Published in final edited form as:

J Intellect Disabil Res. 1998 December ; 42(Pt 6): 472–480.

The latent variable structure of the Compulsive Behaviour Checklist in people with Prader-Willi syndrome

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Abstract

The presence and severity of compulsive behaviours may be evaluated via the Compulsive Behaviour Checklist (CBC) and this instrument has been successfully employed in people with intellectual disability. However, the applicability of the overall CBC scoring system, which entails tallying the number of behavioural categories represented (i.e. five) as well as the number of individual behaviours endorsed (i.e. 25), is not known in the population with Prader-Willi syndrome (PWS). The present investigation examined the latent variable structure of the CBC in people with PWS in order to identify possible population-specific scoring and interpretation considerations. The 25 behaviour-specific items of the CBC were analysed for 75 people with PWS (44 females and 31 males) aged between 4 and 41 years (mean $\pm$ SD = 11.4 $\pm$ 9.4) via factor analysis with principal component extraction and equamax rotation. The most suitable solution was determined on the basis of multiple empirical criteria: (1) the scree test; (2) eigenvalues $>$ 1.00; (3) salient loadings $>$ 0.30; (4) the clarity of item assignment to a single latent dimension; (5) the internal consistency of the latent dimension(s) (coefficient $\alpha \geq 0.70$); and (6) item-total correlations between 0.20 and 0.79. In addition, solutions were examined with respect to psychological theory and previous research. A 'general factor' (i.e. single latent dimension) solution which adhered to all *a priori* criteria was indicated. Twenty-four out of 25 items achieved salient loadings ranging from 0.46 to 0.80 on the general factor. The single item which failed to achieve salience, 'deviant grooming-skin picking', exhibited both substantial unique variance (0.997) and moderate reliability ($r = 0.59$, $P < 0.001$). The internal consistency of the general factor was strong ($\alpha = 0.93$) and all salient items were suitably correlated with the unit-weighted total score ($r_{\text{item-TVtotal}} = 0.41\text{--}0.77$). The traditional CBC scoring system, which includes tallying the number of categories represented, would not be relevant in this PWS sample. In addition, the

recommended tallying of the number of individual behaviours endorsed does not reflect the empirically indicated notion of compulsive behaviour in this special population. These findings indicate that the 24 salient items should be scored as a unit-weighted composite and that the score on the substantially unique item (skin picking) should be considered a separate measure when evaluating compulsive behaviours via the CBC in people with PWS.

Keywords

Compulsive Behaviour Checklist; factor analysis; item analysis; obsessive-compulsive disorder; Prader-Willi syndrome

Introduction

The re-evaluation of an instrument's psychometric properties, including its item characteristics and factor structure, may be warranted when applying it to a special population (Anastasi 1988). Independent of considerations concerning the relevance (or practicality) of establishing separate norms, the identification of population-specific variations in an instrument's psychometric properties, particularly those which impact its scoring and consequent interpretation, are relevant when investigators are interested in examining prevalence, correlating findings with other relevant variables or testing for between-group effects.

Prader-Willi syndrome (PWS) is a genetic disorder characterized by infantile hypotonia and feeding difficulty, hypogonadism, early onset of obesity secondary to hyperphagia, mild intellectual disability, short stature, a particular facial appearance, and a paternal chromosome 15q11-q13 deletion in the majority of cases (Butler *et al.* 1986; Butler 1990; Thompson *et al.* 1996). The prevalence of obsessive-compulsive disorder (OCD) among people with mild to profound intellectual disability has been reported to be 3.5% (Vitiello *et al.* 1989). People with PWS have been reported to have non-food-related compulsions, as evaluated using the Yale-Brown Obsessive Compulsive Scale (Y-BOCS; Goodman *et al.* 1989a,b), which are comparable in both number and severity to an age- and gender-matched comparison group with diagnosed OCD (Dykens *et al.* 1996). In addition, statistically significant elevations in the rate of several types of compulsive behaviours, including checking, hoarding, and the need to tell and/or ask, were observed in the PWS group in comparison to the OCD group (Dykens *et al.* 1996).

The Compulsive Behaviour Checklist (CBC; Gedye 1992) was originally based on an investigation of 15 adults with developmental disability and moderate to profound intellectual disability who met the DSM-III-R (APA 1987) criteria for OCD. The instrument was designed to gather observable information about compulsive behaviours (not to diagnose OCD) through either personal interviews with or completion by caregivers familiar with the person with disability. Gedye (1992) describes the '25 types of compulsions' which comprise the checklist as having been grouped 'loosely into five known categories'. The CBC is scored in two ways by tallying: (1) the number of compulsive behaviours endorsed; and (2) the number of categories represented. In addition, information is collected

concerning the degree to which compulsions interfere with daily living and the client's response to a caregiver's interruption of the behaviours.

Bodfish *et al.* (1995) described the rate of CBC-based prevalence of compulsions among 210 people with severe or profound intellectual disability who resided in a public residential facility as 40%. Among those with any compulsion, the number (mean $\pm$ SD) of CBC items endorsed was 2.6 ± 2.5 and 59% of subjects had items endorsed from more than one category of compulsions (Bodfish *et al.* 1995). The CBC has also been employed to characterize the prevalence of compulsions among people with intellectual disability who exhibit self-injurious behaviour (SIB) (Powell *et al.* 1996). However, a detailed item analysis of the CBC which includes an empirical examination of the theoretical behaviour groupings through latent variable modelling has not been reported. This being the case and in recognition of the fact that people with PWS may exhibit some specific patterns of compulsive behaviour (Dykens *et al.* 1996), the present investigation examines the latent variable (factor) structure of the CBC in people with PWS, and derives an empirically based approach to its scoring and consequent interpretation in this special population.

Materials and methods

Subjects

The present research was approved by the Institutional Review Board of Vanderbilt University. The subjects included 31 males and 44 females with PWS ranging in age from 4 to 41 years (mean $\pm$ SD = 11.4 ± 9.4). Fifty-three PWS subjects (71%) had a paternally derived deletion in the 15q11–q13 chromosome region, 12 (16%) had maternal uniparental disomy of chromosome 15 (both from the mother), and 10 (13%) had PWS but were without conclusive information concerning the genetic subtype. The IQ scores, which were available for 26 subjects, ranged from 46 to 75 and averaged 61.2 ± 7.6 , which is consistent with the level of cognitive disability that is typically associated with PWS.

Instrumentation and data acquisition

The 25 behaviours which comprise the CBC compulsions and their five associated groupings (Gedye 1992) are presented in Table 1. Item abbreviations were created for reference within this report. This reliability and validity of the instrument have been evaluated in 210 people with intellectual disability (Bodfish *et al.* 1995). The interobserver and test-retest reliability, expressed as the average 'occurrence reliability' across rater pairs and testing occasions, were 84.8% and 83.3%, respectively. Validity, expressed as the percentage of cases where a CBC-identified behaviour was directly observed during a predefined period of time, was 91.4% (Bodfish *et al.* 1995).

An ordinal scoring system which reflected a continuum of severity was employed in lieu of the dichotomous CBC checklist approach (i.e. behaviour presence or absence): (0) none, (1) mild, (2) moderate and (3) severe. The ordinal scoring system employed in the present investigation, which differs only slightly from that of the Y-BOCS in that it does not include a fifth category (i.e. 4 = extreme), has been used to evaluate impulsive-aggressive and compulsive symptoms similar to those of the CBC in people with PWS (Stein *et al.* 1994).

The internal-consistency reliability of the instrument using the ordinal scoring system, as evaluated by the Spearman Brown measure, was 0.84.

The CBC was administered to the parents or other primary caregivers of the people with PWS as one aspect of two research projects addressing the biobehavioural characteristics associated with the disorder. Respondents were either interviewed ($n = 26$) or completed a copy of the checklist ($n = 49$). Both mechanisms are recommended for this instrument (Gedye 1992) and the latter has been employed in previous research (Bodfish *et al.* 1995; Powell *et al.* 1996).

Statistical methods

The subject:variable ratio was 3:1, which is appropriate for exploratory purposes given the observed average item intercorrelation coefficient (Baggaley 1982). The use of an ordinal scoring system, as opposed to a dichotomous measure, helped to ensure that potential latent variable solutions would not be compromised by the presence of ‘difficulty’ factors (e.g. McDonald & Ahlawat 1974), i.e. artifacts resulting from non-linear regression of items upon latent dimensions (Mislevy 1986). Bartlett’s (1950) sphericity test and Kaiser’s (1974) measure of sampling adequacy were determined to rule out the likelihood of an identity matrix (which would preclude item factoring because of the absence of item intercorrelations) and to test the likelihood that latent dimensions existed, respectively.

The item data were then analysed via factor analysis with principal component extraction and an orthogonal (equamax) rotation (see Gorsuch 1983). The most suitable solution was determined using multiple empirical criteria (McDermott 1984, 1993; Feuerer 1997). The extracted components: (1) were evaluated with respect to Cattell’s (1966) scree test and related recommendations concerning general factors (Gorsuch 1983; A. R. Baggaley, personal communication); (2) achieved pre-rotated eigenvalues > 1.00 ; (3) comprised at least six items with rotated loadings of > 0.30 (Boyle 1985); (4) exhibited clarity of item assignment to a single latent dimension; (5) exhibited coefficients $\alpha \geq 0.70$ for unit-weighted total scores; and (6) comprised items which exhibited item-total correlations between 0.20 and 0.79 (McDermott *et al.* 1996). An additional consideration was whether the solution made ‘psychological sense in terms of parsimonious coverage of the data and compatibility with previous research’ (McDermott 1984; p. 19).

Results

The summary statistics are presented as the mean $\pm$ SD. The sphericity test was statistically significant ($P < 0.001$) and Kaiser’s (1974) measure of sampling adequacy equalled 0.82. Both findings supported proceeding with latent variable analyses. The item endorsement rates and item skewness (which averaged 1.9 ± 0.9 and ranged from 0.7 to 3.6) demonstrated that the preponderance of data were distributed at the lower end of the severity continuum (Table 1). The scree plot of eigenvalues suggested that two components could be extracted and the eigenvalue > 1.00 criterion suggested that up to five components could be extracted. The five-component and two-component solutions accounted for 68% and 51% of the total item variance, respectively. However, the proportions of item covariance (eigenvalue/total number of items) associated with the (pre-rotated) first and second components were 41%

and 10%, respectively. This precipitous reduction in the explained proportion of covariance, wherein the first latent dimension accounted for greater than 20% and the second accounted for less than half of the first, suggests that a single component (i.e. general factor; Gorsuch 1983) solution would be appropriate (A. R. Baggaley, personal communication).

The magnitude and pattern of item loadings of the five- and two-component solutions did not satisfy the criterion of clarity of item assignment to a single latent dimension. Sixteen items (64%) achieved salient loadings on ≥ 2 components in the five-component solution and the loading pattern did not correspond to the five theoretical item groups. Eight items (32%) were salient on both dimensions of the two-component solution and one variable (GROOM-A, i.e. 'skin picking') was not salient on either dimension. Twenty-four out of 25 items achieved salience (range = 0.46–0.80) on the general factor solution, with GROOM-A being the only item which was clearly not salient (loading = 0.06). The averages of the salient loadings for dimensions one and two of the two-component solution were 0.58 ± 0.17 and 0.61 ± 0.16 , respectively. The average of the salient loadings on the general factor solution was 0.65 ± 0.08 . In addition, the content of the general factor solution (absent GROOM-A) is consistent with the theoretical notion that these items are measures of a single construct (i.e. compulsive behaviours). The general factor solution was considered to be superior for these reasons and additional item characteristics were evaluated from the perspective of a 24-item, unit-weighted composite (with GROOM-A being evaluated as a unique item unto itself). The item loadings and internal consistency measures of the preferred, general factor, solution are presented in Table 2.

Given that GROOM-A was the only item that did not load on the general factor solution, its scoring distribution was inspected to determine whether it was an outlier from that perspective. Figure 1 illustrates that, while it had the highest overall endorsement rate of any item (52% of subjects exhibited the behaviour at some level of severity), its skewness (0.7) was identical to and its overall endorsement rate was comparable to that of item CLEAN-F (47% overall endorsement). This comparably endorsed and skewed item, CLEAN-F (hoarding), was strongly associated (loading = 0.57) with the general factor. GROOM-A was clearly not associated with the general factor (loading = 0.05). Thus, endorsement rates across the severity continuum do not explain the finding that GROOM-A did not load on the general factor.

The uniqueness (i-communality), item-total correlation and coefficient α after deleting a given item are presented for the general factor solution in Table 2. All salient items achieved item-total correlations between 0.41 and 0.77. The observed impact on internal consistency (coefficient α) resulting from deleting the various items in turn was consistent with GROOM-A not being salient on the general factor in that it was the only item which resulted in an increase in α after its deletion. Thus, coefficient α was 0.93 for the 24-item composite. Given Gorsuch's (1983; p. 360) recommendation that a variable exhibiting low communality (i.e. high uniqueness) coupled with high reliability may be an important measure unto itself, the reliability of GROOM-A was evaluated by correlating it with a question (which had been included on a general clinical history survey) addressing whether skin picking was a problematic behaviour. The resultant point-biserial correlation coefficient was 0.59, which

was statistically significant ($P < 0.001$) given the number of observations in the present sample.

Discussion

The present investigation demonstrated that compulsions, as measured by the CBC (Gedye 1992) in people with PWS, constitute a unidimensional set of behaviours with the exception of one item, 'picks at face/hands/legs to point of gouging skin'. The five theoretical CBC item groupings were not evidenced via latent variable analysis. The implications for scoring the CBC in people with PWS are threefold:

1. Tallying the 'number of categories represented' (Gedye 1992) would not be meaningful in these people with PWS.
2. A general notion of compulsive behaviour is implicit in the recommended tallying of the 'number of types of compulsions' (i.e. individual items) endorsed (Gedye 1992). This approach, with the additional recommendation to exclude GROOM-A ('skin picking') from the compulsions composite, was supported by the present investigation.
3. The substantial unique item variance and preliminary evidence of the reliability of GROOM-A support its being treated as a separate item unto itself.

The content validity of the general factor solution is supported by the finding that, independent of its being the only solution which adhered to all *a priori* empirical criteria, it reflects (with the exception of one substantially unique item) the clinical perspective that the CBC items are examples of compulsive behaviours. Evidence in support of item GROOM-A ('skin picking') being considered unique in people with PWS may be drawn from several mathematically based findings. The proportion of unique item variance, i.e. that which is not attributed to the extracted latent dimension(s), is necessarily greater than 0.50 in a general factor solution unless an item loading is ≥ 0.70 . However, the substantial unique variance associated with GROOM-A ($1 - 0.06^2 = 0.996$) reflects the distinctiveness of the item within this data set. Furthermore, the empirically based distinctiveness of this item does not appear to be a function of its skewness (i.e. distribution of endorsement across all levels of the ordinal scale). Given that the 'alternate question' reliability of the item was moderate ($r = 0.59$, $P = 0.001$), considering this item to be a measure unto itself would be warranted (Gorsuch 1983) in people with PWS. Including GROOM-A in the tally of behaviours (or unit-weighted composite via the scoring continuum) would result in the dilution or loss of this item's value in terms of quantifying compulsive behaviours, and in tests of association or between-group differences.

A limitation of the present research is that age (which ranged from 4 to 41 years) did not follow a normal distribution and the preponderance of PWS subjects were younger children. However, it has been noted that behaviour problems that are identified in the pre-school years often persist through adolescence and adulthood in people with PWS (Sulzbacher *et al.* 1981). Given the sample size requirements, an evaluation of the concordance among factor structures across age-based subsamples could not be conducted. However, the intent of the instrument (i.e. the identification of *atypical* behaviours) was emphasized to all respondents

and the use of an ordinal scoring system helped to limit any potential threat to validity at the item level. In addition, it has been demonstrated that CBC-based compulsive behaviours are significantly increased among the younger PWS children included in the present investigation in comparison to typically developing children of comparable age (Dimitropoulos 1998).

Dykens *et al.* (1996) reported a notable prevalence of compulsions via the Y-BOCS in people with PWS in a comparable age range (5–47 years). Leckman *et al.* (1997) evaluated the latent variable structure of the Y-BOCS in subjects with OCD who did not have PWS and the instrument, which includes items characterized as obsessions as well as compulsions and does not include a specific item addressing skin picking, comprised four dimensions (obsessions and checking, symmetry and ordering, cleanliness and washing, and hoarding). The hoarding dimension included two salient items: hoarding obsessions and hoarding compulsions (Leckman *et al.* 1997). McKay *et al.* (1995) evaluated the 10-item severity scale of the Y-BOCS via confirmatory factor analysis in a non-PWS sample of people with OCD and found it to comprise two latent dimensions (obsessions and compulsions) as opposed to its exhibiting a unidimensional or hierarchical structure. In addition to the fundamental consideration that the CBC and Y-BOCS are different instruments, the findings of the present study cannot be directly compared to those of either McKay *et al.* (1995) or Leckman *et al.* (1997) because: (1) the CBC only addresses directly observable compulsions; (2) the CBC only includes one item addressing hoarding behaviour; (3) the Y-BOCS does not include an item specific to skin picking; and (4) McKay *et al.* (1995) did not target the individual Y-BOCS obsessions and compulsions in their analyses.

Bodfish *et al.* (1995) demonstrated a significant association between the overall CBC score and self-injury in people with severe or profound intellectual disability. However, the above authors did not report specific item data, and their sample did not include people with either PWS or mild intellectual disability. Skin picking is the only CBC item which specifically addresses a SIB. (The hair pulling item encompasses inappropriate hair cutting.) The present authors' finding that skin picking was a substantially unique item raises the question of whether it (and possibly SIB in general) may comprise different mechanisms and processes than those involved in other compulsive behaviours in people with PWS. Previous studies have demonstrated that skin picking is a prominent characteristic of PWS (e.g. Clarke *et al.* 1989; Dykens *et al.* 1992; Hellings & Warnock 1994). Whitman & Accardo (1987) found skin picking to be a problematic behaviour for 69% of a sample of adolescents with PWS and Stein *et al.* 1994) reported the presence of the behaviour among 75% of 369 people with PWS. Skin picking is often compared with other forms of SIB within populations with intellectual disability. These behaviours often lead to infection, internal injury, tissue damage or vision loss (Reber & Borcharding 1997) The mechanisms which drive SIB are not well understood, although several neurotransmitters are thought to be involved. One approach to the treatment of SIB involves serotonin replacement through the administration of selective serotonin reuptake inhibitors. Hellings & Wamock (1994) reported a significant decrease in skin picking after administering such agents to two people with PWS.

In summary, the present investigation demonstrated that CBC-based compulsions in people with PWS reflect a unidimensional latent variable structure which is comprised of 24 out of

25 potential compulsive behaviours. The theoretical CBC behaviour groupings were not identified in this sample. The present authors' recommendations for quantifying CBC-based compulsive behaviours beyond the item level in people with PWS are: (1) to consider 'skin picking' to be a unique measure unto itself; (2) to sum the remaining 24 items into a unit-weighted composite score; and (3) not to consider the number of theoretical categories represented to be a valid measure in this special population.

Acknowledgements

This research was supported in part by grants PO1 HD30329 and P30 HD15052 from the National Institute of Child Health and Human Development, and by a grant from the Prader-Willi Syndrome Association of America. This paper was presented in part at the 31st Annual Gatlinburg Conference on Research and Theory in Mental Retardation and Developmental Disabilities, Charleston, South Carolina, USA, 14 March 1998.

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Figure 1. Endorsement rates (per cent of sample) across levels of the severity continuum for 'hoarding' and 'skin picking'. The former loaded on the general factor while the latter was the only item that did not.

Table 1

Compulsive Behaviour Checklist items and theoretical groupings*

Item grouping and abbreviation	Paraphrased item content	Endorsement (%)				Any [†]	Skewness
		Mild	Moderate	Severe			
<i>Ordering compulsions</i>							
ORDER-A	Arranges objects in a certain pattern	20	16	5	41	1.1	
ORDER-B	Arranges certain items in one spot	24	17	5	47	0.9	
ORDER-C	Wants chairs in a fixed arrangement	16	7	5	28	1.9	
ORDER-D	Wants/arranges peers to sit in certain chairs	24	8	5	37	1.5	
ORDER-E	Uses same chair/location in room	23	7	16	45	1.0	
ORDER-F	Insists on activities/chores at a certain time	23	15	3	40	1.2	
<i>Completeness/incompleteness compulsions</i>							
COMPL-A	Insists on closing open doors/cupboards	16	13	3	32	1.5	
COMPL-B	Removes all items from closet, purse etc.	17	9	4	31	1.7	
COMPL-C	Repeatedly removes then replaces items	13	15	5	35	1.3	
COMPL-D	Empties/wants containers emptied	11	3	3	17	3.0	
COMPL-E	Puts garments on and off; hangs up and removes clothes	13	3	1	17	2.9	
COMPL-F	Insists on doing chore; resists letting others	17	16	8	43	1.0	
<i>Cleaning/tidiness compulsions</i>							
CLEAN-A	Fixed hygiene sequence; restarts if interrupted	21	9	3	33	1.6	
CLEAN-B	Cleans body part(s) excessively	17	5	4	27	2.1	
CLEAN-C	Insists on picking up bits; 'lint picks'	17	3	0	20	2.1	
CLEAN-D	Often picks/rips objects if not prevented	17	7	0	24	1.8	
CLEAN-E	Insists certain ('cleaning') activity be done	12	5	0	17	2.3	
CLEAN-F	Hides away, collects/hoards particular objects	15	23	9	47	0.7	
<i>Checking/touching compulsions</i>							
CHECK-A	Repeatedly opens and closes doors/drawers	16	3	1	20	2.6	
CHECK-B	Touches or taps items repeatedly	12	5	1	20	2.5	
CHECK-C	Touching/stepping pattern	7	3	0	10	3.6	
CHECK-D	Unusual sniffing	12	1	7	20	2.5	
<i>Deviant grooming compulsions</i>							

Item grouping and abbreviation	Paraphrased item content	Endorsement (%)			Any [‡]	Skewness
		Mild	Moderate	Severe		
GROOM-A	Picks at face/body to point of gouging skin	20	20	12	52	0.7
GROOM-B	Checks hair, face/excessive mirror checking	8	0	0	8	3.2
GROOM-C	Inappropriately cuts/pulls/calmly pulls out hair	11	1	1	13	3.6

* The items are presented in the order in which they appear in the original instrument (Gedye 1992) and are paraphrased for the sake of brevity. Item abbreviations were created for reference within this report.

[‡]The endorsement rate if behaviours were scored as a dichotomy (present or absent).

Table 2

Item properties of general factor solution

Abbreviated item	Unrotated loading*	Uniqueness	Item-total correlation	α if deleted (versus 0.92)
ORDER-A	0.61	0.62	0.60	0.92
ORDER-B	0.68	0.55	0.69	0.91
ORDER-C	0.77	0.41	0.75	0.91
ORDER-D	0.67	0.56	0.63	0.92
ORDER-E	0.70	0.51	0.68	0.91
ORDER-F	0.58	0.67	0.54	0.92
COMPL-A	0.66	0.54	0.64	0.91
COMPL-B	0.65	0.57	0.60	0.92
COMPL-C	0.70	0.51	0.43	0.92
COMPL-D	0.69	0.59	0.65	0.92
COMPL-E	0.55	0.70	0.46	0.92
COMPL-F	0.80	0.36	0.77	0.91
CLEAN-A	0.52	0.73	0.47	0.92
CLEAN-B	0.63	0.60	0.61	0.92
CLEAN-C	0.69	0.52	0.62	0.92
CLEAN-D	0.60	0.64	0.56	0.92
CLEAN-E	0.71	0.50	0.62	0.92
CLEAN-F	0.57	0.68	0.56	0.92
CHECK-A	0.73	0.47	0.64	0.92
CHECK-B	0.74	0.45	0.45	0.92
CHECK-C	0.53	0.72	0.47	0.92
CHECK-D	0.67	0.56	0.63	0.92
GROOM-A	0.06	10.00	0.05	0.93
GROOM-B	0.59	0.65	0.51	0.92
GROOM-C	0.46	0.79	0.41	0.92

* Rotation is not applicable when a single component is extracted.