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Effectiveness of cognitive-behaviour therapy for hoarding disorder in people with mild intellectual disabilities

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CBT for co-morbid ID and Hoarding

Abstract

Evaluations of cognitive behavioural interventions for hoarding for those with intellectual

disabilities (ID) have not been previously attempted. This investigation therefore examined

the acceptability and effectiveness of cognitive-behavioural therapy (CBT) in a sample of

N=14 adults with mild ID. All participants had hoarding as their primary problem and

received twelve individual CBT sessions, all conducted via domiciliary visits. The primary

outcome measure was an environmental measure (Clutter Image Rating Scale), which was

scored at baseline, end of treatment and at six-month follow-up. Acceptability of CBT was

measured via the treatment refusal and dropout rate. Secondary self-report outcomes

included measures of hoarding, depression and anxiety. Results demonstrate that hoarding

significantly reduced following treatment on both self-report and environmental assessment.

No participants refused or dropped out of treatment and that there was no evidence of relapse

over the follow-up period. No adverse treatment incidences were reported. This open trial

suggests that CBT may be a safe and effective intervention for hoarding difficulties in people

with ID, but that the evidence base in this population needs urgent and detailed attention.

Keywords: hoarding, intellectual disabilities; CBT

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1.0 Introduction

Hoarding Disorder (HD) was a recent new addition to DSM-5 (APA, 2013) and was characterised as a well-defined and distinct disorder, rather than a sub-variant of obsessive-compulsive disorder as has traditionally been the case (Mataix-Cols et al., 2010). HD is typified by the acquisition and failure to discard of a large number of possessions that have little objective value or use. Hoarding behaviour over time creates (and then maintains) sufficient clutter in homes, so that the activities for which the living spaces were originally intended become difficult/impossible and the person experiences significant associated distress or impairment (Frost & Hartl, 1996). The frequently huge amounts of clutter generated tend to significantly impede the basic activities of home living, such as cooking, cleaning, eating and sleeping (Grisham, Frost, Steketee, Kim & Hood, 2006). Chaotic home environments can also create on-going and significant health and safety problems, such as risk of infestation or crush injuries (Frost, Steketee & Williams, 2000). Hoarding tends to emerge around early adolescence, with the clinical course being chronic and progressive without intervention (Grisham, et al. 2006).

The occurrence of mental health problems in people with ID indicates that over 40% of adults develop diagnosable mental health problems such as psychotic or affective disorders (Cooper, Smiley, Morrison, Williamson & Allen, 2010). Whilst the prevalence figures for anxiety disorders in people with ID vary according to methodology, occurrence rates range between 10-39% (Gustafsson et al., 2009). Moss, Prosser, Ibbotson and Goldberg (1996) stated that in ID populations significant proportions of psychiatric disorder goes undetected/untreated and accordingly Williams, Clarke, Fashola and Holt (1998) commented on the profound lack of knowledge concerning hoarding and ID. Whilst there is no extant evidence of the prevalence rate for HD in adults with ID, it is estimated that about 16% of children with ID engage in hoarding that is not linked to either their OCD and/or autism

(Testa, Pantelis & Fontenelle, 2011). People who hoard do not display more autistic tendencies than psychiatric controls (Pertusa, Bejerot, Eriksson, de la Cruz, Bonde, Russell & Mataix-Cols, 2012). When people met diagnostic criteria for Autistic Spectrum Disorder then Klin, Danovitch, Merz and Volkmar (2007) argued that this leaves them vulnerable to pursuing behaviours related to overly and highly circumscribed interests creating associated social isolation.

It is worth noting that hoarding has been identified to be an aspect of the behavioural phenotype of one specific ID syndrome; Prader-Willi Syndrome (Cassidy & Schwartz, 2009). Hoarding is therefore particularly common in PWS with an occurrence rate of 60% and particularly centres on the compulsion to hoard food (Storch et al., 2011). Hoarding in PWS is ego-dystonic and thus a significant cause of distress (Dykens, Leckman & Cassidy 1996). In residential settings, people who hoard can also take possessions from other residents and then hide such possessions in their personal living space (Van Houten & Rolider, 1988). Hoarding for people with ID who live in communal residential settings can significantly interfere with staff/peer relationships and often threatens the viability of the community placement itself (Lane, Wesolowski & Burke, 1989). Staff asked to intervene, often experience the delivery of help as stressful as hoarding clients have poor insight/understanding, with a high frequency of intervention—interfering behaviours and reduced adherence to therapeutic tasks (Tolin, Frost & Steketee, 2012).

Hoarding has been conceptualised from a number of different theoretical models (Gordon, Salkovskis & Oldfield, 2013), with the cognitive-behavioural model receiving the greatest empirical attention. Skirrow, Jackson, Perry and Hare's (2014) cognitive-emotional formulation of hoarding in ID suggests that clutter occurs when those with ID are unable to use emotional cues to differentiate between memories/objects that do and do not need to be remembered/retained. The CBT model characterises hoarding as a product of the interaction

of three factors, (a) deficits in information processing (b) beliefs about and attachments to possessions and (c) avoidance behaviour (Frost & Hartl, 1996). Allied assessment scales being developed and evaluated (e.g. the Saving Cognitions Inventory; Steketee, Frost & Kyrios, 2003) and studies have employed a variety of primary outcome measures including visual ratings of clutter (e.g. Hartl & Frost, 1999), the Yale-Brown Obsessive scale (e.g. Frost et al, 2003) and the Saving Inventory-Revised (e.g. Muroff et al, 2009). Therapy based upon the CBT model has been manualised (Steketee & Frost, 2007). Uncontrolled (Tolin, Frost & Steketee, 2007) and controlled (Steketee, Frost, Tolin, Rasmussen & Brown, 2010) outcome studies do evidence empirical support for the model. However, when the CBT model has been tested in more varied clinical samples (such as in a geriatric hoarding), evidence indicates attenuated outcomes (Ayers et al., 2011), without necessary population specific alterations (Ayers et al., 2014). A recent meta-analysis of HD with the CBT treatment model (Tolin, Frost, Steketee & Muroff, 2015) found large effect sizes for symptom severity reductions, rates of clinically significant change were lower (24-43%).

In terms of ID specific outcome evidence, then the evidence base for hoarding treatment consists of three single case studies and one N=3 study. Each study was based on the application of behaviour therapy. Allyon (1963) used satiation to reduce towel hoarding in a female with ID during a psychotic episode. Van Houten and Rolider (1988) showed reduced hoarding due to movement-suppression timeout and Lane et al. (1989) taught appropriate discard through use of timeout. Berry and Schnell (2006) used a multiple baseline design with N=3 ID hoarders and showed reduced hoarding for each participant when item return procedures were instigated.

In summary, the previous attempts to treat hoarding in ID contexts have been purely behavioural and there have been no prior attempts to test the generalizability and utility of CBT model. There is a growing body of evidence that indicates that CBT can be effectively

adapted to treat emotional disorders including anxiety and depression in people with ID (Taylor, Lindsay & Willner, 2008) and the current study chose to test the effectiveness of CBT based on this evidence. The current study was novel and innovative in attempting to index the safety, effectiveness and durability of CBT for hoarding for the first time in an ID context. Study hypotheses were as follows; home environments will be significantly less cluttered following CBT and will show no evidence of clutter relapse during follow-up; hoarding will reduce following CBT with no relapse over follow-up and finally mental health (anxiety and depression) will improve during CBT and not relapse over the follow-up period.

2. Methods

2.1 Participants

The sample consisted of N=14 adults with ID with hoarding difficulties. To be a potential participant then participants needed to be on the social services case register for people with established intellectual disabilities. Three of the participants lived in a communal house, but had their own rooms; the remaining participants lived independently (alone or with their partners). Inclusion criteria for the study required participants to be 18 years or older and staff to report a primary presenting problem of excessive acquisition of objects, a significantly cluttered home environment and difficulties with discarding possessions.

Participants were required to score 4 or more on the Clutter Image Rating Scale (CIRS; Frost, Steketee, Tolin & Renaud, 2008; see measures section) in at least one room as this is indicative of clinically significant clutter (Steketee & Frost, 2007). Participants were also required to remain on stable doses of psychotropic medication throughout CBT treatment, with no changes for at least 3-months prior to baseline assessment. Participants were excluded if they exhibited or workers reported profound learning disabilities, active psychotic symptoms, unstable bipolar disorder, personality disorder, substance misuse and/or being in

receipt of other on-going psychological interventions or attending a hoarding support group. Other mood and anxiety problems were permitted as long as hoarding was the primary (i.e. most severe) problem. No participants were diagnosed with PWS. The sample included N=9 males and N=5 females, with an average age of 41.78 (SD= 8.52; range 27-56).

2.2 Procedures

Ethical approval was granted from Birmingham City Council to conduct the project in council run facilities (ref WNFSEF/31/3/2012). The project was advertised and key-workers identified potential participants who were invited to an initial meeting to ascertain whether they would like to participate and explain the treatment approach on offer. Prior to treatment, those participants who had said yes at the first meeting met again with a member of the research team to provide informed consent, review inclusion/exclusion criteria and evaluate the severity of the hoarding and the levels of clutter in their home environment. Each participant completed a pre-treatment baseline assessment, with measures repeated again at the end of CBT and at 6-month follow-up. A single therapist provided the CBT across all cases. Levels of clutter were assessed via taking photographic evidence and then applying the CIRS (see measures section). Staff ratings of depression (see measures section) were sought and rated by the participant's key-worker (key-worker age range 20-50). Such staff provided the existing support structure to the participants prior to the intervention and this remained constant throughout the course of each participant's treatment. Staff therefore remained in close contact with the participant throughout the study and provided the keyworker ratings of depression.

2.3 Measures

The following outcome measures were utilised at baseline, end of treatment and at six-month follow-up and psychometric and environmental measures were taken at the same time: (1)

Clutter Image Rating Scale (CIRS; Frost, Steketee, Tolin & Renaud, 2008); this is a valid and reliable measure of the extent of clutter in the home and was the primary outcome measure of the study. The CIRS rates the degree of clutter in the kitchen, living room and bedroom and is scored by an independent assessor. In the current study, photographic assessments of the participants homes were therefore independently rated by N=4 doctoral students (two PhD students and 2 two D Clin Psy trainees) utilising the CIRS scoring procedure after a training session. Participant and stage of therapy randomised images were rated blind to the details of the intervention. Inter-rater reliability was established (Kappa = 0.67, p < 0.001; 95% CI = 0.501, 0.794). An extended CIRS measure was created which also included the bathroom, as well as other miscellaneous areas of the house such as hallways, storerooms and conservatories. This was because these areas were also observed to be places in which clutter had accumulated and therefore a wider assessment of the home environment was sought. This measure is referred to as the CIRS+ in the results. (2) Savings Inventory-Revised (SI-R; Frost, Steketee & Grisham, 2004) is the most commonly used self-report outcome measure used in evaluations of hoarding treatments. It is a valid and reliable measure of the severity of hoarding and is composed of three subscales: (a) difficulty discarding, (b) clutter and (c) acquisition. (3) Glasgow Depression Scale for ID (GDS-LD; Cuthill, Espie and Cooper, 2003) was developed and validated as a self-report measure of the severity of depressive symptoms for people with ID. The Carers Supplement for the Glasgow Depression Scale for ID (GAS-LD-CS; Cuthill, Espie and Cooper, 2003) is a sister version that enables staff to rate for the presence and associated severity of depression. (4) Glasgow Anxiety Scale for LD (GAS-ID; Mindham and Espie, 2003) was developed and validated as a self-report measure of the severity of anxiety in people with ID.

2.4 Treatment

Participants were seen individually for 12 sessions of CBT via weekly domically visits. Each session was typically two hours in duration. Treatment was conducted in accordance to the CBT manual for hoarding developed by Steketee & Frost (2007). This contains modules on treatment planning, enhancing motivation, skills training for organising/problem solving, exposure methods, cognitive strategies, reducing acquiring and preventing relapse. Goals were set by the participants during the treatment-planning module and were typically based on their need to free up space in their property. The home delivery of treatment allowed for the careful monitoring and assistance of in-session change methods and homework compliance, which has previously been seen to be related to treatment outcome in hoarding (Tolin et al., 2007). Where participants could make use of key-workers as co-therapists, these were included where indicated and were involved in assisting treatment from assessment onwards. At termination, a handover meeting provided an overview of what had been achieved, the means of supporting continued progress and also to define hoarding relapse signatures and associated relapse prevention strategies. Several adjustments made to the delivery of the treatment protocol; (a) reducing the amount and complexity of diary keeping, (b) extending the time length of each individual session, (c) keeping written psychoeducation to a minimum and (d) simplified hoarding formulations. These adjustments were in keeping with good practice guidelines for CBT with ID patients (Stenfert Kroese, Dagnan & Loumidis, 1997).

2.5 Adverse events monitoring

As this was the first attempt to study the application of CBT in an ID population, then adverse events monitoring was an integral aspect of this open trial. This was based upon the extant ID hoarding treatment evidence recording significant side effects related to elevated

levels of distress and agitated/aggressive behaviour during treatment. Ayllon (1963) reported shouting, crying, and throwing chairs, whilst Lane et al. (1989) reported yelling, swearing and also throwing objects. Duggan, Parry, McMurran, Davidson, & Dennis (2014) recently defined 'harm' during psychotherapy trials as any sustained deterioration directly caused by treatment. Deterioration needs to be sustained as this enables patients to experience temporary discomfort as an authentic aspect and process of psychological change during therapeutic work. For example, habituation during exposure exercises regarding discard during hoarding treatment. Patient safety was therefore monitored by three mechanisms, (1) incidence rates of any aggressive and agitated responses to treatment, (2) drop out from treatment and (3) a reliable deterioration on the SI-R (see measures section).

2.6 Data collection, completeness and analysis

Treatment was delivered by and outcomes collected by the second author (HM) and outcomes were then analysed separately by the first (SK) and third author (CK). Descriptive statistics were obtained for all variables and then examined for missing values and outliers. Due to the small sample size, all tests were completed with non-parametric statistics. All baseline and post-treatment self-report measures were completed, although N=3 participants did not complete the 6 month follow-up measures. Photographic data was not complete for all participants, therefore complete comparisons between time-points was not always possible. Uncontrolled effect sizes (Cohen's d_+) were calculated on the baseline to end of treatment and baseline to end of follow-up outcomes. Cohen's (1992) power primer defined d_+ = .20 as a "small" effect, d_+ = .50 as a "medium" effect, d_+ = .80 as a "large" effect of treatment. Kruskal-Wallis tests investigated change in the continuous variables; hoarding (SI-R), anxiety (GAS), depression (GDS) and clutter (full CIRS and CIRS+) across baseline, post-treatment and 6-month follow-up and Mann-Whitney tests investigated baseline-termination and baseline-follow-up comparisons. Post-hoc tests were then carried out for

significant results, controlling for Type 1 errors by using Bonferroni corrections. The reliable change index (RCI) was completed to check the extent to which any individual positive change found on the primary outcome measure (i.e. the CIRS) was beyond measurement error (Jacobson & Truax, 1991). This was used therefore to define the presence of a reliable improvement or deterioration in the levels of clutter in the home on a case-by-case basis. As part of the adverse events monitoring, pre-post RCI calculations were also performed on the SI-R outcomes.

3. Results

No participants refused the intervention, with all 14 participants completing the entire course of treatment and so the dropout rate was zero. There were no recorded incidences of aggressive or agitated responses to treatment. Table 1 displays the mean (SD) scores for baseline, post-treatment and follow-up outcome measures and the associated effect size and Mann-Whitney results. Across all the measures there was evidence of a pattern of improvement from baseline to end of treatment and then some further improvement over the follow-up period. The test of hypothesis 1 (reduction in environmental clutter) showed that on the CIRS there was very close to being a significant reduction in the levels of clutter in kitchens, living rooms and bedrooms over time ($X^2(2) = 5.96$, p = 0.05). In terms of significant change between baseline and end of CBT, there was a significant in self-reported hoarding (SI-R; U = 44, p = 0.01). When additional areas of the home (e.g. including bathrooms) were incorporated into the environmental measure (CIRS+), a significant main effect of time was found ($X^2(2) = 8.70$, p = 0.01). This indicates significant reductions to clutter across the wider home environment. The baseline to end of treatment effect size for the CIRS was $d_+0.78$ and for the CIRS+ it was $d_+0.83$. Both scores would represent large

treatment effect sizes in terms of reductions to environmental clutter. The mean scores at follow-up on the CIRS and the CIRS+ would imply that treatment progress gains in terms of environmental clutter were maintained over the follow-up period. Baseline to follow-up comparisons also found significant reductions in self-reported hoarding (U = 35, p = 0.02), clutter (U = 28, p = 0.01) and difficulties with discard (U = 44, D = 0.07).

For the 7 participants with sufficient data to calculate RCIs on the baseline to post-treatment CIRS, 5 achieved a reliable reduction in environmental clutter. These 5 participants tended to then maintain the improvements to the home environment over the follow-up period, as no single patient then went onto further reliably improve (or reliably deteriorate) on end of treatment to follow-up clutter RCI comparisons. No single participant exhibited any reliable deterioration in the home environment during CBT.

The test of the second hypothesis (hoarding would reduce following treatment with no relapse) showed a significant reduction in hoarding over time (SI-R; X^2 (2) = 8.30, p= 0.01). The baseline to post-treatment effect size in hoarding was coded as a moderate reduction (d_+ =0.47). There were no individual reliable deteriorations on pre-post SI-R comparisons. Sub-scale analyses of the SI-R found that there were significant reductions to problems with levels of clutter (X^2 (2) = 7.9, p= 0.02), with an effect size of d_+ =0.34 on baseline to end of treatment comparisons. In contrast, there were no significant effects of time found regarding difficulties with discard (X^2 (2) =4.3, p= 0.12) or problems with acquisition (X^2 (2) =3.63, p= 0.16). To further define both the clinical benefits and potential for relapse, mean percentage reductions to the hoarding specific measures for the study (CIRS, CIRS+, SI-R and SI-R subscales) are displayed in Figure 1. Problems with hoarding fell by 36.60 % (SI-R) and clutter in the home by 49.70 % (CIRS+) on baseline to post-treatment comparisons. Figure 1 also demonstrates that continued progress over the follow-up period (compared to baseline) was

evident in relation to hoarding and clutter. This would further evidence the absence of relapse in hoarding in the current study.

Finally, tests of the third hypothesis (anxiety and depression would improve during treatment and then not relapse) found that the decreases in self-reports of depression (X^2 (2) =3.58, p= 0.17) and anxiety (X^2 (2) =5.20, p= 0.074) were non significant. However, keyworker ratings of depression (i.e. GDS-ID for carers) found that staff working closely with the participants reported them to be exhibiting significantly fewer signs of depression (X^2 (2) = 4.30, p= 0.03). The effect size was d_+ =0.39 (moderate effect) for key-workers based prepost comparisons of depression ratings.

4.0 Discussion

This study represents the first evaluation of the delivery of CBT for hoarding in an ID population. This open-label trial of the CBT model for hoarding in a sample of 14 adults with ID (in which both genders were represented), found statistically significant reductions in the primary outcome measure of environmental clutter across the wider home environment. The reductions in clutter limited to the kitchens, living rooms and bedrooms (i.e. the CIRS measure) were very close to significance (0.05), whereas reductions extended to include bathrooms and other home areas (i.e. the CIRS+) were highly significant. The use of the CIRS+ was reactive to the assessment of the home environments of the participants at the onset of the study indexing a wide distribution of clutter, and so was not reactive to the results on the CIRS. The wider assessment of the home environment in terms of reductions in levels of clutter did appear to capture more change and is a consideration for future ID research. The treatment refusal rate was zero - although no alternative treatment was offered.

No participants dropped out of treatment, indicating that CBT was an acceptable intervention to ID participants that have hoarding issues.

Unlike in previous attempts to psychologically intervene with hoarding in ID populations (Allyon, 1963; Lane et al. 1989), there were no incidences of agitated or aggressive responses to treatment. Also, no participants had a reliable increase in clutter during treatment and there were no individual reliable deteriorations in hoarding. No adverse events were noted during the study, which would suggest that the CBT received was a safe intervention. Berry and Schnell (2006) doubted the generalizability of the CBT hoarding model with ID populations. The current evidence would suggest that such reservations were unfounded. In terms of secondary outcomes, staff observed and experienced the participants to be less depressed in their mood over time. The intervention was focal to hoarding and no help was offered regarding mood management. The reasons for this shift were not explored with staff; improved living conditions may have been the context for staff seeing an improvement in mood, although participants may have felt less depressed due to other factors. It is worth noting that unlike previous hoarding evaluations in ID, participants did not reside in institutions and so initial levels of clutter were relatively high and non-specific. In communal residential settings then constant staff supervision of ID patients tends to keep hoarding to a minimum (Williams et al. 1998).

For those participants where it was possible to calculate the relevant CIRS scores, then a significant proportion (5/7) experienced a reliable reduction in clutter in their homes. The CBT appeared to particularly work on problems with clutter, as the CIRS, CIRS+ and the SI-R subscale analyses showed that levels of clutter particularly responded to treatment. As Pollock, Kellett and Totterdell (2014) have noted the primary outcome measure of choice in hoarding treatment should always pertain to the appearance and functioning and of the home environment. In terms of durability of treatment effects and potential for behavioural relapse,

the outcomes across all the hoarding measures used in the study indicated that gains tended to be at least maintained during follow-up. The current study would therefore challenge the Ayers et al., (2011) evidence which that found that CBT did not generalise well to clinical hoarding populations with additional needs, such as geriatric hoarders.

The 36-50% reduction in hoarding severity achieved in this ID sample is also somewhat different to extant findings from non-disabled adults and older adult samples. Group CBT for hoarding improvement rates range from 10-21% (Muroff et al., 2010; Steketee et al., 2000), whilst individual CBT improvement rates range from 14-28% (Ayers et al., 2011; Steketee et al., 2010; Tolin et al., 2007). This calls into question why this is the case? It may be the case that in the current study all sessions were conducted in the homes of participants. Therefore much of the work was completed in situ with associated close monitoring of between-session tasks and in-session cognitive and behavioural change methods. Participants were therefore kept usefully focussed to the goals of clearing clutter and introducing organisation to the home. This may also explain the non-significant results found on levels of acquisition in the study. The evidence base in terms of the utility of domiciliary treatment for hoarding in non-disabled adults suggests that domiciliary visits do not differentially improve hoarding outcomes (Muroff et al., 2011; Pollock, Kellett & Totterdell, 2014). This may not be the case in ID hoarding and further dismantling trials to study the potential enhanced efficacy of domiciliary hoarding treatment in ID population is indicated.

The current study had many methodological limitations that limit confidence in the validity of the observed results. Perhaps the most pertinent of which was that the study was an open trial and therefore uncontrolled. The presence of a passive or active control to benchmark the CBT outcomes against would have significantly increased the internal validity of the study. The observed reductions to hoarding and clutter in the home might also have

been due to nonspecific factors, such as heightened patient expectations. The fact that the therapist (HM) also collected the outcomes is a study weakness, but the primary outcome measure (CIRS) was usefully independently rated. The use of the SI-R is also questionable, as this outcome measure has not been previously validated in ID populations and so the hoarding results should be therefore treated with caution. As no self-report hoarding outcome measure is available for use with people with ID, this therefore represents a further research goal. Similarly, use of the CIRS+ measure could be criticised, as this was an extension of an already validated clutter measure. If the ID-hoarding evidence base is to develop (e.g. assessing prevalence rates) then valid and reliable measures of hoarding pathology in ID are also required.

Further limitations included the small sample size, the use of only one study therapist and the lack of any treatment integrity checks. Because of the novel and exploratory nature of the current study, we allowed a great deal of individual variability in delivery of the CBT treatment protocol (e.g. individual session duration varying within and across treatments). This represents a study weakness (poor definition of the necessary and sufficient parameters of treatment) and strength (flexibility of manual delivery meeting the individual needs of patients; Kendall, Chu, Gifford, Hayes & Nauta, 1998). Future research concerning developing the treatment evidence base in ID would certainly benefit from more precise definition of and fidelity to treatment procedures. Further outcome research is needed to determine whether CBT for hoarding in ID is superior to passive control (e.g. wait-list) and active treatment comparison. This requires the other psychotherapies for people with ID and mental health problems that have been found to be effective (e.g. such as psychodynamic psychotherapy; James & Stacey, 2014) to produce some evidence with regards to treatment outcome to effectively serve as a valid active comparison arm in an RCT. Outcomes need to

be compared naturalistically between treatments supported by key-worker input and those that are solely therapist-delivered.

In conclusion, the current study suggests that the CBT treatment provided produced clinically meaningful and durable results in ID participants with problematic hoarding. The study demonstrates that people with ID can tolerate and benefit from a full course of CBT for their hoarding, when it is delivered in their own homes. The results from this research are a challenge to the previous opinion that people with ID and hoarding can only be treated via behavioural methods and that cognitively informed interventions 'are likely to be ineffective' (Berry & Schnell, 2006). Much further clinical and research work is required to develop both an understanding of ID hoarding prevalence, phenomenology and aetiology and build a robust evidence base for intervention.

References

- Allyon, T. (1963). Intensive treatment of psychotic behaviour by stimulus satiation and food reinforcement. Behaviour Research and Therapy, 1, 53-61.
- American Psychiatric Association. (2013). Diagnostic and statistical manual of mental disorders (5th ed.). Washington, DC: Author.
- Ayers, C. R., Wetherell , J. L., Golshan, S., & Saxena, S. (2011). Cognitive-behavioral therapy for geriatric compulsive hoarding. Behaviour Research and Therapy, 49, 689-94.
- Ayers, C. R., Saxena, S., Espejo, E., Twamley, E. W., Granholm, E., & Wetherell, L. (2014).

 Novel treatment for geriatric hoarding disorder: An open trial of cognitive rehabilitation paired with behavior therapy. American Journal of Geriatric Psychiatry, 22, 248-252.
- Berry, C. L. & Schnell, R. M. (2006). Reducing hoarding behavior with individualised reinforcement and item return. Behavioral Interventions, 21, 123-135.
- Cassidy, S. B., & Schwartz, S. (2009). Prader-Willi syndrome. In R. A. Pagon, T. Bird, C. R. Dolan, K. Stephens & M. P. Adam (Eds.). Gene reviews. Seattle, WA: University of Washington.
- Cohen, J. (1990). A power primer. Psychological Bulletin, 112, 155-159.
- Cooper, S-A., Smiley, E., Morrison, J., Williamson, A., & Allen, L. (2010). Mental ill-health in adults with intellectual disabilities: prevalence and associated factors. British Journal of Psychiatry, 190, 27-35.

- Cuthill, F. M., Espie, C. A., & Cooper, S-A. (2003). Development and psychometric properties of the Glasgow depression scale for people with a learning disability: individual and carer supplement versions. The British Journal of Psychiatry, 182, 347-353.
- Duggan, C., Parry, G. McMurran, M., Davidson, K., & Dennis, J. (2014). The recording of adverse events from psychological treatments in clinical trials: evidence from a review of NIHR-funded trials. Trials, 15, 335.
- Dykens, E. M, Leckman, J. R., & Cassidy, S. B. (1996). Obsessions and compulsions in Prader-Willi syndrome. Journal of Child Psychology and Psychiatry, 37, 995-1002.
- Frost, R. O., & Hartl, T. L. (1996). A cognitive-behavioral model of compulsive hoarding. Behaviour and Research Therapy, 34, 341-50.
- Frost, R. O., Steketee, G., & Williams, L. (2000). Hoarding: A community health problem. Health and Social Care in the Community, 8, 229–234.
- Frost, R. O., Steketee, G., & Greene, K. (2003). Cognitive and behavioral treatment of compulsive hoarding. Brief Treatment and Crisis Intervention, 3, 323-337.
- Frost, R. O., Steketee, G., & Grisham, J. (2004). Measurement of compulsive hoarding: saving inventory-revised. Behaviour Research and Therapy, 42, 1163-82.
- Frost, R. O., Steketee, G., Tolin, D. F., & Renaud, S. (2008). Development and validation of the clutter image rating scale. Journal of Psychopathology and Behavioral Assessment, 30, 180-192.

- Gordon, O. M., Salkovskis, P. M., & Oldfield, V. B. (2013). Beliefs and experiences in hoarding. Journal of Anxiety Disorders, 27, 328-339.
- Grisham, J., Frost, R. O., Steketee, G., Kim, H. J. & Hood, S. (2006). Age of onset of compulsive hoarding. Journal of Anxiety Disorders, 20, 675-86.
- Gustafsson, C., Öjehagen, A., Hansson, L., Sandlund, M., Nyström, M., Glad, J., Cruce, G., Jonsson A-K., & Fredriksson, M. (2009). Effects of psychosocial interventions for people with intellectual disabilities and mental health problems: A survey of systematic reviews. Research on Social Work Practice, 19, 281-290.
- Hartl, T. L., & Frost, R. O. (1999). Cognitive-behavioral treatment of compulsive hoarding:

 A multiple baseline experimental case study. Behaviour Research and Therapy, 37,
 451–461.
- Jacobson, N., & Truax, P. (1991). Clinical significance: A statistical approach to defining meaningful change in psychotherapy research. Journal of Consulting and Clinical Psychology, 59, 12-19.
- James, C. W. & Stacey, C. M. (2014). The effectiveness of psychodynamic interventions for people with learning disabilities: a systematic review. Tizard Learning Disability Review, 19, 17-24.
- Kendall, P. C., Chu, B., Gifford, A., Hayes, C., & Nauta, M. (1998). Breathing life into a manual: Flexibility and creativity with manual-based treatments. Cognitive and Behavioral Practice, 5, 177-198.

- Klin, A., Danovitch, J.H., Merz, A.B., & Volkmar, F.R. (2007). Circumscribed interests in higher functioning individuals with autistic spectrum disorders: An exploratory study. Research and Practice for Persons with Severe Disabilities, 32, 89-100.
- Lane, I. M., Wesolowski, M. D., & Burke, W. H. (1989). Teaching socially appropriate behaviour to eliminate hoarding in a brain injured adult. Journal of Applied Behavior Analysis, 11, 153-162.
- Luckasson, R., Borthwick-Duffy, S., Buntinx, W. H. E., Coulter, D. L., Craig, E. M., Reeve,
 A., Shalock, R. L., Snell, M. E., Spitalnik, D. M., Spreat, S., & Tassé M. J. (2002).
 Mental retardation: Definition, classification and systems of supports. 10th ed.
 Washington (DC): American Association on Mental Retardation.
- Mataix-Cols, D., Frost, R. O., Pertusa, A., Clark, L. A., Saxena, S., Leckman, J. F., Stein, D. J., Matsunaga, H., & Wilhelm, S. (2010). Hoarding disorder: a new diagnosis for DSM-V? Depression and Anxiety, 27, 556-572.
- Mindham, J., & Espie, C. A. (2003). Glasgow anxiety scale for people with an intellectual disability (GAS-ID): development and psychometric properties of a new measure for use with people with mild intellectual disability. Journal of Intellectual Disability Research, 47, 22-30.
- Moss, S. C., Prosser, H., Ibbotson, B., & Goldberg, D. P. (1996). Respondant and informant accounts of psychiatric symptoms in a sample of patients with learning disability.

 Journal of Intellectual Disability Research, 40, 457-465.
- Muroff, J., Steketee, G., Rasmussen, J., Gibson, A., Bratiotis, C., & Sorrentino, C. M. (2009).

 Group cognitive and behavioral treatment for compulsive hoarding. Depression and

 Anxiety, 26, 634-640.

- Muroff, J., Steketee, G., Himle, J., & Frost, R. O. (2010). Delivery of internet treatment for compulsive hoarding (D.I.T.C.H.). Behaviour Research & Therapy, 48, 79-85.
- Muroff, J., Bratiotis, C., & Steketee, G. (2011). Treatment for hoarding behaviors: A review of the evidence. Clinical Social Work Journal, 39, 406-423.
- Pertusa, A., Bejerot, S., Eriksson, J., de la Cruz, L. F., Bonde, S., Russell, A., & Mataix-Cols, D. (2012). Do patients with hoarding disorder have autistic traits? Depression and Anxiety, 29, 210-218.
- Pollock, L., Kellett, S., & Totterdell, P. (2014). An intensive time-series evaluation of the effectiveness of cognitive behaviour therapy for hoarding disorder: A 2-year prospective study. Psychotherapy Research, 24, 485-495.
- Skirrow, P., Jackson, P. G., Perry, E. P. & Hare, D. J. (2014). I collect therefore I am Autonoetic consciousness and hoarding in Asperger Syndrome. Clinical Psychology & Psychotherapy.
- Steketee, G., Frost, R. O., Wincze, J., Greene, K., & Douglass, H. (2000). Group and individual treatment of compulsive hoarding: A pilot study. Behavioural and Cognitive Psychotherapy, 28, 259-268.
- Steketee, G., Frost, R. O., & Kyrios, M. (2003). Cognitive aspects of compulsive hoarding. Cognitive Therapy and Research, 27, 463-479.
- Steketee, G., & Frost, R. O. (2007). Compulsive hoarding and acquiring; therapist guide.

 NY: Oxford University Press.

- Steketee, G., Frost, R. O., Tolin, D. F., Rasmussen, J., & Brown, T. A. (2010). Waitlist-controlled trial of cognitive behavior therapy for hoarding disorder. Depression and Anxiety, 27, 476-484.
- Stenfert Kroese, S., Dagnan, D., & Loumidis, K. (1997). Cognitive behaviour therapy for people with learning disabilities. London: Routledge.
- Storch, E. A., Muroff, J., Lewin, A. B., Geller, D., Ross, A., Mccarthy, K., Mogan, J., Frost, R. O., & Steketee, G. (2011). Development and psychometric evaluation of the children's saving inventory. Child Psychiatry and Human Development, 42, 166-182.
- Taylor, J., Lindsay, W., & Willner, P. (2008). CBT for people with intellectual disabilities: emerging evidence, cognitive ability and IQ effects. Behavioural and Cognitive Psychotherapy, 36, 723-733.
- Testa, R., Pantelis, C., & Fontenelle, L. F. (2011). Hoarding behaviours in children with learning disabilities. Journal of Child Neurology, 26, 574-9.
- Tolin, D. F., Frost, R. O., & Steketee, G. (2007). An open trial of cognitive-behavioral therapy for compulsive hoarding. Behaviour Research and Therapy, 45, 1461-1470.
- Tolin, D. F., Frost, R. O., & Steketee, G. (2012). Working with hoarding vs. non-hoarding clients: A survey of professionals' attitudes and experiences. *Journal of Obsessive-Compulsive and Related Disorders*, 1, 48-53.
- Tolin, D.F., Frost, R.O., Steketee, G., & Muroff, (2015). Cognitive behaviour therapy for hoarding; a meta-analysis. *Depression and Anxiety*, *32*, 158-166.
- Van Houten, R., & Rolider, A. (1988). Repeating the scene: An effective way to provide

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delayed punishment for inappropriate motor behaviour. Journal of Applied Behaviour Analysis, 21, 187-192.

Williams, H., Clarke, R., Fashola, Y. & Holt, G. (1998). Diogenes' syndrome in patients with intellectual disability: a 'rose by any other name'? Journal of Intellectual Disabilities Research, 42, 316-320.

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Table 1: Baseline, end of treatment and follow-up scores on outcome measure in adults with ID receiving CBT for compulsive hoarding.

Measure	Baseline (N=14)	Post-Treatment (N=14)	6 Month follow-up (N=11)	Baseline to post-treatment	Effect Size	Baseline to 6 month follow-up	Effect Size
CIR	3.4 (0.30)	1.65 (0.18)	1.66 (0.30)	U= 0 (Z= -1.9), p=0.50	0.78	U=0 (Z= -1.7), p=.050	0.70
CIR+	3.4 (1.05)	1.71 (0.19)	1.61 (0.28)	U=0 (Z= -2.50), p=.01**	0.83	U=2 (Z= -1.09), p=.0.27	0.36
SI-R	34.71 (16.45)	22.00 (13.42)	17.27 (15.15)	U= 44 (Z= -2.48), p=.01**	0.47	U=35 (Z= -2.30), p=.02*	0.20
-Clutter	10.86 (8.3)	5.78 (6.62)	2.73 (2.69)	U=59 (Z= -1.80), p=0.70	0.34	U=28 (Z= -2.70), p=.007**	0.54
-Discard	15.21 (11.27)	8.7 (5.9)	7.91 (7.84)	U=62 (Z= - 1.60), p=0.97	0.21	U=44.5 (Z= -1.70), p=.070*	0.02
-Acquire	10.79 (7.21)	7.42 (5)	6.64 (6.6)	U=61 (Z=-1.70), p=.08	0.22	U= 49.5(Z= -1.50), p=.13	0.30
GAS-LD	50.36 (10.30)	47.93 (8.2)	32.82 (19.82)	U=85 (Z=60), p=.50	0.11	U= 38.5(Z=2.11), p=.30	0.42
DS-LD	17.71 (8.11)	13.76 (7)	12.91 (6)	U=67 (Z= -1.43), p=.15	0.27	U=44 (Z= -2.11), p=0.70	0.42
GDS for carers	13.43 (4.51)	10.14 (3.8)	Not administered	U=53 (Z= - 2.08), p=.03*	0.39	Not administered	

^{*} p < 0.05, ** p < 0.01

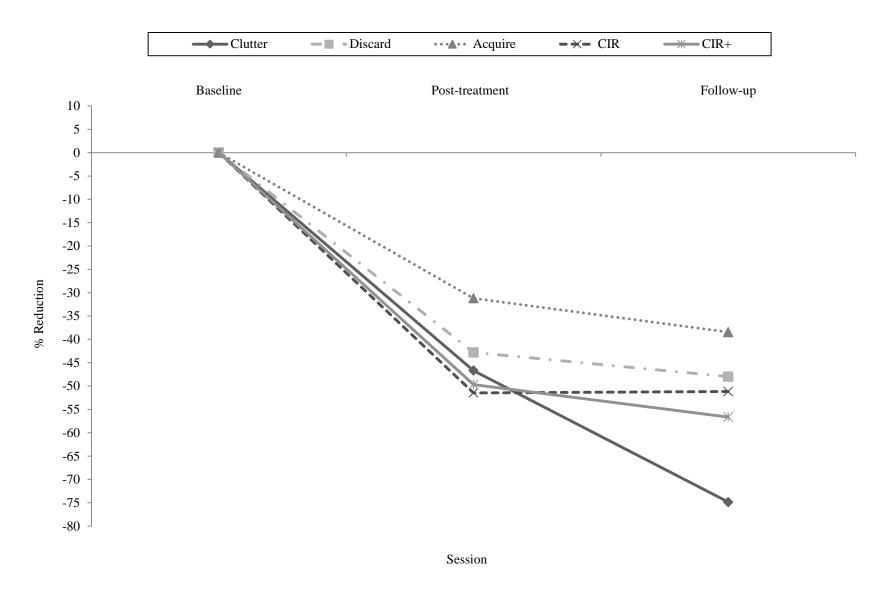


Fig. 1. Mean percent reduction on hoarding outcomes