The assessment of food related problems in individuals with Prader-Willi syndrome.

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Abstract

The development of a twenty item questionnaire, the Food Related Problem Questionnaire (FRPQ), with four subscales (preoccupation with food, impairment of satiety, difficulty with self-control and other food-related 'challenging' behaviour of food-related problems) for use with individuals with Prader-Willi syndrome (PWS) is described. Test-retest and inter-rater reliability for the total score are .78 and .86 respectively. Subscale test-retest and inter-rater reliability are acceptable for three subscales (range .70 to .85) but not for the self control subscale. To establish criterion validity, scores on the FRPQ for a sample of individuals with PWS were compared to scores for individuals with intellectual disability but not PWS. All reliable subscale scores and the total score differentiated between the groups (p<.001 in all cases). Internal consistency for the total scale is good (Cronbach’s Alpha = .87). Finally, some normative data for a limited age range are presented and potential applications of the measure are discussed.
Assessing food related problems

Introduction

There is increasing interest in the concept of behavioural phenotypes within the field of intellectual disability. There is little doubt that in some syndromes specific behaviours are more prevalent than might be expected by chance. Three well documented examples are self-biting in Lesch-Nyhan syndrome (Christie et al., 1982), stereotyped hand-wringing movements in Rett syndrome (Kerr & Stephenson, 1985) and excessive laughing and smiling in Angelman syndrome (Summers et al., 1995). Psychology has an important role to play in this emerging area. For example, examining the effects of the environment on behaviours purported to be part of a behavioural phenotype (see: Hall et al., 2001; Oliver et al., 1993, 2002) and the influence of the concept of behavioural phenotype on the attributions of others (see: Hyman & Oliver, 2001). Additionally, it is important that relevant measures with robust psychometric properties are developed to accurately document behavioural phenotypes and examine with syndrome variability.

Excessive eating (hyperphagia) is a significant feature of Prader-Willi Syndrome (PWS; Holm et al., 1993) and first becomes apparent between the ages of two and six years (Cassidy, 1984; Ehara et al., 1993). Initially, a child's increased appetite may be welcomed by parents, as prior to this, hypotonia (low muscle tone) may have contributed to feeding difficulties and 'failure to thrive' (Greenberg et al., 1987). However, over time a child's interest in food may become debilitating, both for the young person with PWS and for those around them (Greenswag, 1987; Clarke et al., 1995) with two primary consequences of overeating described: obesity and behavioural problems.

Gross obesity has been widely reported in children and adults with PWS if overeating is left unchecked (Butler, 1990; Holm et al., 1993) and individuals are noted to require a lower calorie diet than individuals without the syndrome to stay within normal weight limits (Pipes & Holm,
Assessing food related problems

1973). Decreased physical exercise and lowered energy expenditure have been identified as exacerbating the problem (Butler, 1990; Davies & Joughin, 1993). Physical health problems associated with weight gain have been reported including: diabetes mellitus (Laurance et al., 1981), coronary artery disease (Lamb & Johnson, 1987) and premature death (Laurance et al., 1981). The psychological impact of being overweight may also be detrimental, contributing to difficulties in forming relationships with peers (Dykens et al., 1992) and in turn affecting an individuals' self-worth (Wolfensberger, 2000).

An impaired satiety mechanism has been postulated as responsible for overeating (Laurance, 1993; Holland, 1998) and Dykens et al. (1997) suggests that hyperphagia is a biologically-driven behaviour. Support for a biological basis to voracious eating behaviour has been offered by Holland et al. (1995) who found that individuals with PWS required a larger quantity of food and had greater difficulty recognising when they were satiated compared with a control group. The notion of overeating as a physiologically-driven behaviour is also evident in the ways that a person's food intake is managed. External controls such as locks on fridge doors and carers controlling size of meals have been suggested as effective strategies (Whitman & Greenswag, 1995) and appetite-suppressive medications have been prescribed (Dech & Budow, 1991), although with mixed results (Jun-Bi Tu et al. 1992). However, these approaches to food management have been criticised. Gordon & Clarke-Bruyn (1991) propose that if excessive eating is viewed as an inevitable consequence of PWS, carers may be reluctant to intervene and individuals with PWS may be stigmatised (Levine, 1995). Furthermore, restricting a person's access to food is controversial and has fuelled a debate about the degree to which a person may be viewed as an autonomous individual capable of making food-related decisions (Dykens et al. 1997; Holland & Wong, in press).
A range of food-related behavioural difficulties are also reported in the PWS literature, including taking food from others (Donaldson et al., 1994), stealing money to purchase food (Sherman, 1995), hoarding food items (Dykens et al., 1996) and lifting a kitchen door from its hinges in order to gain access to food (Benjamin & Buot-Smith, 1993). The ways in which individuals seek to obtain food are also described as ‘manipulative’ and 'ingenious' (Donaldson et al. 1994). However, these descriptions are based on anecdotal reports of a few people with PWS. How characteristic such behavioural difficulties are of the broader PWS population is unknown. More recently, Sarimski (1996) systematically investigated food-related behavioural difficulties in a group of twenty-eight children with PWS using the 'Children's Eating Behaviour Inventory' (CEBI; Archer et al., 1991). Sarimski (1996) reported that 'food-seeking behaviours', such as asking for food between meals and hiding food, characterised children with PWS in comparison to children with Williams-Beuren syndrome, although significant variability in food-seeking behaviours of children with PWS was also found. This study employed a relatively small cohort of children within a limited age range and employs an eating behaviour measure used with children from the general population. Food-related behaviours specific to individuals with PWS may therefore be omitted. Consequently, the findings of this study might not comprise the full range of eating-related problems.

Excessive eating has also been described as a feature of people who binge eat (Beglin & Fairburn, 1992; Fairburn & Wilson, 1993; McManus & Waller, 1995) and is reported as a characteristic of Kleine-Levin syndrome (Critchley, 1962) and Tourette's syndrome (Ballard, 1992). A number of measures have been used to assess aspects of eating in the broader eating disorder population (eg. 'Eating Attitudes Test', Garner et al., 1982; 'Mizes Anorectic Cognitions Questionnaire', Mizes & Klesges, 1989; 'Eating Disorders Inventory', Garner, 1991). However, these measures are reliant on
self-report and also do not incorporate all aspects of eating behaviour described in individuals with PWS. They are therefore of limited use within the PWS population.

Clearly, a need exists to systematically investigate the eating behaviour of individuals with PWS. This would be beneficial both in examining global statements of food-related problems associated with PWS, the relationship between food-related problems and other behaviours and characteristics and would offer a measure of treatment efficacy. The principal aim of the current study is to describe the development of a questionnaire for measuring food-related problems in adults with PWS. Test-retest and inter-rater reliability for the questionnaire are reported. As a measure of validity the extent to which the questionnaire is able to differentiate between food-related problems characteristic of individuals with PWS versus individuals from the wider intellectual disability population is reported and internal consistency of the questionnaire is assessed.

**Method**

**Development of the Food Related Problems Questionnaire**

To ascertain the range of difficulties regarding food-related issues, semi-structured interviews were conducted with parents and carers of 12 individuals with PWS, of whom 6 (50%) were female. Families of individuals with PWS were identified via the Prader-Willi syndrome Association (PWSA, UK) and were all members of a local support group. Mean age of females was 9.8 years (age range 8 - 21 years) and mean age of males was 17 years (age range 8 -24 years). During the semi-structured interviews, which focussed on all aspects of food-related behaviour, individual problems reported by informants were noted. Following the interviews, recorded individual problems were clustered according to themes and sixteen questions which captured the breadth of
the difficulties described were constructed. Although no formal reliability analysis of the reported problems was conducted, clustering by themes was achieved by consensus.

To derive subscales the twenty items were grouped under different elements of food-related issues and operational definitions derived for each of these elements. To assess the reliability of the allocation of items to subscales a psychologist then classified each question using the operational definitions provided. 90% inter-rater agreement was established. A second psychologist repeated this procedure and 100% inter-rater agreement was obtained. The operational definitions and principal subscales generated with questions allocated to each subscale are shown in Table 1.

[Insert Table 1 here]

For each item a 7 point Likert scale response format was used with responses ranging from 0 = 'never' to 6 = 'always.' A further response 'does not apply' was included for six items that asked about verbal responses. In addition, three open-ended questions were included that asked about: external control, evidence of self-control and other comments. The maximum possible score for the total FRPQ is 96. The maximum scores for each subscale are shown in Table 1.

Participants

Three groups of participants were recruited (PWS specialist residential group, comparison residential group and PWS community group). The PWS specialist residential group and the comparison residential group were employed for analyses examining inter-rater and test-retest reliability and criterion validity. Participants were drawn from a sample of 65 individuals with intellectual disabilities living in one of six staffed community-based homes. All six homes were located within the same geographical area and existed under one umbrella organisation.
Participants were either: adults with a diagnosis of Prader-Willi syndrome (*PWS specialist residential group*), or adults with intellectual disabilities without PWS (*comparison residential group*). The residential homes were designed around the specific needs of adults with PWS; in particular, food was locked away and the quantity of food restricted. Consent to participate in the study was gained from 38 (58.5%) individuals: 24 (72.7%) adults with PWS and 14 (43.7%) adults with intellectual disabilities without PWS. Prior to analysis questionnaires were screened to identify and exclude potential participants scoring 7 (‘does not apply’) on two or more items that asked about verbal responses. This was done to avoid a low score as a result of limited verbal capacity. The screening procedure led to the exclusion of one individual with PWS and one individual with intellectual disabilities without PWS.

Demographic information was collected concerning the respondent's relationship to the participant, sex and age of the individual, daytime activities and prescribed medication. Descriptors ranging from 'profound' to 'superior' were included to gauge level of intellectual functioning. Keyworkers were also asked about the age at which a diagnosis was made and how this was ascertained.

*PWS Specialist Residential Group.* The mean age of individuals with PWS (*N* = 23) was 27.7 years (SD = 6.5; range 19 to 38.9 years) and 8 (34.8%) participants were female. 10 (43.5%) adults attended college and 7 (30.4%) adults attended onsite workrooms. In the month prior to the study, a range of medication had been prescribed. 11 (47.8%) participants had been prescribed antipsychotic medication and 7 (30.4%) participants had been prescribed antidepressant medication. Insufficient information was available regarding level of intellectual functioning. All participants had received a clinical diagnosis of PWS.
Comparison Residential Group. The mean age of the *residential comparison group* (N = 12) was 43.1 years (SD = 5.2; age range 33 to 51.9 years) and 5 (41.7%) were female. 4 (33.3%) participants attended college and 6 (50%) attended onsite workrooms. 8 (66.7%) of individuals had been prescribed antipsychotic medication and 3 (25%) had been prescribed antidepressant medication. Insufficient information was available regarding level of intellectual functioning.

The *PWS community group* were recruited to increase the sample size in order to evaluate internal consistency and concurrent validity.

Recruitment Procedure

As part of a larger study (Russell and Oliver, submitted) 99 questionnaire packs were sent to carers of individuals with PWS in the United Kingdom. Questionnaires were sent by the Prader-Willi Syndrome Association (PWSA, UK). Carers identified via an earlier postal questionnaire as being willing to participate in future research were contacted. The questionnaires consisted of questions regarding demographic details, three standardised measures and the FRPQ developed in the current study. Included in each pack was a covering letter, participant information and a reply paid envelope. As in study 1, prior to analysis questionnaires were screened to identify and exclude potential participants scoring 7 (‘does not apply’) on two or more items that asked about verbal responses. This was done to avoid a low score as a result of limited verbal capacity. The screening procedure led to the exclusion of two individuals with PWS from 60 respondents.

*PWS Community Group.* 58 individuals with Prader-Willi syndrome were recruited via the Prader-Willi Syndrome Association (PWSA, UK), a parent support group for children diagnosed with PWS. 45 (77.6%) questionnaires were completed by the mother, 7 (12.1%) by the father, 1 (1.7%) by both parents and 5 (8.6%) by another person which included residential care staff and foster carers. Of the 58 participants identified, 31 (53.4%) were female and the total sample ranged in age
Assessing food related problems

from 14.4 to 22.1 years (mean = 18.3, SD = 2.19). The age when the individual was diagnosed with PWS ranged from birth to 17 years (mean = 4.1, SD = 4.7). For 28 participants (48.3%) diagnosis was made based on clinical symptoms alone, 21 (36.2%) following genetic testing and 5 (8.6%) following both procedures. Method of diagnosis was not stated for the remaining 4 participants.

34 (58.6%) carers stated that the individual with PWS had completed an assessment of intelligence and of those whose IQ score was known (N = 27), participants were reported to fall within the mild range of intellectual disabilities. 25 (43.1%) participants attended college and 15 (25.9%) attended a special school. The remaining 18 attended a range of daytime facilities. In the month prior to the questionnaire being completed, a range of medication had been described, notably 4 (6.9%) participants had been prescribed antipsychotic medication and 2 (3.4%) participants had been prescribed antidepressant medication.

Design and Data Analysis

Inter-Rater and Test-Retest Reliability

Two independent groups of raters completed the FRPQ to assess inter-rater reliability. The General Manager of the group homes distributed the FRPQ to the named keyworker of the 35 individuals in the PWS specialist residential group and residential comparison group and to a second carer known to the individual. To ensure that questions were answered independently, respondents were instructed not to confer with other carers. Test-retest reliability was assessed by asking the named keyworker to complete the FRPQ on a two occasions separated by a two to three-week period. To ensure that the second ratings were completed without reference to earlier responses, questionnaires completed at time one were collected prior to distribution of the questionnaires at time two.

Criterion Validity, Concurrent Validity and Internal Consistency
To evaluate the validity of the FRPQ, two strategies were adopted. First, to evaluate criterion validity, subscales and total score were compared for individuals with in the PWS specialist residential group and the residential comparison group residing in the same community-based group homes. Second, to partially evaluate concurrent validity, responses for relevant subscales on the FRPQ were compared with responses to similar question items on the Checklist of Challenging Behaviour (CCB; Harris, Humphreys & Thomson, 1994).

Results
Prior to analysis questionnaires were examined to evaluate the number of participants for whom ‘does not apply’ was indicated. In the PWS specialist residential group this was indicated for two participants on one item, for the residential comparison group this was indicated for one participant on one item. The results were analysed for each pair of raters (inter-rater reliability) and the same raters on two separate occasions (test-retest reliability) using Pearson correlation coefficients. The mean coefficient for inter-rater reliability at item level was .53 (range .05 to .91) and the mean coefficient for test-retest reliability at item level was .57 (range -.01 to .93). The inter-rater and test-retest reliability levels for subscales and total score of the FRPQ are presented in Table 2.

Inter-rater and test-retest reliability varied across subscales. Adequate inter-rater reliability was established for all of the subscales (r = .56 to .89) and adequate test-retest reliability was established for six out of the seven subscales (r = .64 to .85).

Criterion validity
In order to examine whether the FRPQ was able to distinguish between distinct groups, scores obtained for individuals with PWS were compared with those with an intellectual disability without the syndrome. Both cohorts resided within the same community setting; thus it was hypothesised that any differences noted between groups may be attributed to the degree of food-related problems, rather than being attributable to environmental factors. Independent samples t-tests were employed and the results are summarised in Table 3.

+++++++++++ Insert Table 3 here +++++++++++++

In Table 3 it can be seen that the mean scores for the two groups differ on all subscales and total score, with the PWS cohort scoring consistently higher than the comparison group on all subscales.

**Concurrent validity**

A second approach to validating some items of the FRPQ was to examine the level of correspondence between responses to items on the questionnaire with similar items on an existing measure. The Checklist of Challenging Behaviour (CCB; Harris, Humphreys & Thomson, 1994) was used for this purpose. The CCB consists of two parts with items about aggressive behaviours and self-injury (Part I) and other challenging behaviours (Part II). Named keyworkers involved in development of the FRPQ completed the CCB when providing demographic information and rating the FRPQ for the first time. For the purposes of validation, two categories forming part of the subscale 'composite negative behaviour' of the FRPQ were compared with responses to two statements on the CCB. An independent samples t-test was carried out to compare scores on the subscale 'takes/stores food' (comprising items 2, 4 and 8 on the FRPQ) of participants identified as: 'Taking food or drink from others?' on the CCB with those who were not. A significant difference on the FRPQ \((t (52) = 4.6, p < .0001)\) was found between those reported to take food or drink on
Assessing food related problems

the CCB (mean = 13.3, SD = 3.6) and those reported not to take food or drink on the CCB (mean = 7.7, SD = 4.8). A second independent samples t-test was carried out to compare the scores on the subscale 'eats inedible items' (comprising items 12 and 15 on the FRPQ) on the CCB item: 'Eating inappropriate things (eg rubbish, faeces, dangerous objects).' A significant difference on the FRPQ (t (51) = 5.39, p < .0001) was found between those who were reported to show pica on the CCB (mean = 6.7, SD = 2.6) and those for whom pica was not reported on the CCB (mean = .85, SD = 1.3).

Internal Consistency

Independent samples t-tests were carried out to compare scores of individuals with in the PWS specialist residential group with individuals in the PWS community group (see Table 4). This was undertaken to establish whether results obtained during development of the measure were specific to the specialist residential group or typical of the broader PWS population. As can be seen in Table 4, the two groups did not significantly differ on the subscales: impairment of satiety and composite negative behaviour. However, difference on the subscale 'preoccupation with food' was significant (p < .01), with participants in the PWS community group being reported to be more preoccupied with food (mean = 10.63, sd = 3.96) than participants living in PWS-designed group homes (mean = 8.1, sd = 2.8).

+++= Insert Table 4 here +=+=

To establish internal consistency, Cronbach's alpha coefficient was calculated. Participants from all three groups were included in the analysis. Nunnally (1978) states the generally accepted standard for reliability estimates to be above .70 (Huck & Cormier, 1996). In this study, Cronbach's alpha
A coefficient of .87 was determined for the total score of the FRPQ, suggesting good internal consistency.

**Discussion**

The study presents reliability and validity data on an informant-based measure of food-related problems in adults with PWS. Adequate inter-rater and test-retest reliability were demonstrated for all of the subscales. Several items within subscales were found to be unreliable. However, the purpose of each subscale was to measure a specific construct. Hence, reliability for subscales need only be measured. Therefore, it is important that the results of the FRPQ are only considered at the subscale level and not at the item level. The measure was also found to be suitable for assessing food-related problems in people with PWS regardless of setting and to successfully differentiate individuals with PWS from those with intellectual disabilities (criterion validity). Finally, good internal consistency was demonstrated.

This study is a preliminary investigation of difference regarding food-related problems in two distinct groups. Individuals with PWS scored higher on the FRPQ than individuals with intellectual disabilities without the syndrome. However, significant variability in aspects of food-related problems were found between groups. Noticeably, individuals with intellectual disabilities scored significantly lower on subcategories of the subscale 'composite negative behaviour' compared with the PWS group and no behavioural difficulties were reported for some people. These findings are consistent with those reported by Sarimski (1996) in a study investigating 'food-seeking behaviours' in children with Prader-Willi and Williams-Beuren syndrome. Further studies are needed to delineate aspects of food-related problems that characterise individuals with PWS (i.e. syndrome-specific behaviours) and food issues that may be problematic for more than one population.
There are a number of limitations to consider regarding the present study. Firstly, the measure was developed on a relatively small cohort of individuals, of limited age range, living in PWS-designed group homes. However, similar results on the FRPQ were obtained for a cohort of individuals living in the community, suggesting that the measure has wider application. Nevertheless, the utility of the FRPQ for assessing food-related problems in younger children and older adults with PWS is unknown. The finding that individuals recruited via the Prader-Willi Syndrome Association (PWSA, UK) and living in the community were more preoccupied with food than those resident in PWS-designed group homes is interesting and may reflect a difference in environment; access to food is strictly controlled in the PWS-designed group homes and food is not physically present except at meal times. Also, a daily activity schedule is in place for all individuals residing in this specialist facility. Consequently, all responsibility for food issues have been removed from individuals, thus preoccupation with food may be hypothesised to be lower. Conversely, the environmental setting of individuals with PWS residing in the community was not ascertained. Nevertheless, it is likely that food may be more accessible for these individuals, to be more of a predominant feature in the lives of those around them and less likely to be under such strict control. Finally, some items on the measure that require a verbal response by an individual with PWS, allow the respondent to indicate ‘does not apply’ and this might lower the score. In the samples employed in this study this occurred very infrequently. However, it should be noted that the data presented only apply to participants who were rated as ‘does not apply’ on one or no items.

In conclusion, significant within-syndrome variability in aspects of food-related problems were highlighted for individuals with PWS as measured on the FRPQ, suggesting that food-related issues are more problematic for some individuals with PWS than for others (also Sarimski, 1996). The FRPQ may be useful in clinical practice to assess the relationship between food-related problems and environmental factors and to assess the impact of degree of intellectual disability and effect of
Assessing food related problems

age on food-related issues. The FRPQ may also highlight other behaviours such as obsessive and compulsive characteristics described in individuals with PWS (Hodapp & Dykens, 1995; Stein et al., 1994). Therefore, a potential clinical use of the FRPQ is to aid in the implementation of interventions and to evaluate treatment efficacy. Finally, the measure may be adopted by services to highlight an individual's specific food-related needs and to facilitate appropriate support. In this way, the stigma often attributed to individuals with PWS and food-related difficulties (Levine, 1995) may begin to be challenged.
References


### Table 1. Description of the Food-Related Problems Questionnaire (FRPQ) subscales, operational definitions, item inclusion and maximum possible scores

<table>
<thead>
<tr>
<th>Subscale</th>
<th>Operational Definition</th>
<th>Maximum Possible score</th>
</tr>
</thead>
<tbody>
<tr>
<td>Preoccupation</td>
<td>Hypervigilant with regard to food. Excessive reference (by speech) to food. (e.g. How often does the person talk about food?)</td>
<td>18</td>
</tr>
<tr>
<td>Impairment of satiety</td>
<td>Any indication that the person is not satiated and that this is demonstrated in an appropriate way. (e.g. Does the person ever describe 'feeling full'? )</td>
<td>30</td>
</tr>
<tr>
<td>Composite negative behaviour</td>
<td>Inappropriate behaviour with regard to accessing and storing food. Evidence of the person responding in an inappropriate way when access to food is reduced or evidence for external control being required.</td>
<td>48</td>
</tr>
</tbody>
</table>

Table 1. continued

<table>
<thead>
<tr>
<th>Subscale</th>
<th>Operational Definition</th>
<th>Item number</th>
<th>Maximum Possible score</th>
</tr>
</thead>
<tbody>
<tr>
<td>In particular, evidence that the person:</td>
<td></td>
<td></td>
<td></td>
</tr>
</tbody>
</table>
Assessing food related problems

a) takes food and/or stores food inappropriately (eg. Does the person ever hide or hoard food?)

b) eats inedible items (eg. Does the person ever eat non-edible items (eg. dog food, leaves)?)

c) responds in an inappropriate way when food is not available, or when access to food is restricted (eg. Is the person ever able to accept an explanation given to them if a meal is delayed?)

Table 2. Inter-rater and Test-retest Reliability Levels for Subscales and Total score of the Food-Related Problems Questionnaire

<table>
<thead>
<tr>
<th>Subscales and total score</th>
<th>Inter-rater reliability</th>
<th>$N^a$</th>
<th>Test-retest reliability</th>
<th>$N^a$</th>
</tr>
</thead>
</table>

Assessing food related problems
### Principle subscales

<table>
<thead>
<tr>
<th>Subscale</th>
<th>Score 1</th>
<th>Score 2</th>
<th>Score 3</th>
<th>Score 4</th>
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</thead>
<tbody>
<tr>
<td>Preoccupation with food</td>
<td>.67</td>
<td>35</td>
<td>.70</td>
<td>33</td>
</tr>
<tr>
<td>Impairment of satiety</td>
<td>.71</td>
<td>30</td>
<td>.74</td>
<td>22</td>
</tr>
<tr>
<td>Composite negative behaviour</td>
<td>.82</td>
<td>28</td>
<td>.85</td>
<td>27</td>
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</tbody>
</table>

### Composite negative

#### Behaviour subcategories

<table>
<thead>
<tr>
<th>Behaviour</th>
<th>Score 1</th>
<th>Score 2</th>
<th>Score 3</th>
<th>Score 4</th>
</tr>
</thead>
<tbody>
<tr>
<td>Takes/stores food</td>
<td>.89</td>
<td>34</td>
<td>.92</td>
<td>32</td>
</tr>
<tr>
<td>Eats inedible items</td>
<td>.75</td>
<td>30</td>
<td>.71</td>
<td>28</td>
</tr>
<tr>
<td>Inappropriate response</td>
<td>.56</td>
<td>33</td>
<td>.64</td>
<td>32</td>
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</table>

### Food-Related Problems

#### Questionnaire total score

<table>
<thead>
<tr>
<th>Score 1</th>
<th>Score 2</th>
<th>Score 3</th>
<th>Score 4</th>
</tr>
</thead>
<tbody>
<tr>
<td>.86</td>
<td>27</td>
<td>.86</td>
<td>20</td>
</tr>
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</table>
**Table 3. A comparison of individuals with PWS and individuals with intellectual disabilities residing within a community-based group home setting**

<table>
<thead>
<tr>
<th>Subscales and total score</th>
<th>Prader-Willi syndrome</th>
<th>Comparison Group</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>( N^a )</td>
<td>Mean</td>
</tr>
<tr>
<td><strong>Principal subscales</strong></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Preoccupation with food</td>
<td>23</td>
<td>8.10</td>
</tr>
<tr>
<td>Impairment of satiety</td>
<td>19</td>
<td>21.9</td>
</tr>
<tr>
<td>Composite negative behaviour</td>
<td>17</td>
<td>18.88</td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>Composite negative behaviour subcategories</th>
<th>Prader-Willi syndrome</th>
<th>Comparison Group</th>
</tr>
</thead>
<tbody>
<tr>
<td>Takes/stores food</td>
<td>22</td>
<td>11.95</td>
</tr>
<tr>
<td>Eats inedible items</td>
<td>18</td>
<td>1.78</td>
</tr>
<tr>
<td>Inappropriate response</td>
<td>22</td>
<td>6.45</td>
</tr>
</tbody>
</table>

**FRPQ total score**

<table>
<thead>
<tr>
<th>Prader-Willi syndrome</th>
<th>Comparison Group</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>( N^a )</td>
</tr>
<tr>
<td>FRPQ total score</td>
<td>17</td>
</tr>
</tbody>
</table>

\( a \) Subscale scores cannot be calculated for some participants. Consequently \( N \) varies.

**Table 4. A comparison of individuals with PWS recruited via two sources**
### Prader-Willi syndrome: PWSA (UK)

<table>
<thead>
<tr>
<th>Sub-scales and total score</th>
<th>$N^a$</th>
<th>SD</th>
<th>Range</th>
<th>Mean</th>
<th>$N^a$</th>
<th>SD</th>
<th>Range</th>
<th>Mean</th>
<th>$t$</th>
<th>sig</th>
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</thead>
<tbody>
<tr>
<td><strong>Principal subscales</strong></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Preoccupation with food</td>
<td>56</td>
<td>3.96</td>
<td>2 - 18</td>
<td>10.63</td>
<td>23</td>
<td>2.8</td>
<td>4 - 15</td>
<td>8.1</td>
<td>2.75</td>
<td>&lt; .01</td>
</tr>
<tr>
<td>Impairment of satiety</td>
<td>54</td>
<td>5.32</td>
<td>11 - 30</td>
<td>19.91</td>
<td>19</td>
<td>3.18</td>
<td>15 - 26</td>
<td>21.9</td>
<td>1.93</td>
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<td>Composite negative behaviour</td>
<td>53</td>
<td>9.14</td>
<td>1 - 43</td>
<td>19.38</td>
<td>17</td>
<td>7.92</td>
<td>7 - 40</td>
<td>18.88</td>
<td>0.2</td>
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<td><strong>Composite negative behaviour subcategories</strong></td>
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<tr>
<td>Takes/stores food</td>
<td>57</td>
<td>5.17</td>
<td>0 - 18</td>
<td>9.88</td>
<td>22</td>
<td>3.36</td>
<td>4 - 18</td>
<td>11.95</td>
<td>2.10</td>
<td>&lt; .05</td>
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<tr>
<td>Eats inedible items</td>
<td>56</td>
<td>.63</td>
<td>0 - 11</td>
<td>1.79</td>
<td>18</td>
<td>3.25</td>
<td>0 - 12</td>
<td>1.78</td>
<td>0.01</td>
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<tr>
<td>Inappropriate response</td>
<td>55</td>
<td>3.75</td>
<td>0 - 18</td>
<td>7.82</td>
<td>22</td>
<td>3.97</td>
<td>1 - 14</td>
<td>6.45</td>
<td>1.42</td>
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<td><strong>FRPQ Questionnaire total score</strong></td>
<td>52</td>
<td>15.87</td>
<td>19 – 87</td>
<td>50.13</td>
<td>17</td>
<td>12.03</td>
<td>27 – 79</td>
<td>48.41</td>
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</table>

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* Subscale scores cannot be calculated for some participants. Consequently $N$ varies.
Appendix. Items from the Food-Related Problems Questionnaire. (Each rated on a seven point Likert scale: Never to Always)

1. How often does the person compare the size or content of their meal with others? (P)
2. If given the opportunity, how often would the person ‘help themselves’ to food which they should not have? (N, TS)
3. Is the person ever able to accept an explanation given to them if a meal is delayed? (N, IR)
4. Does the person ever hide or hoard food? (N, TS)
5. How often does the person talk about food? (P)
6. If the person was denied food, how often would they respond negatively? (N, IR)
7. Is it necessary to lock food away to stop the person from taking food? (N, TS)
8. After an normal size meal, how often will the person say they still feel hungry? (S)
9. If the person was tired, ill or upset, how often would this result in them going without food? (S)
10. If it was available, would the person eat food not suitable for consumption (eg. frozen food, scraps from dustbins)? (N, EI)
11. If a meal includes an item of food the person does not like or is not expecting, how often would this result in behavioural difficulties? (N, IR)
12. Does the person ever eat non-edible items (e.g. dog food, leaves)? (N, EI)
13. How frequently will the person share food with others? (S)
14. Does the person ever describe ‘feeling full’? (S)
15. Does the person ever associate people and/or places with specific food items or occasions involving food? (P)
16. If given the opportunity, does the person ever eat more than a standard sized meal? (S)

Legend for subscales: P = preoccupation with food, S = Impairment of satiety, N = Composite negative behaviour; N, TS = Negative behaviour, takes and stores food; N, EI = Negative behaviour, eats inedibles.