice recommendations for factor analysis (Costello & Osborne, 2005)

<table>
<thead>
<tr>
<th>Best practice recommendation</th>
</tr>
</thead>
<tbody>
<tr>
<td>1. Method of statistical analysis</td>
</tr>
<tr>
<td>2. Correlation method</td>
</tr>
<tr>
<td>2. Number of factors retained</td>
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<tr>
<td>3. Rotation method</td>
</tr>
<tr>
<td>4. Item loading to factor</td>
</tr>
<tr>
<td>5. Sample size</td>
</tr>
<tr>
<td>6. Case: item ratio</td>
</tr>
</tbody>
</table>

⁰ Based on Wirth and Edwards (2007)

² Eigenvalue

³ Based on Floyd and Widaman (1995)
Table 2: Studies reporting exploratory factor analysis of psychopathology experienced by adults with intellectual disabilities

<table>
<thead>
<tr>
<th>Authors</th>
<th>Sample</th>
<th>Measure of psychopathology</th>
<th>Methods</th>
<th>Number of factors retained (% variance)</th>
<th>Dimension names (eigenvalue, % variance, number of items)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Matson et al. 1984</td>
<td>N= 110, clinic sample; borderline= 8.1%, mild= 47.3%, moderate= 40.9%, severe= 3.7%. Mean age= 45.9 (18-71, SD N/A)</td>
<td>PIMRA^a- 56 psychopathology items derived from DSM-III criteria for schizophrenia, affective, psychosexual, adjustment, anxiety, somatoform and personality disorders. Self-report and informant versions available.</td>
<td>PCA^b, varimax rotation, factor extraction eigenvalue&gt; 1.5, item loading ≥ 0.35.</td>
<td>Self-report: Anxiety (N/A, N/A, 8 items), Social adjustment (N/A, N/A, 5 items); Informant: Affective (N/A, N/A, 14 items), Somatoform (N/A, N/A, 5 items), Psychosis (N/A, N/A, 5 items)</td>
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<tr>
<td>Linaker 1991</td>
<td>N= 169 inpatients; mild= 3.6%, moderate= 20.1%, severe= 50.9%, profound= 15.2%, unknown= 9.7%. Mean age= 40.4 (16-65, SD N/A)</td>
<td>PIMRA- informant version^c</td>
<td>PCA, varimax rotation, factor extraction eigenvalue&gt; 1.5, item loading ≥ 0.35.</td>
<td>9 (49.3%)</td>
<td>Somatoform (5.06, 10.3%, 8), gender identity (3.68, 7.5%, 3), hostility (3.11 6.3%, 4), psychosis (2.66, 5.4%, 5), self-consciousness (2.29, 4.7%, 4), adjustment problem (2.25, 4.6%, 4), anxiety (1.88, 3.8%, 3), autistic traits (1.69, 3.4%, 3), avoidant/ anxious (1.53 3.1%, 3)</td>
</tr>
<tr>
<td>Balboni et al. 2000</td>
<td>N=652 mixed sample-community (411) institution (241); mild= 34%, moderate= 39%, severe/profound=</td>
<td>PIMRA- informant version</td>
<td>PCA, varimax rotation, factor extraction eigenvalue&gt; 1.5, item</td>
<td>7 (34.5%)</td>
<td>Anxiety (6.03, 10.8%, 11), Adjustment problem (3.28, 5.9%, 7), Somatoform (2.74, 4.9%,</td>
</tr>
<tr>
<td>N= 101, mixed sample- community (30), institution (71); mild= 25.7%, moderate= 32.9%, severe/profound= 41.4%. Mean age= 50.2 (24-94, SD= 14.3)</td>
<td>PIMRA- informant version</td>
<td>PCA, varimax rotation, factor extraction eigenvalue&gt; 1.5, item loading ≥ 0.4</td>
<td>5 (51%)</td>
<td>Somatoform (4.29, 16.5%, 5), Psychosis (3.17, 12.2%, 7), Psychosexual (2.37, 9.1%, 4), Adjustment problem (1.85, 7.1%, 5), Anxiety (1.56, 6.0%, 5)</td>
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<tr>
<td>N= 160 mixed sample- living in community (95), institution (65); borderline= 19.4%, mild= 47.5%, moderate= 33.1%. Mean age= 29.4 (18-67, SD= 11.4)</td>
<td>PIMRA</td>
<td>Self &amp; informant version; PCA, varimax rotation, factor extraction eigenvalue&gt; 1.5, item loading ≥ 0.35</td>
<td>4 (N/A)</td>
<td>Self-report: Anxiety; Social adjustment; Identity/ reality concern; Unlabelled Informant: Affective concerns; Social adjustment; Somatoform; Unlabelled (Problem behaviours)</td>
<td></td>
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<tr>
<td>N= 201 community sample. Mean age= 44 (18-83, SD N/A)</td>
<td>PAS-ADD&lt;sup&gt;6&lt;/sup&gt; checklist- 29 item screening instrument, completed by informant to identify possible mental ill-health</td>
<td>Method of analysis not described, quartimax rotation, item loading ≥ 0.5</td>
<td>8 (N/A)</td>
<td>Depression (N/A, N/A, 6), Restlessness (N/A, N/A, 4), Phobic anxiety (N/A, N/A, 5), Psychosis (N/A, N/A, 3), Hypomania (N/A, N/A, 3), Autistic spectrum (N/A, N/A, 3), Depression (N/A, N/A, 2), Non-specific</td>
<td></td>
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<tr>
<td>Study</td>
<td>Sample Size</td>
<td>Sample Characteristics</td>
<td>PAS-ADD Checklist</td>
<td>Methodology</td>
<td>Factor Interpretation</td>
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<tr>
<td>Sturmey et al. 2005</td>
<td>N=226 clinic attendees; mild= 68%, moderate= 20%, severe/profound= 12%. Mean age= 34 (Range N/A, SD= 13.5)</td>
<td>PAS-ADD checklist</td>
<td>PCA, quartimax rotation, factor extraction eigenvalue ≥ 1 and scree plot; item loading ≥ 0.5</td>
<td>9 initially but only 3 factors interpretable (34.6%)</td>
<td>Mood (5.33, 19.7%, 8); Sleep (2.20, 8.1%, 3); Psychosis (1.83, 6.3%, 3)</td>
</tr>
<tr>
<td>Hatton &amp; Taylor 2008</td>
<td>N= 1, 115 administrative sample (98% response rate). Mean age= 44.0 (17-92, SD= 15.19)</td>
<td>PAS-ADD checklist</td>
<td>PCA, varimax rotation, factor extraction eigenvalue&gt; 1.0, rotated factors account &gt; 5% variance, sufficient factors included to account &gt; 60% variance, item. loading ≥ 0.4</td>
<td>7 (61.25%)</td>
<td>Depression 1 (4.19, 15.50%, 7); Sleep problems (2.46, 9.10, 3); Organic problem (2.35, 8.70%, 4); Panic (2.11, 7.80%, 3); Psychosis (2.09, 7.72%, 4); Hypomania (1.72, 6.37%, 3); Depression 2 (1.64, 6.06%, 2)</td>
</tr>
<tr>
<td>Zeilinger et al., 2011</td>
<td>N= 270, convenience sample; mild/moderate= 77%, severe/profound= 33%. Mean age= 40.2 (18-80, SD= 14.7)</td>
<td>PAS-ADD checklist</td>
<td>PCA, varimax rotation, factor extraction by parallel analysis, requirements for item loading to factors not provided.</td>
<td>6 (57.8%)</td>
<td>Depression (2.53, 10.5%, 5), Restlessness (2.53, 10.5%, 6), Anxiety (2.52, 10.5%, 4), Sleep problems (2.44, 10.2%, 3), Psychosis (1.99, 8.3%, 4), Reduced self-care (1.86, 7.7%, 2)</td>
</tr>
<tr>
<td>Gerber &amp; Carmanati, 2013</td>
<td>N= 126, clinic attendees; mild/moderate= 66%, severe/profound= 34%. Mean age= 38.5 (16-71)</td>
<td>PAS-ADD checklist</td>
<td>PCA, quartimax rotation, tested original three factor structure, requirements for item loading to factors not provided.</td>
<td>3 (46.1%)</td>
<td>Factor 1 (N/A, N/A, 6), Factor 2 (N/A, N/A, 6), Factor 3 (N/A, N/A, 12)</td>
</tr>
<tr>
<td>Authors</td>
<td>Sample Description</td>
<td>Instruments and Methods</td>
<td>Factors (N/A)</td>
<td>Sample</td>
<td>Sample 1:</td>
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<tr>
<td>Janssen &amp; Maes, 2013</td>
<td>N= 377, convenience sample; mild= 39%, moderate= 39%, severe= 15%, profound=6%</td>
<td>Mini PAS-ADD 66 items of psychopathology for identification of symptoms suggestive of psychiatric disorder</td>
<td>PCA, varimax rotation, factor extraction based on scree plot, item loading ≥ 0.3.</td>
<td>5 (N/A)</td>
<td>Depression (N/A, N/A, 11), Autism (N/A, N/A, 15), Anxiety (N/A, N/A, 8), Hypomania (N/A, N/A, 5), Psychosis ((N/A, N/A, 2)</td>
</tr>
<tr>
<td>Sturmey et al. 1996</td>
<td>Three samples: Sample 1, n= 180, community sample; Sample 2, n= 102, college sample; Sample 3, n= 71, institutional sample. Age and ability level N/A</td>
<td>RSMB 38 item screening instrument for identification of emotional and behaviour problems</td>
<td>PCA, varimax rotation, factor extraction eigenvalue&gt; 1.0, item loading ≥ 0.3</td>
<td>Sample 1= 1 factor (25.2%); Sample 2 &amp; 3= 3 factors (Sample 2 44.2%; Sample 3 41.5%)</td>
<td>Sample 1: General factor (6.55, 25.2%, 26); Sample 2: Extrapersonal maladaptive behaviour (6.24, 24%, 9), Psychosis (2.94, 11.3%, 8), Intrapersonal maladaptive behaviour (2.31, 8.9%, 6); Sample 3: Extrapersonal maladaptive behaviour (5.62, 21.6%, 11), Psychosis (2.91, 11.2%, 10), Intrapersonal maladaptive behaviour (2.26, 8.7%, 8)</td>
</tr>
<tr>
<td>Gustafson &amp; Sonnander, 2002</td>
<td>N=140, random sample; mild= 27.6%, moderate= 47.0%, severe= 25.4%; Mean age= 43.6 years (Range 21-68, SD=11.9)</td>
<td>RSMB- Swedish translation</td>
<td>PCA, varimax rotation, factor extraction eigenvalue&gt; 1.0, requirements for item loading to factors not provided.</td>
<td>7 (67%)</td>
<td>Aggressive behaviour (N/A, 14.8%, N/A), Avoidant Behaviour (N/A, 13.5%, N/A), Depression B (N/A, 9.3%, N/A), Psychosis (N/A, 8.3%, N/A), Dependant (N/A, 7.7%, N/A), Paranoia (N/A, 7.3%, N/A),</td>
</tr>
<tr>
<td>Study</td>
<td>Sample Description</td>
<td>Measure</td>
<td>Methodology</td>
<td>Factor Loadings</td>
<td>Factors</td>
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<tr>
<td><strong>Matson et al. 1991</strong></td>
<td>N= 506, institutional sample; severe=37%, profound=63%. Mean age=37.7 (Range &amp; SD N/A)</td>
<td>DASH²- 83 items of psychopathology and behaviour</td>
<td>PCA, varimax rotation, factor extraction scree test, item loading ≥ 0.3</td>
<td>6 (39%)</td>
<td>Emotional lability (4.9, 12%, 8), Antisocial (2.7, 6.5%, 8), Language disorder (2.4, 5.8%, 8), Social withdrawal/stereotypy (2.3, 5.5%, 8), Eating disorder (1.9, 4.6%, 6), Sleep disorder (1.8, 4.5%, 3)</td>
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<tr>
<td><strong>Sturmeys et al. 2004</strong></td>
<td>N= 451, institutional sample; severe=11%, profound=89%. Mean age=48 years (Range N/A, SD=15)</td>
<td>DASH-II- 84 items of psychopathology and behaviour</td>
<td>PCA, varimax rotation, factor extraction eigenvalue&gt;1.5, item loading ≥ 0.35</td>
<td>5 (26%)</td>
<td>Emotional lability/antisocial (9.1, 11.1%, 9), Language disorder (3.9, 4.8%, 4), Dementia/anxiety (2.9, 3.6%, 7), Sleep disorder (2.8, 3.4%, 3), Psychosis (2.5, 3.1%, 3)</td>
</tr>
<tr>
<td><strong>Kellett et al. 2004</strong></td>
<td>N=335, clinic attendees with mild intellectual disabilities. Mean age= 33.0 (16-64, SD= 10.65)</td>
<td>BSI¹⁰- 53 item self-report inventory of psychopathology, rated on five point Likert scale</td>
<td>PCA, varimax rotation, factor extraction eigenvalue&gt;1.0, item loading ≥ 0.35</td>
<td>8 (50.26%)</td>
<td>Depression (16.19, 30.56%, 13), Anxiety (2.32, 4.39%, 11), Somatisation (1.91, 3.61%, 10), Cognitive impairment (1.73, 3.27%, 8), Suicidal ideation (1.62, 3.06%, 6), Paranoia (1.44, 2.72%, 5), Hostility (1.39, 2.63%, 7), Anger (1.37, 2.59%, 4)</td>
</tr>
</tbody>
</table>
1 N/A not available from the details provided in the paper
2 PIMRA Psychopathology Instrument for Mentally Retarded Adults
3 PCA Principal Components Analysis
4 The items relating to personality disorder were removed leaving 49 items for inclusion in the EFA
5 The items relating to personality disorder and inappropriate mental adjustment were removed leaving 42 items for inclusion in the EFA
6 PAS-ADD Psychiatric Assessment Schedule for Adults with Developmental Disabilities
7 The exploratory factor analysis included a sub-sample of 543 cases, and a confirmatory factor analysis included a sub-sample of 601 cases
8 RSMB Reiss Screen for Maladaptive Behaviours- the 26 items contributing to total score used in EFA
9 DASH Diagnostic Assessment Schedule for the Severely Handicapped
10 BSI Brief Symptom Inventory
Table 3: Descriptive summary of methods used in studies using factor analysis to examine psychopathology

<table>
<thead>
<tr>
<th>Variable (n)</th>
<th>Mean</th>
<th>SD</th>
<th>Median</th>
<th>Minimum</th>
<th>Maximum</th>
<th>No. of studies (%) meeting recommendation</th>
</tr>
</thead>
<tbody>
<tr>
<td>Sample size (20)</td>
<td>260.6</td>
<td>176.3</td>
<td>190.5</td>
<td>71</td>
<td>652</td>
<td>9 (45)</td>
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<tr>
<td>Case: item (20)</td>
<td>7.0</td>
<td>8.2</td>
<td>5.8</td>
<td>2.0</td>
<td>38.4</td>
<td>13 (65.0)</td>
</tr>
<tr>
<td>Minimum item loading (17)</td>
<td>0.36</td>
<td>0.06</td>
<td>0.35</td>
<td>0.3</td>
<td>0.5</td>
<td>13 (76.5)</td>
</tr>
</tbody>
</table>
Appendix I: Medline (Ovid) search strategy, 1966-2015

<table>
<thead>
<tr>
<th>Search Terms</th>
</tr>
</thead>
<tbody>
<tr>
<td>1. Mental retardation /</td>
</tr>
<tr>
<td>2. Learning disorders /</td>
</tr>
<tr>
<td>3. Mentally disabled persons /</td>
</tr>
<tr>
<td>4. Developmental disabilities /</td>
</tr>
<tr>
<td>5. ((mental$ or learning or intellect$) adj (retard$ or handicap$ or disab$ or impair$)).mp [mp=title, original title, abstract]</td>
</tr>
<tr>
<td>6. Or / 1-5</td>
</tr>
<tr>
<td>7. Psychopathology /</td>
</tr>
<tr>
<td>8. Psychiatry</td>
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<tr>
<td>9. Psychology</td>
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<tr>
<td>10. Or / 7-10</td>
</tr>
<tr>
<td>11. Models, statistical</td>
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<tr>
<td>12. Factor analysis, statistical</td>
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<tr>
<td>13. Data interpretation, statistical</td>
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<tr>
<td>14. Cluster analysis</td>
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<tr>
<td>15. Principal component analysis</td>
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<tr>
<td>16. Or / 11-15</td>
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<tr>
<td>17. 6 and 10</td>
</tr>
<tr>
<td>18. 16 and 17</td>
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