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Hannah King, BSc

Predictors, Moderators and Mediators of Carer Distress and Maintaining Factors in Eating Disorders

Section A

A Review of Predictors of Distress in Carers of People with Eating Disorders, and Moderators and Mediators of These Relationships

Word count: 7,961 (425)

Section B

Moderators and Mediators of Relationships between Eating Disorders and Related Carer Difficulties, in the Context of a Skills Sharing Intervention for Carers of People with Anorexia Nervosa

Word count: 7,997 (1)

Overall word count: 15,958

A thesis submitted in partial fulfilment of the requirements of
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MARCH 2017

SALOMONS
CANTERBURY CHRIST CHURCH UNIVERSITY

Acknowledgements

Dedicated to my beloved co-pilot Zav. Thank you for keeping me fed, watered and grounded. Without your nurture, love and wisdom, none of this would be.

Papa, your gentle encouragement, unfailing belief in me and early lessons in people-watching helped prepare me for this doctorate; your love and support helped me through it. Thank you for always being on hand with good advice and remedies, and for the final push.

To the carers I have worked with, and who contributed their efforts to the data presented in the MRP; your strength, hope and tenacity are a lasting inspiration.

Summary of the Portfolio

This thesis examines the relationships between eating disorders and associated difficulties experienced by carers.

Section A presents a narrative review based on a systematic search of the literature from inception to October 2016. The evidence for predictors of distress experienced by carers of people with eating disorders, and factors that moderate or mediate these predictive relationships, are reported and appraised. This includes predictors, moderators and mediators of changes in carer distress as a proposed outcome of eating disorder interventions for carers.

Section B is an empirical paper reporting the findings from an examination of archival data. Data were obtained from a skills-sharing RCT for carers of people admitted to hospital for treatment of Anorexia Nervosa. Moderators and mediators of intervention outcomes were analysed, with the aim of elucidating for whom, or under what circumstances, the intervention was most likely to be effective, and the processes by which intervention may have affected outcomes. Additionally, longitudinal relationships between eating disorder symptoms and carers' reactions to the illness were examined, to test theoretical models of the ways these are hypothesised to interact.

Limitations and implications from both studies are discussed. Results support the importance of interventions for carers of people with eating disorders.

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Hannah King B.Sc. (Hons)

Major Research Project

Section A: Literature Review

A Review of Predictors of Distress in Carers of People with Eating Disorders, and Moderators and Mediators of These Relationships

Word count: 7,961 (425)

DECEMBER 2016

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Abstract

Background & Objectives: Caring for someone with an eating disorder (ED) is associated with high levels of psychological distress. This review appraised evidence of predictors, moderators and mediators of carer distress in EDs, and changes to distress following intervention.

Method: Electronic databases were searched from inception until October 2016. Thirty selected studies reported at least one predictor of psychological distress in carers of people diagnosed with EDs. Quality of analysis was appraised.

Results: A large number of hypothesised predictors were examined. Results were mixed for most, although there was consensus about direction of relationships. There was reasonable evidence for high burden, expressed emotion, accommodation, and cared-for comorbidity, and low carer coping and skills, as predictors of higher carer distress in cross-sectional data. Evidence of predictors of intervention outcome or mechanisms of treatment action was limited. Burden, carer and cared-for distress predicted carer distress longitudinally.

Limitations & Conclusions: Generalisability was limited by studies mainly sampling mothers and people with Anorexia Nervosa. Methodological issues included validity of measures and recruitment. The evidence is consistent with models in which primary ED-related problems impact on distress through their effect on burden and other secondary factors. Further research, particularly on moderators and mediators, is warranted.

Keywords: Carer, Distress, Predictors, Eating disorders

Introduction

Eating disorders are serious psychiatric illnesses with high mortality rates (Arcelus, Mitchell, Alex, Wales, & Nielsen, 2011). The NICE (2004) guidelines recommend outpatient management as the first form of treatment for all types of eating disorders. This places a burden of care onto parents and close others who often experience their caregiving role as distressing and burdensome (Whitney et al., 2005; Zabala, MacDonald, & Treasure, 2009), with quality of life negatively impacted (Martin et al., 2013; Las Hayas et al., 2014). Many carers report a lack of much needed information on how to manage the problem (Haigh & Treasure 2003), and become discouraged (Treasure et al., 2008), with a large proportion meeting clinical threshold on measures of anxiety and depression (e.g. 70% and 38% respectively, Kyriacou, Treasure, & Schmidt, 2008).

While these difficulties are themselves clinically relevant for carers, the interpersonal aspect of the Cognitive Interpersonal Maintenance Model of Anorexia Nervosa (AN; Schmidt & Treasure, 2006; Treasure & Schmidt, 2013, Figure 1) postulated that carers' emotional and behavioural reactions to the difficulties created by the eating disorder may also maintain the ED. Specifically, psychological distress, expressed emotion (criticism and emotional over-involvement) and accommodating and enabling (going along with the symptomatic behaviours to reduce the impact on family life), can unintentionally serve to maintain the ED. This increases carer distress and unhelpful behavioural responses in a vicious cycle (ibid). There is a growing evidence base for this model (e.g. Anastasiadou, Medina-Pradas, Sepulveda, & Treasure, 2014; Butzlaff & Hooley, 1998; Goddard et al., 2013; Treasure & Nazar, 2016; Treasure & Schmidt, 2013), which can be applied transdiagnostically in eating disorders (Goddard et al., 2011)

Figure 1. Interpersonal Aspect of The Cognitive Interpersonal Maintenance Model of Anorexia Nervosa

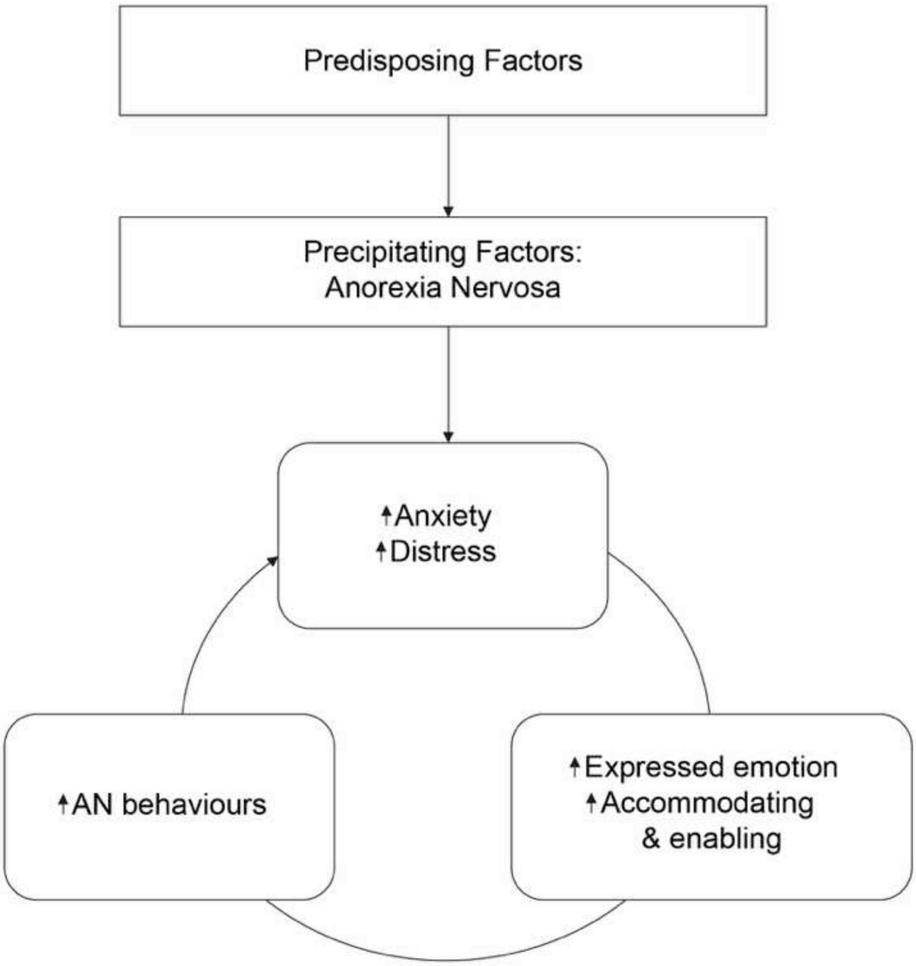

Figure 1. Model describing hypothesised vicious cycle of interactions between the eating disorder symptoms and carer reactions to the illness. Adapted from Treasure and Schmidt (2013).

While there has been much high quality research into predictors of outcomes for people with EDs, including large-scale RCTs (see Vall and Wade's 2015 review), there has been considerably less attention to outcomes for carers. The Model of Carer Coping in eating disorders (Treasure et al., 2007; Treasure & Nazar, 2016, Figure 2) postulated that illness factors, carer factors, and societal factors contribute to carer experiences of distress when coping resources are exhausted.

Figure 2. Model of Carer Coping in Eating Disorders

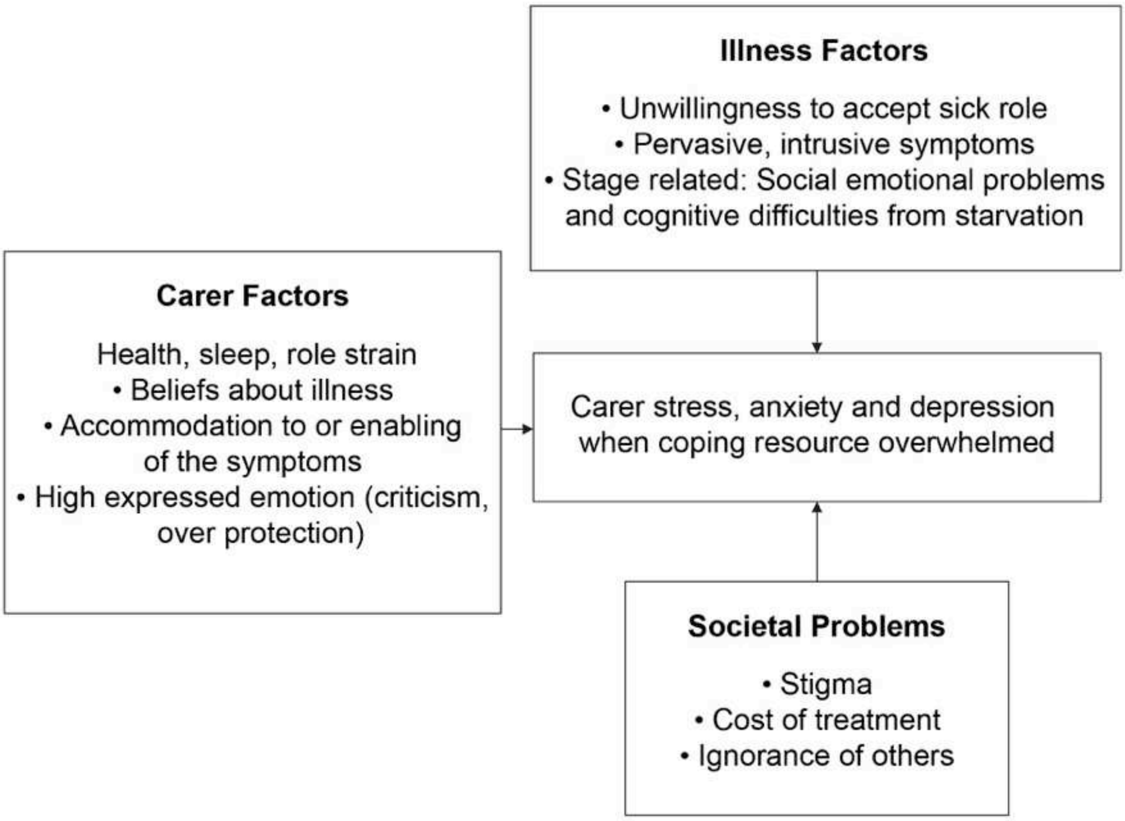

Figure 2. Model describing factors hypothesised to contribute to carer distress when coping resources are overwhelmed. Adapted from Treasure et al. (2007), and Treasure and Nazar (2016).

Szmukler et al. (1996) proposed a model in which caregivers' appraisals about the illness and associated demands leads to perceived stress, which in turn leads to distress; although negative appraisals might be alleviated by social support or feelings of efficacy. Winn et al.'s model (2007, Figure 3) suggested cared-for[1], carer and relationship factors contribute to a negative caregiving experience, hypothesised to lead to distress.

Figure 3. Conceptual Model for Studying Predictors of Negative Experiences of Caregiving and Caregiver Mental Health Status

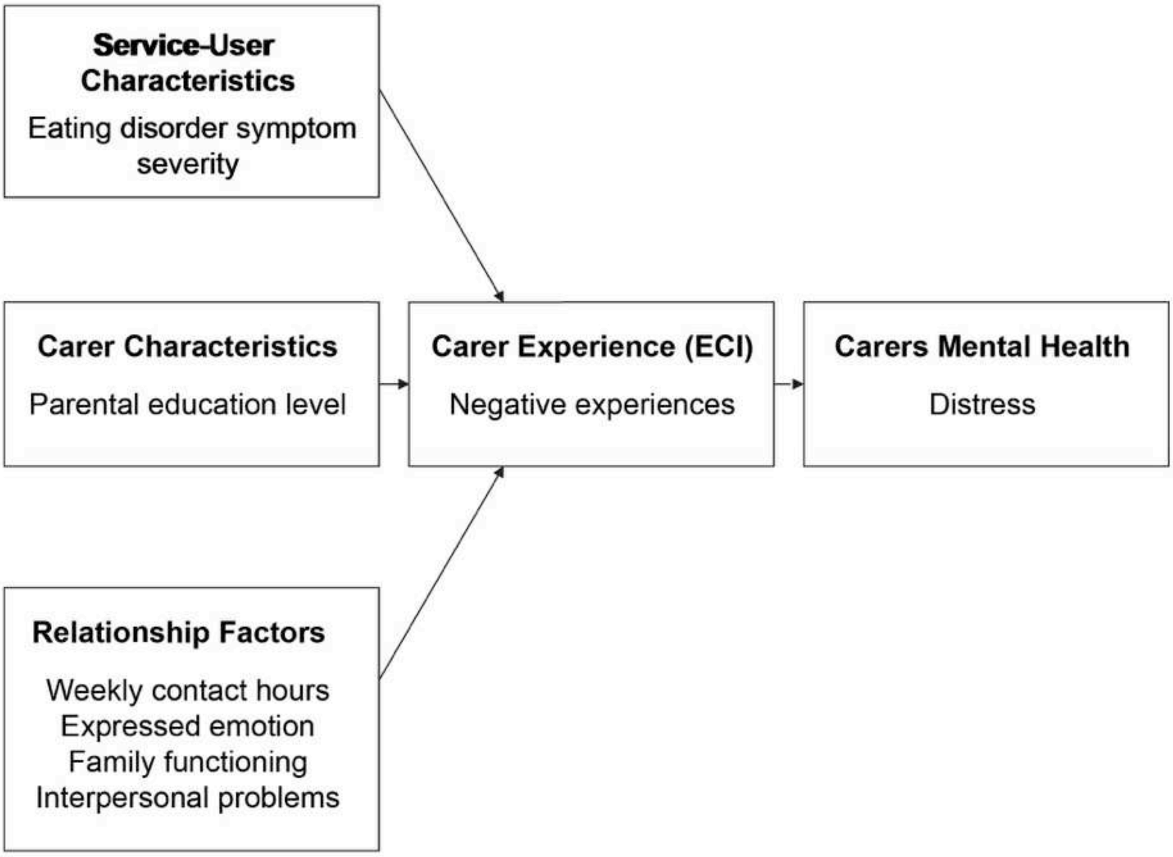

Figure 3. Adapted from Winn et al. (2007).

Carers' skills interventions have been developed (e.g. Goddard, Raenker, & Treasure, 2012; Hibbs et al., 2015a; Treasure & Nazar, 2016) to address these difficulties through seeking to ameliorate modifiable aspects of carer difficulties and interpersonal maintaining factors, sharing skills to increase carers' self-care and adaptive management of the ED. A recent

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[1] This review refers to people with eating disorders as 'cared-for', in respect of their role in relation to carers.

meta-analysis (Hibbs et al., 2015b) reported most interventions for carers of someone with an ED produced a moderate-sized reduction in carer distress, among other outcomes.

While the presence of heightened carer distress in EDs is well-evidenced (Anastasiadou et al., 2014), the growing literature base exploring statistical evidence of predictors of amount of distress in this population, and moderators and mediators of these associations, have not been systematically reviewed. Similarly, while the efficacy of carers' skills interventions has been systematically reviewed (Hibbs et al., 2015b), what predicts, moderates or mediates positive outcomes in terms of carer distress has not. While the latter is an emergent field, appraising available evidence holistically is valuable in the context of rapidly developing theory and clinical interventions.

While examination of ED-related predictors of carer distress can indicate associates of carer distress and identify possible targets for intervention, examination of moderators is useful for identifying who is more likely to experience distress in connection with the ED-related problems, or to benefit from particular interventions. Mediation analysis can suggest explanations for the effect one variable (e.g. time spent caregiving) has on another (e.g. carer distress), and elucidate processes involved.

The aim of this review therefore is to collate and summarise the evidence available regarding hypothesised predictors, moderators and mediators of carer distress to assess progress of the literature towards adequate appraisal of two overarching research questions:

1. What predicts the level of distress experienced by carers of people with eating disorders, and what moderates and mediates these associations?
2. What predicts change in carer distress as an outcome of ED-related interventions, and what moderates and mediates these associations?

In addressing these questions, the review aimed to weigh the evidence concerning the processes that contribute to carer distress in eating disorders, and its amelioration.

Methodology

This is a narrative review based on a systematic literature search guided by the Preferred Reporting Items for Systematic Reviews and Meta Analyses statement (PRISMA, Moher, Liberati, Tetzlaff, & Altman, 2009). Electronic databases Psychinfo, Web of Science, and Medline were searched from inception to October 1st 2016.

Elaboration of search terms for the constructs ‘eating disorder’, ‘carer’, ‘predictors’ and ‘distress’ followed the precedents of Zabala, Macdonald and Treasure (2009), Anastasiadou et al., (2014), Hibbs et al., (2015b) and Vall and Wade (2015). Terms were expanded to include correlates of carer distress frequently suggested by the literature, to support retrieval of evidence of correlation for which distress was not conceptualised as the response variable of interest. Search terms employed are presented in Table 1.

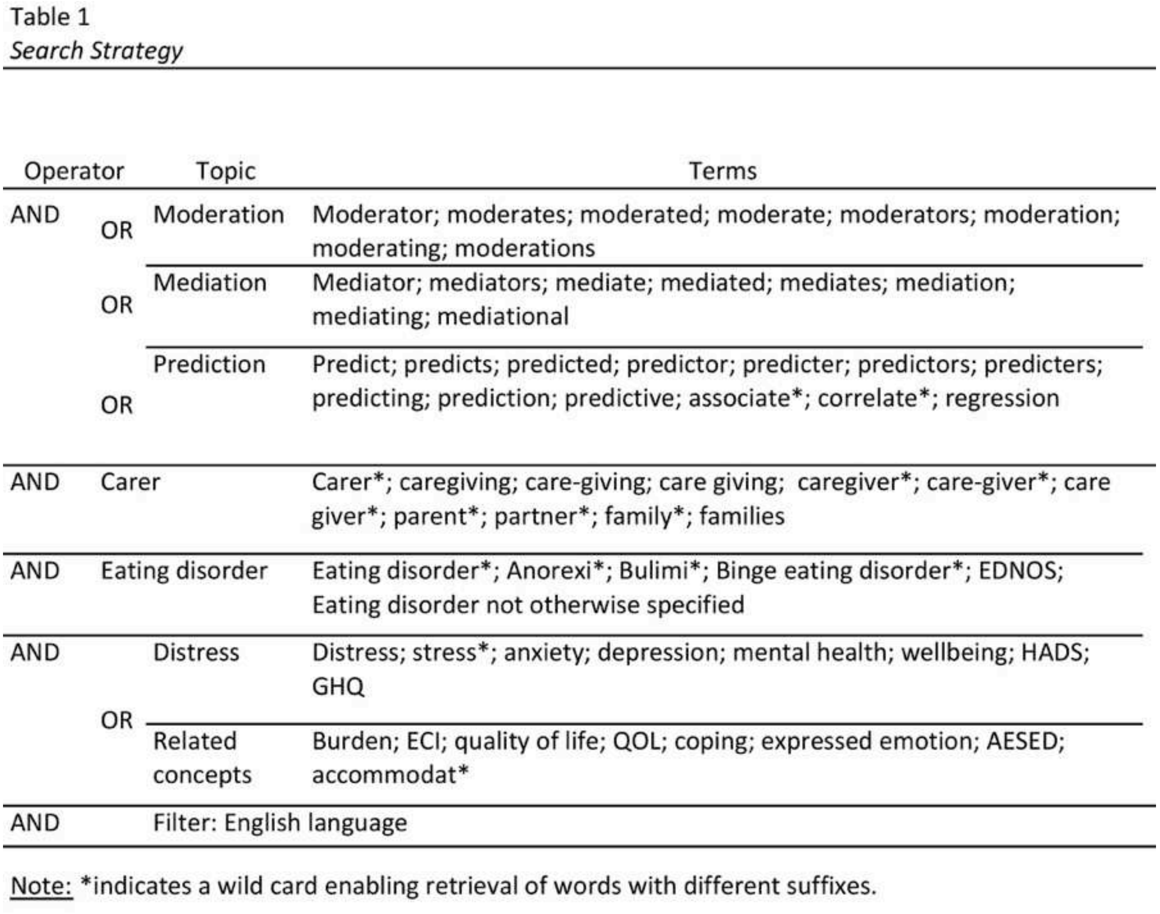

Table 1
Search Strategy

Operator		Topic	Terms
AND	OR	Moderation	Moderator; moderates; moderated; moderate; moderators; moderation; moderating; moderations
	OR	Mediation	Mediator; mediators; mediate; mediated; mediates; mediation; mediating; mediational
	OR	Prediction	Predict; predicts; predicted; predictor; predicter; predictors; predicters; predicting; prediction; predictive; associate*; correlate*; regression
AND		Carer	Carer*; caregiving; care-giving; care giving; caregiver*; care-giver*; care giver*; parent*; partner*; family*; families
AND		Eating disorder	Eating disorder*; Anorexi*; Bulimi*; Binge eating disorder*; EDNOS; Eating disorder not otherwise specified
AND	OR	Distress	Distress; stress*; anxiety; depression; mental health; wellbeing; HADS; GHQ
		Related concepts	Burden; ECI; quality of life; QOL; coping; expressed emotion; AESED; accommodat*
AND		Filter: English language	

Note: *indicates a wild card enabling retrieval of words with different suffixes.

Inclusion Criteria

- Peer reviewed, English language, quantitative study.
- Participants defined as having a current caregiving role (e.g. Department of Health, 2014) for someone with a current, medically diagnosed ED[2]. This included people self-identifying as carers.
- Included a measure of carer distress that was completed by the carer, clinician or interviewer with the carer. For the purposes of this review, level of distress is

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[2] Anorexia Nervosa, Bulimia Nervosa, Binge Eating Disorder, or Eating Disorder Not Otherwise Specified (EDNOS).

defined by scores on measures of anxiety, depression, general psychological distress and mental health.

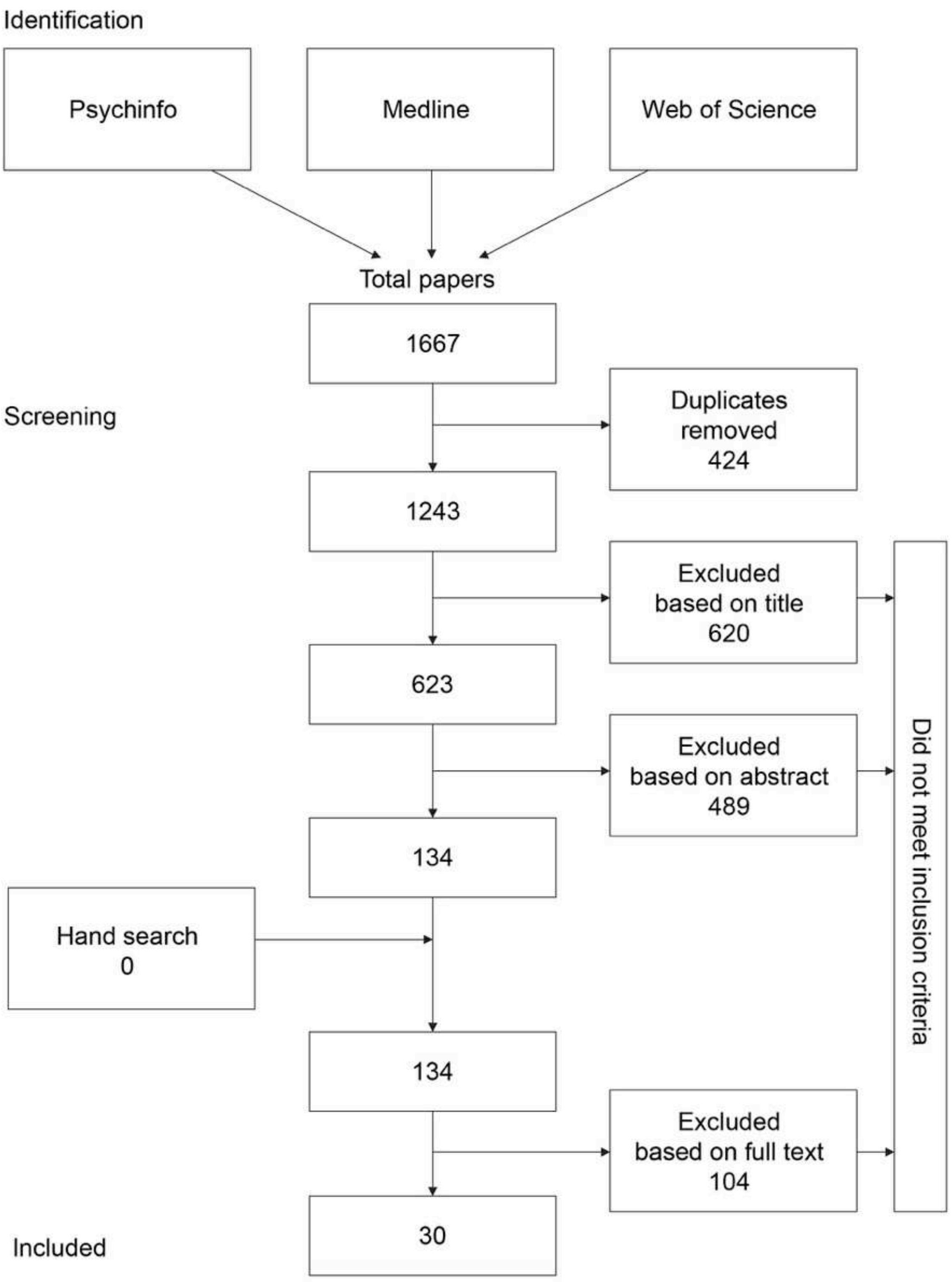
- Analysis addressed predictors of carer distress and attempted to quantify strength of relationship between predictor variables and carer distress. Specifically, the following analyses qualified for inclusion:
    - Predictors (or correlates) of carer distress. Moderators or mediators of associations between predictor variables and carer distress.
    - Predictors of change in carer distress as a proposed treatment outcome. Moderators and mediators of these associations.

Exclusion Criteria

- Studies in which the only otherwise relevant analysis was whether levels or changes in carer distress were predicted by treatment itself, or by the cared-for having an eating disorder, to avoid replicating recent comprehensive reviews by Anastasiadou et al. (2014) and Hibbs et al. (2015b).
- Studies that did not separate carers of someone with and without EDs (e.g. control groups) for analysis so that evidence of prediction of distress in carers of people with EDs specifically was unobtainable.
- Studies that analysed the same cohort of participants as presented by previous studies, and presented no new analysis.

A flow diagram of the selection process based on the PRISMA guidelines is presented in Figure 4.

Figure 4. Flow Chart of Systematic Search and Appraisal of Literature



Quality of Evidence

Assessment of studies was guided by CASP (2013a, 2013b) and STROBE (Vandenbroucke et al., 2014) checklists. While suitability of analytical approach is described numerically for individual predictors (Tables 9 and 11), a single numeric rating is not provided for studies overall to avoid oversimplification and misrepresentation of a diverse range of studies. As informed by Booth, Papaioannou and Sutton (2012), reliability and validity of such overall quality scores is poor.

Levels of statistical evidence.

Statistical analyses undertaken by included studies were categorised in this review by the way they related to carer distress and the extent to which they were able to offer insight into possible contributory mechanisms. Four levels of evidence of prediction (Chalmers et al., 2009) are described below, followed by brief explanation of moderation and mediation.

Correlation and prediction.

In terms of prediction, the most basic level of evidence is analyses of the simple association between the proposed predictor and carer distress in cross-sectional data (indicated in Tables 9 and 11 as '4'). This is followed by longitudinal design ('3'), more compelling due to the predictor occurring in time before carer distress. The nature of these equations mean 'predictor' is misleading, as relationships between the predictor and carer distress are observed equally in both directions.

The more robust approach is to include hypothesised predictor variables in multivariate regression with other potential predictors, as it allows at least some confounds to be controlled for ('2'). Most ideal is multivariate longitudinal design with distress measured at a later time-point than predictors (‘1’). Results from higher levels of evidence are afforded greater weight in corresponding evidence synthesis, as informed by Coren and Fisher (2006).

Given the increased risk of a Type I error when multiple statistical tests are run (Field, 2013), the number of comparisons (of each type, at each time-point) undertaken within studies and the same sample are provided (Tables 9-12), to contextualise levels of significance reported.

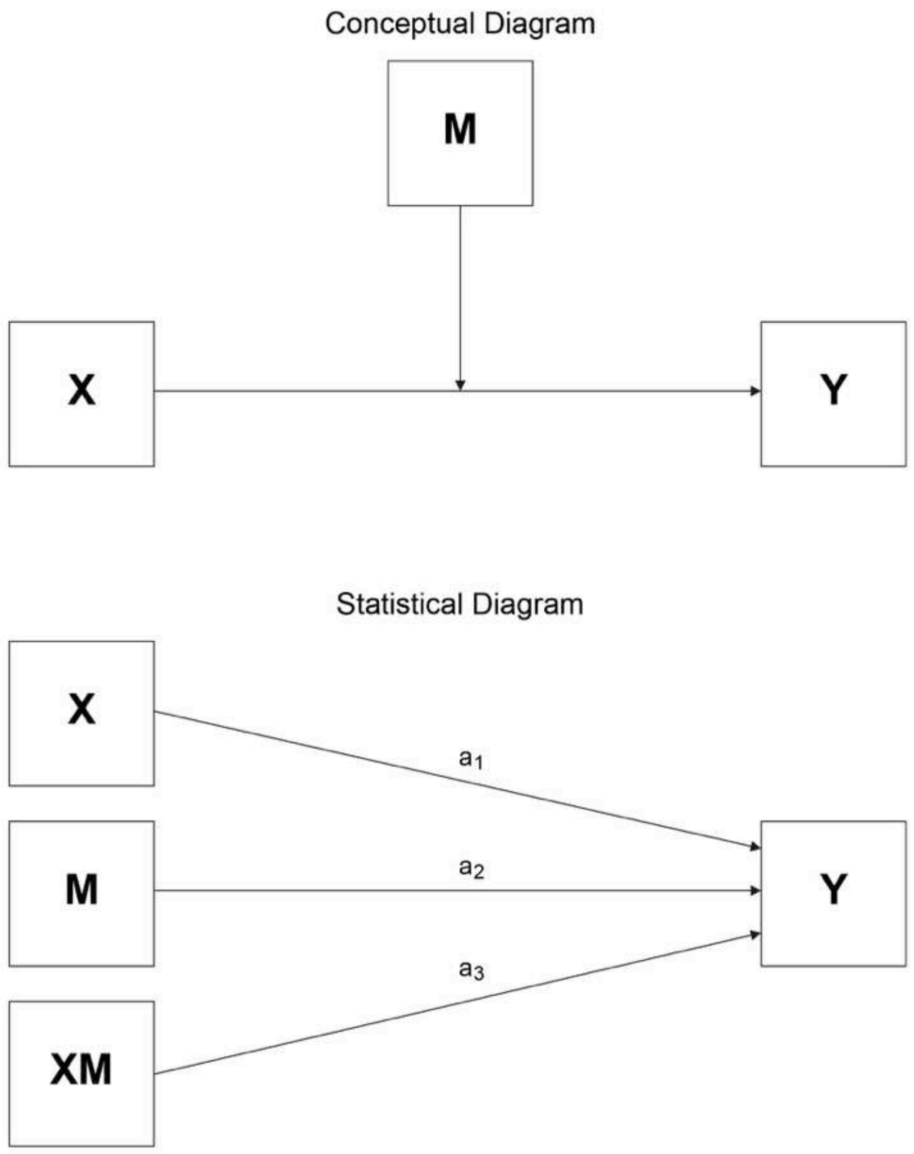
Strength of associations.

Associations between variables are described in terms of the correlation coefficient (*r,* and non-parametric r_s*)*, standardised Beta (β; where unavailable, unstandardized *b* is given) and regression coefficient R^2. Standardised β describes strength of relationship in terms of the regression slope, indicating the change in the dependent variable for each standard deviation (SD) change in the predictor, hence providing a basis for comparison across differently scaled measures. The higher the β value, the stronger relationship it has with the dependent variable (Field, 2013).

Moderators.

Moderation analysis determines whether the strength of relationship between two variables changes at different levels of the moderating variable. Analysis of moderators yields an R^2 change to the relationship as a result of the interaction (Hayes, 2013). Figure 5 illustrates the simple moderation model commonly tested.

Figure 5. Conceptual and Statistical Diagrams Showing Simple Moderation



Conceptual Diagram

Statistical Diagram

Adapted from Hayes (2013).

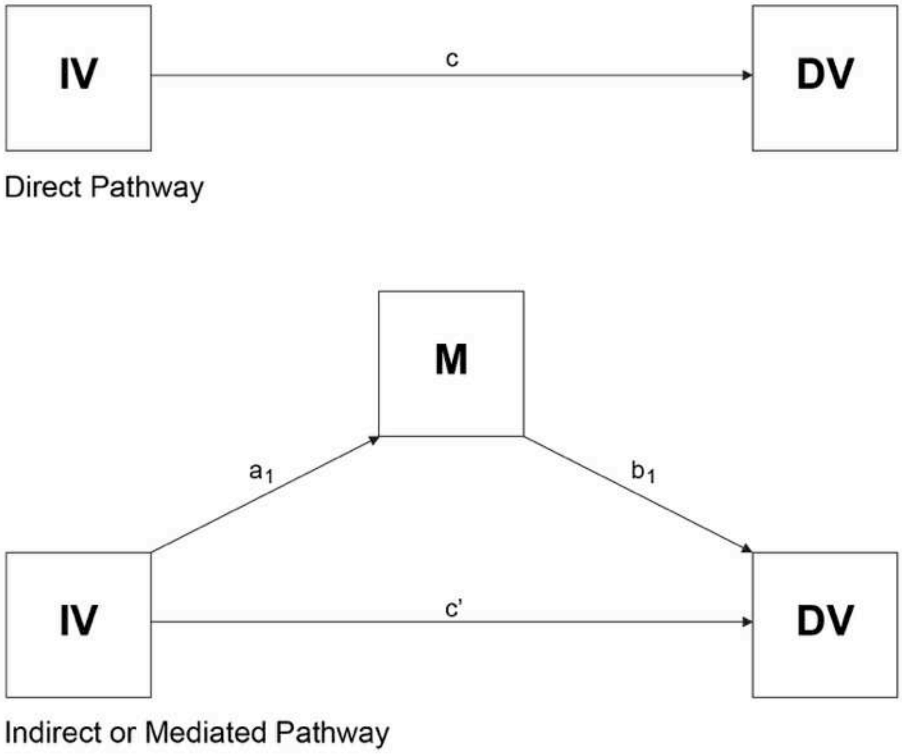
Predictors of change in distress over the course of an intervention could also be potential moderators of the effect of intervention on distress. However, while a pre-post design can be employed to test whether variables moderate the relationship between distress at baseline (predictor) and distress following an intervention (dependent variable), analysis of moderation of a relationship between intervention and carer distress requires a controlled design.

Mediators.

Ideally, mediators occur temporally between the predictor and outcome variable (Kraemer, Wilson, Fairburn, & Agras, 2002). However, cross-sectional mediation analysis remains valuable despite reduced potential to suggest mechanism of action (Hayes, 2013).

All predictors of carer distress could potentially mediate the relationships between carer distress and its other proposed predictors. Baron and Kenny's (1986) commonly employed steps for mediation analysis (see Figure 6) require the predictor to be significantly related to the mediator variable (*a*), the mediator to be significantly related to the dependent variable (*b*), and the relationship between the independent and dependent variable (total effect; *c*) to be significantly reduced when the mediator is included in the regression equation (direct effect; *c'*). The mediation is 'partial' if there remains a significant direct effect (c') when the mediator is included, and 'full' if c' is non-significant.

Figure 6. Conceptual and Statistical Diagram Showing Simple Mediation



Adapted from Hayes (2013).

Results

Characteristics, strengths and limitations of included studies are described below, followed by presentation of results from included studies.

Characteristics of Included Studies

The 30 included studies are presented by type in Tables 2 to 5; first, cross-sectional designs (12 included studies; Table 2), followed by cross-sectional analysis of intervention baseline data (10 studies; Table 3), before prospective cohort studies (four studies; Table 4) and intervention trials (five studies; Table 5).

Table 2

Studies of Cross-sectional Design

			Design - Cross-sectional			Sample										
						Cared-for					Carer					
Sample	Author	Area of focus	SU involvment	Setting	Sample	n	Age mean (SD)	Female %	Diagnosis	Duration illness, Y mean (SD)	n	Age mean (SD)	Female %	Relation-ship %	Living with % Contact h/w	Analysis in this review
b	Coomber & King, 2012	Predictors of carer distress and burden (part of longitudinal study).	None	Australia. Community (mail & online)	Advertised study via internet & local media, flyers at support group and ED treatment service. Self-selecting.	56	21.48 (8.67)	89	AN: 67.86 BN: 12.50 AN + BN: 10.71 ?: 7.14	5.09 (5.03)	56	45.79 (11.84)	78.57	M+F: 75 P: 14.29 S: 7.14 O: 3.57	78.5	MP, Med
	Dimitropoulos, Carter, Schachter, & Woodside Woodside, 2008	Predictors of family functioning and carer distress	None	Toronto. Inpatient ED hospital	Family (biological/legal) caregivers knowing of ED for ≥ 1 year, of people hospitalised for AN with a BMI ≤ 18.5. Carers recruited at cared-for's admission, or via advertising (self-selecting).	45	26.4 (8.2)	93	AN:100		63	49.5 (range: 22-70)	57.1	M: 44.4 F: 27.0 S: 7.9 P: 12.7 O: 7.9	- x̄ 30.7 (±35.6)	UP, MP
	Graap et al., 2008	Distress and need for support in AN and BN carers	I, M	Germany, Hospital	Adult (≥18yo) ED service-users and carers living with them or having frequent contact.	32	23.0 (6.0)	97	AN: 50 BN: 50	5.0 (5.2)	32	41 (12)	50	M: 43.8 F: 15.6 P: 34.4 O: 6.2	62.5	UP

Table 2 continued

Studies of Cross-sectional Design

			Design - Cross-sectional		Sample											
						Cared-for					Carer					
Sample	Author	Area of focus	SU involvment	Setting	Sample	n	Age mean (SD)	Female %	Diagnosis	Duration illness, Y mean (SD)	n	Age mean (SD)	Female %	Relation-ship %	Living with % Contact h/w	Analysis in this review
d	Haigh & Treasure, 2003	Development of carers' needs measure (CaNAM)	None	UK. Conference	Convenience sample of carers of people with AN. Recruited at carers' conference.	28	23 (8.6)	100	AN:100	5 (5.5)	28	52 (7.5)	57	Parents: 82 P: 7 O:8	64	UP
d	Kyriacou, Treasure & Schmidt, 2008	Predictors of carer distress and self/other-related and individual strains	None	UK. Community (mail)	Parents currently caring for child with AN, from IoP (NHS) carer volunteer database (self-selecting) and parents of inpatients approached.	91	23.3 (6.6)	100	AN: 100	7.8 (5.5)	151	54.3 (7.2)	91	M: 60 F: 40	61.5 >21: 62%	MP
f	Martin et al., 2013	Predictors of quality of life and burden in mothers & fathers (part of longitudinal study).	I, M	Spain. Community (mail)	Parents of 16-65yo outpatients of people with AN/BN invited to participate. *	113	23.16 (7.14)	98	AN: 47.79 BN: 19.47 EDNOS: 32.74	-	181	53.38 (7.98)	61	M: 61 F: 39	86.59 ≥32: 44%	UP
	Ohara, Komaki, Yamagata, Hotta, Kamo, & Ando, 2016	Factors associated with AN caregiving burden and carers' mental health	I	Japan. Community (mail).	Primary carers of outpatients with AN, recruited through	79	26.6 (7.9)	-	AN-R: 60.0 AN-BP: 39.2	8.8 (6.1)	79	56.0 (8.0)	-	M: 88.6 F: 6.3 O: 5.1	91.1 H/day: <3: 17.1, 3-6: 45.6,	UP, MP

Table 2 continued

Studies of Cross-sectional Design

			Design - Cross-sectional		Sample											
						Cared-for					Carer					
Sample	Author	Area of focus	SU involvment	Setting	Sample	n	Age mean (SD)	Female %	Diagnosis	Duration illness, Y mean (SD)	n	Age mean (SD)	Female %	Relation-ship %	Living with % Contact h/w	Analysis in this review
a	Sepulveda, Anastasiad-ou, del Rio,	Spanish validation of Level of Expressed Emotion scale	None	Spain. National health Hospital.	Primary and secondary carers. Recruited through ED treatment services and carers' association. *	186	19.7 (5.7)	100	AN: 70 BN: 30	2.6 (3.4)	270	48.8 (7.22)	63	Relatives	- -	UP
a	Sepúlveda et al., 2012a	Factors associated to wellbeing for primary and seconday carers	None	Spain. National health ED treatment hospitals	Primary and secondary carers for family member with ED, recruited through EDUs and carers' asociation.	185	19.2 (5.5)	94	AN:71.6 BN:28.4	2.6 (3.4)	277	48.7 (7.4)	63.5	M/F:96.8 P:.7 S:2.5	85.1 >21: 73.4	UP, MP
a	Sepulveda et al., 2014a	Predictors of distress in carers of purging and non-purging service-users	None	Spain. National health ED treatment service	Primary family carers of consecutive admisssions of outpatient services. *	177	19.37 (5.58)	93	AN-Restrictrive: 36.16 AN- P: 15.82 BN:15.25 EDNOS: 32.20	3.94 (4.38)	177	48.8 (7.4)	91.53	-	- -	UP, MP, Mod
	Sepulveda et al., 2014b	Spanish validation of expressed emotion measure (FQ)	None	Spain. National health Eating disorders service	Recruited through EDU. *	203	15.8 (3.41)	-	AN:71.0 BN:7.5 EDNOS: 21.5	Months:15.9	382	M:46.2 (5.1) F: 48.7 (5.55)	53.1	M:53.1 F: 46.9	- M>21: 86.2 F>21: 58.9	UP

Table 2 continued

Studies of Cross-sectional Design

			Design - Cross-sectional		Sample											
						Cared-for					Carer					
Sample	Author	Area of focus	SU involvment	Setting	Sample	n	Age mean (SD)	Female %	Diagnosis	Duration illness, Y mean (SD)	n	Age mean (SD)	Female %	Relation-ship %	Living with % Contact h/w	Analysis in this review
d	Whitney, Haigh, Weinman, & Treasure, 2007	Predictors of carer distress and negative caregiving appraisals	None	UK. Community (mail)	Primary carer from IoP (NHS) carers' volunteer database. Self-selecting.	115	24.0 (9.7)	74	AN: 88.5 BN: 11.5 BED: 0.89 O: 1.77	8.0 (8.0)	115	51.7 (7.6)	82	M: 80 F: 10.4 P: 7.8 S: 1.7	73 >21: 78%	MP

Note: Power: For multi-variate analysis, size of effect the study is powered to find is given. *Carers attending some Spanish treatment centres were routinely offered professional counselling as part of holistic ED treatment programme. **Only baseline cross-sectional data & TAU group longitudinal analysis included in this review. ***: Some patients recruited during/immediately after inpatient admission met all criteria for AN diagnosis except low weight. F%: Percentage of sample female. C: Carer. SU: Service-user. I: Carer identified for recruitment to study by cared-for. C: Cared-for consent gained. M: Cared-for completed measures/demographic details. AN: Anorexia Nervosa. BN: Bulimia Nervosa. EDNOS: Eating disorder not otherwise specified. BED: Binge eating disorder. ?: ED diagnosis not known or data missing. M: Mother. F: Father. P: Partner. S: Sibling. O: Other (friend, relation). EDU: Eating disorder treatment unit. Trmt: treatment. Y: years. Yo: years old. FU: Follow-up. UP: Univariate prediction. MP: Multivariate prediction. Mod: Moderation. Med: Mediation.

Table 3

Studies of Cross-sectional Design at Baseline of Intervention

						Sample										
						Cared-for					Carer					
Sample	Study	Area of focus	SU involvment	Setting	Sample	n	Age mean (SD)	Female %	Diagnosis	duration illness yrs mean (SD)	n	Age mean (SD)	Female %	Relationship %	Living with % Contact h/w	Analysis in this review
c	Goddard et al., 2013	Modelled relationships between carer and patient variables (part of carer skills RCT).	M	UK. Hospital EDUs (predominantly specialist NHS).	Carers' of SUs over 12 with AN presenting for in/daypatient treatment.	152	25.4 (8.5)	95	AN: 100	6.7 (6.9)	152	51.5 (9.9)	81	M: 79 F: 6 P: 14 O: 1	70 >21: 55%	UP, MP
e	Hibbs et al., 2015c	Development and validation of carer's skills measure; CASK (part of a carer's skills RCT).	I, M	UK. NHS Hospital EDU.	Carers taking part in skills workshops.	-	-	-	-	-	325	49.4 (7.6)	76	M: 41.54 F: 17.23 StepF: 1.54 S: 1.54 O: 0.62 ?: 37.53	-	UP
-	Quiles, Quiles, Pamies, Sepulveda, & Treasure, 2016	Spanish validation of accommodating and enabling scale; AESED (part of larger carers' skills intervention trial).	None	Spain. National health outpatient ED service.	Recruited through national health hospital EDU. All C offered professional counselling.	90	18.8 (6.0)	92	AN-R: 55.7% AN-P:9.8% BN-P: 6.6 BN-nonP: 1.6 EDNOS1 3.1%	-	90	47.8 (8.1)	61.10%	M:57.8 F:32.2 P:4.4 S:3.3 O:2.2	100 M>21: 48.1 F>21:42.9	UP
c	Raenker et al., 2013	Time spent caregiving, distress and social support in carers of AN hospital patients (part of trial of carer's skills intervention).	M	UK. Community (mail & telephone).	Parents and P of consecutive AN hospital admissions.	178	25.8 (9.2)	95	AN: 100	Parent as caregiver 6.2 (6.1) P as caregiver 14.4 (12.3)	267	M: 53.3 (7.3) F: 54.9 (8.6) P: 39.3 (12.1)	53.93	M: 53.93 F: 29.96 P: 10.49	M: x̄ 90.6 F: x̄ 39.0 P: x̄ 87.8	Med

Table 3 continued

Studies of Cross-sectional Design at Baseline of Intervention

					Sample											
						Cared-for					Carer					
Sample	Study	Area of focus	SU involvment	Setting	Sample	n	Age mean (SD)	Female %	Diagnosis	duration illness yrs mean (SD)	n	Age mean (SD)	Female %	Relationship %	Living with % Contact h/w	Analysis in this review
-	Rienecke, Richmond, & Lebow, 2016	Service-user and parent therapeutic alliance, correlates of parent alliance, and alliance-treatment relationship. Pre-post deisgn, baseline measure 2 weeks into treatment.	M	USA. Hospital EDU.	Parents of adolescent patients of AN hospital treatment programme, recruited through EDU.	56	15.78 (2.89)	93	AN: 73.21 EDNOS-AN:26.79	1.36	78	-	-	-	M:62.82 F: 37.18	UP
e	Rhind et al., 2016	Mediators of relationship between time spent caregiving and distress (part of carer skills RCT).	I, M	UK, outpatient hospital.	Parents of adolescents (13-21y) presenting for outpatient treatment of AN.	144	16 (2.14)	92	AN: 74 EDNOS-AN: 26	<3: 79%	196	M: 48.3 (4.9) Step/F : 49.5 (5.7)	68.88	M: 68.8 F: 31.1 StepF: 3.06	100 >21 face to face: 75%	UP, Med
g	Sepulveda, Whitney, Hankins, Treasure, 2008	Development and validation of scale measuring ED-specific burden (EDSIS) Pre- and post- carer skills intervention.	None	UK. NHS Hospital & Community (mail & telephone).	Carers who enrolled for workshops advertised through beat, EDU and hospital referrals, IoP volunteer database.	156	23.5 (8.3)	93	AN: 76.13 BN:23.86	-	190	51.1 (8.6)	73.1	M+F: 67.90 P: 6.3 S: 3.5 O: 0.7	-	UP

Table 3 continued

Studies of Cross-sectional Design at Baseline of Intervention

					Sample											
						Cared-for					Carer					
Sample	Study	Area of focus	SU involvment	Setting	Sample	n	Age mean (SD)	Female %	Diagnosis	duration illness yrs mean (SD)	n	Age mean (SD)	Female %	Relationship %	Living with % Contact h/w	Analysis in this review
g	Sepulveda, Kyriacou, & Treasure, 2009	Development and validation of accommodation and enabling scale (AESED) pre-and post carers' skills intervention (DVD & workshops).	None	UK. NHS Hospital & Community (mail & telephone).	Primary carers who enrolled for workshops advertised through beat, EDU and hospital referrals, IoP volunteer database.	193	21.3 (6.8)	96	AN: 70.5 BN:25.5 EDNOS: 5	-	193	49.1 (8.4)	85.5	M+F: 91.0 P: 5.2 S/O: 3.8	-	UP
	Winn et al., 2007	Predictors of carer distress (part of RCT evaluating CBT for BN).	I, M	UK. NHS hospital EDU.	Carers of adolescents (13-20) referred for treatment for BN/EDNOS(BN criteria except purging frequency).	68	17.7 (1.7)	97	BN-P: 66.2 BN-nonP: 5.95 EDNOS: 27.9	median: 2 (0.17)	112	S/P: 18.6 (4.6) M/F: 46.2 (5.7)	62.5	M: 44.6 F:18.8 P: 5.4 S: 17.0 O: 14.6	86.6	MP

Note: Sample a-g; please see Table 6. Power: For multi-variate analysis, size of effect the study is powered to find is given. *1: Carers attending some Spanish treatment centres were routinely offered professional counselling as part of holistic ED treatment programme. F%: Percentage of sample female. C: Carer. SU: Service-user. I: Carer identified for recruitment to study by cared-for. M: Cared-for completed measures/demographic details. AN: Anorexia Nervosa. BN: Bulimia Nervosa. EDNOS: Eating disorder not otherwise specified. BED: Binge eating disorder. ?: ED diagnosis not known or data missing. M: Mother. F: Father. P: Partner. S: Sibling. O: Other (friend, relation). EDU: Eating disorder treatment unit. Trmt: Treatment. Y: Years. Yo: Years old. FU: Follow-up. UP: Univariate prediction. MP: Multivariate prediction. Mod: Moderation. Med: Mediation.

Table 4

Studies of Prospective Cohort Design

			Design - Prospective cohort			Sample											
							Cared-for					Carer					
Sample	Study	Area of focus	Timepoints & design	SU involvment	Setting	Sample (attrition by final timepoint)	n	Age mean (SD)	F%	Diagnosis	duration illness yrs mean (SD)	n	Age mean (SD)	F%	Relationship %	Living with % / Contact h/w	Analysis
b	Coomber & King., 2013	Predictors of carer's distress and burden across 9m.	B, 4.5m, 9m. Part of larger longitudinal study.	None	Australia: Community - mail & internet.	Carers recruited via web, media, carer's groups, local EDU advertising. Self-selecting. (23.33%)	56	21.48 (8.67)	89	AN: 79 BN: 13 BED: 2 ?: 11	5.09 (5.03)	56	45 (11.84)	79	M+F:75.0 P: 14.29 S: 7.14 O: 3.57	- x̄ 78.9 (± 63.83)	UP, MP
f	Gonzalez et al., 2012	Spanish validation of involvement evaluation questionnaire (IEQ).	B, 1y	I? C, M	Spain: Community (mail).	Primary carers recruited through ED outpatient service	246	25.73	99	AN: 44.14 BN:24.83 EDNOS: 31.3	>5y: 56.24	246	≤45: 33.33 46-60: 51.63 >60: 15.04	53	M: 45.12 F: 28.46 P: 13.82 O: 12.60	85.66 >32: 45%	UP
f	Las Hayas et al., 2014	Changes in caregivers' quality of life and distress over 1 year.	B, 1y	I, C, M	Spain: National health outpatient ED service.	1 carer per family, 18-65yo outpatients. (50.2% C & 45.7% CF 1y, 78.63% C to y2 [no analysis])	127	25.88 (9.50)	100	AN: 53.62 BN: 18.84 EDNOS: 27.54	-	219	49.35 (11.73)	54	M: 45.87 F: 29.36 P: 12.84 O: 11.93	90.74 ≥32: 53.33%	UP, MP, Mod

Table 4 continued

Studies of Prospective Cohort Design

			Design - Prospective cohort			Sample											
							Cared-for					Carer					
Sample	Study	Area of focus	Timepoints & design	SU involvment	Setting	Sample (attrition by final timepoint)	n	Age mean (SD)	F%	Diagnosis	duration illness yrs mean (SD)	n	Age mean (SD)	F%	Relationship %	Living with % Contact h/w	Analysis
f	Orive et al., 2013	Anxiety and depression among caregivers of patients with eating disorders and their change over 1 year.	B, 1yr	I, C, M	Spain: Community (mail).	Caregivers of outpatients 16-65. *1. (42% C & CF).	84	26.46 (9.50)	100	AN/BN/ED NOS:100 Purgative: Restrictive : 47		143	49.92 (11.21)	55	M:47.55 F:31.47 P:11.19	87 ≥32: 49%	UP, MP

Note: Sample a-g; please see Table 6. Power: For multi-variate analysis, size of effect the study is powered to find is given. *1: Carers attending some Spanish treatment centres were routinely offered professional counselling as part of holistic ED treatment programme. F%: Percentage of sample female. C: Carer. SU: Service-user. I: Carer identified for recruitment to study by cared-for. M: Cared-for completed measures/demographic details. AN: Anorexia Nervosa. BN: Bulimia Nervosa. EDNOS: Eating disorder not otherwise specified. BED: Binge eating disorder. ?: ED diagnosis not known or data missing. M: Mother. F: Father. P: Partner. S: Sibling. O: Other (friend, relation). EDU: Eating disorder treatment unit. Trmt: Treatment. Y: Years. Yo: Years old. FU: Follow-up. UP: Univariate prediction. MP: Multivariate prediction. Mod: Moderation. Med: Mediation.

Table 5

Studies Addressing Distress as an Intervention Outcome

			Design				Sample											
								Cared-for					Carer					
Sample	Study	Area of focus	Timepoints & design	Intervention	SU involvment	Setting	Sample*	n	Age mean (SD)	Female %	Diagnosis	duration illness yrs mean (SD)	n	Age mean (SD)	Female %	Relationship %	Living with % Contact h/w	Analysis in this review
Pre-post designs																		
-	Goddard et al., 2011	Outcomes, moderators and mediators of a carer skills intervention	Pre-post design. Assessment of change over preliminary waiting period and 3m follow-up. B, EoT (6wks), FU (EoT+3m).	Carers skills intervention: 1 book, 5 DVDs, 1/2 sample recieved additional telephone coaching (no effect of coaching found).	None	UK. Community (post and telephone).	Community sample, self-selected for intervention in response to advertising. 18.54% attrition by EoT.	153	20.8	95.4	AN: 79.74 BN: 7.84 EDNOS: 3.27 ?: 1.96	Median: 3	153	49.64	88.9	M: F: 5.88 P: 5.23 O: 3.92	76.47 face to face> 21:	UP, Mod, Med
g	Sepulveda et al., 2010	Expressed emotion, distress and negative appraisals of caregiving following carer's psychoedu-cation.	Pre-post design. B, EoT (3m), FU (EoT+3m).	6x2hr carers psychoeduca tion workshops over 3, plus 1 FU workshop at 3m.	None	UK. NHS Hospital & Community (mail & telephone).	Primary caregivers of AN patients who completed workshops advertised through beat, EDU and hospital referrals. 1.74%	46	21.5 (5.3)	93.5	AN: 77.8 BN: 22.2	3.07 (3.1)	46	53.33 (7.7)	91.5	M+F: 95.6 O: 4.4	76.1 >21: 62.2% <21: 37.8%	MP
-	Slater, Treasure, Schmidt, Gilchrist & Wade, 2014	Moderators of impact of treatment for patient on carer distress.	Pre-post design. B, sessions five, 10, 25 (EoT), FU (EoT+3m), FU (EoT+12m)	10-month AN outpatient treatment (predominan-tly individual) for adult service-user.	I, M	UK. NHS outpatient hospital EDU.	Carers of service-user who were willing to have regular physical checkups, no acute issues.	12	-	89	AN: 100	-	12	-	67	M: P: 25.0 S:17.67	- -	Mod

Table 5 continued

Studies Addressing Distress as an Intervention Outcome

			Design				Sample											
								Cared-for					Carer					
Sample	Study	Area of focus	Timepoints & design	Intervention	SU involvment	Setting	Sample*	n	Age mean (SD)	Female %	Diagnosis	duration illness yrs mean (SD)	n	Age mean (SD)	Female %	Relationship %	Living with % Contact h/w	Analysis in this review
Randomised Controlled Trials																		
-	Grover et al., 2011	Internet-based systemic CBT intervention for carers.	RCT. Baseline, EoT (4m), FU (B+6m).	RCT of internet-based CBT for carers compared to ED charity ad-hoc support.	None	UK. Community (Online + email/telephone).	Carers of pepole with AN, advertised through beat website, carers groups and clinical departments.	63	20.4 (6.2)	-	AN-R: 60.3 AN-P: 17.5 EDNOS:2 0.6 ?: 1.6	4.3 (4.5)	63	48.2 (7.6)	-	M: 79.4 F: 7.9 P: 7.9 O: 4.8	77.8 -	Mod
e	Salerno et al., 2016	Relationships between baseline and post-treatent distress in patient and carer dyads.	Part of large multi-site RCT. B, 1y.	Carers' skills intervention with guided/self-help.	I, M	UK: NHS hospital EDUs.	Primary carers living with adolescents (12-21yo) presenting for outpatient trmt with primary diagnosis of AN/atypical AN.	50	16.86 (2.06)	96	AN:100	1.90 (1.88)	50	48.52 (4.92)	100	M: 98 S: 2	100 -	MP

Note: Sample a-g; please see Table 6. Power: For multi-variate analysis, size of effect the study is powered to find is given. *1: Carers attending some Spanish treatment centres were routinely offered professional counselling as part of holistic ED treatment programme. F%: Percentage of sample female. C: Carer. SU: Service-user. I: Carer identified for recruitment to study by cared-for. M: Cared-for completed measures/demographic details. AN: Anorexia Nervosa. BN: Bulimia Nervosa. EDNOS: Eating disorder not otherwise specified. BED: Binge eating disorder. ?: ED diagnosis not known or data missing. M: Mother. F: Father. P: Partner. S: Sibling. O: Other (friend, relation). EDU: Eating disorder treatment unit. Trmt: Treatment. Y: Years. Yo: Years old. FU: Follow-up. UP: Univariate prediction. MP: Multivariate prediction. Mod: Moderation. Med: Mediation.

Most (95%) of the participants with eating disorders were female (data calculated from the 26/30 studies in which it was available). The average age was 21.72 for cared-fors (range = 16 - 26, calculated from 28/30 studies), and 49.54 for carers (range = 41 - 56, calculated from 26/30 studies). Among carers, 57.99% (calculated from 20 studies) were mothers and 28.46% fathers (calculated from 20/30 studies; 7.42% of fathers were step-fathers, calculated from 2/30 studies), 6.21% were partners (calculated from 27 studies) and 1.48% siblings (calculated from 26/30 studies). Other carers were extended family, offspring, and friends. In the 19 studies including this information, 83.61% carers lived with the cared-for. Fifteen studies were conducted in the UK, nine in Spain, two in Australia, two in the USA, two in Germany, and one in Japan. The participants with ED were diagnosed (calculated from 29/30 studies) with 72.21% AN, 15.23% Bulimia Nervosa, 10.16% EDNOS, 1.54% Binge Eating Disorder, and 0.52% other/unknown, with a mean illness duration of 4.41 years (range = 1.33 – 8.8, calculated from 17/30 studies). Twenty two studies recruited carers through treatment settings, and 11 relied on cared-fors identifying carers. Most studies excluded participants with significant physical or psychiatric comorbidities, and many included only primary caregivers, defined by the cared-for, caregiver, or number of hours spent caregiving.

Five (three pre-post and two RCT) studies investigating predictors, moderators and/or mediators of distress as a proposed intervention outcome, three involved carers' skills programmes, one an online carer's CBT trial and one a treatment for cared-fors. Cared-fors were young (mean age 20′, calculated from 4/5 studies), and predominantly had diagnoses of AN (79.56%). Most (80.46%) lived with carers, who were mostly mothers (84.89%). As effects of carers interventions have recently been reviewed (Hibbs et al., 2015b), predictors of carer distress are presented for treatment as usual (TAU), but not intervention groups (where applicable) as change in relationships between risk factors and distress may be confounded by intervention. Predictors of the effect of intervention on distress as an outcome are presented separately. Salerno et al.'s (2016) experimental design investigated predictors of change in distress in the intervention and treatment as usual (TAU) groups, presented in Table 6 (intervention) and appendices E and F (TAU), with between-group differences discussed below.

Repeated testing.

Several studies analysed participants from the same cohort (private correspondence with A. Sepulveda, Treasure, R. King and Martín, Oct. 2016, appendix A). These samples are described in appendix B, and marked in Tables 2-5 as *a* to *g.* Where analyses have utilised many of the same participants' data, duplicated analyses have been excluded, with only the most robust (see 'levels of evidence', above) of each included in results (where quality of evidence did not differ, the most detailed is presented, or where otherwise equal, the first published). Therefore, 13 analyses have not been presented (see appendix C) across four included studies. One paper (Carral-Fernandez et al., 2013), which contributed only two replicated analyses, was excluded.

Measures

A heterogeneous selection of measures had been used by the studies (see appendix D) complicating comparison of results. The validity of several measures for this sample was questionable (DBS, Cockell, Geller, & Linden, 2003; FCS, Semple, 1992; J-ZBI_8, Zarit, Reever, & Bach-Peterso, 1980), and authors sometimes altered validated measures when suitable instruments were unavailable (CNA, Wancata et al., 2006; DCCF, Struening et al., 2001; SNQ, Magliano et al., 1998; WAI-S, Horvath & Greenberg, 1989). New measures created specifically for this population were used (AESED, Sepulveda, Kyriacou, & Treasure, 2009; EDSIS, Sepulveda et al., 2008) although some were not yet thoroughly validated (CaNAM, Haigh & Treasure, 2003; Care-ED, Raenker et al., 2013). However, all studies assessed distress using well-validated measures with good psychometric properties.

Results of Included Analyses

Results are presented verbally below, with further detail provided in tables (6, 7 and 8, and appendices E and F). A brief overview of the studies which included multivariate analysis is presented in appendix E, showing the amount of variance accounted for by significant predictors in the final models. For brevity, data from subscales is given only when differing in significance to the total scale score, or when the strength of relationship is notably different. Generally, models accounted well for the variance in distress (R^2 = 24.0 - 60.2%, where given) with the most common predictors in significant models being aspects of burden and carer distress. Quality of evidence is considered below.

Predictors of carer distress.

Predictors (non-intervention-related) of carer distress are presented by type below and in appendix F. Unless otherwise mentioned, predictive relationships were positive.

Demographic characteristics of the person with the ED, and clinical information.

The cared-for being older was associated with greater carer distress (Goddard et al., 2013, Level 2 analysis), but not change in distress over time (Salerno et al., 2016, level 1 analysis). As age of onset is typically adolescence (Currin, Schmidt, Treasure, & Jick, 2005; Micali, Hagberg, Petersen, & Treasure, 2013) age is related to illness duration. However, illness duration did not persuasively predict carer distress, although a longer duration predicted lower anxiety in secondary carers[3] (Sepúlveda et al., 2012a). A longer illness predicted less improvement in anxiety over a year in one study with level 2 analysis, presumably due to the likelihood that the chronic illness persisted (ibid), but this disappeared in multivariate analysis, possibly suggesting the univariate finding had been confounded. The cared-fors' rejection of having the ED and the carers' help, and the cared-fors' ratings of importance of, and confidence in recovery did not predict carer distress.

Orive et al. (2016) reported cared-for suicidal intent (clinician-rated) predicted carer depression (level 2 analysis) and cared-for drug addiction predicted carer anxiety in multivariate analyses. Beta values for relationships between cared-for comorbidities and carer distress were larger than for other predictors reviewed. These comorbidities did not predict carer distress longitudinally.

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[3] Defined as carers with less involvement with the cared-for than a 'primary carer' involved in the study.

Eating disorder symptoms.

Interestingly, the only study reporting ED symptoms as a significant predictor of carer distress was Ohara et al. (2016), which used carer observations of AN symptoms; conceivably more vulnerable to subjective changes linked with the carer's level of distress than cared-for or clinician ratings. Multivariate and univariate analyses both described non-significant findings, although power may have been an issue. Salerno et al.'s (2016) high quality analysis (level 1) reported that BMI did not predict change in carer distress, and the analyses by Goddard et al. (2013, level 2) and Rhind et al. (2016, level 4) also found no effect. Treatment received by the cared-for and their compensatory behaviour (purgative/restrictive) were not predictors. However, treatment received may not be an adequate measure of severity (Sepulveda et al., 2014a) considering cared-for unwillingness for treatment is associated with EDs (Goldner, 1989). Results for purging, discussed further below, were mixed.

Cared-for distress and quality of life.

At baseline, level 4 evidence suggested small-moderate correlations between cared-for and carer distress (Goddard et al., 2013; Rhind et al., 2016). Cared-for distress at baseline predicted change in carer distress at one-year follow-up (Salerno et al., 2016), and improvement in cared-for depression and mental health predicted improvements in carer depression and anxiety, respectively, after a year (Las Hayas et al., 2014) in high quality analysis (level 1). Investigating in more detail, Orive et al. (2013, level 3) reported only possible, not clinical, depression at baseline predicted improvement in carer depression after a year, and was only associated with cared-for depression, not anxiety. Las Hayas et al.'s (2014) longitudinal univariate analyses indicated a relationship between cared-for quality of life and change in carer anxiety over a year. However, there were a large number of comparisons for this sample, and the mixed results are inconclusive, despite well-validated measures.

Carer demographics and clinical information.

Gender and relationships.

Being a mother compared to a father predicted higher levels of distress in level 2 analysis (Kyriacou, Treasure, & Schmidt, 2008), albeit with a small effect size (R^2 = .035). Orive et al. (2013, level 2) expanded upon this, finding fathering the cared-fors to be predictive of higher anxiety but not depression. Also, being a sibling or offspring predicted lower depression compared to mothers, with a comparatively high Beta compared to other predictors in this review (ibid). No significant differences were found between partners and mothers (ibid) and type of relationship between carer and cared-for did not predict change in carer distress over time (level 3, Las Hayas et al., 2014). Generally, contact time was not found to be a predictor of distress, with the exception of Rhind et al.’s (2016) sample of mothers, and Goddard et al.'s (2013) sample, of whom 79% were mothers.

Carer distress, quality of life and eating difficulties.

Unsurprisingly, carer anxiety and depression (sub-scales of carer distress measures), were strongly linked, positively predicting each other in cross-sectional and longitudinal analysis. Additionally, higher carer distress at baseline significantly predicted more change in carer distress over a year. These unanimous effects were reported by four studies, three of which included carer distress (anxiety/depression) as a predictor in multivariate analyses (levels 2, 1).

Martin et al. (2013) reported mental quality of life for parents, and physical quality of life for mothers but not fathers, to negatively correlate with distress, although a large number of comparisons were made. Goddard et al. (2013) reported carer history of ED to positively predict distress. These single level 4 and 2 analyses, respectively, require replication.

Hypothesised reactions to the eating disorder.

Expressed emotion.

With the exception of Coomber and King (2012) who may have been under-powered to find a medium-sized correlation (as reported by Hibbs et al., 2015a), and some of the Levels of Expressed Emotion measure not predicting distress in their Spanish translation validation study (Sepulveda et al., 2012b), the data clearly indicate a relationship between expressed emotion and distress in both mothers and fathers in cross-sectional studies. Size of associations were comparatively large compared to others in this review, however, expressed emotion was a non-significant predictor in longitudinal analyses.

Family functioning and psychological control.

Regarding family functioning and conflict, evidence of a relationship was unconvincing, with both multivariate analyses (level 2) failing to show significant relationships with carer distress.

In the single investigating study, the psychological control that carers rated themselves as having over the cared-fors (Goddard et al., 2013) positively predicted carer distress, including in multivariate analysis.

Accommodating and enabling.

There were a wealth of significant cross-sectional correlations between accommodation and distress. The only study to address this with a multivariate design however (level 2, Goddard et al., 2013), found it not to be a significant predictor.

Experience of caregiving.

The clearest predictor of carer distress was burden, as shown by seven studies' univariate analysis and three studies' multivariate analysis, accounting for between 6.5% and 31% of variance in distress (see appendix E) with positive associations mainly small to moderate-sized (see appendix F). The only outcome not fitting this pattern was Dimitropoulos et al. (2008) showing p =.06 in level 2 analysis not powered to detect anything smaller than medium-large effect sizes, and using a partially-validated measure. Outcomes were significant to highly significant across the GHQ-12, and HADS. In high quality analysis (level 1) with fully-validated measures, Las Hayas et al. (2014) reported higher caregiving burden at baseline to predict less improvement over a year, while change in burden positively correlated with change in distress. A much less decisive picture was presented by studies investigating suggested components of burden, using subscales and components of subscales, although there was good evidence for the contribution of nutritional problems to all but primary carer depression in this sample (see appendix F), with comparatively high Beta values ($\beta = 0.3 – 0.5$).

Positive aspects of caregiving were assessed using ECI subscales by two studies of relatives (predominantly parents) of adolescents with short illness duration, with cross-sectional, multivariate analysis. Positive experiences predicted lower depression in secondary carers, but not primary carers, or anxiety in either (Winn et al., 2007; Sepulveda et al., 2012a). Good relationships were found to predict lower carer anxiety, but not depression (ibid). One multivariate analysis (Whitney et al., 2007) found stigma to have a positive association with distress.

Burden, expressed emotion and purging.

Sepulveda et al. (2014a, level 2 analysis) found differences in relationships between distress and subscales of both expressed emotion and burden for caregivers of individuals who purged compared with those who did not purge. For caregivers of someone who purged, impact of nutrition (β = .44, *p* <.001) and carer intrusiveness (β = .24, *p* <.05), significantly predicted carer distress, while for caregivers of someone who did not purge, the significant predictors were impact of nutrition (β = .47, *p* <.001), guilt (β = .28, *p* <.01) and attitude towards the illness (β = .18, *p* <.05, see appendix E for details of non-significant predictors in the models). Therefore, intrusiveness predicted distress only for carers of individuals who purged, and guilt and attitude towards the illness predicted distress only for carers of individuals who did not, while impact of nutritional problems was a significant predictor in both.

Needs and coping.

Carers’ needs for themselves, for example for information and support, were not found to be predictors of distress at the same or later time-points, from all levels of analysis. From one level 4 analysis, carers’ reports of unmet needs in terms of unresolved ED problems requiring interventions significantly positively correlated with carer distress, although this relied on a measure adapted for the study and not fully-validated (Graap et al., 2008).

Maladaptive coping styles at baseline were found to predict distress, cross-sectionally (Ohara et al., 2016) and 4.5 months, but not 9 months, later; Coomber and King (2013) described a fluctuating pattern of distress over time, whereas rates of maladaptive coping remained high, possibly explaining the inability to predict the levels of distress longer-term.

Protective factors.

Using multivariate analysis (level 2) Sepulveda et al. (2012a) found a higher level of education to predict lower distress, particularly depression. However, this was not the case for secondary carers, who had received significantly higher education and were less distressed than primary carers (ibid). Orive et al. (2013, level 2) found the highest level of education being university (but not secondary) to predict lower depression.

Rhind et al. (2016) found social support, and Ohara et al. (2016) found affective, but not practical, social support to predict lower distress cross-sectionally. Dimitropolous et al.'s (2008) non-significant findings for social support with level 4 analysis may have been due to measures that were not validated and potentially unsuitable. Professional support or therapeutic alliance were not demonstrated as predictors. Distress was not predicted longitudinally by any suggested protective factors.

In two studies' univariate analyses (level 4), caregiving skills were significantly correlated with lower distress, for both mothers and fathers. Sepulveda et al. (2012a, level 2) found positive aspects of caregiving protective only for secondary caregivers.

Mediators of relationships between predictors and carer distress.

The simple mediational model described in Figure 6 analysed cross-sectional relationships between predictors (non-intervention-related) and carer distress in three studies (Table 6).

Rhind et al. (2016) tested accommodation, carers' skills, and expressed emotion as mediators of the effect of time spent caregiving on carer distress, estimating models for mothers and fathers separately. The only significant (partial) mediation occurred for mothers; greater time spent caregiving predicted greater accommodation, which in turn predicted greater distress. Coomber and King (2012) found the relationship between expressed emotion and psychological distress was mediated by maladaptive coping with positive relationships between all three variables, but quality of social support was not a mediator. Raenker et al. (2013) reported that more time spent caregiving predicted higher carer distress, with burden fully mediating the relationship. Social support was a partial mediator of the relationship between burden and distress, and was negatively related to both (ibid).

Predictors, Moderators and Mediators of Distress as a Proposed Treatment Outcome

Predictors of proposed treatment outcome.

Only three studies appraised predictors of change in carer distress before vs. after an intervention. Two employed pre-post designs (Goddard et al., 2011; Sepulveda et al., 2010) and one an RCT (Salerno et al., 2016) which investigated predictors of change in distress in the intervention group without employing the treatment condition (intervention/no intervention) as a predictor, meaning the experimental design was not harnessed for this analysis. Although the proposed predictors of change presented in Table 7 could be candidates for moderation and/or mediation analysis, the only study of these three to do this was Goddard et al. (2011; Tables 8, 9). Salerno et al.'s (2016) high quality (level 1) analysis reported cared-for factors not to predict change in carer distress, but higher carer distress at baseline predicted greater improvement in carer distress at one-year follow-up after a skills intervention (ibid) with a higher Beta (0.43) than for many other predictors. Increase in carers skills, and reduction in accommodation and expressed emotion correlated with change in carer distress (Goddard et al. 2011, level 3). The latter was supported by Sepulveda et al.'s (2010) finding from level 1 analysis with fully validated measures that higher baseline expressed emotion accounted for 19% of variance in change to carer distress at end of treatment, with the subscale Critical Comments accounting for 33%.

Moderators of proposed treatment outcome.

Three studies, presented in Table 8, performed simple moderation analysis (Figure 5).

Table 8. Moderators of Proposed Effect of Intervention on Change in Carer Distress

Only Grover et al. (2011) used a controlled design, meaning this was the only set of analyses to assess moderation of the relationship between intervention and carer distress. They did not identify any moderators, but were underpowered (ibid). The remaining two studies used a pre-post design to assess change in carer distress before vs. after an intervention. Slater et al. (2014) reported a significant difference between carers whose cared-for had lower (≤ 17.5) vs. higher BMI at baseline, with the former showing a decrease in distress between three and 12 months after cared-for treatment, and vice versa. They did not find living together or duration of illness moderated this change in distress over the same time-period. Goddard et al. (2011) reported both expressed emotion and accommodation as moderators, with a greater decrease in distress following intervention for people with high expressed emotion and accommodation at baseline.

Mediators of the proposed effect of intervention on carer distress.

Only Goddard et al. (2011) addressed mediators of change in carer distress as a proposed outcome (Table 9). Reduction in distress between baseline and the end of a carers' skills intervention was partially mediated by reduction in expressed emotion and accommodation, and increase in carers' self-efficacy. Analysis of change in contact time as a mediator was not undertaken as assumptions were not met.

Table 6

Mediators of the Relationship bewteen Carer Distress and its Predictors

											Total effect		Direct effect		Predictor on mediator		Mediator on outcome		Total indirect effect		
Sample	Author	Predictor	Who	Measure	Time	Mediator	Measure	Time	Outcome measure	Time	β	*p*	β	*p*	β	*p*	β	*p*	Size	Sig.(BCI)	Mediation
b	Coomber & King, 2012	Expressed emotion	C	FQ	0	Maladaptive coping	Brief COPE	0	GHQ-12	0	0.4	<.01	0.25	>.05	0.4	<.05	0.36	<.01	-	0.02 - 0.23	Full
			C	FQ	0	Social support	SSQ6	0	GHQ-12	0	-	-	-	-	-	>.05	-	>.05	-	-	none
c	Raenker et al., 2013	Time spent caregiving	C	Care-ED h/m	0	ED-specific burden	EDSIS	0	DASS	B	0.2	0	0.07	0.26	0.21	0	0.46	<.001	-	-0.03 - -0.13	full
		ED specific burden	C	EDSIS	0	Social support	WHOQOL Social relation-ships ss	0	DASS	B	0.3	<.001	0.41	<.001	-0.24	<.001	-0.3	<.001	-	0.001-0.003	partial
e	Rhind et al., 2016	Time spent caregiving	M	Care-ED	B	Accomm-odating	AESED	B	DASS	B	0.2	<.01	0.08	>.05	0.32	<.001	0.38	<.001	-	-	full
			F	Care-ED	B	Accomm-odating	AESED	B	DASS	B	0.1	>.05	-0.07	>.05	0.22	>.05	0.39	<.01	-	-	none
			M	Care-ED	B	Expressed Emotion	FQ, BDSEE	B	DASS	B	0.2	<.01	0.19	<.05	0.11	>.05	0.51	<.001	-	-	none
			F	Care-ED	B	Expressed Emotion	FQ, BDSEE	B	DASS	B	0.1	>.05	-0.02	>.05	0.2	>.05	0.47	<.001	-	-	none
			M	Care-ED	B	Carer's Skills	CASK	B	DASS	B	0.2	<.01	0.22	<.05	-0.17	>.05	-0.4	<.001	-	-	none
			F	Care-ED	B	Carer's Skills	CASK	B	DASS	B	0.1	>.05	0.07	>.05	0.03	>.05	-0.5	<.01	-	-	none

Key: 0: Cross-sectional analysis. B: Baseline. C: Carer. M: Mother. F: Father. Sig:Significance statistic. BCI: Bootstrapped confidence interval; significant to 95% if does not cross 0. ss: subscale. h/w: hours per week. h/m: hours per month.

Table 7

Predictors of Change in Distress Following Intervention

Category	Sample	Study	Analysis type	Lv. Evidence	Predictor	Measure	Time	Distress Measure	Change period	Test	Statistic	*p*	Power	# study comparisons
Patient Clinical & Demograpics														
	e	Salerno et al., 2016	MP	1	Patient BMI	Clinical	B	DASS	B-1 year	β	–.06	0.54	L	
	e	Salerno et al., 2016	MP	1	Duration of illness	Clinical	B	DASS	B-1 year	β	-0.17	0.99	L	
	e	Salerno et al., 2016	MP	1	Patient age	SD-Q	B	DASS	B-1 year	β	0.002	0.21	L	
	e	Salerno et al., 2016	MP	1	Patient distress	DASS	B	DASS	B-1 year	β	0.14	0.17	L	
Carer distress														
	e	Salerno et al., 2016	MP	1	Carer distress	DASS	B	DASS	B-1 year	β	0.43	<.000	L	
Subjective Burden														
	g	Sepulveda et al., 2010	MP	1	Negative appraisal of caregiving	ECI-ss	B	GHQ-12	B-EoT (3m)	b	*0.01*	0.8	L	
Accommodating and Enabling														
		Goddard et al., 2011	Co	3	Accommodation and Enabling	AESED	B-EoT (6wks)	HADS	B-EoT (6wks)	*r*	0.5	<.0001		3
Carer Skills														
		Goddard et al., 2011	Co	3	Carer self-efficacy	CSE	B-EoT (6wks)	HADS	B-EoT (6wks)	*r*	-0.5	<.0001		3
Expressed emotion														
		Goddard et al., 2011	Co	3	Expressed emotion	FQ	B-EoT (6wks)	HADS	B-EoT (6wks)	*r*	0.5	<.0001		3
	g	Sepulveda et al., 2010	MP	1	Expressed emotion	FMSS	B	GHQ-12	B-EoT (3m)	b, R^2	5.3, 0.19	0.01	L	
	g	Sepulveda et al., 2010	MP	1	Criticism	FMSS-ss	B	GHQ-12	B-EoT (3m)	b, R^2	6.6, 0.33	0.01	L	
	g	Sepulveda et al., 2010	MP	1	Emotional over-involvement	FMSS-ss	B	GHQ-12	B-EoT (3m)	b, R^2	2.6, 0.06	0.06	L	

Table 8

Moderators of the Proposed Effect of Intervention on Carer Distress

Sample	Author	Predictor	Measure	Moderator (baseline)	Measure	Outcome	Measure	R2 Change	*p*
-	Slater et al., 2014	Distress (3m)	GHQ-28	BMI	SD-Q	Distress 12m	GHQ-28	-	<.01
				Living together	SD-Q	Distress 12m	GHQ-28	-	>.05
				Duration of illness	SD-Q	Distress 12m	GHQ-28	-	>.05
-	Goddard et al. ,2011	Distress (B)	HADS	Expressed emotion	FQ	Distress	HADS	-0.02	0.04
				Accommodating	AESED	Distress	HADS	-0.01	<.001
				Carer Self-efficacy	CSE	Distress	HADS	-	>.05
				Contact time	h p/w	Distress	HADS	-	>.05
				length of illness	duration	Distress	HADS	-	>.05
				living together	SDQ	Distress	HADS	-	>.05
-	Grover et al., 2011	Reciept of carer's skills intervention, Y/N		Expressed emotion	LEE-C	Ch. HADS B-		-	>.05
				Carer psychological profile; cared-for characteristics (inc. previous hospitalisation & ED treatment)	carer-report	Ch. HADS B-EoT		-	>.05

Note: B: Baseline. 3m: Three-month follow-up timepoint. 12m: 12-month follow-up timepoint. EoT: End of treatment.

Table 9

Mediators of the Proposed Effect of Intervention on Carer Distress

											Total effect		Direct effect		Predictor on mediator		Mediator on outcome		Total indirect effect		
Sample	Author	Predictor	Who	Measure	Time	Mediator	Measure	Time	Outcome measure	Time	β	*p*	β	*p*	β	*p*	β	*p*	Size	Sig.	Mediation
-	Goddard et al., 2011	Distress	C	HADS	B	Ch. in Expressed emotion	FQ	B-EoT	HADS	EoT	-	<.0	-	>.0	-	-	-	-	0.5	*p*<.0001	Partial
			C	HADS	B	Ch. in Accommodating	AESED	B-EoT	HADS	EoT	-	<.0	-	>.0	-	-	-	-	0.5	*p*<.0001	Partial
			C	HADS	B	Ch. in Carer self-efficacy	CSE	B-EoT	HADS	EoT	-	<.0	-	>.0	-	-	-	-	-1	*p*<.0001	Partial
			C	HADS	B	Ch. in contact time	h/w (< / > 21)	B-EoT	HADS	EoT	-	>.0	-	n/a	-	-	-	-	-	-	None

Note: 0: Cross-sectional analysis. B: Baseline. C: Carer. M: Mother. F: Father. Sig:Significance statistic. ss: subscale. h/w: hours per week. h/m: hours per month.

Discussion

Firstly, this review appraised the evidence for predictors of distress experienced by carers of people with eating disorders, and mediators of these associations; no studies investigated moderators. Secondly, the evidence for predictors of change in carer distress as an outcome of ED-related intervention was appraised, alongside the moderators and mediators of these associations. These two aims are considered in turn below.

In total, 30 studies were included. The majority of carers were mothers, the majority of people with eating disorders were diagnosed with AN, and a high proportion were living together. Findings regarding most predictors were mixed, possibly related to variations in sampling, measures employed, and inclusion of possible co-variates in analysis. Power issues may additionally have added to lack of significant findings, especially in longitudinal data, as analyses with lower power were more frequently non-significant. As each moderation and mediation analysis was assessed by no more than two studies, and each significant moderation and mediation result was reported by no more than one study, these initial findings should be taken with some caution. Reassuringly however, there was consensus amongst all significant results concerning whether predictors were positively or negatively associated with distress.

Predictors of Carer Distress and Mediators of these Associations

Addressing the first aim, the variable most robustly evidenced as a predictor of carer distress was caregiving burden. A positive relationship was found using a range of measures and designs, which included multivariate and longitudinal analysis. This supports the hypotheses of the models proposed by Treasure and Nazar (2016) and Winn et al. (2007). Beta values from univariate analyses were at the higher end of the range reported by included studies, meaning a one standard deviation increase in burden was associated with a larger increase in carer distress compared to most other predictors reviewed. This is concordant with Grunfield et al. (2004) who also reported burden as the strongest predictor of distress in family carers of cancer patients.

Overall, assessment of longitudinal predictors of carer distress was limited by there being few studies, of which most had only enough power for large effects. In addition to burden, several variables were assessed longitudinally. The only other predictors emerging as significant in longitudinal analysis were cared-for distress and carer distress measured at an earlier time-point. Higher levels of cared-for depression at baseline predicted greater reduction in carer depression over a year when the cared-for was moderately depressed, but not clinically depressed. This possibly indicated greater opportunity for recovery where depression was less entrenched (Teasdale, 1988). Similarly, multivariate analysis found reduction in cared-for distress to positively predict reduction in carer distress, with one of the largest Betas in this review. This was supported by findings of cared-for distress as a positive predictor of carer distress from multivariate cross-sectional analyses. With regard to carer distress as a predictor, higher baseline carer anxiety and distress were strongly associated with greater reduction in carer anxiety and distress (respectively) a year later. Again, this may be due to greater scope for change.

Cared-for quality of life was tested as a longitudinal predictor using only univariate analysis, with mixed results, and illness duration was a significant predictor in univariate but not multivariate longitudinal analysis. The majority of cross-sectional analyses for these variables were non-significant, and the former was part of a large battery of comparisons, indicating these may not be reliable predictors.

Carer coping, expressed emotion and cared-for suicidal intent and drug addiction were significant positive predictors in the majority of cross-sectional analysis (including multivariate), but non-significant when tested longitudinally. In particular, expressed emotion was well-evidenced as a predictor cross-sectionally, and longitudinal analyses were underpowered to detect anything but large effects, indicating this as an important target for further investigation. Similarly, accommodation was a significant positive predictor in a large majority of a large number of cross-sectional studies, but was not tested longitudinally. This was also the case for carer skills, which unanimously negatively predicted distress in univariate cross-sectional analyses. The findings regarding expressed emotion, accommodation and carer skills support the hypotheses of the Cognitive Interpersonal Model (Treasure & Schmidt, 2013) and carers' intervention models proposed by Goddard et al. (2011) and Hibbs et al. (2015a), discussed further below in addressing the second aim.

Arguably, it might be invalid to claim variables are predictors when not significant longitudinally in multivariate analysis. However, the limitations of the longitudinal studies discussed are likely to have affected these findings, and imply that longitudinal studies require replication with more participants. Similarly, the cross-sectional findings need further investigation using multivariate analysis with more co-variates to determine what is leading to these contradictory findings.

Some univariate, or single examples of multivariate cross-sectional analysis of the following variables found these to be significant positive predictors of carer distress; ED symptoms; carers being single (relationship status); spending longer caregiving or in contact with the cared-for; carers' lower quality of life and own history of eating difficulties; shorter cared-for illness duration; higher perceived stigma; highest level of carer education; having a worse relationship with the cared-for or a less positive experience of caregiving; lower family functioning and higher family conflict; higher levels of carer psychological control; and being a mother or female caregiver. However, results regarding these variables were mixed and will require replication with multivariate and ideally longitudinal analysis.

The following variables were generally found not to be predictive of carer distress: Carer and cared-for age; time spent by the cared-for in treatment; compensatory behaviour (purgative vs. restrictive); carer needs and employment. This was also the case for support received, with the exception of the sub-scale ‘affective social support received’, which was significant in both univariate and multivariate cross-sectional analysis. These non-significant variables were predominantly tested by weaker analysis and, as most are implicated in models of carer distress, it would be useful to investigate these with more robust analysis.

Most univariate, and all multivariate and longitudinal analysis investigating whether ED symptoms predicted carer distress (including problems which seem likely to be closely associated to symptoms such as the cared-for’s rejection of having ED and of the carer’s help), were non-significant. This was interesting in light of Anastasiadou et al.’s (2014) finding that carer distress is associated with caring for someone with an ED, and reports from research in PTSD (Calhoun, Beckham, & Bosworth, 2002) and Schizophrenia (Schene, van Wijngaarden, & Maarten, 1998) of symptom severity positively predicting higher severity of carer distress. The model proposed by Kyriacou, Treasure, and Schmidt (2008, based on empirical findings) suggested that negative/difficult ED behaviours and the cared-for’s rejection of carer help predicted strain related to the carer and role; this in turn predicted carer distress. Similarly, the Model of Carer Coping (Treasure & Nazar, 2016) suggested carer distress arises as a product of ED-related stressors only when carers’ coping resources are overwhelmed. This may explain why direct relationships between these cared-for variables and carer distress were not found by the studies investigating only predictors; these relationships did not include the proposed intermediary variables of coping or carer/role-related strain. Coomber and King (2012), one of the few studies to address mediators of the relationships between predictors and carer distress, reported the relationship between expressed emotion and carer distress to be statistically positively mediated by maladaptive coping. This again supports the assertion of Treasure and Nazar's (2016) model. Coomber and King's (2012) finding that coping difficulties account for 20% of the variance in distress provides further evidence for this hypothetically key link.

In other cross-sectional mediation analyses, expressed emotion, accommodation (Rhind et al., 2016) and ED-specific burden (Raenker et al., 2013) positively mediated the relationship between time spent caregiving and carer distress, and social support negatively mediated the same relationship (Raenker et al., 2013). This is not inconsistent with the idea that spending more time caregiving is likely to be associated with increased emotionally difficult and burdensome aspects of caregiving, which may subsequently result in greater carer distress, but that social support (see Szmuckler et al., 1996) may alleviate this.

In relation to possible differences between sub-groups of participants, several within-study findings from analyses with primary and secondary carers found notable differences, in absence of a clear pattern. Secondary carers in this review were generally fathers, in line with mothers generally being more involved in childcare (Connell and Goodman, 2002), potentially taking greater responsibility (von Essen et al., 2004). This raises interesting questions about how differences in gender, role and involvement might affect caregiver experience. Sepulveda et al.'s (2014a) finding that different aspects of expressed emotion and burden were predictors dependent on whether the cared-for purged or not may be related to Anastasiadou et al.’s (2014) finding of differences in distress between carers of people with BN and AN. However, no other findings of this review could contribute to the interesting questions this poses about qualitative differences in relationships between service-users, carers and distress in these sub-groups.

Predictors of Proposed Intervention Outcome, and Moderators and Mediators of these Associations

In respect of the second aim, only a handful of studies tested predictors of proposed treatment outcome. The only significant results from multivariate analyses were that higher expressed emotion and carer distress at baseline positively predicted greater reduction in carer distress over a time-period in which carers’ skills interventions took place.

It might be that these predictors moderated or mediated a relationship between the intervention and change in distress; however, an experimental design in which these are entered as moderators (moderation x group interaction) or mediators (indirect effect of group on distress through the proposed mediating variable) would need to be conducted in order to provide evidence for this. In lieu of such a design, two pre-post studies tested moderators of the change in carer distress between two time-points in the course of intervention. It was found that carers of people with higher compared to lower BMI reported less improvement in distress between three and 12-month follow-ups, possibly related to initial hopes for improvement in the former and greater relief at small changes in the latter (Slater et al., 2014). Expressed emotion and accommodation were similarly identified as potential moderators of intervention outcome, with higher levels at baseline predicting greater reduction in distress by the post-intervention time-point (Goddard et al., 2011). Interestingly, the only study to address moderation of intervention outcome with an experimental design identified no moderators, although power appeared insufficient.

The above findings of carer distress, expressed emotion, skills, and accommodation as either positive predictors or moderators of change in distress over a time period in which an intervention takes place is likely related to greater scope for change for carers who present at baseline of an intervention with higher levels of the difficulties intervention aims to reduce. Consistent with the hypothesis that improvements in accommodation, expressed emotion and caregiving skills may explain reduction in distress (Goddard et al., 2011; Treasure & Nazar, 2016; Szmukler et al., 1996; Winn et al., 2007), these variables statistically mediated change in carer distress.

Limitations of Included Studies

There were several limitations of the studies reviewed, including the wide variety of measures and research designs meaning results were less directly comparable and that the literature is not fit for meta-analysis at this stage.

Additionally, most longitudinal studies suffered from high attrition rates, even preventing follow-up analysis (e.g. Las Hayas et al., 2014). The contribution of pre-post designs, and prospective cohort studies, is limited by the inability to control for treatment and other support, professional or otherwise, received by the carer and cared-for over the course of the trial. For example, in Goddard et al.'s otherwise well-controlled (2011) study, it was not possible to separate unrelated changes, such as those observed over the waiting period (ibid) from any impact of intervention, meaning proposed intervention effects may instead have related to unobserved influences such as clinical improvement in cared-fors, or family therapy. Analysis of proposed predictors, moderators or mediators of intervention outcome, therefore, must be interpreted cautiously.

Repeated testing of samples meant that the number of people the data were collected from was more limited than if each sample had been novel, potentially reducing generalisability of the findings. For the univariate analyses from these samples there is also an increased risk of Type I errors due to the number of analyses that were conducted without correction for multiple comparisons. However, large-scale studies such as these have been instrumental in gaining momentum in this area and provided foundations for further research.

Several issues potentially compromised generalisability of findings (CASP, 2013b). These included the majority of studies recruiting carers through self-selection, usually via contact with services. It is therefore arguable that the sample was more empowered, and actively involved in seeking support for themselves and their cared-for (Kyriacou, Treasure & Schmidt, 2008). It could be speculated that carers whose situation was more limiting may not have had time or been too distressed to participate. Additionally, the results may be biased towards carers of cared-fors who had a higher severity of illness than would typically be seen, as many were recruited through inpatient settings. Furthermore, several studies accessed carers through referral for the study by cared-fors, which likely meant under-representation of carers of people who choose not to involve their carer in treatment, a factor that may be meaningfully linked to carer distress. For example, 61% of the possible participant sample in Slater et al.'s (2015) study were excluded, as cared-fors chose not to identify them, giving reasons such as considering carers part of the problem or not wanting to burden them further. Fathers, partners and other carers were under-represented by the studies as a whole, as were carers of people with Bulimia.

Research Implications

This review has reported several predictors of carer distress for which there are relatively robust findings, and several areas of mixed results for which further examination using multivariate and longitudinal analysis will be important. These include the longitudinal role of expressed emotion, accommodation and coping styles, and differences between primary and secondary carers, and between carers of people who do and do not purge, together with the role that relationships with the cared-for and carer gender may play in this. As the majority of studies were with mothers of people with AN, further research with all types of non-professional carers and all EDs would be of interest.

Using well validated measures would be helpful, and potentially provide basis for future meta-analysis. The development of ED-specific measures have supported specificity and progression in this area of study, and their continued use and validation will be of further benefit.

Salerno et al. (2016) reported that carer distress did not predict cared-for distress at 1 year follow-up, but maternal depression has been found to predict less favourable cared-for ED treatment outcomes (Vall & Wade, 2015). Goddard et al. (2013) found cared-for distress fully mediated a cross-sectional relationship between carer distress and ED psychopathology. It would therefore be useful to examine whether reductions in carer distress as an outcome of intervention for carers has secondary benefits for those that they care for, and the process by which, and whether, carer distress may impact cared-for distress and ED symptoms longitudinally.

There is a conspicuous lack of evidence on moderators and mediators of carer distress, especially as a treatment outcome. This promising area has potential to guide intervention choice and development on the basis of what will be most effective for which carers, and through which mechanisms benefits from interventions occur. Variables of interest from theory and this review's results include cared-for distress, carer coping, burden and/or strains, accommodation, expressed emotion and carer skills. Ideally, moderators and mediators would be tested in longitudinal designs to support better understanding of how these variables interact. Investigation using RCT design would enable clarification of whether these factors explained mechanisms of treatment efficacy, or were related in other ways. Although such trials may be challenging to obtain funding for, further moderation and mediation analysis using cross-sectional designs would nevertheless contribute meaningfully to this area. As carers interventions represent a rapidly evolving approach to the treatment of eating disorders, such evidence would be valuable in clinical and commissioning choices and to refine treatment, improving efficacy for both carer and cared-for.

Conclusion

Eating disorders appear to impact on carer distress in a variety of ways which unsurprisingly seem complex and inter-related. A number of studies have undertaken investigation of predictors of carer distress, and the findings present a complex, mixed picture. Despite several limitations, there are some key conclusions.

There is evidence for multiple predictors of carer distress, with the strongest evidence available for higher burden, cared-for distress and earlier carer anxiety and depression as predictors of greater carer distress, including longitudinally. Additionally, higher expressed emotion and accommodation, and lower carer skills were positive predictors of carer distress in a large number of cross sectional analyses, including multivariate studies. ED symptoms were found not to directly predict carer distress. However, the data provide support for models in which the ED impacts on carers' distress through its effects on carers' behavioural responses, burden and coping (ways in which it becomes personally relevant for the carer).

Results indicated expressed emotion, accommodation and carers' skills may be implicated in change in distress over the course of an intervention. However, evidence of predictors of carer intervention outcome, and moderators and mediators of these associations, is insufficient to guide decisions as to who, or under what circumstances, different approaches to intervention for carers would be most effective. Further investigation of these research questions will be necessary. In particular, analysis of moderators and mediators of outcomes from interventions for carers would further inform refinement of theory and intervention, for the benefit of both carers and those that they care for.

References

Anastasiadou, D., Medina-Pradas, C., Sepulveda, A. R., & Treasure, J. (2014). A systematic review of family caregiving in eating disorders. *Eating behaviors*, *15*(3), 464-477. doi: 10.1016/j.eatbeh.2014.06.001

Arai, Y., Tamiya, N., & Yano, E. (2003). The short version of the Japanese version of the Zarit Caregiver Burden Interview (J-ZBI_8): Its reliability and validity. *Japanese Journal of Geriatrics, 40*(5)*,* 497–503. doi:10.3143/geriatrics.40.497

Arcelus, J., Mitchell, A. J., Wales, J., & Nielsen, S. (2011). Mortality rates in patients with Anorexia Nervosa and other eating disorders: A meta-analysis of 36 studies. *Archives of General Psychiatry*, *68*(7), 724-731. doi:10.1001/archgenpsychiatry.2011.74

Barber, B. K. (1996). Parental psychological control: Revisiting a neglected construct. *Child Development, 67*(6), 3296–3319. doi:10.1111/j.1467-8624.1996.tb01915.x

Baron, R. M., & Kenny, D. A. (1986). The moderator–mediator variable distinction in social psychological research: Conceptual, strategic, and statistical considerations. *Journal of Personality and Social Psychology*, *51*(6), 1173. http://psycnet.apa.org/doi/10.1037/0022-3514.51.6.1173

Bauer, S., Winn, S., Schmidt, U., & Kordy, H. (2005). Construction, scoring and validation of the Short Evaluation of Eating Disorders (SEED). *European Eating Disorders Review*, *13*(3), 191-200. doi:10.1002/erv.637

Booth, A., Papaioannou, D., & Sutton, A. (2012). *Systematic Approaches to a Successful Literature Review*. London: Sage.

Butzlaff, R. L., & Hooley, J. M. (1998). Expressed emotion and psychiatric relapse: a meta-analysis. *Archives of General Psychiatry*, *55*(6), 547-552. doi:10.1001/archpsyc.55.6.547

Calhoun, P.S., Beckham, J.C., & Bosworth, H.B. (2002). Caregiver burden and psychological distress in partners of veterans with chronic post-traumatic stress disorder. *Journal of Traumatic Stress, 15*, 205–212. doi:10.1023/A:1015251210928

Carral-Fernandez, L., Sepulveda, A. R., Gomez del Barrio, A., Graell, M., & Treasure, J. (2013). The Spanish validation of an Eating Disorders Symptom Impact Scale (EDSIS) among caregivers. *Psychiatry Research*, *209*(3), 626–631. doi:10.1016/j.psychres.2013.02.019

Carver, C. S. (1997). You want to measure coping but your protocol’s too long: Consider the Brief COPE. *International Journal of Behavioural Medicine.* *4*:(1), 92-100. doi:10.1207/s15327558ijbm0401_6

Centre for Evidence Based Medicine – Levels of Evidence (2009). Chalmers, I., Glasziou, P., Greenhalgh, T., Heneghan, C., Howick, J., Liberati, A., Moschetti, I., Phillips, B., & Thornton, H. (2009). Steps in finding evidence (‘Levels’) for different types of question. Retrieved from: http://www.cebm.net/mod_product/design/files/CEBM-Levels-of-Evidence-2.pdfs

Cole, J. D., & Kazarian, S. S. (1988). The Level of Expressed Emotion Scale: A new measure of expressed emotion. *Journal of Clinical Psychology, 44*, 392–397. doi:10.1002/ 1097-4679(198805)44:3<392::AID-JCLP2270440313> 3.0.CO;2-3

Connell, A. M., & Goodman, S. H. (2002). The association between psychopathology in fathers versus mothers and children's internalizing and externalizing behavior problems: A meta-analysis. Psychological bulletin, 128(5), 746. doi:10.1037/0033-2909.128.5.746

Coomber, K., & King, R. M. (2012). Coping strategies and social support as predictors and mediators of eating disorder carer burden and psychological distress. *Social Psychiatry and Psychiatric Epidemiology*, *47*(5), 789-796. doi:10.1007/s00127-011-0384-6

Coomber, K., & King, R. M. (2013). A longitudinal examination of burden and psychological distress in carers of people with an eating disorder. *Social Psychiatry and Psychiatric Epidemiology*, *48*(1), 163-171. doi:10.1007/s00127-012-0524-7

Coren, E. and Fisher, M. (2006) *The Conduct of Systematic Research Reviews for SCIE Knowledge Reviews.* Report. London, UK: Social Care Institute for Excellence. Retrieved from https://create.canterbury.ac.uk/id/eprint/5088

Critical Appraisal Skills Programme (CASP). (2013). *Cohort Study Checklist.* Retrieved from: http://media.wix.com/ugd/dded87_e37a4ab637fe46a0869f9f977dacf134.pdf

Critical Appraisal Skills Programme (CASP). (2013). *Randomised Controlled Trials Checklist.* Retrieved from: http://media.wix.com/ugd/dded87_40b9ff0bf53840478331915a8ed8b2fb.pdf

Currin, L., Schmidt, U., Treasure, J., & Jick, H. (2005). Time trends in eating disorder incidence. *The British Journal of Psychiatry*, *186*(2), 132-135. doi:10.1192/bjp.186.2.132

Cutrona, C. E., & Russell, D. W. (1987). The Provisions of Social Relationships and Adaptation to Stress. *Advances in Personal Relationships, 1*(1), 37-67. Retrieved from: https://www.researchgate.net/profile/Daniel_Russell4/publication/271507385_THE_PROVISIONS_OF_SOCIAL_RELATIONSHIPS_AND_ADAPTATION_TO_STRESS/links/54c960b40cf2807dcc265ce2.pdf

Department of Health. (2004). Carers Strategy: Actions for 2014 to 2016. London: Department of Health. Retrieved from: https://www.gov.uk/government/publications/carers-strategy-actions-for-2014-to-2016

Derogatis, L. R., & Melisaratos, N. (1983). The brief symptom inventory: an introductory report. Psychological Medicine, 13(03), 595-605. doi:10.1017/S0033291700048017

Dimitropoulos, G., Carter, J., Schachter, R., & Woodside, D. B. (2008). Predictors of Family Functioning in Carers of Individuals with Anorexia Nervosa. *International Journal of Eating Disorders, 41*(8), 739–747. doi:10.1002/eat.20562

Endler, N.S., & Parker, J.D. (1990). *Coping Inventory for Stressful situations (CISS): Manual*. Toronto: Multi-Health Systems.

Engel, S. G., Adair, C. E., Hayas, C. L., & Abraham, S. (2009). Health-related quality of life and eating disorders: A review and update. *International Journal of Eating Disorders*, *42*(2), 179-187. doi:10.1002/eat.20602

Epstein, N. B., Baldwin, L. M., & Bishop, D. S. (1983). The McMaster family assessment device. *Journal of Marital and Family Therapy*, *9*(2), 171-180. doi: 10.1111/j.1752-0606.1983.tb01497.x

Field, A. (2013). *Discovering Statistics using IBM SPSS Statistics*. Los Angeles, London, New Delhi, Singapore, Washington DC: Sage.

Furukawa, T., Suzuki-Moor, A., Saito, Y., & Hamanaka, T. (1992). Reliability and validity of the Japanese version of the coping inventory for stressful situations (CISS): A contribution to the cross-cultural studies of coping. Seishin shinkeigaku zasshi Psychiatria et neurologia Japonica, 95(8), 602-620. Retrieved from http://europepmc.org/abstract/med/8234537

Gandek, B., Ware, J. E., Aaronson, N. K., Apolone, G., Bjorner, J. B., Brazier, J. E., ... & Sullivan, M. (1998). Cross-validation of item selection and scoring for the SF-12 Health Survey in nine countries: results from the IQOLA Project. *Journal of Clinical Epidemiology*, *51*(11), 1171-1178. doi:10.1016/S0895-4356(98)00109-7

Goddard, E., Macdonald, P., Sepulveda, A. R., Naumann, U., Landau, S., Schmidt, U., & Treasure, J. (2011). Cognitive interpersonal maintenance model of eating disorders: Intervention for carers. *The British Journal of Psychiatry, 199*(3), 225–231. doi:10.1192/bjp.bp.110.088401

Goddard, E., Raenker, S., & Treasure, J. (2012). Involving carers: A skills-based learning approach. In J. Alexander, & J. Treasure (Eds.), *A Collaborative Approach to Eating Disorders (pp.*149-162). London & New York: Routledge.

Goddard, E., Salerno, L., Hibbs, R., Raenker, S., Naumann, U., Arcelus, J., … Treasure, J. (2013). Empirical examination of the interpersonal maintenance model of Anorexia Nervosa. *International Journal of Eating Disorders, 46*(8), 867–874. doi:10.1002/eat.22172

Goldberg, D., & Williams, P. (1998). *A User's Guide to the GHQ*. Windsor: NFER-Nelson.

Goldberg, D.F., & Hillier, V.F. (1979). A scaled version and the General Health Questionnaire. Psychological Medicine, 9(1), 139–145. doi:10.1017/S0033291700021644

Goldner, E. (1989). Treatment refusal in Anorexia Nervosa. *International Journal of Eating Disorders*. doi:10.1002/1098-108X

González de Rivera, J.L., Derogatis, L.R., De las Cuevas, C., Gracia Marco, R., Rodríguez Pulido, F., … & Henry Benítez, M. (1989). The Spanish version of the SCL-90/R. Normative data in the general population. Towson: Clinical Psychometric Research.

González, N., Bilbao, A., Padierna, A., Martín, J., Orive, M., & Quintana, J. M. (2012). Validity and reliability of the Spanish version of the Involvement Evaluation Questionnaire among caregivers of patients with eating disorders. *Psychiatry research*, *200*(2), 896-903. doi:10.1016/j.psychres.2012.07.033

Graap, H., Bleich, S., Herbst, F., Trostmann, Y., Wancata, J., de Zwaan, M., … Zwaan, and M. de. (2008). The needs of carers of patients with anorexia and bulimia nervosa. *European Eating Disorders Review, 16*(1), 21–29. doi:10.1007/s00127-008-0364-7

Grover, M., Naumann, U., Mohammad-Dar, L., Glennon, D., Ringwood, S., Eisler, I., … Schmidt, U. (2011). A randomized controlled trial of an Internet-based cognitive-behavioural skills package for carers of people with Anorexia Nervosa. *Psychological Medicine, 41*(12), 2581–91. doi:10.1017/S0033291711000766

Grunfeld, E., Coyle, D., Whelan, T., Clinch, J., Reyno, L., Earle, C. C., ... & Glossop, R. (2004). Family caregiver burden: Results of a longitudinal study of breast cancer patients and their principal caregivers. *Canadian Medical Association Journal*, *170*(12), 1795-1801. doi: 10.1503/cmaj.1031205

Haigh, R., & Treasure, J. (2003). Investigating the needs of carers in the area of eating disorders: Development of the Carers' Needs Assessment Measure (CaNAM). *European Eating Disorders Review, 11*(2), 125–141. doi:10.1002/erv.487

Harper, A. (1998). Development of the World Health Organisation WHOQOL-BREF quality of life assessment. Psychological Medicine, 28(3), 551-558. Retrieved from https://www.cambridge.org/core/article/development-of-the-world-health-organization-whoqol-bref-quality-of-life-assessment/0F50596B33A1ABD59A6605C44A6A8F30

Hayes, A. F. (2013). *Introduction to mediation, moderation, and conditional process analysis: A regression-based approach*. New York, NY: Guilford Press.

Herrmann, C. (1997). International experiences with the hospital Anxiety and Depression Scale: a review of validation data and clinical results. *Journal of Psychosomatic Research 42(*1), 17–41. doi:10.1016/S0022-3999(96)00216-4

Hibbs, R., Magill, N., Goddard, E., Rhind, C., Raenker, S., Macdonald, P., … Treasure, J. (2015a). Clinical effectiveness of a skills training intervention for caregivers in improving patient and caregiver health following in- patient treatment for severe Anorexia Nervosa : pragmatic randomised controlled trial. *British Journal of Psychiatry,* 1, 56–66. doi:10.1192/bjpo.bp.115.000273

Hibbs, R., Rhind, C., Leppanen, J., & Treasure, J. (2015b). Interventions for caregivers of someone with an eating disorder: A meta-analysis. *International Journal of Eating Disorders*, *48*(4), 349-361. doi:10.1002/eat.2229

Hibbs, R., Rhind, C., Salerno, L., Lo Coco, G., Goddard, E., Schmidt, U., … Treasure, J. (2015c). Development and validation of a scale to measure caregiver skills in eating disorders. *International Journal of Eating Disorders, 48*(3), 290–297. doi:10.1002/eat.22362

Horvath, A. O., & Greenberg, L. S. (1989). Development and validation of the Working Alliance Inventory. *Journal of Counseling Psychology*, *36*(2), 223. doi:10.1037/0022-0167.36.2.223

Instituto Nacional de la Salud, 1995. Protocolo de trastornos del comportamiento alimentario. Instituto Nacional de la Salud. Insalud, Madrid, pp. 125–126.

Kraemer, H. C., Wilson, G. T., Fairburn, C. G., & Agras, W. S (2002). Mediators and moderators of treatment effects in a randomized clinical trials. *Archives of General Psychiatry, 59*(10), 877-883. doi:10.1001/archpsyc.59.10.877

Kyriacou, O., Treasure, J., & Schmidt, U. (2008). Understanding how parents cope with living with someone with Anorexia Nervosa: Modelling the factors that are associated with carer distress. *International Journal of Eating Disorders*, *41*(3), 233-242. doi:10.1002/eat.20488

Las Hayas, C. C., Padierna, Á. J., Bilbao, A., Martín, J., Muñoz, P., Quintana, J. M. M., … Quintana, J. M. M. (2014). Eating disorders: predictors of change in the quality of life of caregivers. *Psychiatry Research, 215*(3), 718–26. doi: 10.1016/j.psychres.2013.12.028

Las Hayas, C., Padierna, J. Á., Bilbao, A., Martín, J., Muñoz, P., & Quintana, J. M. (2014). Eating disorders: Predictors of change in the quality of life of caregivers. *Psychiatry Research*, *215*(3), 718-726. doi:10.1016/j.psychres.2013.12.028

Liddle, E. B., Batty, M. J., & Goodman, R. (2009). The social aptitudes scale: an initial validation. *Social Psychiatry and Psychiatric Epidemiology, 44*(6), 508-513. doi:10.1007/s00127-008-0456-4

Lobo, A., &Muñoz, P. (1996). Versiones en lengua Española validadas. In Goldberg, D. P., & Williams, P. (Eds.), Cuestionario de Salud GeneralGHQ(GeneralHealthQuestionnaire),Guía para el usuario de las distintas vesiones. Barcelona, Spain:Masson.

Lovibond, S.H., & Lovibond, P.F. (1993). *Manual for the Depression Anxiety and Stress Scales (DASS)*. NSW, Australia: Psychology Foundation Monograph.

Magana AB, Goldstein JM, Karno M, Miklowitz DJ, Jenkins J, Fal- loon IR. A brief method for assessing expressed emotion in rel- atives of psychiatric patients. Psychiatry Res 1986;17:203–212.doi:10.1016/0165-1781(86)90049-1

Magaña, A. B., Goldstein, M. J., Karno, M., Miklowitz, D. J., Jenkins, J., & Falloon, I. R. (1986). A brief method for assessing expressed emotion in relatives of psychiatric patients. Psychiatry Research, 17(3), 203-212. doi:10.1016/0165-1781(86)90049-1

Magliano, L., Fadden, G., Economou, M., Xavier, M., Held, T., Guarneri, M., ... & Maj, M. (1998). Social and clinical factors influencing the choice of coping strategies in relatives of patients with schizophrenia: results of the BIOMED I study. *Social Psychiatry and Psychiatric Epidemiology*, *33*(9), 413-419. doi:10.1007/s001270050074

Martin, J., Padierna, A., Aguirre, U., Quintana, J. M., Las Hayas, C., & Munoz, P. (2013). Quality of life among caregivers of patients with eating disorders. *Quality of Life Research,20* (9) 1359-1369. doi: 10.1007/s11136-011-9873-z

Medina-Pradas, C., Navarro, J. B., Lopez, S. R., Grau, A., Obiols, J. E., López, S. R., … Obiols, J. E. (2011). Dyadic view of expressed emotion, stress, and eating disorder psychopathology. Appetite, 57(3), 743–748. doi:10.1016/j.appet.2011.08.016

Meltzer, H. (2003). Development of a Common Instrument for Mental Health. Chap. 4 In A. Nosikov, & C. Gudex (Eds.). *EUROHIS: Developing common instruments for health surveys*, *57*, pp. 35-60. Amsterdam: IOS.

Micali, N., Hagberg, K. W., Petersen, I., & Treasure, J. L. (2013). The incidence of eating disorders in the UK in 2000–2009: findings from the General Practice Research Database. *British Medical Journal Open*, *3*(5), e002646. doi:10.1136/bmjopen-2013-002646

Moher, D., Hopewell, S., Schulz, K. F., Montori, V., Gøtzsche, P. C., Devereaux, P. J., ... & Altman, D. G. (2010). CONSORT 2010 explanation and elaboration: Updated guidelines for reporting parallel group randomised trials. British Medical Journal, 340 (c869). doi: 10.1136/bmj.c869

Ohara, C., Komaki, G., Yamagata, Z., Hotta, M., Kamo, T., & Ando, T. (2016). Factors associated with caregiving burden and mental health conditions in caregivers of patients with Anorexia Nervosa in Japan. *BioPsychoSocial Medicine*, *10*(1), 21. doi:10.1186/s13030-016-0073-5

Orive, M., Padierna, A., Martin, J., Aguirre, U., González, N., Muñoz, P., & Quintana, J. M. (2013). Anxiety and depression among caregivers of patients with eating disorders and their change over 1 year. *Social Psychiatry and Psychiatric Epidemiology*, *48*(9), 1503-1512. doi:10.1007/s00127-013-0662-6

Pearlin, L. I., Mullan, J. T., Semple, S. J., & Skaff, M. M. (1990). Caregiving and the stress process: An overview of concepts and their measures. *The Gerontologist*, *30*(5), 583-594. doi:10.1093/geront/30.5.583

Piacentini, J., Langley, A., & Roblek, T. (2007). *Cognitive-Behavioural Treatment of Childhood OCD: it's Only a False Alarm, Therapist Guide.* New York: Oxford University Press.

Quiles Marcos, Y., Quiles Sebastian, M. J., Pamies Aubalat, L., Sepulveda Garcia, A. R., & Treasure, J. (2016). The Spanish Validation of the Accommodation and Enabling Scale for Eating Disorders Among Carers: A Pilot Study. European Eating Disorders Review, 24(1), 62–68. article. doi:10.1002/erv.2378

Raenker, S., Hibbs, R., Goddard, E., Naumann, U., Arcelus, J., Ayton, A., … Treasure, J. (2013). Caregiving and coping in carers of people with Anorexia Nervosa admitted for intensive hospital care. *International Journal of Eating Disorders, 46*(4), 346–354. doi:10.1002/eat.22068

Reinhard, S. C., Gubman, G. D., Horwitz, A. V., & Minsky, S. (1994). Burden assessment scale for families of the seriously mentally ill. Evaluation and Program Planning, 17(3), 261-269. doi:10.1016/0149-7189(94)90004-3

Rhind, C., Salerno, L., Hibbs, R., Micali, N., Schmidt, U., Gowers, S., ... & Lo Coco, G. (2016). The Objective and Subjective Caregiving Burden and Caregiving Behaviours of Parents of Adolescents with Anorexia Nervosa. European Eating Disorders Review. 24 (4) 310-319. doi:10.1002/erv.2442

Rienecke, R. D., Richmond, R., & Lebow, J. (2016). Therapeutic alliance, expressed emotion, and treatment outcome for Anorexia Nervosa in a family-based partial hospitalization program. Eating Behaviors, 22, 124-128. doi:10.1016/j.eatbeh.2016.06.017

Saeki T, Asukai N, Miyake Y, Miguchi M, Yamawaki S. (1997) Reliability and Validity of the Japanese Version of the Family Assessment Device. *Archives of Psychiatric Diagnosis and Clinical Evaluation, 8,*181-92.

Salerno, L., Rhind, C., Hibbs, R., Micali, N., Schmidt, U., Gowers, S., ... & Treasure, J. (2016). An examination of the impact of care giving styles (accommodation and skilful communication and support) on the one year outcome of adolescent Anorexia Nervosa: Testing the assumptions of the cognitive interpersonal model in Anorexia Nervosa. *Journal of Affective Disorders*, *191*, 230-236. doi:10.1016/j.jad.2015.11.016

Sarason, B. R., Sarason, I. G., & Pierce, G. R. (1990). *Social Support: An Interactional View*. Oxford, England: John Wiley & Sons.

Schene, A.H., van Wijngaarden, B., & Maarten, W.J.K. (1998). Family caregiving in schizophrenia: Domains and distress. *Schizophrenia Bulletin, 24* (4) 609–618. Retrieved from http://schizophreniabulletin.oxfordjournals.org/content/24/4/609.full.pdf+html

Schmidt, R., Tetzlaff, A., & Hilbert, A. (2016). Validity of the Brief Dyadic Scale of Expressed Emotion in Adolescents. *Comprehensive Psychiatry*, *66*, 23-30. doi:10.1016/j.comppsych.2015.12.002

Schmidt, U., & Treasure, J. (2006). Anorexia Nervosa: Valued and visible. A cognitive-interpersonal maintenance model and its implications for research and practice. *British Journal of Clinical Psychology, 45*(3), 343–366. doi:10.1348/014466505X53902

Semple, S. J. (1992). Conflict in Alzheimer's caregiving families: Its dimensions and consequences. *The Gerontologist, 32*(5), 648-655. doi:10.1093/geront/32.5.648

Sepulveda, A. R., Anastasiadou, D., Pellegrin, Y., Andres, P., Graell, M., Carrobles, J. A., & Morande, G. (2014a). Impact of caregiving experience on mental health among caregivers: A comparison of eating disorder patients with purging and non-purging behaviours. *Eating and Weight Disorders, 19*(1), 31–39. doi:10.1007/s40519-013-0064-5

Sepúlveda, A. R., Anastasiadou, D., del Río, A. M., & Graell, M. (2012a). The Spanish validation of level of expressed emotion scale for relatives of people with eating disorders. The Spanish Journal of Psychology, 15(02), 825-839. doi:10.5209/rev_SJOP.2012.v15.n2.38894

Sepulveda, A. R., Anastasiadou, D., Rodríguez, L., Almendros, C., Andrés, P., Vaz, F., … & Graell, M. (2014b). Spanish validation of the Family Questionnaire (FQ) in families of patients with an eating disorder. *Psicothema, 26*(3), 321–327. doi:10.7334/psicothema2013.310

Sepúlveda, A. R., Berbel, E., Anastasiadou, D., Botella, J., Carrobles, J. A., & Morandé, G. (2012b). Factors associated with emotional well-being in primary and secondary caregivers of patients with eating disorders. *European Eating Disorders Review, 20*(1), e78–e84. doi:10.1002/erv.1118

Sepulveda, A. R., Kyriacou, O., & Treasure, J. (2009). Development and validation of the accommodation and enabling scale for eating disorders (AESED) for caregivers in eating disorders. *BMC Health Services Research, 9*, 171. doi:10.1186/1472-6963-9-171

Sepulveda, A. R., Todd, G., Whitaker, W., Grover, M., Stahl, D., & Treasure, J. (2010). Expressed emotion in relatives of patients with eating disorders following skills training program. *The International Journal of Eating Disorders, 43*(7), 603–10. doi:10.1002/eat.20749

Sepulveda, A. R., Whitney, J., Hankins, M., & Treasure, J. (2008). Development and validation of an Eating Disorders Symptom Impact Scale (EDSIS) for carers of people with eating disorders. Health and Quality of Life Outcomes, 6(1), 28. doi:10.1186/1477-7525-6-28

Slater, J., Treasure, J., Schmidt, U., Gilchrist, P., & Wade, T. D. (2014). A pilot study of associations between treatment for Anorexia Nervosa and carers' distress. *Clinical Psychology & Psychotherapy, 22*(4), 372–376. doi:10.1002/cpp.1895

Steffen, A. M., McKibbin, C., Zeiss, A. M., Gallagher-Thompson, D., & Bandura, A. (2002). The revised scale for caregiving self-efficacy reliability and validity studies. *The Journals of Gerontology Series B: Psychological Sciences and Social Sciences*, *57*(1), P74-P86. doi:10.1093/geronb/57.1.P74

Struening, E. L., Perlick, D. A., Link, B. G., Hellman, F., Herman, D., & Sirey, J. A. (2001). Stigma as a barrier to recovery: The extent to which caregivers believe most people devalue consumers and their families. *Psychiatric Services, 52*(12),1633-1638. http://dx.doi.org/10.1176/appi.ps.52.12.1633

Szmukler, G. I., Burgess, P., Herrman, H., Bloch, S., Benson, A., & Colusa, S. (1996). Caring for relatives with serious mental illness: The development of the Experience of Caregiving

Inventory. *Social Psychiatry and Psychiatric Epidemiology*, *31*(3-4), 137-148. doi:10.1007/BF00785760

Szmukler, G. I., Burgess, P., Herrman, H., Bloch, S., Benson, A., & Colusa, S. (1996). Caring for relatives with serious mental illness: The development of the Experience of Caregiving Inventory. *Social Psychiatry and Psychiatric Epidemiology*, *31*(3-4), 137-148. doi: 10.1007/BF00785760

Teasdale, J. D. (1988). Cognitive vulnerability to persistent depression. *Cognition & Emotion*, *2*(3), 247-274. doi:10.1080/02699938808410927

Treasure, J., & Nazar, B. P. (2016). Interventions for the Carers of Patients With Eating Disorders, *Current Psychiatry Reports, 18*(16), 1–7. doi: 10.1007/s11920-015-0652-3

Treasure, J., & Schmidt, U. (2013). The cognitive-interpersonal maintenance model of Anorexia Nervosa revisited: a summary of the evidence for cognitive, socio-emotional and interpersonal predisposing and perpetuating factors. Journal of Eating Disorders, 1(1), 13. doi:10.1186/2050-2974-1-13

Treasure, J., Sepulveda, A. R., MacDonald, P., Whitaker, W., Lopez, C., Zabala, M., ... & Todd, G. (2008). Interpersonal maintaining factors in eating disorder: Skill sharing interventions for carers. International Journal of Child and Adolescent Health, 1(4), 331-338. Retrieved from http://s3.amazonaws.com/academia.edu.documents/42001112/Interpersonal_maintaining_factors_in_eat20160203-4839-fuspnz.pdf?AWSAccessKeyId=AKIAJ56TQJRTWSMTNPEA&Expires=1479825877&Signature=JV0fekKxl1qJfVRATd%2FcgYZNeg0%3D&response-content-disposition=inline%3B%20filename%3DInterpersonal_Maintaining_Factors_in_Eat.pdf

Treasure, J., Sepulveda, A. R., Whitaker, W., Todd, G., Lopez, C., & Whitney, J. (2007). Collaborative Care between Professionals and Non-Professionals in the Management of Eating Disorders: A Description of Worksho, *15*, 24–34. doi:10.1002/erv ps Focussed on Interpersonal Maintaining Factors. *European Eating Disorders Review*

Uehara, T., Takeuchi, K., Ohmori, I., Kawashima, Y., Goto, M., Mikuni, M., & Vandereycken, W. (2002). Factor-analytic study of the Anorectic Behavior Observation Scale in Japan: comparisons with the original Belgian study. Psychiatry Research, 111(2), 241-246. doi:10.1016/S0165-1781(02)00143-9

Vall, E., & Wade, T. D. (2015). Predictors of treatment outcome in individuals with eating disorders: A systematic review and meta-analysis. *International Journal of Eating Disorders*, *48*(7), 946-971. doi:10.1002/eat.22411

Vandenbroucke, J. P., von Elm, E., Altman, D. G., Gøtzsche, P. C., Mulrow, C. D., Pocock, S. J., ... & STROBE Initiative. (2014). Strengthening the reporting of observational studies in epidemiology (STROBE): Explanation and elaboration. International Journal of Surgery, 12(12), 1500-1524. doi: 10.1016/j.ijsu.2014.07.014

Vandereycken, W. (1992). Validity and reliability of the Anorectic Behavior Observation Scale for parents. *Acta Psychiatrica Scandinavica*, *85*(2), 163-166. doi:10.1111/j.1600-0447.1992.tb01462.x

vanWijngaarden, B., Schene, A.H., Koeter, M., Vazquez-Barquero, J.L., Knudsen, H.C., Lasalvia, A., & McCrone, P. (2000). Caregiving in schizophrenia: Development, internal:10.1192/bjp.177.39.s21 consistency and reliability of the Involvement Evaluation Questionnaire - European Version. *British Journal of Psychology 177*(S39) 21–27. doi von Essen, L., Olow Sjödén, P., & Mattsson, E. (2004). Swedish mothers and fathers of a child diagnosed with cancer A Look at Their Quality of Life. Acta Oncologica, 43(5), 474-479. http://dx.doi.org/10.1080/02841860410029348

Wancata, J.,Krautgartner, M., Berner, J., Scumaci, S., Freidl, M., Alexandrowicz, R., & Rittmannsberger, H. (2006). The 'Carers'Needs Assessment for Schizophrenia'—an instrument to assess the needs of relatives caring for schizophrenia patients. *Social Psychiatry and Psychiatric Epidemiology, 41*(3), 221–229. doi:10.1007/s00127-005-0021-3

Ware Jr, J. E., Kosinski, M., & Keller, S. D. (1996). A 12-Item Short-Form Health Survey: construction of scales and preliminary tests of reliability and validity. *Medical Care*, *34*(3), 220-233. Retrieved from http://journals.lww.com/lww-medicalcare/pages/default.aspx

Whitney, J., Haigh, R., Weinman, J., & Treasure, J. (2007). Caring for people with eating disorders: factors associated with psychological distress and negative caregiving appraisals in carers of people with eating disorders. *The British Journal of Clinical Psychology / the British Psychological Society, 46*(4), 413–428. doi:10.1348/014466507X173781

Whitney, J., Murray, J., Gavan, K., Todd, G., Whitaker, W., & Treasure, J. (2005). Experience of caring for someone with Anorexia Nervosa: A qualitative study. *The British Journal of Psychiatry*, *187*(5), 444-449. doi:10.1192/bjp.187.5.444

Wiedemann, G., Rayki, O., Feinstein, E., & Hahlweg, K. (2002). The Family Questionnaire: development and validation of a new self-report scale for assessing expressed emotion. Psychiatry Research, 109(3), 265-279. doi:10.1016/S0165-1781(02)00023-9

Winn, S., Perkins, S., Walwyn, R., Schmidt, U., Eisler, I., Treasure, J., … & Yi, I. (2007). Predictors of mental health problems and negative caregiving experiences in cares of adolescents with Bulimia Nervosa. *International Journal of Eating Disorders, 40*(2), 171–178. doi:10.1002/eat

Winn, S., Perkins, S., Walwyn, R., Schmidt, U., Eisler, I., Treasure, J., … & Yi, I. (2007). Predictors of mental health problems and negative caregiving experiences in cares of adolescents with Bulimia Nervosa. *International Journal of Eating Disorders, 40*(2), 171–178. doi:10.1002/eat

Yokoyama, K, & Araki, S. (1991). *POMS Japanese version II*. Tokyo, Japan: Kaneko Shobo.

Zabala, M. J., Macdonald, P., & Treasure, J. (2009). Appraisal of caregiving burden, expressed emotion and psychological distress in families of people with eating disorders: A systematic review. *European Eating Disorders Review, 17*(5), 338–349. doi:10.1002/erv.925

Zarit, S.H., Reever, K.E., & Bach-Peterson, J. (1980). Relatives of the impaired elderly: correlates of feelings of burden. *Gerontologist, 20*(6):649–55. doi:10.1093/geront/20.6.649

Zigmond, A.S., & Snaith, R.P. (1983). The hospital anxiety and depression scale. *Acta Psychiatrica Scandinavica 67*(6), 361–370. doi:10.1111/j.1600-0447.1983.tb09716.x

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Major Research Project

Section B: Empirical Study

Moderators and Mediators of Relationships between Eating Disorders and Related Carer Difficulties, in the Context of a Skills Sharing Intervention for Carers of People with Anorexia Nervosa

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CANTERBURY CHRIST CHURCH UNIVERSITY

Abstract

Background: Carers of people with eating disorders (EDs) have heightened levels of distress, expressed emotion, burden and accommodation to the ED; factors implicated in maintenance of the ED. Although carers' skills interventions are helpful, how they effect change, and the processes involved in hypothesised relationships between ED symptoms and carer distress, is unclear. **Aims:** To determine the processes involved in the beneficial carer and service-user outcomes from a carers' skills RCT, including by longitudinally examining relationships between ED symptoms and carer distress. **Method:** This study utilised archival data from a multi-site large-scale carers' skills RCT**.** Primary carers (159) of people presenting for Anorexia Nervosa hospital treatment were randomly allocated to the intervention or treatment as usual. Moderators and mediators of intervention outcomes, and of longitudinal relationships between ED symptoms and carer distress, were examined. **Results:** The positive association between intervention and reduction in expressed emotion was significant only when more than one carer was involved per service-user. Reductions in expressed emotion and burden statistically mediated positive relationships between intervention and reduction in carer and ED outcomes. Findings from longitudinal mediation models were consistent with positive indirect associations between ED symptoms (at discharge) and carer distress (at six-month follow-up), and between carer distress (at baseline) and ED symptoms (at 12-month follow-up), mediated by carer and service-user factors. **Limitations & Implications**: Missing data may have introduced bias. Accessing only primary carers of severely unwell service-users limited generalisability. Results support the importance of carers' skills interventions in addressing ED-related service-user and carer difficulties.

Keywords: Carer, Anorexia Nervosa, Mediation, Distress, Intervention

Introduction

Anorexia Nervosa (AN) is a serious psychiatric illnesses with a mortality rate of 10-20% (Arcelus, Mitchell, Alex, Wales, & Nielsen, 2011; Harris & Barraclough, 1998) and lifetime prevalence of 0.7–1.0% (Fairburn & Harrison, 2003; Hoek & van Hoeken, 2003). The NICE guidelines (2004) recommend outpatient management as the first-line treatment, meaning parents and close others can take on a caregiving role often experienced as burdensome and associated with heightened rates of clinically relevant depression and anxiety (herein conceptualised as distress; Anastasiadou, Medina-Pradas, Sepulveda, & Treasure, 2014).

Models have been proposed to understand the ways in which the eating disorder (ED) and carer psychological distress and emotionally driven behaviours may impact one another. Two prominent models are described in turn. Seeking to explain how the ED may lead to carer distress, the Model of Carer Coping (Treasure et al., 2007; Treasure & Nazar, 2016, Figure 1) proposed that the impact of the ED symptoms and behaviours, related socio-economic consequences and the carer’s behavioural responses to the ED can result in reduced coping, leading to psychological distress for carers when resources are overwhelmed.

Seeking to explain how carer responses may support maintenance of the ED, the interpersonal aspect of the Cognitive-Interpersonal Maintenance Model of AN (Schmidt & Treasure, 2006; Treasure & Schmidt, 2013, Figure 2) proposed that the ED can lead to anxiety and distress in carers, which contribute to high expressed emotion and accommodating and enabling (Sepulveda, Kyriacou, & Treasure, 2009; Treasure et al., 2008).

Figure 1. Model of Carer Coping in Eating Disorders

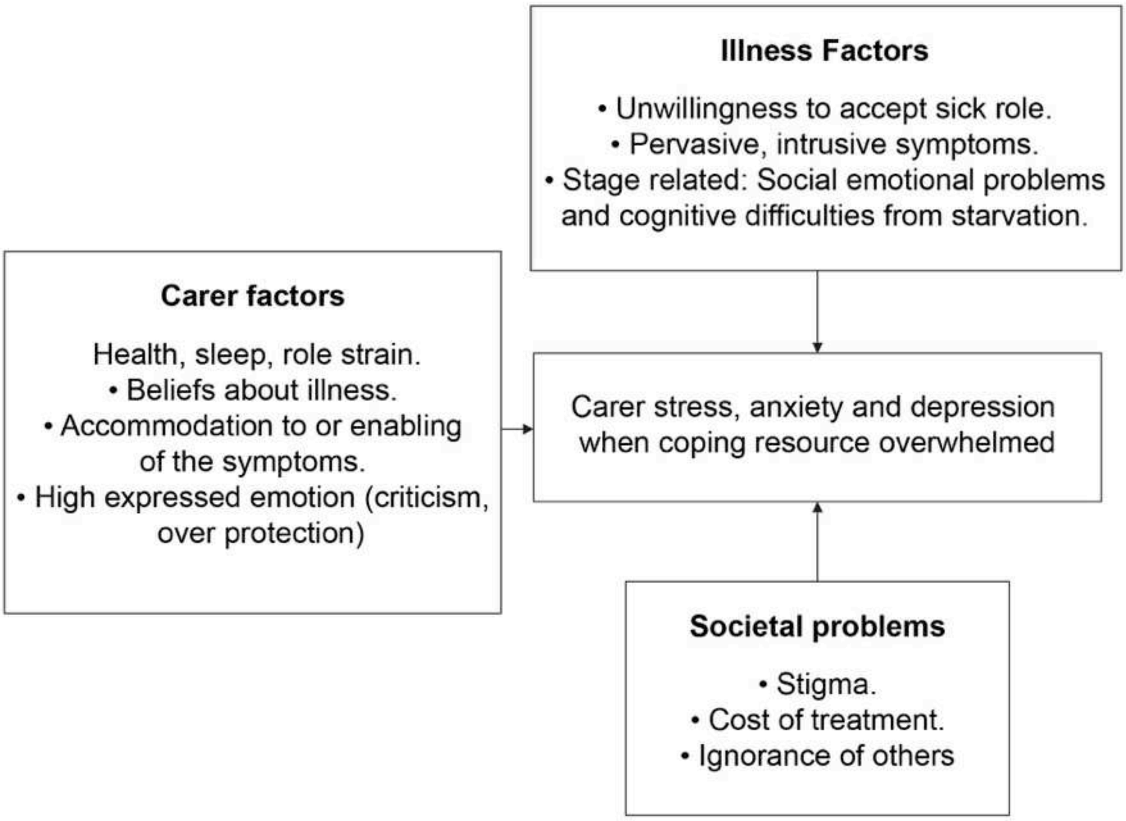

Figure 1. Model adapted from Treasure et al. (2007), showing the impact of factors associated with caregiving in eating disorders on carer distress.

Expressed emotion refers to caregivers’ criticism and emotional over-involvement. However, rather than being a one-way behaviour, it is proposed to describe the relational interaction between carer and cared-for factors, as carers attempt to care for their unwell loved one (Hooley & Campbell, 2002). Accommodating and enabling behaviours (herein ‘accommodation’) refers to carers’ attempts to accommodate family life to the demands of the ED, and to enable ED behaviours in an effort to reduce the negative impact of the illness (Sepúlveda, Kyriacou, & Treasure, 2009). Expressed emotion and accommodation are hypothesised (Treasure & Schmidt, 2013) to inadvertently lead to heightened ED symptoms and maintenance of the disorder. Empirical support for this model includes Vall and Wade's (2015) review finding that maternal depression predicted less favourable ED treatment outcomes, and Goddard et al. (2013c) found service-user[4] distress fully mediated a cross-sectional relationship between carer distress and ED symptoms, while carer distress was associated with expressed emotion. There is evidence for expressed emotion as a predictor of psychiatric relapse (Butzlaff & Hooley, 1998; Hooley, 2007), and service-user distress (Medina-Pradas et al., 2011). Furthermore, service-user distress has been found to predict ED symptoms (ibid; Goddard et al., 2013c; Vall & Wade et al., 2015). Salerno et al. (2016) reported that parental accommodation was predictive of poorer outcomes for adolescents with AN. However, the proposed processes by which carer distress may impact service-user distress and ED symptoms have not been examined longitudinally.

Support for the Model of Carer Coping (Treasure & Nazar, 2016) includes significant associations between ED carer distress and burden (e.g. Las Hayas et al., 2014), expressed emotion (e.g. Goddard et al., 2013c), accommodation (e.g. Rhind et al., 2016), stigma (e.g. Whitney, Haigh, Weinman, & Treasure, 2007), and carer coping (e.g. Coomber & King, 2012). However, the relationship between ED symptoms and carer distress, hypothesised by both the models discussed, is challenged by findings of non-significant associations (e.g. Kyriacou,

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[4] 'Service-users' is herein used to reference the participants with ED.

Figure 2. Interpersonal Aspect of The Cognitive Interpersonal Maintenance Model

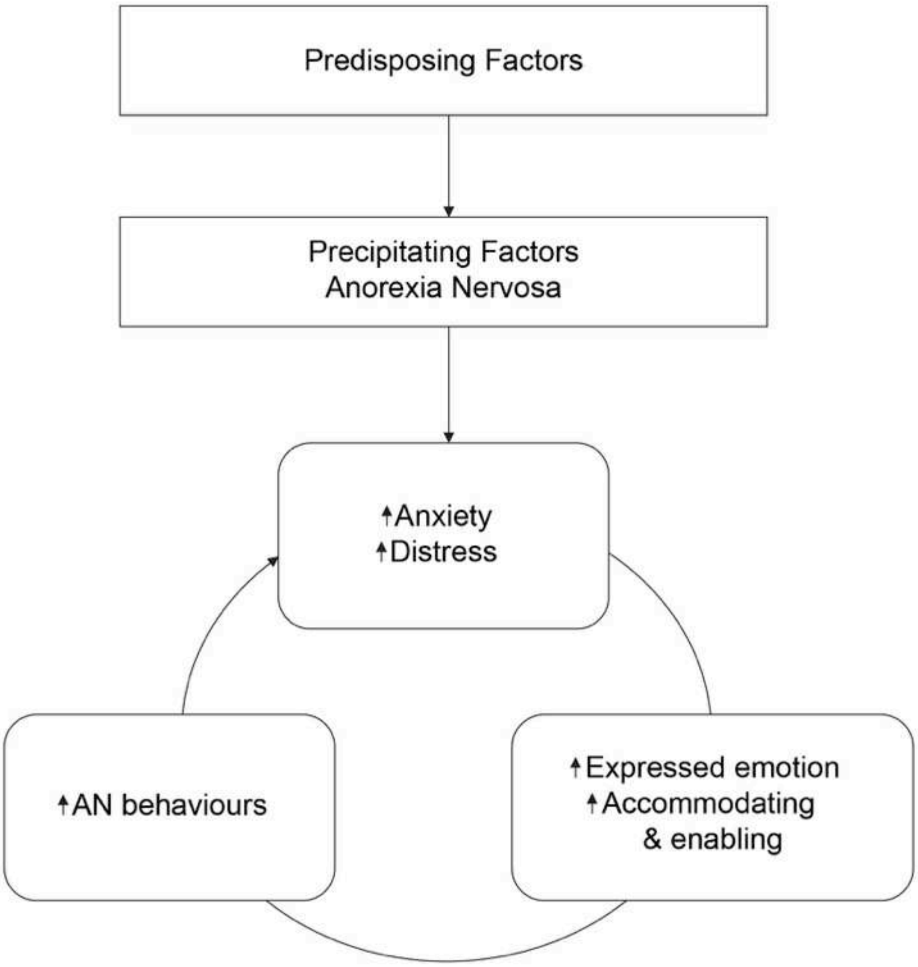

Figure 2. The model, adapted from Treasure and Schmidt (2013), can be applied transdiagnostically in eating disorders.

Treasure & Schmidt, 2008; Salerno et al., 2016; Sepulveda et al., 2014a). Carer burden[5], accommodation and expressed emotion are associated with each other (Sepulveda,

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[5] Demands, strains and negative experiences related to caregiving.

Whitney, Hankins, & Treasure, 2008) and the eating disorder (Anastasiadou et al., 2014), and are predictive of carer distress (Hibbs et al., 2015; Las Hayas et al., 2014), suggesting these as possible mediators of relationships between ED symptoms and carer distress. This is in keeping with models such as Kyriacou, Treasure and Schmidt (2008**),** Winn et al. (2007**)** and Szmukler et al. (1996), which suggest a role for intermediary variables concerning the way in which the ED symptoms become personally relevant for the carer. However, whether these variables mediate relationships between ED symptoms and carer distress has not been tested.

As a consequence of the wide-reaching ED-related difficulties and interpersonal aspects implicated in illness maintenance, developing interventions targeting effects of eating disorders on caregivers is imperative for both carers and service-users. The Model of Carer Coping (Treasure & Nazar, 2016) and Cognitive Interpersonal Maintenance Model (Treasure & Schmidt, 2013) provide a theoretical basis for such interventions employing skills-sharing approaches (e.g. Hibbs et al., 2015a; Treasure et al., 2008), most of which have reported outcomes of moderate-sized reductions in carer distress and small-moderate reductions in carer burden and expressed emotion (Hibbs et al., 2014). In contrast to literature regarding interventions for people with EDs (see Vall and Wade's 2015 meta-analysis), there has been little attention to moderators and mediators of carer and cared-for outcomes from interventions for carers.

Moderation ('*who*' or '*when*') and mediation ('*how*' or '*why*') analyses potentially have wide-reaching clinical and theoretical implications (Baron & Kenny, 1986; Hayes, 2013). Moderation occurs when the relationship between two variables differs at levels of the moderating variable, therefore indicating for who, or under what circumstances, differential treatments may be most beneficial. Mediation analysis may suggest mechanisms through which one variable effects another. For example, Goddard et al.’s (2011) pre-post study found reduction in carer distress following a carer-skills intervention was statistically mediated by reductions in expressed emotion and accommodation.

Mediators of outcomes from carers’ ED interventions have not been tested longitudinally with experimental designs, so robust empirical support for proposed mechanisms of action is lacking. Similarly, moderators of carers’ outcomes have been tested by only four studies (Goddard et al., 2011; Grover et al., 2011; Slater et al., 2015; Rhodes, Baillee, Brown, & Madden, 2008), while moderators of outcomes for the person with the ED from carer-skills interventions have not been tested.

Therefore, the current study examines moderators and mediators of carer and service-user outcome from a large-scale skills-sharing randomised controlled trial (RCT) for carers of people with a diagnosis of AN who were admitted to hospital for ED treatment (Hibbs et al., 2015a). The RCT reported multiple outcomes, but the focus of this study is on those most keenly implicated in ED maintenance and carer distress. The hypotheses were designed to address gaps in the literature above, and were guided by the RCT findings of Hibbs et al. (2015a), who reported that, compared to those receiving treatment as usual, those receiving the intervention showed significantly larger improvements in burden, expressed emotion and ED symptom severity, but not accommodation, carer or service-user distress. In addition to examining intervention outcomes, longitudinal processes contributing to, and resulting from, carers’ distress are investigated.

Hypotheses

Moderators of intervention outcome.

Potential moderators of the relationship between experimental group (intervention vs. control) and outcome variables (Hypotheses 1a to 1e) were selected on the basis of existing empirical support and the specifics of the intervention. These are presented in Table 1. With the exception of expressed emotion, which was found by Grover et al.'s (2011) notably under-powered study not to moderate change in carer distress following carers' ED intervention, these hypotheses have not previously been tested in experimental designs addressing outcomes of a carers' ED intervention.

Mediators of intervention outcome.

Hypotheses related to mediators of intervention outcome (H2, H3) were based on the empirical literature described above, and the interpersonal aspect of the Cognitive Interpersonal Model (Treasure & Schmidt, 2013), upon which the intervention was based. These are presented in Table 2.

It is arguable that the change processes involved in such complex disorders are likely to involve a cascading sequential mediation (Prins, Ollendick, Maric, & MacKinnon, 2015). The greater reduction of ED symptoms in the intervention condition (Hibbs et al., 2015a) was hypothesised to be positively sequentially mediated by greater reductions in first burden then expressed emotion (H4, Figure 3, Table 2). The contribution of these variables in this order is theoretically sound, and supported by significant improvements in burden occurring earlier during the course of the intervention follow-up period than changes in expressed emotion (Hibbs et al., 2015a).

Table 1
Hypotheses Regarding Moderation of Intervention Outcomes

Hyp. #	Proposed moderator/s	DV/s^1	Direction of moderation	Theoretical basis	Empirical basis
1a	i. Duration of ED ii. Service-user age	ED symptom severity	Older service-users with longer illness duration will benefit less from the intervention in terms of ED symptom reduction.	Service-user age and illness duration are associated (Fisher, 2010) due to the protracted prognosis of EDs (Beumont, 2000). Longer duration may indicate a more entrenched, treatment resistant illness. Older service-users may be more independent from carers and thus less affected by carer changes.	Service-user age and illness duration have both been implicated as moderators of treatment outcome in adolescents (Murray, Loeb, & Le Grange, 2015). Younger service-users and those with shorter illness duration have been found to respond to interventions including service-users more favourably (Russell, Szmuckler, Dare, & Eisler, 1987, Vall & Wade, 2015).
1b	Service-user BMI	ED symptom severity	Service-users with lower BMI will benefit less from the intervention in terms of ED symptom reduction.	Lower BMI is an indicator of greater illness severity (e.g. Rhind et al., 2014), which may be harder to ameliorate.	Lower BMI at baseline is a robust predictor of less favourable treatment outcome (Steinhausen et al., 2009).
1c	Carer's own history of eating difficulties	i. Accommodation ii. Carer distress iii. ED symptom severity	Carers with their own history of eating difficulties will benefit less from the intervention in terms of reductions in carer distress and accommodation. The service-users they care for will benefit less in terms of reduction in ED symptoms.	Carers having their own eating difficulties could be hypothesised to make the situation more distressing and make it more challenging for them to change accommodating to the symptoms as they struggle to manage difficulties related to food, weight and shape for both the service-user and themselves. The intervention may also be less effectively targeted to the difficulties they experience as carers. According to the CIMM (Treasure & Schmidt, 2013), less reduction in carer distress and accommmodation could be expected to reduce reduction in ED symptoms.	Goddard (2013b) found carer ED history was associated with accommodating and carer distress. Carers having their own ED difficulties can reduce efficacy of ED treatment involving parents on patient ED symptoms (Lock & Le Grange, 2013).

Table 1 continued

Hypotheses Regarding Moderation of Intervention Outcomes

Hyp. #	Proposed moderator/s	DV/s^1	Direction of moderation	Theoretical basis	Empirical basis
1d	Number of carers per patient (1 or >1)	i. Expressed emotion ii. Accommodation	Where more than 1 carer for each service-user takes part in the intervention, those carers will benefit more from the intervention in terms of reduced expressed emotion and accommodation	Multiple carers per service-user were encouraged to take part in the intervention. Number of carers per service-user was hypothesised to moderate behavioural outcomes expressed emotion and accommodation as carers supported each other to change and maintained consistency.	This is a speculative hypothesis, supported by findings that consistency is important in behaviour-change interventions (e.g. McClean et al., 2005).
1e	i. Accommodation ii. Expressed emotion	Carer distress	Carers showing higher accommodation and expressed emotion at baseline will benefit more from the intervention in terms of reduction in carer distress.	According to the Model of Carer Coping (Treasure & Nazar, 2016), accommodation and expressed emotion can lead to carer distress. As targets of the intervention (Rhind et al., 2014), higher levels of these factors at baseline is expected to increase the opportunity for their reduction, with this contributing to greater reduction in carer distress.	Goddard et al. (2011) reported carers with higher expressed emotion and accommodation at baseline to show significantly greater reductions in carer distress following a carer skills intervention. However, this was not addressed within an experimental design, meaning it was not possible to ascertain whether the change in distress was related to the intervention or was coincidental.

Note:-^{1} Dependent variable is amount of change in variable between baseline and six months after the service-user's discharge. Hyp. #: Hypothesis reference number.

Table 2

Hypotheses Regarding Mediation of Intervention Outcomes

Hyp. #	Independent variable	Hypothesised mediator	Dependant variable[2]	Hypothesis
2a	Group[1]	Expressed emotion	ED symptoms	The relationship between receipt of ECHO and reduction in ED symptoms will be mediated by reduction in expressed emotion.
2b	Group[1]	Accommodation	ED symptoms	The relationship between receipt of ECHO and reduction in ED symptoms will be mediated by reduction in accommodation.
3a	Group[1]	Expressed emotion	Carer distress	The relationship between receipt of ECHO and reduction in carer distress will be mediated by reduction in expressed emotion.
3b	Group[1]	Burden	Carer distress	The relationship between receipt of ECHO and reduction in carer distress will be mediated by reduction in burden.
3c	Group[1]	Expressed emotion	Accommodation	The relationship between receipt of ECHO and reduction in accommodation will be mediated by reduction in expressed emotion
3d	Group[1]	Burden	Accommodation	The relationship between receipt of ECHO and reduction in accommodation will be mediated by reduction in burden.
3e	Group[1]	Burden	Expressed emotion	The relationship between receipt of ECHO and reduction in expressed emotion will be mediated by reduction in burden
3f	Group[1]	Expressed emotion	Burden	The relationship between receipt of ECHO and reduction in burden will be mediated by reduction in expressed emotion
3g	Group[1]	Accommodation	Carer distress	The relationship between receipt of ECHO and reduction in carer distress will be mediated by reduction in accommodation.
4	Group[1]	1. Burden 2. Expressed emotion	ED symptoms	The relationship between receipt of ECHO and reduction in ED symptoms will be sequentially mediated by first reduction in burden, then reduction in expressed emotion.

Note: [1] Intervention vs. control group. [2] Dependent variable relates to reduction in variable between baseline and 6-months follow-up. Hyp. #: Hypothesis reference number.

Figure 3. Sequential Mediation of the Association between Intervention and ED Symptoms

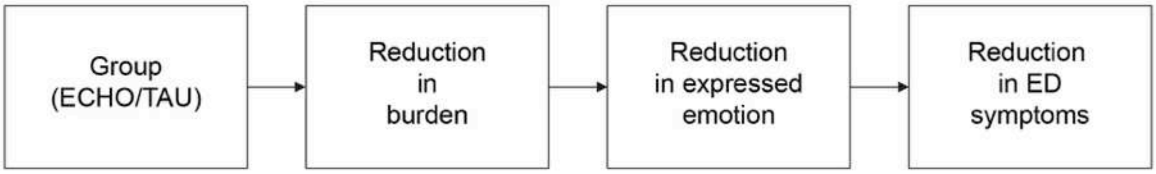

Figure 3. Hypothesised mediational model to be tested (H4) describing sequential mediation of the positive association between intervention and reduction in ED symptoms between baseline and six months (compared to carers in the TAU condition) sequentially positively mediated by first reduction in burden, then reduction in expressed emotion, both between baseline and 6-month follow-up time period.

Longitudinal examination of the role of carer distress.

We hypothesised that burden, accommodation and expressed emotion would mediate the hypothesised relationship between ED symptoms and carer distress in a longitudinal model consistent with the hypothesised causal order (H5).

Consistent with the literature, a positive relationship between carer distress and ED symptom severity was hypothesised to be mediated sequentially in a positive longitudinally-measured chain from carer distress, to expressed emotion, to service-user distress, to ED symptom severity (H6, Figure 4). Additionally, a positive longitudinal relationship between carer distress and ED symptoms was hypothesised (H7) to be positively mediated by accommodation. These hypotheses are summarised in Table 3.

Figure 4. Longitudinal Sequential Mediation of the Association between Carer Distress and ED Symptoms

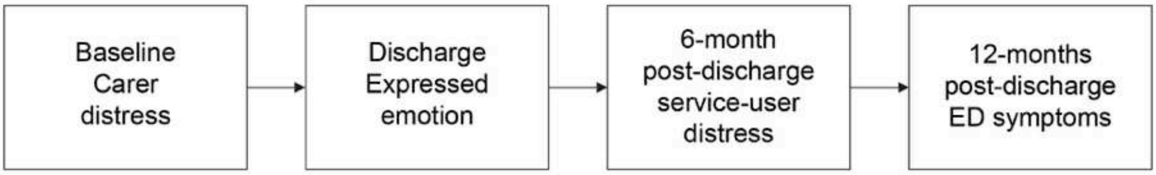

Figure 4. Hypothesised longitudinal model to be tested (H6) describing sequential mediation of the positive association between carer distress at baseline and ED symptoms at 12-month follow-up (compared to carers in the TAU condition) sequentially positively mediated by first expressed emotion (discharge time-point), then expressed emotion (6-month time-point).

Table 3

Hypotheses Regarding Longitudinal Mediation of Relationships between ED Symptoms and Carer Distress

Hyp. #	Independent variable		Hypothesised mediator/s		Dependant variable		Hypothesis
	Name	T^1	Name	T^1	Name	T^1	
5a	ED symptoms	D	Burden	6m	Carer distress	12m	The positive relationship between ED symptoms and carer distress at will be positively mediated by burden.
5b	ED symptoms	D	Accom-odation	6m	Carer distress	12m	The positive relationship between ED symptoms and carer distress at will be positively mediated by accommodation.
5c	ED symptoms	D	Expressed emotion	6m	Carer distress	12m	The positive relationship between ED symptoms and carer distress at will be positively mediated by expressed emotion.
6	Carer distress	B	1. Expressed emotion 2. Service-user distress	1: D 2: 6m	ED symptoms	12m	The positive relationship between carer distress and ED symptoms at will be sequentially mediated by first expressed emotion, then service-user distress.
7	Carer distress	D	Accom-odation	6m	ED symptoms	12m	The positive relationship between carer distress and ED symptoms will be mediated by accommodation.

Note: T^1: Time-point of variable measurement. B: Baseline time-point. D: Discharge time-point. 6m: 6-month follow-up time-point. 12m: 12-month follow-up time-point. Hyp. #: Hypothesis reference number.

Method

This project used archival data from a completed RCT, which had not yet been analysed in relation to the above hypotheses. Therefore, the procedure has been published in detail (Goddard et al., 2013b). For clarity, key elements are reported below.

Design and Ethical Considerations

Data were obtained from a multi-centre, parallel-group, pragmatic RCT of an intervention for carers of AN service-users with two arms; intervention versus a treatment as usual (TAU) control. The current study analysed archival data in a way consistent with the remit of the consent originally given by participants, and with the ethical application for the trial, for which approval was granted by the Royal Free Hospital Ethics Committee (08/H0720/41, see Appendices G to I and Q). The trial minimised risks to this vulnerable group by not involving service-users in the intervention. The intervention was designed not to replace any aspect of individual or family therapy for carers or service-users, and was administered as an additional resource alongside usual treatment offered by clinical teams. Carers allocated to the TAU condition were given contact details of the ED charity Beat and offered the intervention following completion of the trial.

Participants

Participants were service-users (aged ≥ 12) admitted for inpatient or day-patient[6] treatment at one of fifteen NHS hospitals in England with a primary diagnosis of AN[7], and up to three of each service-user's caregivers. Carers were defined as someone who gives unpaid help

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[6] non-residential intensive specialist ≥ 4 days a week

[7] /Eating Disorder Not Otherwise Specified with anorexic symptoms

for a child, partner, sibling or friend (Department of Health, 2014). Where there was more than one carer per service-user, primary carers self-identified as the person spending most time caregiving. This study analysed data from dyads of service-users and their primary caregivers (*n* = 178*)*. The Consort diagram (Moher et al., 2010) for participant flow is presented in Figure 3. Participants needed to be able to speak and understand English, give informed consent, and not have a severe comorbidity.

Procedure

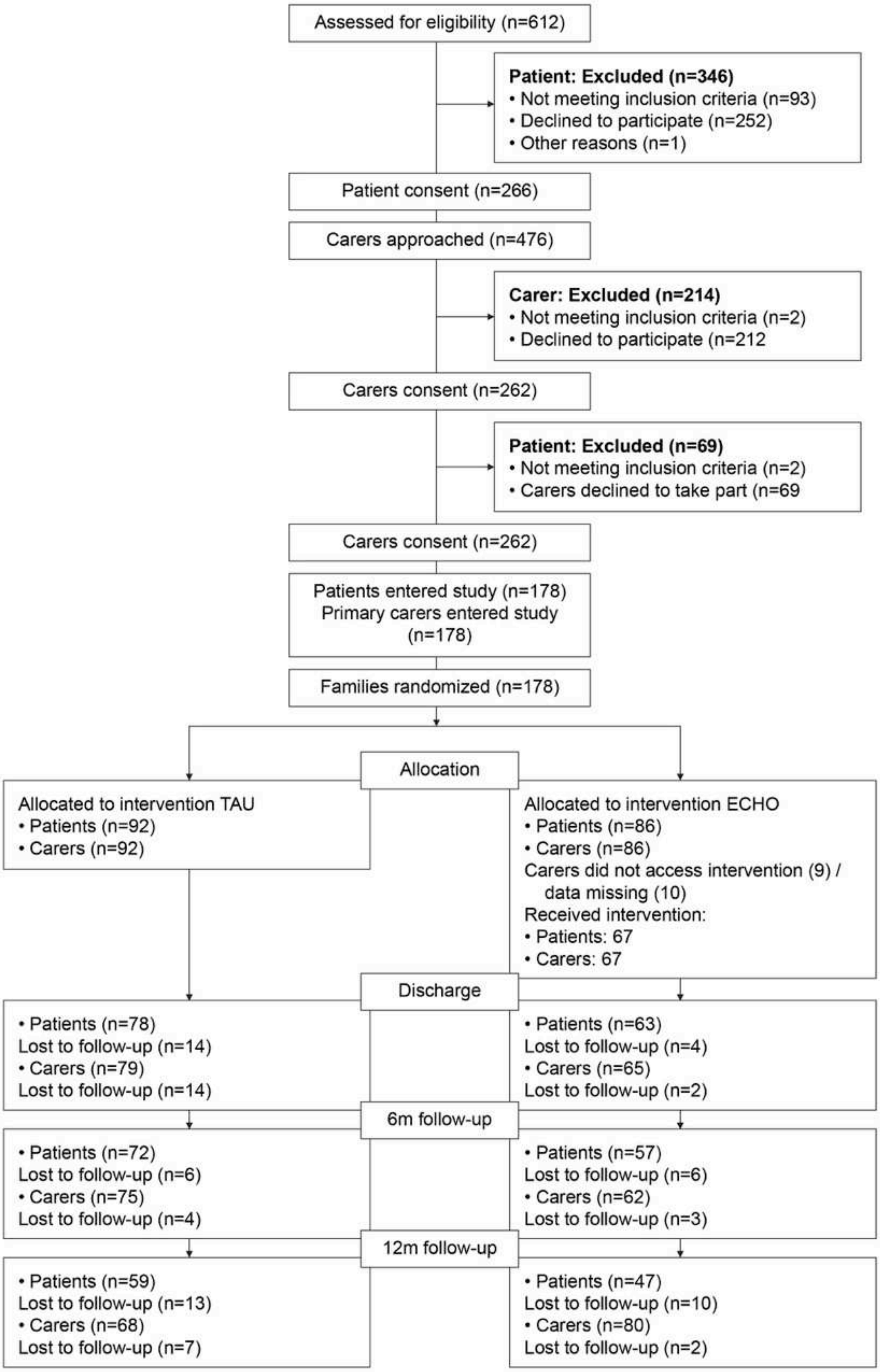
Measures.

Data were gathered via postal questionnaire from service-users and their carers at baseline (admission + 2 weeks), discharge from hospital, and at six and 12-month post-discharge follow-up. The measures used by this study (see Table 4) are validated in eating disorder samples with good psychometric properties, and have high internal consistency in this sample (α between .87 and .96; Goddard et al., 2013c). The number of participants giving questionnaire responses at each time-point is shown in Table 5.

Intervention.

'Expert Carers Helping Others' (ECHO) was a New Maudsley collaborative care intervention for carers of people with eating disorders. The approach (Treasure, Rhind, MacDonald, & Todd, 2015) and specifics of the intervention (Goddard et al., 2013b) are described in detail elsewhere. In brief, the intervention involved sharing theory and practical examples via a book (Treasure, Smith, & Crane, 2007), five purpose-made DVDs, and telephone coaching sessions for carers (up to 10 per service-user). Coaching was delivered bi-weekly where possible.

Figure 5. Consort Diagram for Participant Flow



Adapted from Moher et al. (2010).

Table 4

Details of Psychometric Measures Used by This Study

Name of measure	Authors & date	Construct	Domains, scoring & comments
Issued to carers			
Family Questionnaire (FQ)	Weidemann, Rayki, Feinstein, & Hahlweg, 2002	Expressed emotion present in carer's interactions with service-user	Subscales Criticism and Emotional Over-Involvement. A frequently used measure with excellent psychometric properties. Good convergent validity with more time-consuming expressed emotion interview measures, and sound psychometric properties (Sepulveda, Whitnet, Hankins, & Treasure, 2008). Higher scores indicate presence of higher expressed emotion.
Accommodation and Enabling Scale for Eating Disorders (AESED)	Sepulveda, Kyriacou, & Treasure, 2009	Behaviour accommodating to, and enabling of eating disorder demands	Developed and validated for use with ED carers. 33 items on a 5-point likert scale. Domains: Avoidance and Modifying Routines, Providing Reassurance, Accepting Rituals around mealtimes, Turning a Blind Eye to unwanted behaviours and allowing ED to Control Family functioning. High scores indicate a greater degree of carer behaviour that accommodates to or enables the ED.
Eating Disorders Symptom Impact Scale (EDSIS)	Sepulveda et al., 2008	Burden specifically related to the impact of ED symptoms	Developed for use in eating disorders, validated with good psychometric properties. Self-report, 24 items with a 1-4 likert scale. Domains: problems related to cared-for's Nutrition and Dysregulated behaviour, Guilt (carer assumption of responsiblity for illness), and Social isolation (for both carer and cared-for). High scores indicate high carer self-perception of ED-related burden.
Issued to both service-users and carers			
Depression, Anxiety and Stress Scale (DASS)	Lovibond & Lovibond, 1993.	Psychological distress	A 21-item self-report scale with good psychometric properties. Validated in clinican and non-clinical populations. Domains Depression, Stress, Anxiety. Higher scores indicate higher distress.
Socio-demographic & clinical questionnaire	-	Socio-demographics, clinical and relationship information	The following variables were gathered via a purpose-made questionnaire: Demographics and clinical characteristics of illness and treatment, and details about the caregiving relationship. Carer eating history (binary yes/no) was based on carer disclosure of a life-time history of difficulties with eating/shape/weight.
Issued to service-users			
BMI (body-mass index)	-	BMI at baseline	Where service-users were unable to give BMI, consent was sought and their treatment team was contacted. Lower scores indicate lower weight for height.
Eating Disorders Examination Questionnaire (EDEQ)	Luce & Crowther (1999)	Eating disorder symptoms	Self-report measure of eating disorder symptoms with good validity and reliability. Assesses psychological and behavioural ED symptoms over the 28 days prior to administration. Domains: Eating, Shape, Weight concerns. Higher score indicates greater symptomology.

Aims of the intervention included reduction in carers' distress, burden, expressed emotion and accommodation, and, through carers' more highly skilled caregiving efforts and reduction in interpersonal maintaining factors, improvements in service-user distress and ED symptoms.

Service-User Involvement

The department which collected the data had a strong presence of service-user and carer involvement, including as coaches and co-authors of intervention materials. Service-users were consulted in the development of this study's design.

Data analysis

Data were analysed using IBM SPSS version 22. Extensive work was undertaken prior to analysis of the large raw datasets; data were combined, cleaned and re-coded before use. Dyads allocated to the intervention arm for whom treatment information was missing (n=10) or who did not access any treatment (n=9) were excluded.

Moderation and Mediation.

Data were analysed in line with Hayes' (2013) contemporary approach, using the PROCESS plug-in for SPSS (Hayes, 2012), chosen as the superior approach uniquely providing details on the size of indirect effects, and enabling bootstrapping (Hayes, 2013). While causal language is employed in this and the results section, both for clarity and in keeping with convention (ibid), this is not intended to suggest these correlational analyses imply causality (Field, 2013).

Moderation.

Moderation analysis using linear regression tested whether relationships between the predictor (condition; ECHO/TAU) and intervention outcomes significantly differed at levels of the proposed moderator variables, as described in Figure 6.

Mediation.

The test of simple mediation conducted using ordinary least squares path analysis is described in Figure 7. In the present data, two approaches to variable selection were used to optimise validity of observed indirect effects. Firstly, to explore mediators of the effect of intervention on outcomes (hypotheses 2-3), the amount of change between baseline and six-month measures was used for proposed mediator and outcome variables (Hayes, 2013). Secondly, as longitudinal data exist, time-points were entered in sequence of proposed causality to test hypotheses 5-7 (Deković, Asscher, Manders, Prins, & van der Laan, 2012).

To disentangle order effects and increase the validity of suggesting causality beyond only theoretical assumptions, Hayes (2013) recommends following-up mediation analysis by testing mediator and outcome variables in reverse order. This was actioned for hypotheses 2, 3, 5 and 7. For hypothesis 4, mediators and outcome were tested in all positions, and for hypothesis 6, an alternative position was tested in line with theory (see results). Non-significant findings are listed in Appendix J.

Sequential mediation.

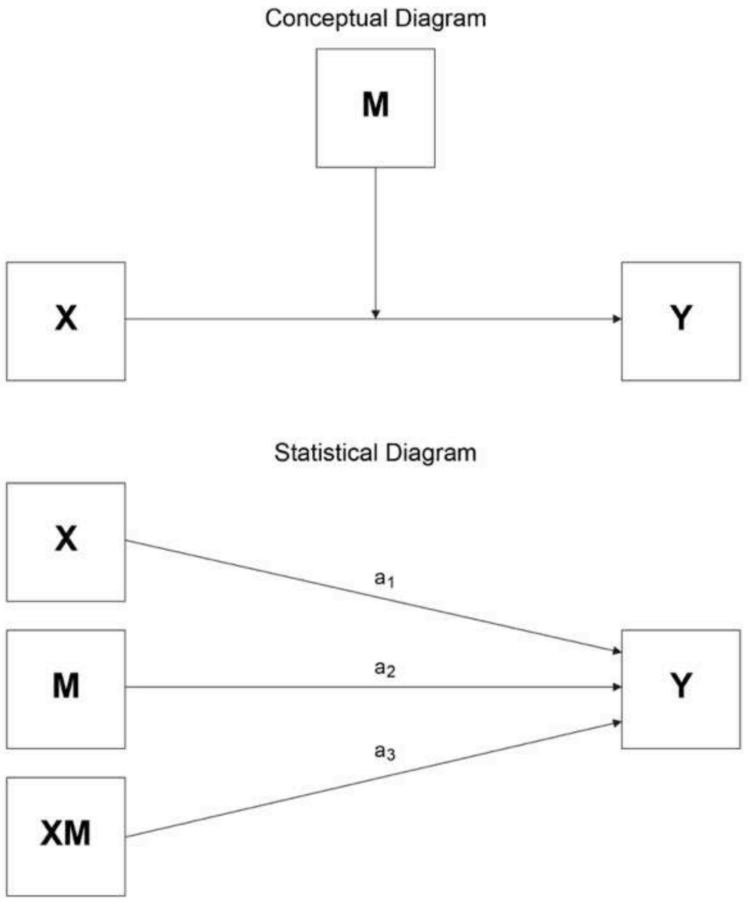
Hypotheses 4 and 6 regarding indirect effects involving two mediators were tested using the model described in Figure 8.

Table 5

Number of Questionnaire Respondents by Time-Point

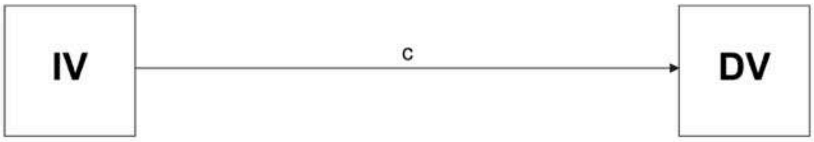
	Number of respondents (% of sample) at timepoint			
Measure	Baseline	Discharge	6 Months	12 Months
Patient variables				
Distress, Anxiety and Stress Scale	151 (94.97)	119 (74.84)	115 (72.33)	106 (66.67)
Eating Disorder Examination Questionnaire	147 (92.45)	118 (74.21)	116 (72.33)	105 (66.04)
Carer Variables				
Distress, Anxiety and Stress Scale	156 (98.11)	125 (78.62)	124 (77.99)	112 (76.73)
Family Questionnaire	156 (98.11)	126 (79.25)	123 (77.36)	112 (76.73)
Accommodation and Enabling Scale for Eating Disorders	147 (92.45)	125 (78.62)	119 (74.84)	118 (74.21)
Eating Disorder Symptom Impact Scale	154 (96.86)	110 (69.18)	113 (71.07)	105 (66.04)

Figure 6. Conceptual and Statistical Diagrams for Simple Moderation

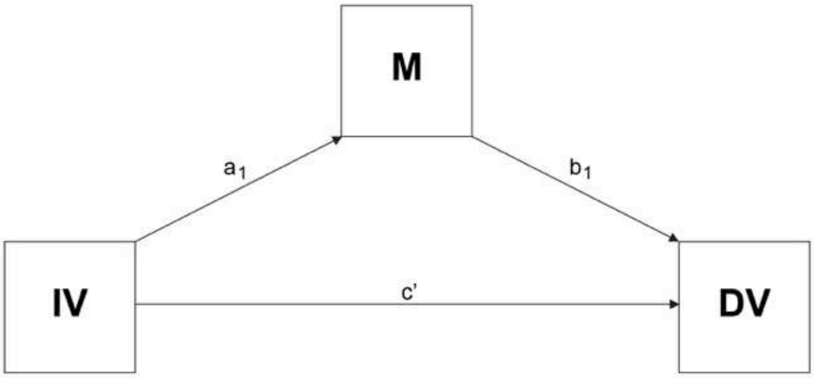


Adapted from Hayes (2013).

Figure 7. Simple Mediation Model to be Tested



Direct Pathway



Indirect or Mediated Pathway

Figure 7. Adapted from Hayes (2013). Simple mediation is said to occur when there is an indirect effect (ab) of the independent variable (IV) on the dependent variable (DV) through one or more mediating (M) variables. This requires the combination of the relationship between the independent and mediating variable (a) and the relationship between the mediating and dependent variable (b) to reach significance. Through development of understanding since the Baron and Kenny approach (1986), evidence of a simple association between the IV and DV is no longer a precondition of modern mediation analysis (Hayes, 2013; MacKinnon, 2008). When the total effect (c; the effect of the combined IV and mediator variable/s on the DV), is non-significant, or significance does not markedly reduce after partialing out the effect of the mediator (c'; the 'direct effect' of the IV on the DV), partial mediation has occurred. This contrasts with 'full' mediation where a significant total effect becomes a non-significant direct effect.

Figure 8. Sequential Mediation Model to be Tested

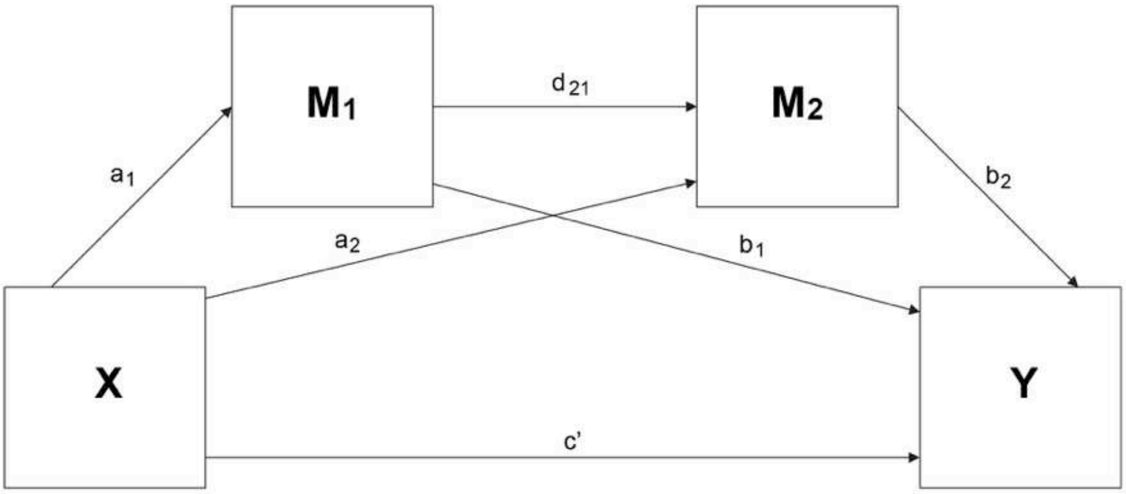
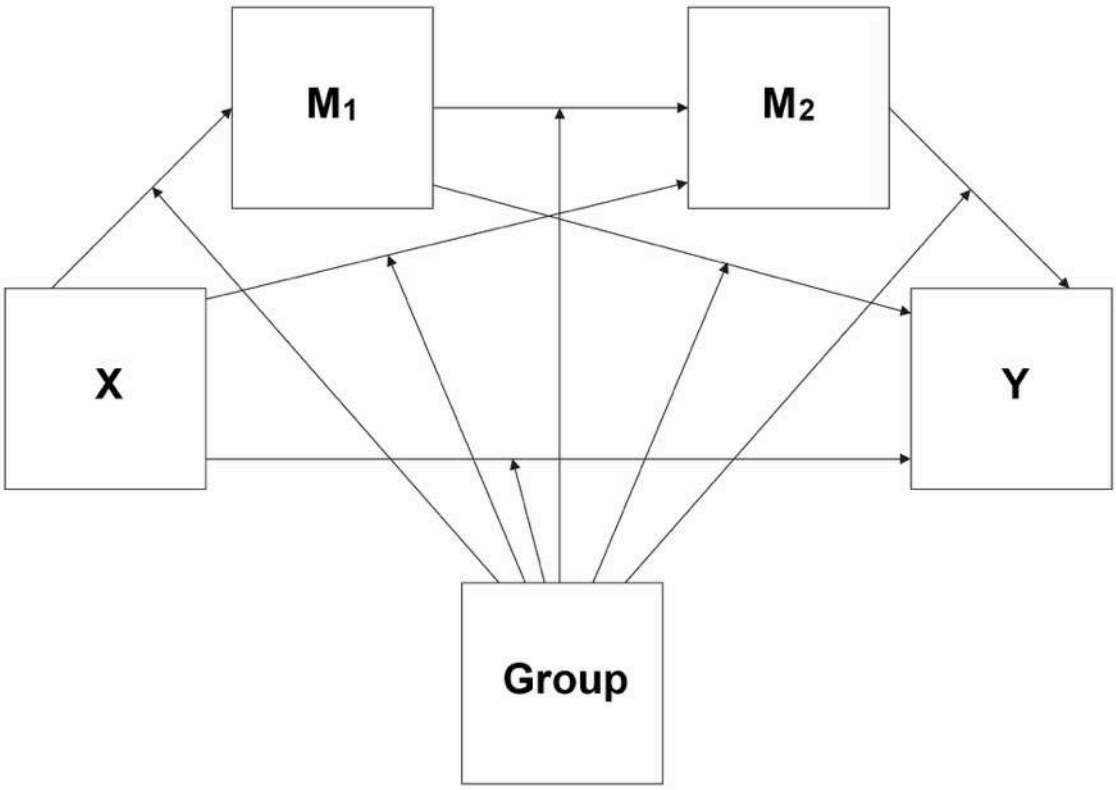


Figure 8. Adapted from Hayes (2013). This model simultaneously assesses indirect effects through the first (a1b1) and second (a2b2) mediator, and sequential mediation through the combined indirect through both the first and second mediator, additionally requiring a relationship between the first and second mediator (a1d21b2).

Moderated mediation.

Expanding upon these ideas, moderated mediation can occur when an indirect effect differs at different values of a moderator. Conditional process analysis was utilised pre-hoc (hypotheses 6-8,) to simultaneously assess whether experimental group (ECHO/TAU) moderated the relationships to be tested (e.g. Figure 9). All participants would be analysed together if condition was not a moderator, and separately if significant moderated mediation was observed.

Figure 9. Simultaneous Test of Group as a Moderator of Direct and Indirect Effects to be Tested in Hypothesised Model of Sequential Mediation



Adapted from Hayes (2013).

Missing data.

As with many large-scale, longitudinal RCTs (Van Buuren, 2012), there were significant missing data. Multiple Imputation is suitable for imputation of small quantities of data that are missing at random (MAR). However, missing questionnaire data were found not to be MAR (Appendix K) and large proportions of data (considerably greater than 10%, see Appendix L) were missing for most variables. For these reasons, Multiple Imputation was not appropriate (Van Buuren, 2012). Therefore, missing data were handled by omitting it listwise on an analysis-by-analysis basis. The benefit of this is maximum validity of results which are based only on data collected (Scheffer, 2002). Number of participants therefore differed by, and are described with, each analysis.

Power calculations.

Bootstrapping within PROCESS meant there were no recommendations on sample size to consider (Hayes, 2009). However, as a guide, the work of Fritz and Mckinnan (2007) suggests the sample size should have sufficient power (.8) to allow small-to-medium effects to be detected.

Assumptions and bootstrapping.

Bootstrapping is a robust, modern, multiple resampling technique not reliant on assumptions regarding sample distribution, therefore precluding the need to test multicollinearity, homogeneity of variance, outliers and deviations from normal distribution (Hayes, 2013). Bootstrapping also copes well with heteroscedasticity and PROCESS' standard error estimator was selected wherever deviations from homoscedasticity (assessed using scatter and PP plots) were suspected. Violation of the assumption of independence was avoided by only utilising data from primary carers in analyses and entering carers and service-users as related data.

Bias-corrected, bootstrapped confidence intervals based on 1000 bootstrapped samples (herein BCI) were used to assess the significance of indirect effects, with BCIs that did not cross zero indicative of significance at the 5% level. Standardised effect sizes are reported as these are more comparable across studies (Field, 2013), with the exception of analysis with mediators with dichotomous independent variable (e.g. group), where the standardised effects are not meaningful (Hayes, 2013, p. 188). In such cases, unstandardised effects are instead reported. Where available, kappa-squared ($ƙ^2$) indirect effect sizes are given. $ƙ^2$ is comparable to R^2 (0.01 = small; 0.09 = medium; >0.25 = large; Field, 2013).

For clarity, statistical methods are described further with results. Where not mentioned below, all assumptions were met; where violated, alternative analysis is presented using non-parametric tests.

Results

Demographics

Table 6 presents sociodemographics and clinical characteristics for the 159 included dyads of service-users and their primary carers. See Appendix M for sociodemographics and clinical characteristics for the excluded group (*n* = 19). A detailed summary of descriptive and clinical information for the total sample of service-users has been published (Goddard et al., 2013a).

The mean age of service-users was 25.53 (*SD* = 8.55, range 13-57), with 16 aged under 18 years at baseline. The majority of service-users had been admitted to hospital due to being significantly underweight (mean BMI a t baseline = 15.07, *SD* = 2.30) or having electrolyte problems; 30.20% (*n* = 45) used vomiting as a compensatory method. Mean illness duration was 9.21 years (*SD* = 8.11), with almost half having been unwell for at least six years (49.7%, *n* = 83; enduring AN). The service-users' hospital stay was 180.89 days on average (*SD* = 120.27, range 28-991), with one person remaining inpatient for the two years study duration. Roughly half the service-users (49.65%, *n* = 70) also had a diagnosis of depression.

Table 6

Sociodemographics and Clinical Characteristics of Service-Users and Carers at Baseline

	ECHO (*n* = 67)		TAU (n = 92)		Tests of difference: ECHO/TAU	
	Carer	SU	Carer	SU	Carer	SU
Demographics						
Age, years: mean (SD)	51.56 (9.82)	25.31 (8.94)	51.24 (9.96)	25.69 (8.29)	*t*(143) = 0.196, *p* = .85	U = 2896, *p* = .52
Female: n (%)	60 (89.55)	65 (97.01)	72 (78.26)	86 (93.48)	C^2 = 3.51, p = .06	*F*, *p* = .47
Ethnic origin: n (%)					*F* = 2.05, p = .63	*F* = .52, *p* = .88
White British	59 (88.06)	62 (92.54)	80 (86.96)	79 (85.87)		
White other	4 (5.97)	0 (0.00)	5 (5.43)	1 (1.09)		
Asian/mixed Asian	3 (4.48)	3 (4.48)	4 (4.35)	5 (5.43)		
Other	1 (1.49)	1 (1.49)	0 (6.00)	1 (1.09)		
Employment: n (%)					C^2 = 0.43, p = .51	C^2 = .74, *p* = .39
Full/part time employed	41 (61.19)	8 (11.94)	59 (64.13)	15 (16.30)		
Unemployed[a]	26 (38.81)	59 (88.06)	33 (35.87)	74 (80.43)		
Highest level of education: n (%)					*F* = 1.98, p = .59	*F* = 1.90, *p* = .60
No qualification	4 (5.97)	2 (2.99)	8 (8.70)	5 (5.43)		
O/A Levels	25 (37.31)	32 (47.76)	24 (26.09)	48 (52.17)		
University/higher degrees	31 (46.27)	26 (38.81)	46 (50.00)	26 (28.26)		
Other	7 (10.45)	7 (10.45)	10	9 (9.78)		
Marital status: n (%)					C^2 = 0.13, *p* = .72	C^2 = 4.3, *p* = .04*
Married/living together	51 (76.12)	8 (11.94)	67 (72.83)	23 (25.00)		
Single/divorced/widowed/seperated	16 (23.88)	59 (88.06)	24 (26.09)	68 (73.91)		
> 1 caregiver n (%)	41 (61.19)		38 (41.30)		C^2 = 6.135, *p* = .01*	

Note: (%) Indicates percentage of respondents by group. Contact hours is face to face. TAU: Treatment as usual.

[a] Unemployed/sick/student/retired/homemaker/other

Table 6 continued

Sociodemographics and Clinical Characteristics of Service-Users and Carers at Baseline

	ECHO (*n* = 67)		TAU (n = 92)		Tests of difference: TAU vs ECHO
	Carer	SU	Carer	SU	
Relationship					
Relationship: n (%)					C^2 = 5.87, *p* = .12
Mother	57 (85.07)		69 (75.00)		
Father	0 (0.00)		5 (5.43)		
Partner/spouse	7 (10.45)		15 (16.30)		
Sibling/friend	3 (4.48)		3 (3.26)		
Living together: y/n (% yes)		43/23 65.15 (65.15)		66/21 (75.86)	C^2 = 2.1, *p* = .15
Contact h/w: n (%)					F = 7.73, p = .13
0-7		20 29.85 (29.85)		22 (26.19)	
8–14		13 19.40 (19.40)		7 (8.33)	
15–21		2 2.99 (2.99)		8 (9.52)	
>21		31 46.27 (46.27)		46 (54.76)	
Other		1 1.49 (1.49)		1 (1.19)	
Clinical characteristics					
Duration of illness, months: mean (SD)		81.87 (84.74)		91.83 (97.82)	U = 2200.0, *p* = .88
BMI: mean (SD)		15.43 (2.35)		14.30 (12.40)	U = 3283.0, *p* = .09
Length of admission, days: mean (SD)		179.36 (135.83)		182 (108.02)	U = 2855.0, *p* = .56
compensatory vomiting: y/n (% yes)		19/46 29.2 (29.23)		26/58 (30.95)	C^2 = 0.03, *p* =.87
Comorbidity: n (%)					F = 3.36, *p* = .51
None		20 33.33 (33.33)		33 (40.74)	
Depression		34 56.67 (56.67)		36 (44.44)	
Anxiety		0 0.00 (0.00)		2 (2.47)	
OCD		6 10.00 (10.00)		9 (11.11)	
Borderline		0 0.00 (0.00)		1 (1.23)	
Carer ED Hx y/n (%yes)	20/47 (29.85)		18/71 (20.22)		C^2 = 1.92, *p* = .17

Note: SU: Service-user. (%) Indicates percentage of respondents by group. Contact hours is face to face. T: TAU: Treatment as usual. Compulsive Disorder. Carer ED Hx: Carer has history of eating difficulties. SPSS only gives Fishers's Exact Test p-value for 2x2 tables. OCD: Obsessive

Of the service-user group, 19.50% (*n* = 31) were in a relationship, and 14.47% (*n* = 23) were employed. Of the carers, 74.21% (*n* = 118) were in a relationship and 62.89% (*n* = 100) were employed. Overall, the sample had a high level of education, with 48.43% (*n* = 77) carers and 32.70% (*n* = 52) service-users having received university level equivalent or higher education, and 7.55% (*n* = 12) carers and 4.40% (*n* = 7) service-users having no qualifications. The majority of the sample were of White British ethnic origin (service-users, 88.70%; carers, 87.42). Most carers (83.02%, *n* = 132) and service-users (94.97%, *n* = 151) were female.

Carers were predominantly mothers (79.25%, *n* = 126), with the remaining sample of carers comprised of partners (13.84%, *n* = 22), fathers (3.15%, *n* = 5), siblings (1.89%, *n* = 3) and friends (1.89%, *n* = 3). Carers' mean age was 51.37 (*SD* = 9.87, range 22-76). Most carers and service-users lived together (71.24%, *n* = 109) and had a high level of contact; for roughly half (50.99%, *n* = 77) this was more than 21 hours each week. The number of caregivers taking part was one for 80 service-users (50.32%), two for 77 service-users (48.43%), and three for two service-users (1.26%). Some carers reported their own history of difficulties with eating (24.36%, *n* = 38).

Differences between intervention and treatment as usual groups, and between intervention and excluded (excluded/missing participants) groups were assessed using Pearson's chi^2, Fishers Exact Test, Independent samples t-test and Mann Whitney U test. Bonferroni's correction was not applied to the 2-tailed tests of difference as Type II errors were more of a concern than Type I errors when identifying possible confounding variables. The difference between service-user marital status of participants in the ECHO group compared to the TAU group was statistically significant c^2 (1, *n* = 154) = 4.30, *p* =.04, with a greater proportion of single service-users in the ECHO group. Additionally, there were significantly fewer carers involved in the intervention per service-user in the ECHO compared to the TAU group, c^2 (1, *n* = 159) = 3.51, *p* =.01. Service-user marital status and number of carers were therefore entered as covariates to all relevant models, but in no cases did this make a significant difference. All other differences in demographic variables between groups were found to be non-significant (*p* >.05), meaning there was no evidence for the introduction of bias through the exclusion of the 19 participants.

Descriptive Statistics

The means and standard deviations for the measures from included service-users and primary carers are presented in Table 7 by time-point and experimental condition. As might be expected, a general downwards trend in the means can be observed between baseline and 12 months (see Hibbs et al., 2015).

Correlations

The assumption that moderators are not significantly correlated with the dependent variable (Kraemer, Wilson, Fairburn, & Agras, 2002; Hayes, 2013) was checked pre-hoc using Pearson's bivariate correlations (Table 8). Results indicated it was acceptable to proceed with all planned moderation analyses.

Table 7

Values of Service-User and Carer Outcome Measures by Time-Point

		Timepoint							
		Baseline		Discharge		6 Months		12 Months	
Measure; mean (SD)	Max^{1}	TAU	ECHO	TAU	ECHO	TAU	ECHO	TAU	ECHO
Patient variables									
Distress, Anxiety and Stress Scale (DASS)	126	78.54^{a} (27.81)	77.66^{a} (28.00)	64.28 (33.17)	62.45 (30.10)	65.24 (32.86)	62.07 (30.97)	62.07^{b} (31.52)	61.27b (31.76)
Eating Disorder Examination Questionnaire (EDEQ)	6	4.15 (1.21)	4.33 (1.28)	3.26 (1.51)	3.46 (1.46)	3.57 (1.56)	3.35 (1.69)	3.39 (1.58)	3.29 (1.81)
Carer Variables									
Distress, Anxiety and Stress Scale (DASS)	126	34.85^{c} (28.95)	29.59^{d} (27.34)	36.53 (29.76)	33.65 (26.58)	35.46 (28.02)	30.15 (24.93)	32.17^{c} (30.80)	27.94^{e} (25.65)
Family Questionnaire (FQ)	60	48.55 (9.79)	48.94 (8.64)	48.12 (9.34)	45.86 (9.30)	47.56 (8.80)	45.26 (9.16)	45.28^{f} (9.64)	43.78^{f} (10.15)
Accommodation & Enabling Scale for Eating Disorders (AESED)	132	50.15 (25.35)	46.66 (21.27)	45.67 (25.55)	37.75 (20.86)	43.27 (25.55)	36.33 (23.01)	38.01 (26.43)	31.25 (21.09)
Eating Disorder Symptom Impact Scale (EDSIS)	96	43.03 (14.03)	41.53 (12.70)	38.78 (14.09)	32.09 (15.14)	36.55 (14.93)	29.75 (15.11)	34.84 (16.54)	29.37 (16.36)

Note: [1] Maximum score possible for scale, minimum score was 0 in all cases. TAU: Treatment as usual group. ECHO: Intervention. [a] Within the 'severe' range. [b] Within the 'moderate' range. [c] Within the 'mild' range. [d] At the top end of the 'normal' range ('mild' is 30) [e] Within the 'normal' range. [f] Remained above cut-off for 'high' expressed emotion (27). Verbal descriptions or cut-offs not available for remaining measures. High scores on all measures indicate greater presence of the factor measured.

Table 8

Pearson's Correlations between Outcome Variables, Experimental Group, and Proposed Moderating Variables

#		1	2	3	4	5	6	7	8	9	10
1	Group (ECHO/TAU)	-	-	-	-	-	-	-	-	-	-
Carer Variables^{a}											
2	Distress	-.019	-	-	-	-	-	-	-	-	-
3	Expressed emotion	-.259**	.354**	-	-	-	-	-	-	-	-
4	Accommodation	-.136	.269**	.405**	-	-	-	-	-	-	-
Patient variables^{a}											
5	Distress	.013	.137	.193	.116	-	-	-	-	-	-
6	ED symptoms	-.147	-.103	.240*	.107	.199	-	-	-	-	-
Moderator variables											
7	Carer ED history	-.095	.081	.104	.015	-.035	-.069	-	-	-	-
8	Patient BMI	.041	.080	.034	-.080	-.021	.043	-.003	-	-	-
9	Patient age	-.114	-.084	-.066	.146	.078	.004	.011	.014	-	-
10	Illness duration	-.058	-.033	-.088	.175	.016	-.078	.045	-.053	.778**	-
11	Number of carers	.142	-.135	.021	.051	-.059	-.151	.044	-.133	-.315**	-.206*

Note: ^{a}Time period is change over baseline to six-month post-discharge. *significant to <.05. ** significant to <.01

Moderation and Mediation Analysis

Moderation and mediation analyses and results are presented below, followed by a summary of results by hypothesis (Table 27).

Moderation and mediation of intervention outcomes.

H1. Moderators of intervention outcomes.

We assessed whether carer, service-user, illness and intervention factors moderated the intervention effects using linear regression examining the interaction between group (ECHO/TAU) and the proposed moderator as a predictor of outcome. The number of carers taking part in the intervention per service-user significantly moderated the reduction in expressed emotion (H1d.i) between baseline and six months (BCI [-11.764, -1.486]). When there was one carer per service-user, the relationship between group and reduction in expressed emotion was non-significant (*b* = -.048, BCI [-4.060, 3.094], *t* = -0.27, *p* =.79). With more than one carer per service-user, there was a significant positive relationship between ECHO and reduction in expressed emotion (*b* = -7.11, BCI [-10.789, -3.418], *t* = -0.38, *p* <.001), as shown in Tables 9a and 9b. Number of carers remained a significant moderator when controlling for service-user and carer relationship status, service-user age and illness duration (*b* =-7.29, BCI [-12.489, -2.081], *t* = -2.77, *p* <.01). Therefore, the intervention significantly reduced expressed emotion only for carers who attended with another of the service-user's carers. As seen in Table 10, number of carers per service-user did not moderate the effect of group on change in accommodation (H1d.ii, BCI [-20.405, 11.108]). The remaining moderators, namely service-user age, illness duration, BMI and carer history of eating difficulties were also non-significant in eight further analyses (Table 10), meaning no further moderators could be identified.

Table 9a

H1d.i: Results from a Regression Analysis. The Effect of Intervention on Reduction in Expressed Emotion Moderated by Number of Carers Involved in Intervention Per Service-User

Moderator	Outcome variable	Predictor		Coeff.	*SE*	*t*	*p*
Number of Carers	Expressed emotion[a]	Intercept		-2.05	1.20	-1.71	.09
		IV (Group; ECHO/TAU)	*a1*	-0.48	1.81	-0.27	.79
		M (# carers)	*a2*	3.83	1.85	2.07	.04
		Interaction (Group x # carers)	*a3*	-6.63	2.60	-2.55	.01
				R^2 = 0.12, F(3, 118) = 4.87, p = <.01			

Note: *n* = 122. M: Moderator variable.

Table 9b

H1d.i: Conditional Effect of Intervention on Reduction in Expressed Emotion at Values of the Moderator (Number of Carers Involved in Intervention Per Service-User)

Number of carers	Coeff.	*SE*	*t*	p	LBCI	UBCI
1	-0.48	1.81	-0.27	.79	-4.060	3.094
>1	-7.11	1.86	-0.38	<.001	-10.798	-3.418

Note: LBCI, UBCI: Lower and upper bootstrapped 95% confidence interval.

Table 10

H1: Moderators of the Effect of Group on Outcomes. Non-significant P-values and Bootstrapped Confidence Intervals for R^2 Change as a Result of the Moderator x Group Interactions

Hyp.#	Moderator (baseline)	Outcome variable^a	*p*	*95% BCI*	
				LL	UL
a.i	Patient age	ED Psychopathology	.52	-0.037	0.073
a.ii	Duration of illness	ED Psychopathology	.97	-0.068	0.071
b	Baseline BMI	ED Psychopathology	.56	-0.436	0.238
c.i	Carer history of ED	ED Psychopathology	.92	-1.483	1.650
c.ii		Accommodation	.43	-29.036	12.360
c.iii		Carer distress	.94	-20.580	22.173
d	Number of Carers	Accommodation	.51	-0.838	1.688
e.i	Accommodation	Carer distress	.80	-0.735	0.949
e.ii	Expressed emotion	Carer distress	.48	-0.412	0.197

Note: ^{a} Time period is change between baseline and 6 months post-discharge. Hyp. #: Hypothesis reference number. BCI: Bootstrapped confidence interval LL: Lower limit, UL: Upper limit.

H2. The indirect effect of ECHO on ED symptoms mediated by changes in interpersonal maintaining factors.

From a simple mediation analysis, group indirectly influenced change in ED symptoms through its effects on expressed emotion. As can be seen in Table 11 and Figure 10, carers in the ECHO (compared to TAU) group showed significantly greater baseline to six-month reduction of expressed emotion (*a* = -4.64, *p* <.01), and carers who had greater reduction in expressed emotion cared for service-users with greater reduction in ED symptoms (*b* = 0.04, *p* =.03). The BCI for the indirect effect (*ab* = -0.19) was entirely above zero (-0.450, -0.047). There was no evidence that group affected change in ED symptoms independent of its effect on expressed emotion (*c'* = -0.09, *p* =.77).

However, as seen in Table 12, the effect of group on change in ED symptoms was not mediated by accommodation (*ab* = -0.03, BCI [-0.263, 0.030]). There was also no significant direct effect of group on the ED (*p* =.45).

Table 11

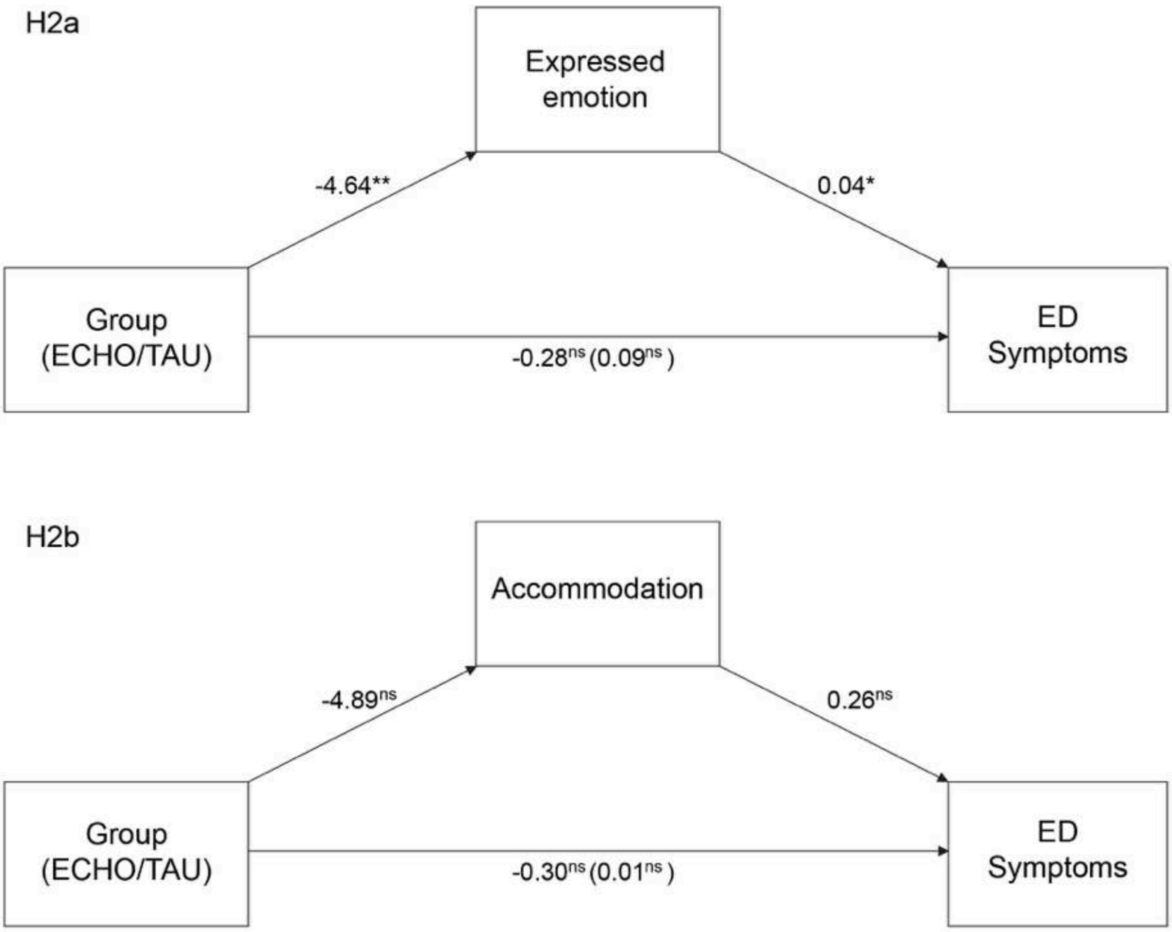
H2a: Results from Mediation Analysis. The Effect of Intervention on Reduction in ED Symptoms Mediated by Reduction in Expressed Emotion

		Consequent						
		M (Expressed emotion^{a})				DV (ED symptoms^{a})		
Antededent		Coeff.	*SE*	*p*		Coeff.	*SE*	*p*
IV (Group; ECHO/TAU)	*a*	-4.6387	1.56	<.01	*c′*	-0.09	0.29	.77
M (Expressed emotion^{a})		-	-	-	*b*	0.04	0.02	.03
Constant		0.03	1.09	.98		-0.50	0.18	<.01
		$R^2 = 0.31$ $F(1, 88) = 8.82, p < .01$				$R^2 = 0.06$ $F(2, 87) = 2.76, p = .07$		

	Unstandardised effects	*SE*	LBCI	UBCI
Total effect of group on carer distress	-0.28 *c*	0.30	-0.865	0.310
Total indirect effect of group on carer distress	-0.19 *ab*	1.90	-0.450	-0.047

Note: *n* = 90. ^{a}Change between baseline and six-month follow-up time-point. M: Mediator variable. LBCI, UBCI: Lower and upper bootstrapped 95% confidence interval.

Figure 10. H2: Outcomes of Two Mediation Analyses of Reduction in Expressed Emotion and Accommodation as Mediators of Indirect Effects of Intervention on Reduction in ED Symptoms



* $p \leq .05$. ** $p \leq .01$. *** $p \leq .001$. **** $p \leq .0001$.

Table 12

H2b: Results from Mediation Analysis. The Effect of Intervention on Reduction in ED Symptoms Mediated by Reduction in Accommodation

		Consequent						
		M (Accommodation^a)				DV (ED symptoms^a)		
Antededent		Coeff.	*SE*	*p*		Coeff.	*SE*	*p*
IV (Group; ECHO/TAU)	*a*	-4.89	4.48	.28	*c'*	0.01	0.01	.45
M (Accommodation^a)		-	-	-	*b*	-0.26	0.31	.40
Constant		-4.49	2.72	.10		-0.49	0.18	.01
		R^2 = 0.02				R^2 = 0.02		
		F(1, 79) = 1.19, *p* = .28				*F*(2, 78) =0.60 , *p* = .55		

	Unstandardised effects	*SE*	LBCI	UBCI
Total effect of group on carer distress	-0.30 *c*	0.32	-0.924	0.334
Total indirect effect of group on carer distress	-0.03 *ab*	0.07	-0.263	0.030

Note: *n* = 81. ^{a}Change between baseline and six-month follow-up time-point. M: Mediator variable. LBCI, UBCI: Lower and upper bootstrapped 95% confidence interval.

H3. The indirect effect of ECHO on carer outcomes mediated by changes in carer factors.

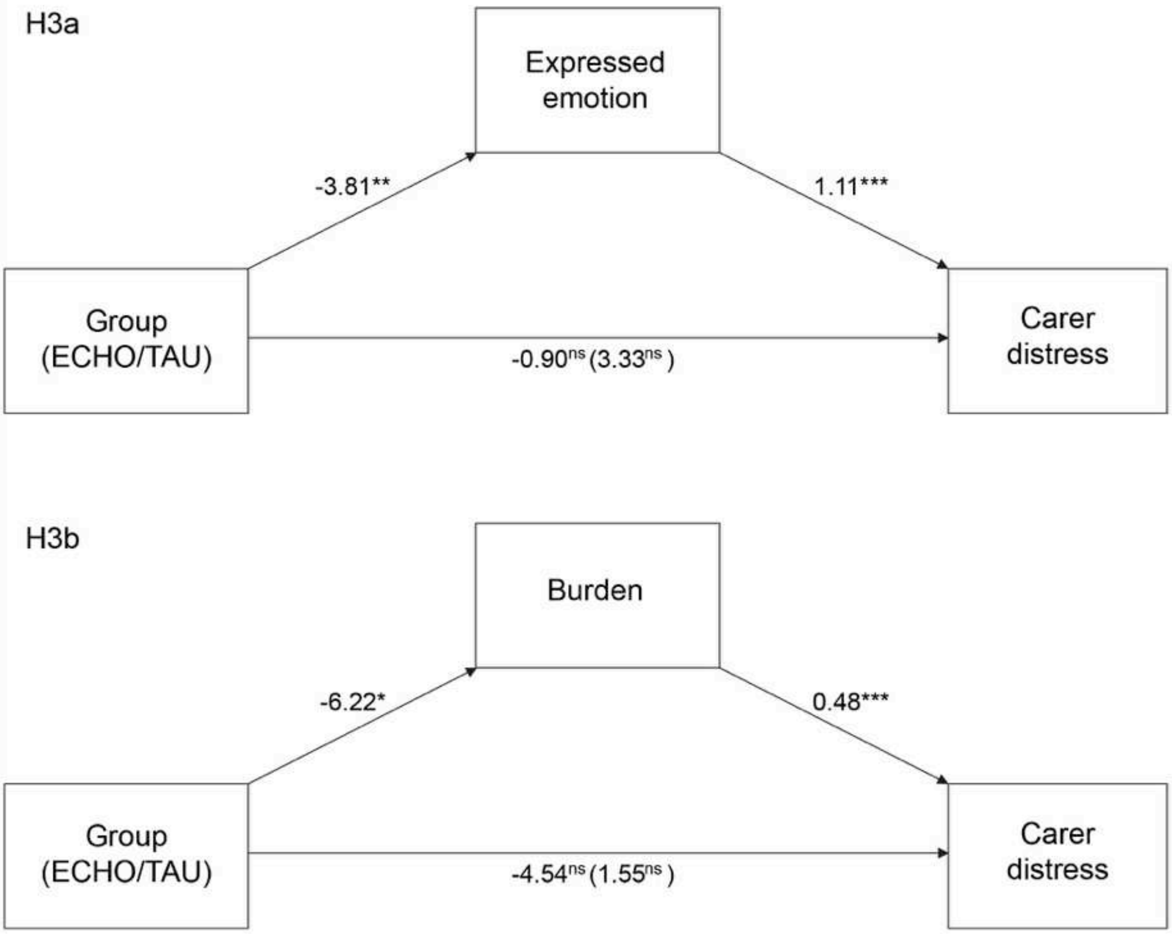
Seven analyses were undertaken to assess potential mediators of the relationships between intervention and carer outcomes.

Two mediation analyses (Figure 11) showed indirect effects of intervention on change in carer distress mediated by change in expressed emotion and burden. Tables 13 and 14, respectively, show that carers who received ECHO were more likely than those who received TAU to have larger reductions in expressed emotion (a = -3.81) and caregiving burden (a = -6.22), and carers with greater reduction in these outcomes were also more likely to show greater reductions in distress (b = 1.11 and 0.48, respectively). The significant indirect effects of ECHO compared to TAU through expressed emotion (ab = -4.22, BCI [-8.788, -1.085]) and burden (ab = -2.99, BCI [-6.559, -0.745]) were entirely above zero with effects of around medium size (K^2 = 0.09 and 0.07, respectively). In these analyses, there was no evidence that the intervention influenced distress independent of the indirect effects of expressed emotion (c' = -3.33, p =.41) and burden (c' = 1.55, p =.70).

As seen in Figure 12 and Tables 15 and 16, carers who received the intervention showed greater reductions in expressed emotion (a = -3.49, p =.02) and caregiving burden (a = -5.47, p =.02), and carers with greater reduction in these outcomes were more likely to show greater reductions in accommodation (b = 1.01, p <.0001, and 1.09, p <.001, respectively).

Therefore, the indirect effects of intervention on accommodation through expressed emotion (*ab* = -3.52, BCI [-7.350, -0.914], K^2 = .09) and burden (*ab* = -5.94. BCI [-12.009, -1.280], K^2 = .02) were significant. There was no evidence that the intervention influenced accommodation independent of the indirect effects of expressed emotion (*p* = .63) or burden (*p* =.48).

Figure 11. H3: Outcomes of Two Mediation Analyses of Reduction in Expressed Emotion and Burden as Mediators of Indirect Effects of Intervention on Reduction in Carer Distress



* p ≤.05. ** p ≤.01. *** p ≤.001. **** p ≤.0001.

Table 13

H3a: Results from Mediation Analysis. The Indirect Effect of Intervention on Reduction in Carer Distress Mediated by Reduction in Expressed Emotion

		Consequent						
		M (Expressed emotion[a])				DV (Carer distress[a])		
Antededent		Coeff.	*SE*	*p*		Coeff.	*SE*	*p*
IV (Group; ECHO/TAU)	*a*	-3.81	1.30	<.01	*c'*	3.33	4.02	.41
M (Expressed emotion[a])		-	-	-	*b*	1.11	0.30	<.001
Constant		0.31	0.93	.74		2.08	2.94	.48
		$R^2 = 0.07$				$R^2 = 0.13$		
		F(1, 120) = 8.55, *p* = .004				*F*(2, 119) =7.14 , *p* = .001		
			Unstandardised effects			*SE*	LBCI	UBCI
Total effect of group on carer distress			-0.90 *c*			3.97	-8.759	6.965
Total indirect effect of group on carer distress			-4.22 *ab*			1.90	-8.788	-1.085

Note: *n* = 122. [a]Change between baseline and six-month follow-up time-point. M: Mediator variable. LBCI, UBCI: Lower and upper bootstrapped 95% confidence interval.

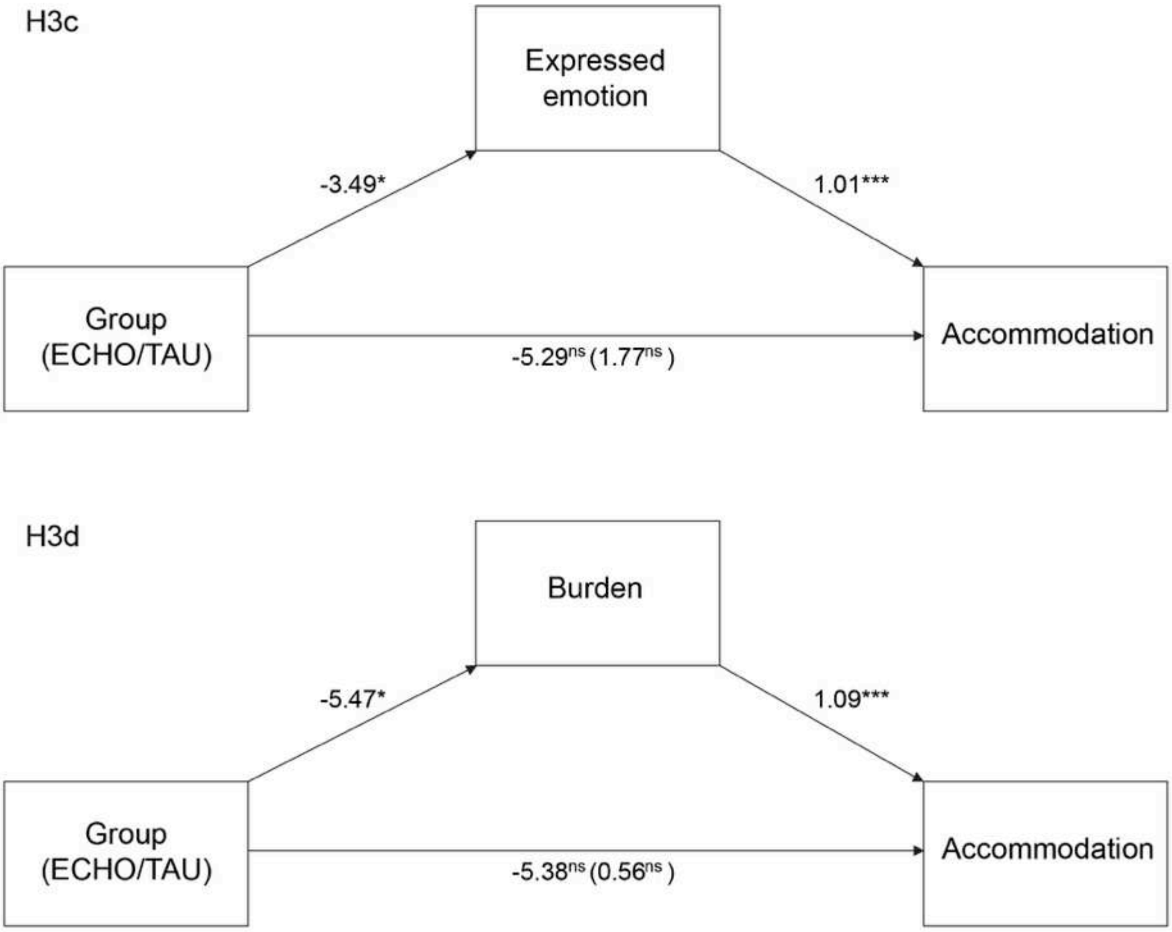
Table 14

H3b: Results from Mediation Analysis. The Indirect Effect of Intervention on Reduction in Carer Distress Mediated by Reduction in Burden

		Consequent						
		M (Burden^a)				DV (Carer distress^a)		
Antededent		Coeff.	*SE*	*p*		Coeff.	*SE*	*p*
IV (Group; ECHO/TAU)	*a*	-6.22	2.39	.01	*c′*	-1.55	3.98	.70
M (Burden^a)		-	-	-	*b*	0.48	0.14	<.001
Constant		-5.69	1.28	>.0001		7.04	2.89	.02
			$R^2 = 0.06$				$R^2 = 0.09$	
			F(1, 108) = 6.77, *p*= .01				*F*(2, 107) = 7.52, *p*= <.001	
			Unstandardised Effects			*SE*	LBCI	UBCI
Total effect of group on carer distress			-4.54 *c*			3.87	-12.206	3.124
Total indirect effect of group on carer distress			-2.99 *ab*			1.45	-6.559	-0.745

Note: *n* = 110. ^{a}Change between baseline and six-month follow-up time-point. M: Mediator variable. LBCI, UBCI: Lower and upper bootstrapped 95% confidence interval.

Figure 12. H3: Outcomes of Two Mediation Analyses of Reduction in Expressed Emotion and Burden as Mediators of Indirect Effects of Intervention on Reduction in Accommodation



* $p \leq .05$. ** $p \leq .01$. *** $p \leq .001$. **** $p \leq .0001$.

Table 15

H3c: Results from Mediation Analysis. The Indirect Effect of Intervention on Reduction in Accommodation Mediated by Reduction in Expressed Emotion

		Consequent						
		M (Expressed emotion[a])				DV (Accommodation[a])		
Antededent		Coeff.	*SE*	*p*		Coeff.	*SE*	*p*
IV (Group; ECHO/TAU)	*a*	-3.49	1.44	.02	*c'*	-1.77	3.64	.63
M (Expressed emotion[a])		-	-	-	*b*	1.01	0.22	<.0001
Constant		-0.64	1.01	.53		-4.13	2.14	.06
		$R^2 = 0.05$ $F(1,107) = 5.86, p= 0.02$				$R^2 = 0.17$ $F(2, 106) = 11.4, p <.0001$		

	Unstandardised effects	*SE*	LBCI	UBCI
Total effect of group on accommodation	-5.29 *c*	3.80	-12.826	2.252
Total indirect effect of group on accommodation	-3.52 *ab*	1.61	-7.350	-0.914

Note: *n* = 109. [a]Change between baseline and six-month follow-up time-point. M: Mediator variable. LBCI, UBCI: Lower and upper bootstrapped 95% confidence interval.

Table 16

H3d: Results from Mediation Analysis. The Indirect Effect of Intervention on Reduction in Accommodation Mediated by Reduction in Burden

		Consequent						
		M (Burden^a)				DV (Accomodating^a)		
Antededent		Coeff.	*SE*	*p*		Coeff.	*SE*	*p*
IV (Group; ECHO/TAU)	*a*	-5.47	2.54	0.02	*c'*	0.56	2.90	.85
M (Burden^a)		-	-	-	*b*	1.09	0.11	<.001
Constant		-6.49	1.32	<.001		1.50	2.13	.48
		$R^2 = 0.05$				$R^2 = 0.48$		
		F(1, 97) = 4.65, *p* = .02				F(2, 96) = 52.36, *p* <.001		

	Unstandardised Effects	*SE*	LBCI	UBCI
Total effect of group on accommodating	-5.38 *c*	3.97	-13.257	2.505
Total indirect effect of group on accommodating	-5.94 *ab*	2.70	-12.009	-1.280

Note: *n* = 99. ^{a}Change between baseline and six-month follow-up time-point. M: Mediator variable. LBCI, UBCI: Lower and upper bootstrapped 95% confidence interval.

Two further analyses showed the indirect effect of intervention on expressed emotion to be statistically mediated by burden (*ab* = -1.30, BCI [-2.630, -0.361], K^2 = .09) and the indirect effect of intervention on burden to be mediated by expressed emotion (*ab* = -2.55, BCI [-5.190, -0.941], K^2 = .10), with medium effect sizes. The intervention was associated with greater reductions in expressed emotion and burden, and changes in each of these variables were positively associated with changes in the other (see Tables 17 and 18, Figure 13). The significant total effect of intervention on expressed emotion (*c* = -4.22, *p* <.01) remained significant after the addition of the mediator burden (*c′* = -2.92, *p* =.03). However, the significant total effect of intervention on burden (*c* = -6.22, *p* =.01) became non-significant on addition of the mediator expressed emotion (*c′* = -3.67, *p* =.11), making this the only 'full' mediation found; all others reported are 'partial', meaning either the total effect was non-significant, or the direct effect did not lose significance on addition of the mediator.

Accommodation was not found to be a significant mediator of the relationship between intervention and change in distress (Table 19). Although the relationship between baseline to six-month change in accommodation and distress was significant (*b* = 1.63, *p* <.01), the relationship between intervention and accommodation was not (*a* = -5.27, *p* =.17), and there was no significant indirect effect (*ab* = -1.61, BCI [-5.384, 0.349]).

Figure 13. H3: Outcomes of Two Meditation Analyses Exploring Relationships between Expressed Emotion, Burden, and the Indirect Effects of Intervention

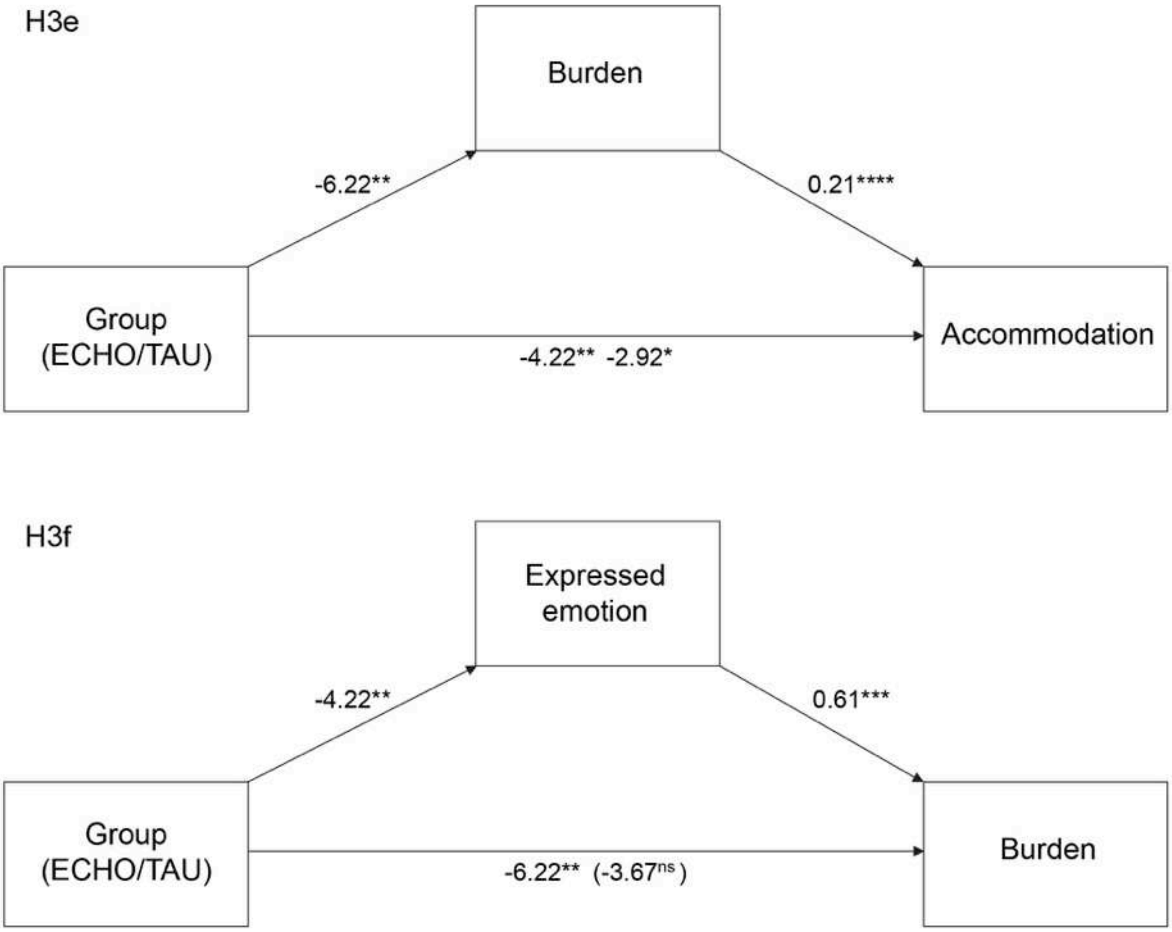


Figure 13. Two mediation analyses explored firstly whether greater reduction in expressed emotion mediated an effect of intervention on reduction in burden, and secondly whether greater reduction in burden mediated an indirect effect of reduction in expressed emotion.
* $p \leq .05$. ** $p \leq .01$. *** $p \leq .001$. **** $p \leq .0001$.

Table 17

H3e: Results from Mediation Analysis. The Indirect Effect of Intervention on Reduction in Expressed Emotion Mediated by Reduction in Burden

	Consequent							
		M (Burden[a])				DV (Expressed emotion[a])		
Antededent		Coeff.	*SE*	*p*		Coeff.	*SE*	*p*
IV (Group; ECHO/TAU)	*a*	-6.22	2.39	.01	*c'*	-2.92	1.32	.03
M (Burden[a])		-	-	-	*b*	0.21	0.05	<.0001
Constant		-5.68	1.28	<.0001		0.9	0.93	.33
		$R^2 = 0.06$				$R^2 = 0.20$		
		F(1, 108) = 6.77, *p*= .01				*F*(2, 107) = 15.52, *p*= <.0001		

	Unstandardised Effects	*SE*	LBCI	UBCI
Total effect of group on expressed emotion	-4.22 *c*	1.37	-6.929	-1.517
Total indirect effect of group on expressed emotion	-1.30 *ab*	0.56	-2.630	-0.361

Note: *n* = 110. [a] Change between baseline and six-month follow-up time-point. M: Mediator variable. LBCI, UBCI: Lower and upper bootstrapped 95% confidence interval.

Table 18

H3f: Results from Mediation Analysis. The Indirect Effect of Intervention on Reduction in Burden Mediated by Reduction in Expressed Emotion

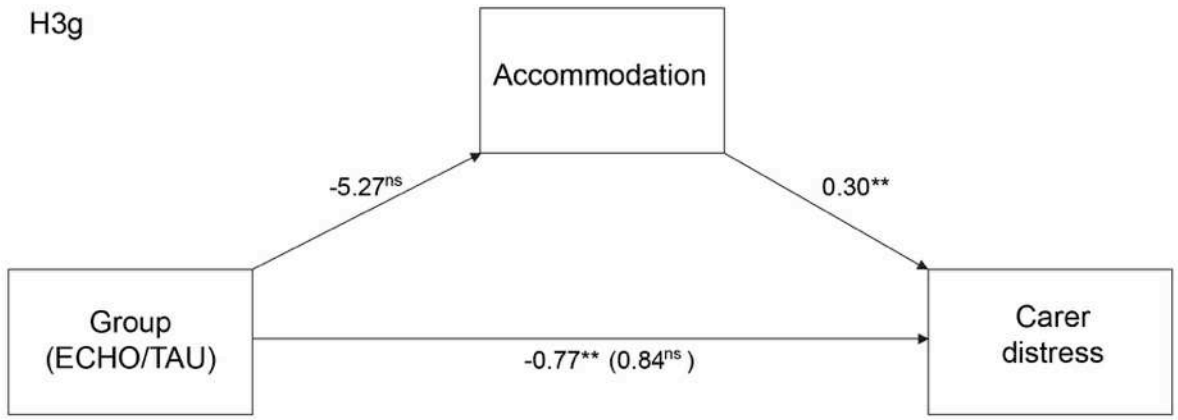
		Consequent						
		M (Expressed emotion[a])				DV (Burden[a])		
Antededent		Coeff.	*SE*	*p*		Coeff.	*SE*	*p*
IV (Group; ECHO/TAU)	*a*	-4.22	1.37	.002	*c'*	-3.67	2.28	.11
M (Expressed emotion[a])		-	-	-	*b*	0.61	0.13	<.001
Constant		-0.29	0.94	.76		-5.51	1.19	<.001
		$R^2 = 0.08$				$R^2 = 0.18$		
		$F(2, 108) = 9.57$, $p = .002$				$F(2, 107) = 14.51$, $p < .0001$		

	Unstandardised Effects	*SE*	LBCI	UBCI
Total effect of group on burden	-6.22 *c*	2.39	-10.958	-1.483
Total indirect effect of group on burden	-2.55 *ab*	1.03	-5.190	-0.941

Note: $n = 110$ [a]Change between baseline and six-month follow-up time-point. M: Mediator variable. LBCI, UBCI: Lower and upper bootstrapped 95% confidence interval.

[[[[

Figure 14. H3: Outcome of a Mediation Analysis of Reduction in Accommodation as a Mediator of an Indirect Effect of Intervention on Reduction in Carer Distress



* p ≤.05. ** p ≤.01. *** p ≤.001. **** p ≤.0001.

Table 19

H3g: Results from Mediation Analysis. The Indirect Effect of Intervention on Reduction in Carer Distress by Reduction in Accommodation

		Consequent						
		M (Accommodation^a)				DV (Carer distress^a)		
Antededent		Coeff.	*SE*	*p*		Coeff.	*SE*	*p*
IV (Group; ECHO/TAU)	*a*	-5.27	3.8	.17	*c'*	0.84	4.12	.84
M (Accommodation^a)		-	-	-	*b*	0.30	0.09	<.01
Constant		-4.77	2.32	.04		1.63	3.05	.59
		R^2 = 0.02 *F*(1, 107) = 1.93, *p* =.17				R^2 = 0.07 *F*(2, 106) = 5.47, p < .01		

	Unstandardised Effects	*SE*	LBCI	UBCI
Total effect of group on Carer distress	-0.77 *c*	4.18	-9.054	7.520
Total indirect effect of group on Carer distress	-1.61 *ab*	1.43	-5.384	0.349

Note: *n* = 109. ^{a}Change between baseline and six-month follow-up time-point. M: Mediator variable. LBCI, UBCI: Lower and upper bootstrapped 95% confidence interval.

H4. The indirect effect of ECHO on ED symptoms sequentially mediated by burden and expressed emotion.

A mediation analysis was conducted to examine whether the effect of intervention on ED symptoms was sequentially mediated by burden and expressed emotion. For clarity, only key associations are described (see Table 20 and Figure 15 for full details). Change in variables was between baseline and six-month follow-up. In contrast to the significant indirect effect of the intervention on reduction in ED symptoms mediated by reduction in expressed emotion (also described above, H2), the indirect effect of intervention on reduction in ED symptoms through burden did not reach significance ($a1b1$ = 1.01, BCI [-0.149, 0.178]). However, greater change in burden was associated with greater change in expressed emotion ($d21$ = 0.24, $p < .001$). The combined indirect effects through burden and expressed emotion amounted to a significant indirect effect of intervention on ED symptoms, sequentially mediated through reduction in burden then expressed emotion, leading to reduction in ED symptoms ($a1d21b2$ = 0.06, BCI [-0.205, -0.006]). There was no evidence that the intervention influenced ED symptoms independent of its effects on burden and expressed emotion (c' = -0.01, p =.76). Testing the model in all possible configurations with intervention as the independent variable (five, Appendix J) found no significant effects other than those already hypothesised and tested above.

Table 20

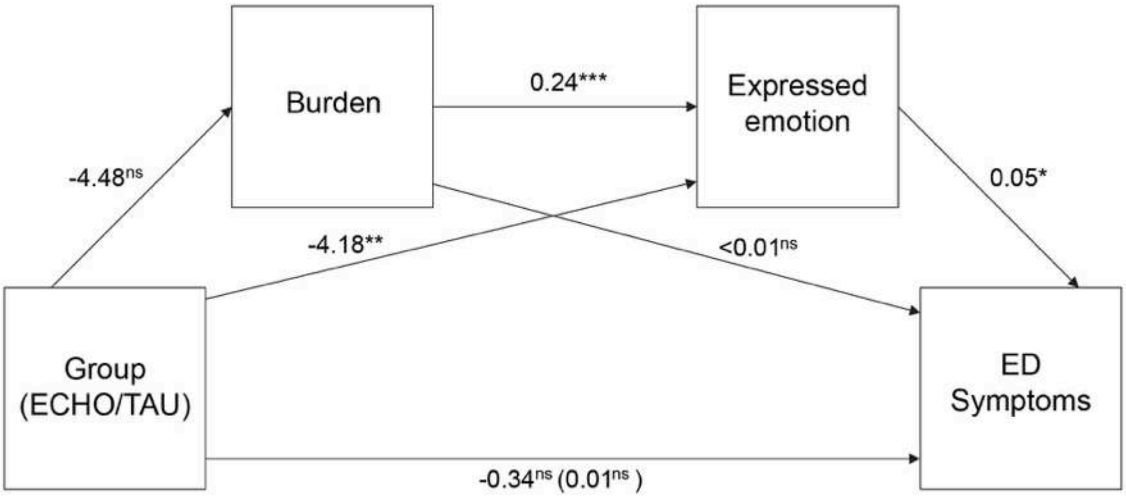
H4: The Indirect Effect of Intervention on Reduction in ED Symptoms Sequentially Mediated Through First Burden Then Expressed Emotion

		Consequent										
		M1 (Burden^{a})				M2 (Expressed emotion^{a})				DV (ED symptoms^{a})		
Antededent		Coeff.	*SE*	*p*		Coeff.	*SE*	*p*		Coeff.	*SE*	*p*
IV (Group)	*a1*	-4.82	2.72	.08	*a2*	-4.18	1.6	0.01	*c'*	-0.10	0.33	.76
M1 (Burden^{a})		-	-	-	*d21*	0.24	0.06	<.001	*b1*	<0.01	0.02	.98
M2 (Expressed emotion^{a})		-	-	-		-	-	-	*b2*	0.05	0.02	.03
Constant		-1.07	6.96	.88		4.03	4.17	0.34		-1.20	0.60	.05
		R2 = .06				R2 = .26				R2 = .10		
		F(3, 75) = 1.27, *p* = .29				F(4, 74) = 9.30, *p* <.001				F(5, 73) = 2.05, *p* = .08		

Observed effects		Unstandardised effect	*SE*	LBCI	UBCI
Total Effect of Group on ED	*c*	-0.34	0.33	-0.983	0.311
Indirect effect of Group on ED through Burden	*a1b1*	0.01	0.08	-0.149	0.178
Indirect effect of Group on ED through EE	*a2b2*	-0.22	0.13	-0.562	-0.031
Indirect effect of Group on ED through burden and EE	*a1d21b2*	-0.06	0.05	-0.208	-0.006

Note: *n* = 81. [a]Change between baseline and six-month follow-up time-point. M1, M2: Mediator variable 1 and 2. LBCI, UBCI: Lower and upper bootstrapped 95% confidence interval.

Figure 15. H4: Outcome of a Mediation Analysis. The Indirect Effect of Intervention on Reduction in ED Symptoms Sequentially Mediated by Reduction in Burden and Expressed Emotion



* p ≤.05. ** p ≤.01. *** p ≤.001. **** p ≤.0001.

Longitudinal investigation.

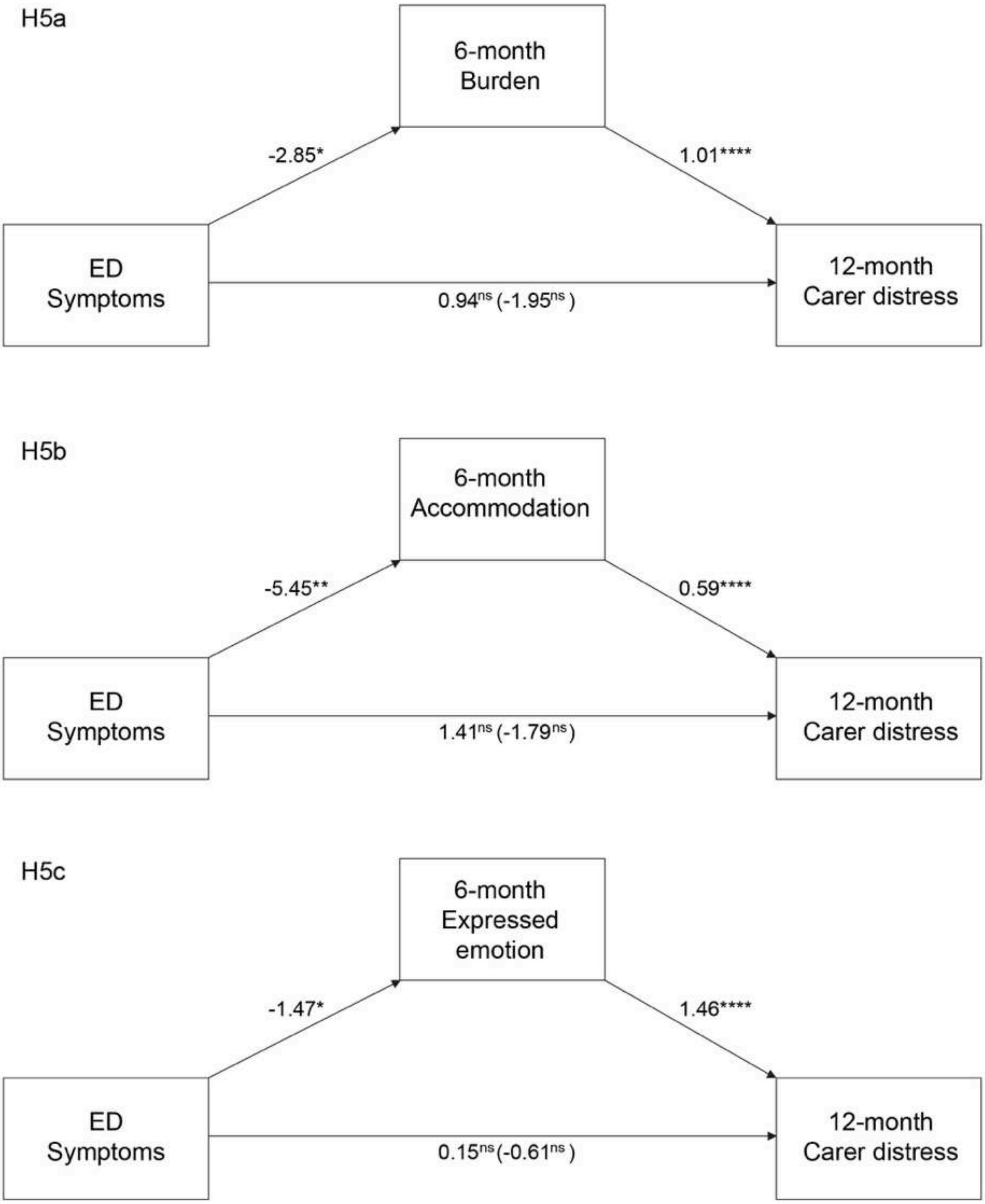
Experimental group (ECHO/TAU) did not moderate the relationships to be tested (H5-7) (Appendix N), indicating it was appropriate to test these mediational models with ECHO and TAU participants combined. As a check, group was included as a confounder in each analysis, but it made no material difference to significance.

H5. The indirect effect of ED symptoms on carer distress.

As can be seen in Tables 21, 22 and 23, and Figure 16, ED-specific burden, accommodation, and expressed emotion at six months mediated the relationship between ED symptoms at discharge and carer distress at 12 months: More severe ED symptoms predicted higher carer burden (*a* = 2.85, *p* =.02), accommodation (*a* = 5.45, *p* <.01), and expressed emotion (*a* = 1.47, *p* =.03), while higher carer distress was predicted by higher burden (*b* = 1.01, *p*

<.0001), accommodation (*b* = 0.59, *p* <.0001) and expressed emotion (*b* = 1.46, *p* <.0001). The three analyses showed positive indirect relationships between ED symptoms and carer distress positively mediated by burden (*ab* = 0.16, BCI [0.050, 0.300], K^2 =.17), accommodation (*ab* = 0.17, BCI [0.068, 0.302], K^2 =.18) and expressed emotion (*ab* = .011, BCI [0.027, 0.205], K^2 =.12), all with medium effect sizes. The indirect effects through burden (*ab* = 0.11, BCI [0.027, 0.253]), accommodation (*ab* = 0.12, BCI [0.018, 0.284]) and expressed emotion (*ab* = 0.08, BCI [0.016, 0.157]) remained significant when controlling for experimental group and carer distress at discharge. There was no evidence the ED symptoms directly predicted carer distress independent of its effects on burden (*p* =.70), accommodation (*p* =.35), or expressed emotion (*p* =.75).

Figure 16. H5: Outcomes of Three Longitudinal Mediation Analyses of the Indirect Effect of ED Symptoms on Carer Distress Mediated by Expressed Emotion, Burden and Accommodation



* p ≤.05. ** p ≤.01. *** p ≤.001. **** p ≤.0001.

Table 21

H5a: Results from Mediation Analysis. The Effect of ED Symptoms on Carer Distress Mediated by ED-Specific Burden

		Consequent						
		M (6m Burden)				DV (12m Carer distress)		
Antededent		Coeff.	*SE*	*p*		Coeff.	*SE*	*p*
IV (D ED symptoms)	*a*	2.85	1.17	.02	*c'*	-1.95	20.1	.36
M (6m Burden)		-	-	-	*b*	1.01	0.21	<.0001
Constant		21.8	4.3	<.0001		3.48	8.89	.70
		R2 = 0.08				R2 = 0.29		
		F(1, 79) = 5.96, *p* = .02				*F*(2, 78) = 12.22, *p* = .<.0001		

	Standardised effects	*SE*	LBCI	UBCI
Total effect of ED on carer distress	0.94 *c*	2.31	-3.648	5.532
Total indirect effect of ED on carer distress	0.16 *ab*	0.06	0.045	0.300

Note: *n* = 81. D: Patient discharge time. 6m: Six months post-patient discharge time. 12m: Twelve months post-patient discharge time. M: Mediator variable. LBCI, UBCI: Lower and upper bootstrapped 95% confidence interval.

Table 22

H5b: Results from Mediation Analysis. The Effect of ED Symptoms on Carer Distress Mediated by Accommodation

		Consequent						
		M (6m Accommodation)				DV (12m Carer distress)		
Antededent		Coeff.	*SE*	*p*		Coeff.	*SE*	*p*
IV (D ED symptoms)	*a*	5.45	1.79	<.01	*c'*	-1.79	1.91	.35
M (6m Accommodation)		-	-	-	*b*	0.59	0.12	<.0001
Constant		20.55	6.18	<.01		13.13	7.7	.09
		R2 = 0.10				R2 = 0.26		
		$F(1, 85) = 9.22$, $p = <.01$				$F(2, 84) = 12.96$, $p = <.0001$		

	Standardised effects	*SE*	LBCI	UBCI
Total effect of ED on carer distress	1.41 *c*	2.25	-3.053	5.875
Total indirect effect of ED on carer distress	0.17 *ab*	0.06	0.068	0.302

Note: $n = 87$. D: Patient discharge time. 6m: Six months post-patient discharge time. 12m: Twelve months post-patient discharge time. M: Mediator variable. LBCI, UBCI: Lower and upper bootstrapped 95% confidence interval.

Table 23

H5c: Results from Mediation Analysis. The Effect of ED Symptoms on Carer Distress Mediated by Expressed Emotion

		Consequent						
		M (6m Expressed emotion)				DV (12m Carer distress)		
Antededent		Coeff.	*SE*	*p*		Coeff.	*SE*	*p*
IV (D ED symptoms)	*a*	1.47	0.64	.03	*c'*	-0.61	1.90	0.75
M (Expressed emotion 6m)					*b*	1.46	0.31	<.0001
Constant		40.72	2.41	<.0001		-34.31	15.19	.03
		$R^2 = 0.06$				$R^2 = .021$		
		F(1, 89) = 5.22, *p* = .03				*F*(2, 88) = 11.73, *p* = <.0001		

	Standardised effects		*SE*	LBCI	UBCI
Total effect of group on carer distress	0.15	*c*	2.18	-2.814	5.875
Total indirect effect of group on carer distress	0.11	*ab*	0.04	0.027	0.205

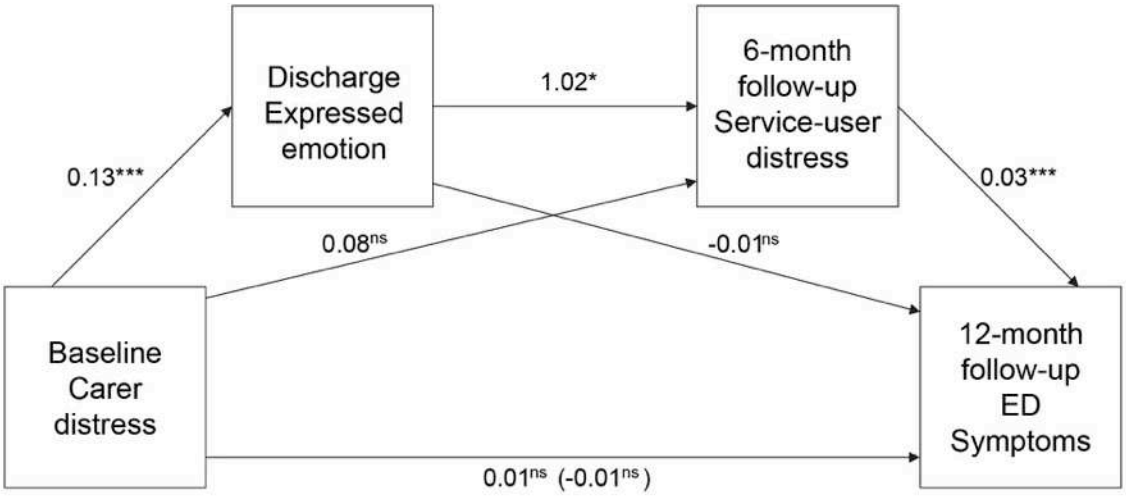
<u>Note:</u> *n* = 91. D: Patient discharge time. 6m: Six months post-patient discharge time. 12m: Twelve months post-patient discharge time. M: Mediator variable. LBCI, UBCI: Lower and upper bootstrapped 95% confidence interval.

H6. The indirect effect of carer distress on ED symptoms sequentially mediated by expressed emotion and service-user distress.

From the mediation analysis described in Figure 17, carer distress at baseline positively predicted eating disorder symptoms at 12 months, mediated sequentially with a positive indirect effect through first expressed emotion at discharge, then service-user distress at six months (*a1d21b2* = 0.07, BCI [0.016, .0163]). Interestingly, the indirect effects of carer distress on ED symptoms through expressed emotion (*a1b1* = -0.01, BCI [-0.091, 0.084]) or service-user distress (*a2b2* = 0.04, BCI [-0.104, 0.168]) individually were non-significant, only reaching significance when combined. This sequentially positively mediated indirect effect remained significant when controlling for baseline ED symptoms and experimental group (*a1d21b2* = 0.04, BCI [0.007, 0.108]), and when additionally controlling for carer eating history (*a1d21b2* = 0.04, BCI [0.009, 0.106]). There was no evidence that carer distress influenced ED symptoms independent of its effects on expressed emotion and service-user distress (*c'* = -0.01, *p* =.27). See Table 24 for full details.

The model was subsequently re-ordered to assess the hypothesised order for validity. With baseline expressed emotion as the independent variable and carer distress at discharge as the initial mediator (with service-user distress and ED symptoms unchanged), the indirect effects disappeared as predicted (*a1d21b2* = 0.027, BCI [-0.010, 0.086]).

Figure 17. H6: Outcome of a Longitudinal Mediation Analysis of the Indirect Effect of Carer Distress on ED Symptoms, Sequentially Mediated by Expressed Emotion and Service-User Distress



* p ≤.05. ** p ≤.01. *** p ≤.001. **** p ≤.0001.

H7. The indirect effect of carer distress on ED symptoms through accommodation.

Before controlling for ED symptoms at baseline, accommodation at six months was found to be a significant mediator of the relationship between carer distress at discharge and ED symptoms at 12 months (*ab* = .020, BCI [0.091, 0.337], see Table 25). However, when controlling for ED symptoms at discharge this lost significance (*ab* = 0.09, BCI [-0.032, 0.023], see Figure 18, Table 26). Although the relationship between carer distress and accommodation remained significant (*b* = 1.63, *p* <.01), the relationship between accommodation and ED symptoms did not (*a* = -5.27, *p* =.17), and there was no significant indirect effect (*ab* = -1.61, BCI [-5.384, 0.349]). There was also no evidence of a relationship between carer distress and ED symptoms before (*c* = 0.01, *p* =.26) or after (*c′* =<0.00, *p* =.79) accounting for ED symptoms at discharge.

Table 24

H6: The Indirect Effect of Carer Distress at Baseline on ED Symptoms at 12-Month Follow-Up Sequentially Mediated through Expressed Emotion at Discharge and Patient Distress at Six-Monthm Follow-Up, Controlling for ED Symptoms at Baseline

		Consequent										
		M1 (D Expressed emotion)				M2 (6m service-user distress)				DV (12m ED symptoms)		
Antededent		Coeff.	*SE*	*p*		Coeff.	*SE*	*p*		Coeff.	*SE*	*p*
IV (B Carer distress)	*a1*	0.13	0.03	<.001	*a2*	0.08	0.15	0.59	*c'*	-0.01	0.01	0.27
M1 (D Expressed emotion)		-	-	-	*d21*	1.02	0.46	0.03	*b1*	-0.01	0.02	0.8
M2 (SU 6m distress)		-	-	-		-	-	-	*b2*	0.03	0.01	<.001
Constant		42.93	1.83	<.001		14.56	20.63	0.48		1.44	0.89	0.11
		R2 = 0.15				R2 = 0.13				R2 = .32		
		F(2, 75) = 7.83, *p* <.001				F(3, 74) = 2.35, *p* = .08				F(4, 73) = 11.55, *p* <.001		

	Standardised Effects		*SE*	LBCI	UBCI
Total Effect of Group on ED	*c*	0.01	0.01	-0.002	0.025
Indirect effect of carer distress on ED through EE	*a1b1*	-0.01	0.05	-0.091	0.084
Indirect effect of carer distress on ED through patient distress	*a2b2*	0.04	0.07	-0.104	0.168
Indirect effect of carer distress on ED through EE and patient distress	*a1d21b2*	0.07	0.03	0.016	0.163

Note: *n = 81. B: Baseline. D: Discharge time-point. 6m: Six-month follow-up time-point. 12m: 12-month follow-up time-point. M1, M2: Mediator variable 1 and 2. LBCI, UBCI: Lower and upper bootstrapped 95% confidence interval.*

Table 25

H7: Results from a Mediation Analysis. The Effect of Carer Distress on ED Symptoms Mediated by Accommodation

		Consequent						
		M (6m Accommodation)				DV (12m ED symptoms)		
Antededent		Coeff.	*SE*	*p*		Coeff.	*SE*	*p*
IV (D Carer distress)	*a*	0.44	0.10	<.0001		<-0.01	0.01	.61
M (6m Accommodation)		-	-	-	*b*	0.03	0.01	<.01
Constant		26.09	3.84	.0001	*c'*	2.34	0.39	<.0001
		$R^2 = 0.24$				$R^2 = 0.14$		
		$F(1, 79) = 17.58$, $p = .0001$				$F(2, 78) = 5.35$, $p < .01$		

	Standardised effects	SE	LBCI	UBCI
Total effect of carer distress on ED symptoms	0.01 *c*	0.01	-0.009	0.023
Total indirect effect of group on ED symptoms	0.2 *ab*	0.06	0.09	0.337

Note: $n = 81$. D: Patient discharge time. 6m: Six months post-patient discharge time. 12m: Twelve months post-patient discharge time. M: Mediator variable. LBCI, UBCI: Lower and upper bootstrapped 95% confidence interval.

Table 26

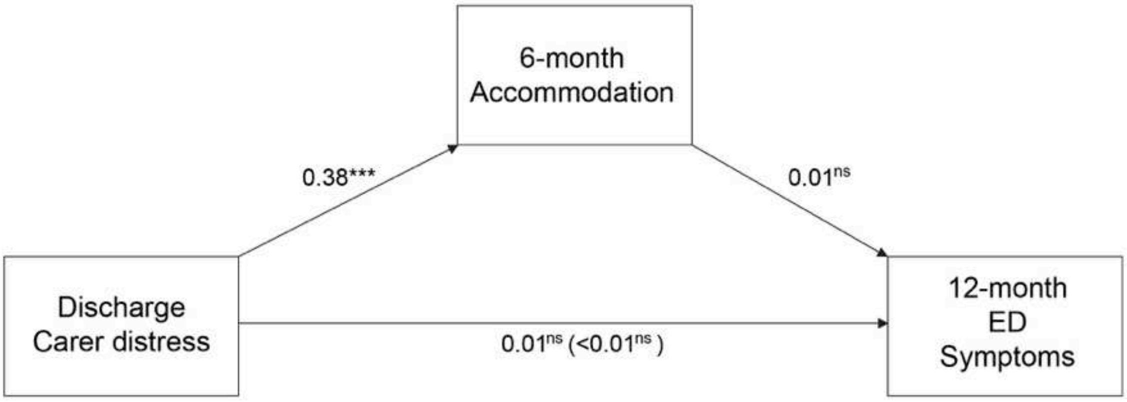
H7: Results from a Mediation Analysis. The Effect of Carer Distress on ED Symptoms Mediated by Accommodation, Controlling for ED Symptoms at Discharge

		Consequent						
		M (6m Accommodation)				DV (12m ED symptoms)		
Antededent		Coeff.	*SE*	*p*		Coeff.	*SE*	*p*
IV (Discharge Carer distress)	*a*	0.38	0.10	<.001	*c'*	<0.01	0.27	.79
M (Accommodation)		-	-	-	*b*	0.01	1.35	.18
Constant		12.55	6.48	0.06		0.54	0.33	.10
		$R^2 = 0.32$				$R^2 = 0.52$		
		$F(2, 69) = 17.70$, $p < .0001$				$F(3, 68) = 37.15$, $p < .0001$		

	Standardised effects	*SE*	LBCI	UBCI
Total effect of carer distress on ED symptoms	0.01 *c*	0.01	-0.004	0.015
Total indirect effect of group on ED symptoms	0.09 *ab*	0.07	-0.032	0.023

Note: *n* = 72. D: Patient discharge time. 6m: Six months post-patient discharge time. 12m: Twelve months post-patient discharge time. M: Mediator variable. LBCI, UBCI: Lower and upper bootstrapped 95% confidence interval.

Figure 18. H7: Outcome of a Longitudinal Mediation Analysis of the Indirect Effect of Carer Distress at Discharge on ED Symptoms at 12-Month Follow-Up, Mediated by Accommodation at Six-Month Follow-Up, Controlling for ED Symptoms at Discharge



* p ≤.05. ** p ≤.01. *** p ≤.001. **** p ≤.0001.

Summary of Support for Hypotheses

A summary of results, and how these relate to the hypotheses is presented in Table 27.

Table 27

Summary of Findings and Support for Hypotheses

Hyp. #	Hypothesis	Hypothesis supported?
Moderation of outcomes		
1a	Service-users with lower BMI will benefit less from the intervention in terms of ED symptom reduction	No
1b	Older service-users with longer illness duration will benefit less from the intervention in terms of ED symptom reduction.	No
1c	Cares of service-users with lower BMI will show greater reduction in carer distress.	No
1d	Where more than 1 carer for each service-user takes part in the intervention, those carers will benefit more from the intervention in terms of reductions in	
	i. Expressed emotion	Yes
	ii. Accommodation	No
1e	Carers with their own history of eating difficulties will benefit less from the intervention in terms of reductions in carer distress, accommodation and ED symptoms.	No
1f	Carers showing higher accommodation and expressed emotion at baseline will benefit more from the intervention in terms of reduction in carer distress.	No

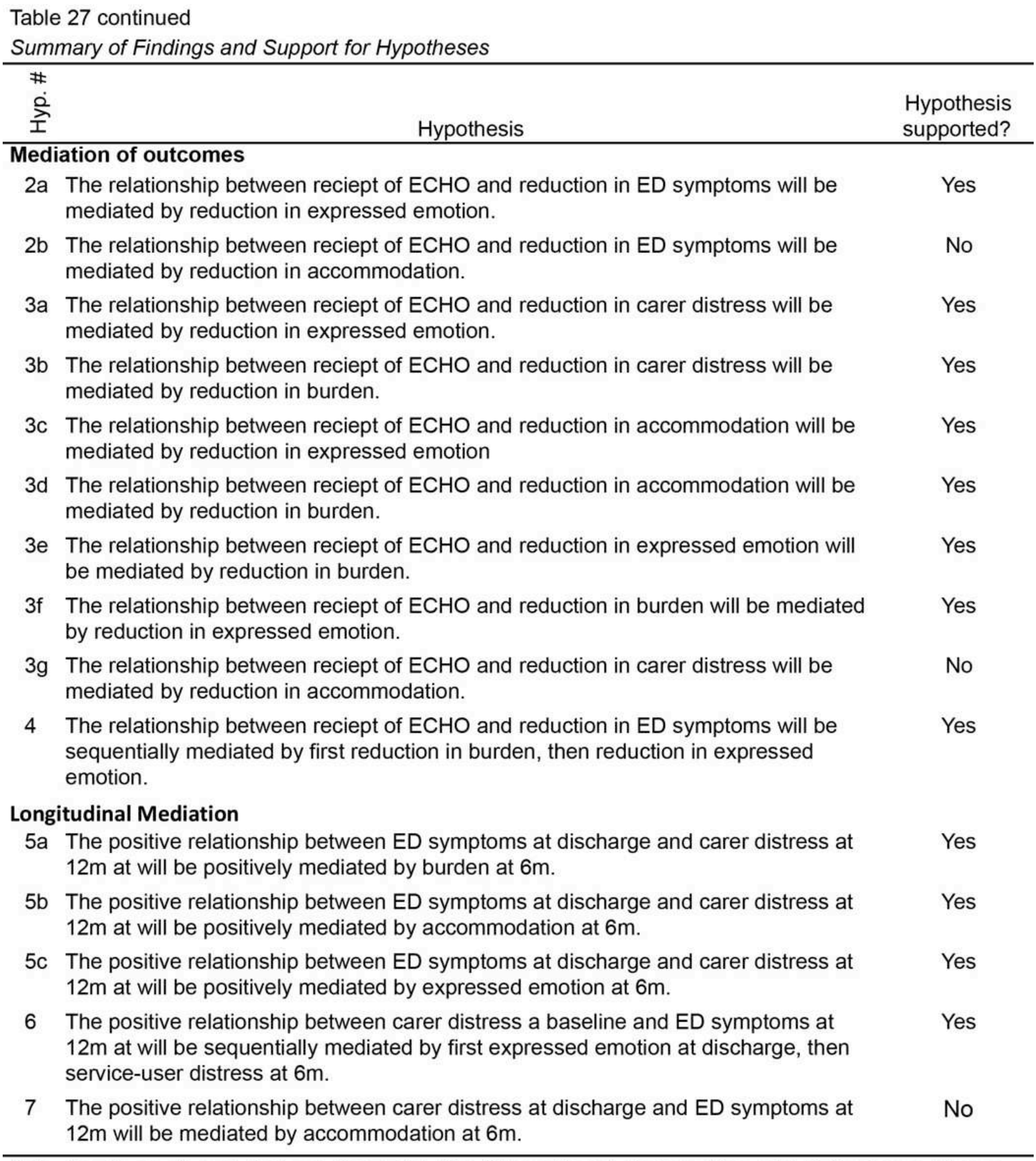


Table 27 continued

Summary of Findings and Support for Hypotheses

Hyp. #	Hypothesis	Hypothesis supported?
Mediation of outcomes		
2a	The relationship between reciept of ECHO and reduction in ED symptoms will be mediated by reduction in expressed emotion.	Yes
2b	The relationship between reciept of ECHO and reduction in ED symptoms will be mediated by reduction in accommodation.	No
3a	The relationship between reciept of ECHO and reduction in carer distress will be mediated by reduction in expressed emotion.	Yes
3b	The relationship between reciept of ECHO and reduction in carer distress will be mediated by reduction in burden.	Yes
3c	The relationship between reciept of ECHO and reduction in accommodation will be mediated by reduction in expressed emotion	Yes
3d	The relationship between reciept of ECHO and reduction in accommodation will be mediated by reduction in burden.	Yes
3e	The relationship between reciept of ECHO and reduction in expressed emotion will be mediated by reduction in burden.	Yes
3f	The relationship between reciept of ECHO and reduction in burden will be mediated by reduction in expressed emotion.	Yes
3g	The relationship between reciept of ECHO and reduction in carer distress will be mediated by reduction in accommodation.	No
4	The relationship between reciept of ECHO and reduction in ED symptoms will be sequentially mediated by first reduction in burden, then reduction in expressed emotion.	Yes
Longitudinal Mediation		
5a	The positive relationship between ED symptoms at discharge and carer distress at 12m at will be positively mediated by burden at 6m.	Yes
5b	The positive relationship between ED symptoms at discharge and carer distress at 12m at will be positively mediated by accommodation at 6m.	Yes
5c	The positive relationship between ED symptoms at discharge and carer distress at 12m at will be positively mediated by expressed emotion at 6m.	Yes
6	The positive relationship between carer distress a baseline and ED symptoms at 12m at will be sequentially mediated by first expressed emotion at discharge, then service-user distress at 6m.	Yes
7	The positive relationship between carer distress at discharge and ED symptoms at 12m will be mediated by accommodation at 6m.	No

Note: 6m: Six-month post-discharge time-point. 12m: 12-month post-discharge timepoint. B-6m change: Change between baseline and 6-months follow-up timepoints.

Discussion

The aims of this study were to test possible moderators and mediators of the relationships between receipt of a carers' skills intervention and reduction of outcome variables, and to examine the ways in which carer distress and proposed interpersonal maintaining factors may be associated with eating disorder symptoms. Following presentation of key overall findings, these two related aims will be considered in turn, before discussion of limitations and implications.

Summary and Implications of Key Findings

The key findings of this study are as follows. Firstly, greater reduction in expressed emotion following intervention only occurred for the group of carers who took part with another of the service users' carers, not for carers who took part alone. This indicates that it would be useful for carers to attend with another carer of the same service-user, where possible. Although a non-significant direct effect of intervention on carer distress and accommodation has been reported (Hibbs et al., 2015a), the findings of the present study suggest that intervention was associated with greater reductions in both carer distress and accommodation, but that this was indirectly, through greater reductions in burden and expressed emotion. Therefore, the current findings increase the known efficacy of the intervention, which is potentially valuable for supporting future funding. The greater reductions in burden and expressed emotion following intervention also mediated the greater reduction in ED symptoms; it appears that the intervention reduced burden, which reduced expressed emotion, which in turn reduced ED symptoms. This highlights the importance of interventions targeting carer burden and expressed emotion as these appear to be potentially key processes in the amelioration of clinically relevant problems for both service user (ED symptoms) and carer (distress). Results from the longitudinal analysis are consistent with the hypothesis that more severe ED symptoms lead to greater accommodation, burden and expressed emotion, which in turn lead to greater carer distress over time. This offers support and clarification to the Cognitive Interpersonal Model (Treasure & Schmidt, 2013) and Model of Carer Coping (Treasure & Nazar, 2016), both of which are underpinned by a hypothesised link between ED symptoms and carer distress that is otherwise poorly supported by the literature (that predominantly addresses only direct associations, e.g. Goddard et al., 2013; Kyriacou, Treasure, & Schmidt, 2008; Rhind et al., 2016). Finally, results were consistent with the hypothesis that carer distress at baseline was positively and indirectly associated with ED symptom severity following intervention, through first expressed emotion, then service-user distress. These results indicate that carers who were less distressed were likely to show less expressed emotion, which was therefore less distressing for the person with the eating disorder, which supported greater reductions in ED symptoms following hospital treatment. This is the first time that carer distress has been statistically implicated as a predictor of eating disorder symptoms, supporting the case for improved services aimed at reducing carer distress. Discussion of results in greater detail is presented below.

Moderators and Mediators of Intervention Outcome

The hypothesised moderators of the effect of intervention on outcomes (ED symptoms, carer distress, expressed emotion and accommodation) were tested in 10 analyses. Nine were non-significant. This is in keeping with Grover et al. (2011) and Rhodes, Baillee, Brown and Madden (2008) who similarly struggled to identify moderators of carers' ED intervention outcomes. To understand the lack of moderating action by variables found to predict outcome in previous studies, it may be helpful to consider the symmetry in moderation (Hayes, 2013) whereby moderators can equally be conceived as the independent variable, with the intervention not observed to alter relationships between these variables and outcome. This study's non-replication of Goddard et al.'s (2011) pre-post study finding of baseline accommodation and expressed emotion as moderators of change in carer distress may be due to design differences and suggest these variables may predict longitudinal change in distress, but that intervention does not moderate this relationship.

The exception was finding that the number of carers taking part in the intervention statistically moderated the relationship between intervention and reduction in expressed emotion, consistent with the possibility that the intervention only reduced expressed emotion when more than one carer took part. This might be due to increased opportunity for carers to enhance their learning through sharing the experience; or to the conceivably higher level of support carers received in their role when more than one attended. Raenker et al. (2013), Kyriacou, Treasure and Schmidt (2008), and Coomber and King (2012) reported that greater social support predicted lower carer distress, which could hypothetically lead to reduction in emotionally driven behaviours. However, the number of carers involved did not moderate change in accommodation, which, by this rationale, might be similarly impacted. As expressed emotion was more strongly associated with intervention than accommodation was (Hibbs et al., 2015a), power of the moderation analysis may have been insufficient to detect a smaller effect.

This moderator was interesting in light of the subsequent finding that expressed emotion statistically mediated an indirect relationship between receiving the intervention and reduction in ED symptoms at 6-month follow-up compared to baseline. As high expressed emotion predicts psychiatric relapse (Hooley, 2007), it follows that lowered expressed emotion would predict improved outcomes following hospitalisation. This finding, which is consistent with the hypothesis that the intervention was effective at reducing expressed emotion which in turn influenced improvement in ED symptoms, provided support for expressed emotion as an interpersonal maintaining factor (Treasure & Schmidt, 2013).

In contrast to expressed emotion, accommodation was not found to be a statistical mediator of the relationships between intervention and ED symptoms, suggesting it may not help to explain how or why the intervention predicted reduced ED symptoms (Hibbs et al., 2015a).

Next, the question of whether changes in some carer outcomes may mediate changes in others was addressed. Greater reductions in expressed emotion and burden in the ECHO group (compared to TAU) statistically mediated greater reductions over the same time in carer distress and accommodation. Hibbs et al. (2015a), who did not investigate mediators of outcome, reported failure of the intervention to reduce carer distress and accommodation. The present findings from mediation analysis contrast with this, and are consistent with the hypothesis that intervention did reduce these outcomes, but that this was indirectly, through the reduction of expressed emotion and burden. In line with the interpersonal element of the Cognitive Interpersonal Maintenance Model (Treasure & Schmidt, 2013) upon which the intervention was based, reductions in expressed emotion and burden may subsequently enable further changes for carers through their role in cycles which either help maintain, or alleviate difficulties.

Further exploration of these two variables found that greater reduction in expressed emotion statistically mediated the relationship between intervention and greater reduction in burden, while greater reduction in burden statistically mediated the relationship between intervention and greater reduction in expressed emotion. It could be hypothesised that this represents a virtuous cycle of reduction in burden allowing reduction in expressed emotion, allowing further reduction in burden, etcetera. Additionally, burden and expressed emotion statistically mediated a sequential indirect relationship between the intervention and reduction in ED symptoms. This was consistent with the hypothesis that ECHO reduced burden, which subsequently enabled carers to benefit more from the intervention in terms of reduction of expressed emotion, cascading to ultimately support greater reduction in ED symptoms. The relative strength of burden and expressed emotion across all these analyses may be because the intervention primarily affected these variables, and/or indicate the importance of burden and expressed emotion in maintenance of problematic carer responses and the ED.

This study found the association between intervention and greater reduction in carer distress not to be mediated by greater reduction in accommodation. This appears to be in contrast to Goddard et al.’s (2011) finding that accommodation mediated change in carer distress following intervention. However, this may relate to study design; Goddard et al.’s (2011) pre-post design precluded examination of accommodation as a mediator of differences between experimental group. Therefore, it is possible these findings may have related to mediation of changes in carer distress over time that were not related to intervention.

Longitudinal Investigation of Carer Distress

Addressing the second aim using longitudinal models, higher burden, expressed emotion and accommodation at six months were found to each statistically mediate indirect relationships between higher ED symptoms at discharge and higher carer distress at 12-month follow-up. This remained significant when controlling for carer distress at discharge, and ED symptoms did not predict carer distress independently of these indirect effects. These findings are consistent with models in which the impact of the symptoms on carers are mediated by carers' emotional, interpersonal and behavioural reactions to the ED (Kyriacou, Treasure, & Schmidt, 2008; Szmukler et al., 1996; Winn et al., 2007). Suggesting a similar process, Rhind et al. (2016) reported that accommodation, expressed emotion and carers' skills mediated the relationship between time spent caregiving and carer distress.

Analysis of the second longitudinal model suggested a significant positive indirect relationship between carer distress at baseline and ED symptoms at 12-month follow-up that was statistically mediated by first expressed emotion at discharge then service-user distress at six-month follow-up, including when controlling for baseline ED symptoms. This is consistent with the hypotheses that higher carer distress could lead to higher carer expressed emotion (Treasure & Schmidt, 2013), which could negatively impact service-user distress, and in turn negatively impact their ED symptoms (Goddard et al., 2013c). These findings are consistent with the Cognitive Interpersonal Maintenance Model (Treasure & Schmidt, 2013).

The finding that accommodation at six months statistically mediated the relationship between carer distress at discharge and ED symptoms at 12-month follow-up only when ED symptoms at discharge were not controlled for could have been due to loss of power.

However, it seems more likely that the former was instead indicative of covariance between ED symptoms and accommodation at discharge, rather than showing evidence of mediation. This finding, like those of Goddard et al. (2013c) and Kyriacou, Treasure and Schmidt's (2009), failed to support the role of accommodation suggested by the Interpersonal Maintenance Model (Treasure & Schmidt, 2013). This is in contrast to the significant effect of expressed emotion, conceptualised here as a predictor of both carer and service-user distress and as a consequence of carer distress. Considering expressed emotion as a relational interaction between carer and service-user factors (Hooley & Campbell, 2002), and accommodating and enabling as more distinctly carer behaviours, the latter in isolation may be insufficient to produce change in ED. Alternatively, non-significant findings regarding accommodation might indicate the recently developed accommodation measure (Sepulveda, Kyriacou, & Treasure, 2009) may benefit from development, in contrast to the extremely well-validated expressed emotion measure (e.g. Sepulveda et al., 2014b).

A model consistent with the present findings is presented in Figure 19. With the exception of a link between illness factors and carer factors, and the interpolation of service-user distress between expressed emotion and ED symptoms, this model is consistent with elements of both the Model of Carer Coping (Treasure & Nazar, 2016), and the interpersonal element of the Cognitive Interpersonal Maintenance Model (Treasure & Schmidt, 2013). Figure 19 therefore represents a development of how these two models may combine to contribute to understanding how the ED effects carer distress and how this in turn contributes to maintenance of the ED. Although this is perhaps most easily conceptualised as a vicious cycle, it is not inconsistent with this study's findings regarding beneficial changes associated with intervention, where such a process may occur as a virtuous cycle.

Figure 19. Maintenance Cycle Involving Carer and Patient Factors

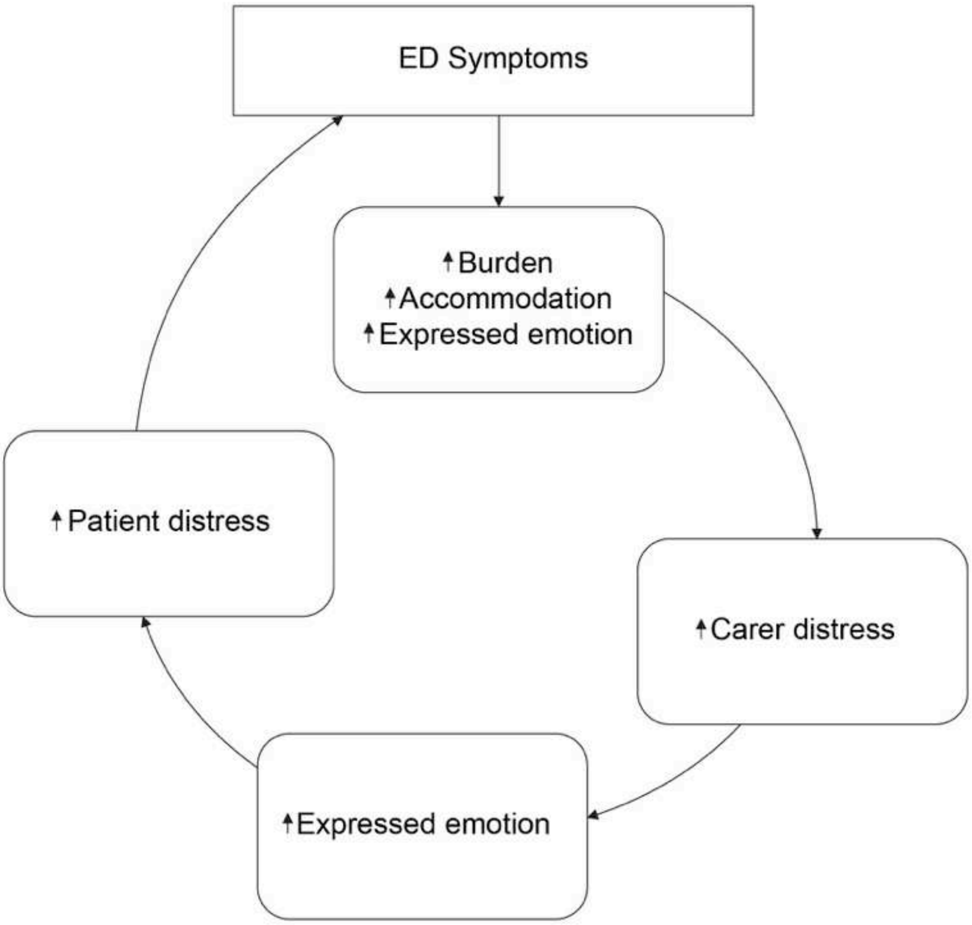


Figure 19. In this model, the eating disorder symptoms impact upon carer distress through the ways in which these become personally relevant for the carer, measured in terms of ED-specific burden, accommodation and enabling behaviours, and expressed emotion. Carer distress then further increases expressed emotion, which increases patient distress, which in turn supports maintenance of ED symptoms and feeds back into the cycle.

Limitations

The study has a number of limitations which are described in turn. Generalisability of the findings is limited as the sample represented only service-users with severe AN. All of the participants were admitted for hospital treatment, thereby receiving more intensive intervention than recommended for most people with EDs (NICE, 2004), and conceivably at a time of unusually high stress for the family. Self-report measures, particularly of ED symptoms, may have reduced validity of observations. It is also noteworthy that this study did not investigate positive aspects of caregiving, which may play important roles as protective factors (Sepulveda et al., 2012a). As most primary carers were mothers, fathers and other secondary carers were not well-represented. Issues related to the high proportions of missing data, which was not missing at random, may have introduced bias. For example, primary caregivers not adhering to ECHO was predictive of loss-to-follow up (Hibbs et al., 2015a) and those with no ECHO adherence were excluded from this study. This raises the question of why some ECHO-allocated carers did not complete the intervention or questionnaires, and whether missing data meaningfully reduced representativeness and validity of findings. It would therefore be useful for these analyses to be replicated in a study with lower attrition rates.

Although choice of moderators was based on existing literature, it necessarily remained reasonably speculative in line with Grover et al. (2011), due to the dearth of previously identified moderators. While choice of mediators was more confidently grounded in existing theoretical and empirical literature, this, too, is a relatively novel field. Together with the number of comparisons made, this may have inflated the possibility of Type I errors. In particular, the single significant finding of the moderation analyses requires replication to increase confidence that it is not merely an artefact of data-mining.

A strength of mediation analysis is that, if perfectly employed, it can indicate causality (Hayes, 2013). However, perfect design is challenging, and was not achieved by this study. Ideally, the design would ensure the independent variable was not influenced by the mediator or outcome variables. Randomisation to experimental group was valuable in this aim, however, for analyses in which group was the independent variable, data missing post-randomisation prevented this condition being met. In contrast, the design of the longitudinal mediation analyses provided assurance that later variables did not influence those measured before them. As with any correlational design however, results could be caused by latent variables, including those hypothesised as important, such as coping (Treasure & Nazar, 2016), but not tested. However, several variables conceived as likely confounders were observed not to meaningfully alter results, affording additional confidence in findings. The longitudinal models, for which it was unfeasible to test all configurations of time-point and order of measurement, are not necessarily the only valid representation of the data (see research implications). However, confidence in the supported hypotheses was increased by non-significant findings for the models in the alternative configurations tested, which were conceived as the most theoretically likely alternatives. Therefore, while this study's design made it relatively well-equipped to suggest possible processes involved, findings are proposed as suggestions for future testing and exploration, rather than as evidence of cause and effect.

Research Implications

Ideally, the research hypotheses would be tested longitudinally with Structural Equation Modelling (SEM). However, SEM is highly sensitive to missing data, meaning such a project would represent a significant challenge, particularly in light of high attrition rates in this area (e.g. Coomber & King, 2013). The findings of this study would nevertheless benefit from replication with other methodologies that may be more achievable, and with a wider variety of participants.

Murray, Loeb and Le Grange (2015) suggested that family-based therapy outcomes may be mediated by service-user's anxiety reduction. Further to the findings of this study regarding service-user distress as a mediator between expressed emotion and ED symptoms, it would be useful to test whether service-user anxiety, specifically, is similarly implicated.

Despite being centrally theoretically implicated in maintenance of EDs, accommodation has sometimes failed to gain empirical support (Goddard et al., 2013c; Sepulveda, Kyriacou, & Treasure, 2009). Exploration of hypothesised associated factors such as psychological control (Goddard et al., 2013c), anxiety and frustration (Treasure & Schmidt, 2013), may prove useful.

Clinical Implications

The results of this study may indicate that addressing expressed emotion and burden, where change might be more accessible, could indirectly improve ED symptoms and carer distress and accommodation.

The finding that reduction in expressed emotion statistically mediated the relationship between the intervention and reduction in ED symptoms, and that reduced expressed emotion was only associated with the intervention when more than one carer per service-user attended, indicate that encouraging multiple carers to attend for each service-user may increase efficacy. This is in line with anecdotal reports from clinicians administering the intervention. Adding peer-support may be helpful (Leggatt, 2007), especially for carers taking part in the intervention alone, for example through online moderated forums (see Binford, Le Grange, Moessner, & Bauer, 2013).

Our support for the hypothesis that the relationship between ED symptoms and carer distress would be mediated by factors alterable through intervention, and that ECHO appears to have had positive repercussions throughout service-user and carer factors, encourages optimism. This is especially considering the noted drive of carers to help their loved one (Treasure, Schmidt, & Macdonald, 2009), and the opportunity to engage and support carers when the cared-for declines treatment.

This study's findings, alongside others (e.g. Hibbs et al., 2015a; Magill et al., 2015) imply that carers can, and do, have a positive impact on the outcome of their cared-for's ED, with the ECHO intervention assisting them to do this. The present findings being consistent with the hypothesis that carer distress, burden and expressed emotion play a key role in maintenance and are alterable through intervention supports an argument for greater professional support for carers including through interventions such as ECHO.

Conclusion

This was the first study to test moderators and mediators of carer and cared-for outcomes from a large-scale carers skill-sharing RCT. Overall, results provided support for the interpersonal aspect of the Cognitive Interpersonal Maintenance Model (Treasure & Schmidt, 2013), the Model of Carer Coping (Treasure & Nazar, 2016), and the effectiveness of the ECHO intervention (Hibbs et al., 2015a), including for reducing carer distress and accommodation. This study's results suggested the importance of burden and expressed emotion in statistically mediating intervention outcomes, including ED symptoms. This was also the first study to longitudinally examine mediators of the relationships between ED symptoms and carer distress, and between carer distress and ED symptoms. A model summarising findings has been proposed, suggesting an indirect relationship between ED symptoms and carer distress mediated by burden, accommodation and expressed emotion; and an indirect relationship between carer distress and ED symptoms sequentially mediated by expressed emotion and service-user distress. There is therefore a strong argument for the utility and importance of interventions, such as ECHO, addressing carer distress and other responses to the ED.

References

Anastasiadou, D., Medina-Pradas, C., Sepulveda, A. R., & Treasure, J. (2014). A systematic review of family caregiving in eating disorders. *Eating behaviors*, *15*(3), 464-477. doi:10.1016/j.eatbeh.2014.06.001

Arcelus, J., Mitchell, A. J., Wales, J., & Nielsen, S. (2011). Mortality rates in patients with Anorexia Nervosa and other eating disorders: a meta-analysis of 36 studies. *Archives of General Psychiatry*, *68*(7), 724-731. doi:10.1001/archgenpsychiatry.2011.74

Arkowitz, H., Miller, W. R., & Rollnick, S. (Eds.). (2015). *Motivational interviewing in the treatment of psychological problems*. Guilford Publications.

Baron, R. M. & Kenny, D. A. (1986). The moderator - mediator variable distinction in social psychological research: Conceptual, strategic, and statistical considerations. Journal of Personality and Social Psychology, 51(6), 1173-1182. doi:10.1037/0022-3514.51.6.1173

Binford Hopf, R. B., Grange, D. L., Moessner, M., & Bauer, S. (2013). Internet-based chat support groups for parents in family-based treatment for adolescent eating disorders: A pilot study. *European Eating Disorders Review*, *21*(3), 215-223. doi: 10.1002/erv.2196

Butzlaff, R. L., & Hooley, J. M. (1998). Expressed emotion and psychiatric relapse: A meta-analysis. *Archives of General Psychiatry*, *55*(6), 547-552. doi:10.1001/archpsyc.55.6.547

Coomber, K., & King, R. M. (2012). Coping strategies and social support as predictors and mediators of eating disorder carer burden and psychological distress. *Social Psychiatry and Psychiatric Epidemiology*, *47*(5), 789-796. doi:10.1007/s00127-011-0384-6

Coomber, K., & King, R. M. (2013). A longitudinal examination of burden and psychological distress in carers of people with an eating disorder. *Social Psychiatry and Psychiatric Epidemiology*, *48*(1), 163-171. doi:10.1007/s00127-012-0524-7

Deković, M., Asscher, J. J., Manders, W. A., Prins, P. J., & van der Laan, P. (2012). Within-intervention change: Mediators of intervention effects during multisystemic therapy. *Journal of consulting and clinical psychology*, *80*(4), 574. doi:10.1037/a0028482

Fairburn, C.G., & Harrison, P.J. (2003). Eating disorders. *Lancet, 361,* 407–416. doi:10.1016/S0140-6736(03)12378-1

Field, A. (2013). *Discovering Statistics using IBM SPSS Statistics*. Los Angeles, London, New Delhi, Singapore, Washington DC: Sage.

Fritz, M. S., & MacKinnon, D. P. (2007). Required sample size to detect the mediated effect. Psychological science, 18(3), 233-239. doi:10.1111/j.1467-9280.2007.01882.x

Goddard E, Hibbs R, Raenker S, Salerno L, Arcelus J, Boughton N, … & Moore, K.(2013a). A multi-centre cohort study of short term outcomes of hospital treatment for Anorexia Nervosa in the UK. *BioMed Central Psychiatry, 13*(1), 287. doi:10.1186/1471-244X-13-287

Goddard, E., Macdonald, P., Sepulveda, A. R., Naumann, U., Landau, S., Schmidt, U., & Treasure, J. (2011). Cognitive interpersonal maintenance model of eating disorders: Intervention for carers. *The British Journal of Psychiatry, 199*(3), 225–231. doi:10.1192/bjp.bp.110.088401

Goddard, E., Raenker, S., Macdonald, P., Todd, G., Beecham, J., Naumann, U., … Treasure, J. (2013b). Carers' assessment, skills and information sharing: Theoretical framework and trial protocol for a randomised controlled trial evaluating the efficacy of a complex intervention for carers of inpatients with Anorexia Nervosa. *European Eating Disorders Review : The Journal of the Eating Disorders Association, 21*(1), 60–71. doi:10.1002/erv.2193

Goddard, E., Salerno, L., Hibbs, R., Raenker, S., Naumann, U., Arcelus, J., … Treasure, J. (2013c). Empirical examination of the interpersonal maintenance model of Anorexia Nervosa. *The International Journal of Eating Disorders, 46*(8), 867–74. doi:10.1002/eat.22172

Grover, M., Naumann, U., Mohammad-Dar, L., Glennon, D., Ringwood, S., Eisler, I., … Schmidt, U. (2011). A randomized controlled trial of an Internet-based cognitive-behavioural skills package for carers of people with Anorexia Nervosa. *Psychological Medicine, 41*(12), 2581–91. doi:10.1017/S0033291711000766

Harris, E. C., & Barraclough, B. (1998). Excess mortality of mental disorder. *The British Journal of Psychiatry*, *173*(1), 11-53. doi: 10.1192/bjp.173.1.11

Hayes, A. F. (2009). Beyond Baron and Kenny: Statistical mediation analysis in the new millennium. *Communication Monographs*, *76*(4), 408-420. doi:10.1080/03637750903310360

Hayes, A. F. (2012). *PROCESS: A versatile computational tool for observed variable mediation, moderation, and conditional process modeling* (White paper). Retrieved from http://www.afhayes.com/ public/process2012.pdf

Hayes, A. F. (2013). *Introduction to mediation, moderation, and conditional process analysis: A regression-based approach*. New York, NY: Guilford Press

Hibbs, R., Magill, N., Goddard, E., Rhind, C., Raenker, S., Macdonald, P., … Treasure, J. (2015a). Clinical effectiveness of a skills training intervention for caregivers in improving patient and caregiver health following in- patient treatment for severe Anorexia Nervosa : pragmatic randomised controlled trial. British Journal of Psychiatry, 1(September), 56–66. doi:10.1192/bjpo.bp.115.000273

Hibbs, R., Rhind, C., Leppanen, J., & Treasure, J. (2015b). Interventions for caregivers of someone with an eating disorder: A meta-analysis. *International Journal of Eating Disorders,* 48(4), 349-361. doi: 10.1002/eat.22298

Hoek, H. W., & Van Hoeken, D. (2003). Review of the prevalence and incidence of eating disorders. *International Journal of Eating Disorders*, *34*(4), 383-396. doi:10.1002/eat.10222

Hooley, J. M. (2007). Expressed emotion and relapse of psychopathology. *Annual Review of Clinical Psychology, 3,* 329-352. doi:10.1146/annurev.clinpsy.2.022305.095236

Hooley, J. M.,& Campbell, C. (2002). Control and controllability: Beliefs and behaviour in high and low expressed emotion relatives. *Psychological Medicine, 32*(6), 1091–1099. doi:10.1017/S0033291702005779

Kraemer, H. C., Wilson, G. T., Fairburn, C. G., & Agras, W. S (2002). Mediators and moderators of treatment effects in a randomized clinical trials. *Archives of General Psychiatry, 59*(10), 877-883. doi:10.1001/archpsyc.59.10.877

Kyriacou, O., Treasure, J., & Schmidt, U. (2008). Understanding how parents cope with living with someone with Anorexia Nervosa: Modelling the factors that are associated with carer distress. *International Journal of Eating Disorders*, *41*(3), 233-242. doi:10.1002/eat.20488

Las Hayas, C., Padierna, J. Á., Bilbao, A., Martín, J., Muñoz, P., & Quintana, J. M. (2014). Eating disorders: Predictors of change in the quality of life of caregivers. *Psychiatry Research*, *215*(3), 718-726. doi:10.1016/j.psychres.2013.12.028

Leggatt, M. S. (2007). Minimising collateral damage: Family peer support and other strategies. *Medical Journal of Australia*, *187*(7), 61-63. Retrieved from: https://www.mja.com.au/system/files/issues/187_07_011007/leg10280_fm.pdf

Lovibond, S.H., & Lovibond, P.F. (1993). *Manual for the Depression Anxiety and Stress Scales (DASS)*. NSW, Australia: Psychology Foundation Monograph.

Luce, K. H., & Crowther, J. H. (1999). The reliability of the eating disorder examination—Self-report questionnaire version (EDE-Q). *International Journal of Eating Disorders*, *25*(3), 349-351. doi:10.1002/(SICI)1098108X(199904)25:3<349::AID-EAT15>3.0.CO;2-M

Macdonald, P., Rhind, C., Hibbs, R., Goddard, E., Raenker, S., Todd, G., ... & Treasure, J. (2014). Carers' assessment, skills and information sharing (CASIS) trial: A qualitative study of the experiential perspective of caregivers and patients. European Eating Disorders Review, 22(6), 430-438. doi:10.1002/erv.2320

Magill, N., Rhind, C., Hibbs, R., Goddard, E., Macdonald, P., Arcelus, J., ... & Treasure, J. (2016). Two-year follow-up of a pragmatic randomised controlled trial examining the effect of adding a carer's skill training intervention in inpatients with Anorexia Nervosa. *European Eating Disorders Review*, *24*(2), 122-130. doi:10.1002/erv.2422

Medina-Pradas, C., Navarro, J. B., Lopez, S. R., Grau, A., Obiols, J. E., López, S. R., … Obiols, J. E. (2011). Dyadic view of expressed emotion, stress, and eating disorder psychopathology. *Appetite, 57*(3), 743–748. doi:10.1016/j.appet.2011.08.016

Moher, D., Hopewell, S., Schulz, K. F., Montori, V., Gøtzsche, P. C., Devereaux, P. J., ... & Altman, D. G. (2010). CONSORT 2010 explanation and elaboration: Updated guidelines for reporting parallel group randomised trials. British Medical Journal, 340 (c869). doi: 10.1136/bmj.c869

Murray, S. B., Loeb, K. L., & Le Grange, D. (2015) Moderators and mediators of treatments for youth with eating disorders. In M. Maric, P.J.M. Prins, & T.H. Ollendick (Eds.) *Moderators and Mediators of Youth Treatment Outcomes* (chap. 9). Oxford: Oxford University Press.

National Institute of Clinical Excellence (2004). Eating disorders: Core interventions in the treatment and management of Anorexia Nervosa, Bulimia Nervosa and related eating disorders. CG9. Retrieved from http://guidance.nice.org.uk/CG9

Prins, P. J., Ollendick, T. H., Maric, M., &, MacKinnon D. P. (2015). Moderators and Mediators in Treatment Outcome Studies of Childhood Disorders: the What, Why and How. In P.J. Prins, T.H. Ollendick, M. Maric, & D.P. MacKinnon (Eds), *Moderators and Mediators of Youth Treatment Outcomes* (pp.1-20). Oxford: Oxford University Press.

Raenker, S., Hibbs, R., Goddard, E., Naumann, U., Arcelus, J., Ayton, A., … Treasure, J. (2013). Caregiving and coping in carers of people with Anorexia Nervosa admitted for intensive hospital care. *International Journal of Eating Disorders, 46*(4), 346–354. doi:10.1002/eat.22068

Rhind, C., Salerno, L., Hibbs, R., Micali, N., Schmidt, U., Gowers, S., ... & Lo Coco, G. (2016). The Objective and Subjective Caregiving Burden and Caregiving Behaviours of Parents of Adolescents with Anorexia Nervosa. European Eating Disorders Review. 24 (4) 310-319. doi:10.1002/erv.2442

Rhodes, P., Baillee, A., Brown, J., & Madden, S. (2008). Can parent-to-parent consultation improve the effectiveness of the Maudsley model of family-based treatment for Anorexia Nervosa? A randomized control trial. *Journal of Family Therapy*, *30*(1), 96-108. doi:10.1111/j.1467-6427.2008.00418.x

Salerno, L., Rhind, C., Hibbs, R., Micali, N., Schmidt, U., Gowers, S., ... & Treasure, J. (2016). An examination of the impact of care giving styles (accommodation and skilful communication and support) on the one year outcome of adolescent Anorexia Nervosa: Testing the assumptions of the cognitive interpersonal model in Anorexia Nervosa. *Journal of Affective Disorders*, *191*, 230-236. doi:10.1016/j.jad.2015.11.016

Scheffer, J. (2002). Dealing with missing data. Research Letters in the Information and Mathematical Sciences, 3, 153–160. doi: 10179/4355

Schmidt, U., & Treasure, J. (2006). Anorexia Nervosa: Valued and visible. A cognitive-interpersonal maintenance model and its implications for research and practice. *British Journal of Clinical Psychology, 45*(3), 343–366. doi:10.1348/014466505X53902

Sepulveda, A. R., Anastasiadou, D., Pellegrin, Y., Andres, P., Graell, M., Carrobles, J. A., & Morande, G. (2014a). Impact of caregiving experience on mental health among caregivers: A comparison of eating disorder patients with purging and non-purging behaviours. *Eating and Weight Disorders, 19*(1), 31–39. doi:10.1007/s40519-013-0064-5 1

Sepúlveda, A. R., Anastasiadou, D., del Río, A. M., & Graell, M. (2012a). The Spanish validation of level of expressed emotion scale for relatives of people with eating disorders. The Spanish Journal of Psychology, 15(02), 825-839. doi:10.5209/rev_SJOP.2012.v15.n2.38894

Sepulveda, A. R., Anastasiadou, D., Rodríguez, L., Almendros, C., Andrés, P., Vaz, F., … & Graell, M. (2014b). Spanish validation of the Family Questionnaire (FQ) in families of patients with an eating disorder. *Psicothema, 26*(3), 321–327. doi:10.7334/psicothema2013.310

Sepulveda, A. R., Kyriacou, O., & Treasure, J. (2009). Development and validation of the accommodation and enabling scale for eating disorders (AESED) for caregivers in eating disorders. *BMC Health Services Research, 9*, 171. doi:10.1186/1472-6963-9-171

Sepulveda, A. R., Whitney, J., Hankins, M., & Treasure, J. (2008). Development and validation of an eating disorder symptom impact scale (EDSIS) for carers of people with eating disorders. *Health and Quality of Life Outcomes, 6*(28). doi:10.1186/1477-7525-6-28

Slater, J., Treasure, J., Schmidt, U., Gilchrist, P., & Wade, T. D. (2014). A pilot study of associations between treatment for Anorexia Nervosa and carers' distress. *Clinical Psychology & Psychotherapy, 22*(4), 372–376. doi:10.1002/cpp.1895

Szmukler, G. I., Burgess, P., Herrman, H., Bloch, S., Benson, A., & Colusa, S. (1996). Caring for relatives with serious mental illness: The development of the Experience of Caregiving Inventory. *Social Psychiatry and Psychiatric Epidemiology*, *31*(3-4), 137-148. doi: 10.1007/BF00785760

Treasure, J., & Nazar, B. P. (2016). Interventions for the Carers of Patients With Eating Disorders Carer Coping, *Current Psychiatry Reports, 18*(16), 1–7. doi: 10.1007/s11920-015-0652-3

Treasure, J., & Schmidt, U. (2013). The cognitive-interpersonal maintenance model of Anorexia Nervosa revisited: a summary of the evidence for cognitive, socio-emotional and interpersonal predisposing and perpetuating factors. Journal of Eating Disorders, 1(1), 13. doi:10.1186/2050-2974-1-13

Treasure, J., Rhind, C., Macdonald, P., & Todd, G. (2015). Collaborative care: The New Maudsley model. Eating Disorders: The Journal of Treatment & Prevention, 23(4), 366–376. doi: 10.1080/10640266.2015.1044351

Treasure, J., Schmidt, U., & Macdonald, P. (Eds.). (2009). *The Clinician's Guide to Collaborative Caring in Eating Disorders: The New Maudsley Method*. London and New York: Routledge.

Treasure, J., Sepulveda, A. R., MacDonald, P., Whitaker, W., Lopez, C., Zabala, M., ... & Todd, G. (2008). Interpersonal maintaining factors in eating disorder: Skill sharing interventions for carers. International Journal of Child and Adolescent Health, 1(4), 331-338. Retrieved from http://s3.amazonaws.com/academia.edu.documents/42001112/Interpersonal_maintaining_factors_in_eat20160203-4839-1fuspnz.pdf?AWSAccessKeyId=AKIAJ56TQJRTWSMTNPEA&Expires=1479825877&Signature=JV0fekKxl1qJfVRATd%2FcgYZNeg0%3D&response-content-disposition=inline%3B%20filename%3DInterpersonal_Maintaining_Factors_in_Eat.pdf

Treasure, J., Sepulveda, A. R., Whitaker, W., Todd, G., Lopez, C., & Whitney, J. (2007). Collaborative Care between Professionals and Non-Professionals in the Management of Eating Disorders: A Description of Workshops Focussed on Interpersonal Maintaining Factors. *European Eating Disorders Review*, *15*, 24–34. doi: 10.1002/erv

Treasure, J., Smith, G., & Crane, A. (2007). *Skills-based learning for caring for a loved one with an eating disorder*. New York, NY: Routledge.

Vall, E., & Wade, T. D. (2015). Predictors of treatment outcome in individuals with eating disorders: A systematic review and meta-analysis. International Journal of Eating Disorders, 48 (7), 946-971. doi:10.1002/eat.22411

Van Buuren, S. (2012). *Flexible Imputation of Missing Data*. New York, NY: CRC press.

Whitney, J., Haigh, R., Weinman, J., & Treasure, J. (2007). Caring for people with eating disorders: factors associated with psychological distress and negative caregiving appraisals in carers of people with eating disorders. *The British Journal of Clinical Psychology* *46*(4), 413–428. doi:10.1348/014466507X173781

Wiedemann, G., Rayki, O., Feinstein, E., & Hahlweg, K. (2002). The Family Questionnaire: development and validation of a new self-report scale for assessing expressed emotion. Psychiatry research, 109(3), 265-279. doi:10.1016/S0165-1781(02)00023-9

Winn, S., Perkins, S., Walwyn, R., Schmidt, U., Eisler, I., Treasure, J., … & Yi, I. (2007). Predictors of mental health problems and negative caregiving experiences in cares of adolescents with Bulimia Nervosa. *International Journal of Eating Disorders, 40*(2), 171–178. doi:10.1002/eat

Section C

Appendices of Supporting Material

Appendix A: Private Correspondence with Authors

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Appendix B: Samples of Participants Analysed in Multiple Studies

Samples of Participants Analysed in Multiple Studies

Sample	Description	Included studies accessing sample
a	Sample recruited from 2 ED services at Spanish hospitals, and the Spanish Eating Disorders Carers Association (ADANER).	Sepúlveda et al., 2012a; Sepulveda, Anastasiadou, del Rio, & Graell, 2012b; Sepulveda et al., 2014a
b	Part of a large longitudinal trial in Australia investigating caring for someone with an ED	Coomber & King, 2012, 2013
c	Part of a large carer's skills intervention trial for carers of people admitted to hospital for AN treatment	Goddard et al., 2013; Raenker et al., 2013
d	Participants recruited from IoP carer volunteer register. Each study accessed the majority of carers on the register at the time, although some change in carers in the register is to be assumed over time.	Haigh & Treasure, 2003; Kyriacou, Treasure, & Schmidt, 2008; Whitney, Haigh, Weinman, & Treasure, 2007
e	Part of a large carers skills intervention trial for carers of adolescents with AN	Hibbs et al., 2015; Rhind et al., 2016; Salerno et al., 2016
f	Part of a large-scale study recruiting all patients diagnosed and treated at 2 ED Outpatient Clinics as part of the Basque Health Care Service in Spain.	Gonzalez et al., 2012; Las Hayas et al., 2014; Martin et al., 2013; Orive et al., 2013
g	Carers recruited through a set of IoP carers skills workshops	Sepulveda, Whitney, Hankins, & Treasure, 2008; Sepulveda, Kyriacou, & Treasure, 2009; Sepulveda et al., 2010

Note: IoP: Instutute of Psychiatry, London.

Appendix C: Repetition of Analysis Removed from Studies

Table showing Repetition of Analysis Removed from Studies

Sample	Study	Respondent	type	Predictor	Measure	Time	Distress measure	Time	test	statistic	P	Reason
a	Carral-Fernandez et al., 2013	Carers	Co	ED-specific burden	EDSIS	1	GHQ-12	1	*r*	0.5	<.01	Already presented in greater detail by Sepulveda et al. 2012a. Study removed from review as no additional relevant analysis.
			Co	ED-specific burden (all subscales)	EDSIS-ss	1	GHQ-12	1	*r*	.32-.42	<.01	
a	Sepulveda et al., 2014a	Carers (all)	MP	Guilt	EDSIS subscale	0	GSI	0	β	0.21	<.01	Multivariate analysis already addressed for this sample in greater detail by Sepulveda et al. 2012a.
			MP	Social Isolation	EDSIS subscale	0	GSI	0	-	-	0.07	
		Carers of non-purging	UP	ED-specific burden (all subscales)	EDSIS-ss	0	GSI	0	β	0.44 - 0.67	<.01	Univariate relationships between LEE and EDSIS subscales already addressed in greater detail for this sample by Sepulveda 2012a and 2012b. The differences between purging and non-purging sub-groups are presented as discussion below tables instead, for clarity, as no other studies have split groups in this way to allow comparison.
			UP	Expressed emotion	LEE-ss	0	GSI	0	β	0.25 - 0.45	<0.5	
		Carers of purging	UP	ED-specific burden (all subscales)	EDSIS-ss	0	GSI	0	β	0.31 - 0.48	<.01	
			UP	Expressed emotion	LEE-ss	0	GSI	0	β	-	>.05	
			UP	Expressed emotion	LEE-total scale	0	GSI	0	β	0.25 - 0.35	<0.05	
d	Kyriacou, Treasure, & Schmidt, 2008		MP	Difficult behaviours	ECI	0	HADS	0	-	-	>.05	Already presened by Whitney et al., 2007
a	Sepulveda et al., 2012b	Carers	Co	Demo- graphics	SD-Q	0	GHQ-12	0	*rs*	-	>.05	Already presented in greater detail by Sepulveda 2012a (C & service-user age, C gender, C marital status, C educational level, C employment status, living with, age of onset, illness duration, current treatment)
e	Rhind et al., 2016	Carers	Co	Accommodation & Enabling	AESED	1	DASS	1	*rs*	0.41	<.001	Already presented by Hibbs et al. 2015
			Co	Expressed Emotion	FQ & BDSEE	1	DASS	1	*rs*	0.55	<.001	

Note: C: Carer

Appendix D: Measures

Measures

Name of Measure	Abbrev-iation	Authors & Date	Admin	Construct measured	Subscales, Scoring & Comments	Studies Using Measure	# Used	Validation
Construct								
Distress								
General Health Questionnaire	GHQ-12; GHQ-28	Goldberg & Williams, 1998; Goldberg & Hillier, 1979; Spanish: Lobo & Muñoz, 1996	SR	Psychological health over the past 4 weeks	Respondents compare their mental state over the past few weeks to how they usually feel. Subscales; Somatic symptoms, Anxiety/Insomnia (GHQ-A/I), Social Dysfunction, Severe Depression (GHQ-SD). Higher scores indicate more severe distress. Likert scale (0-3) give scores out of 26 (GHQ-12) and 84 (GHQ-28). A score of 12 (GHQ-12) or 28 (GHQ-28) or less indicates ≥ mental health compared to usual. Good psychometric properties.	GHQ-12: Coomber & King, 2012, 2013; Dimitropoulos et al., 2008; Graap et al. 2008; Haigh & Treasure, 2003; Hibbs et al. 2015; Quiles et al. 2016; Sepulveda et al. 2008; 2010, 2012b; Whitney et al. 2007. GHQ-28: Ohara et al. 2016; Slater et al. 2016	13	F
Hospital Anxiety and Stress Scale	HADS	Zigmond & Snaith, 1983; Spanish: Herrmann, 1997	SR	Anxiety and depression	Subscales: Anxiety (HADS-A), Depression (HADS-D). Higher scores indicate higher distress. Well-validated in a non-psychiatric sample with good psychometric properties, including in Spanish.	Goddard et al., 2011; Golzalez et al., 2012; Grover et al., 2011; Kyriacou, Treasure & Schmidt, 2008; Las Hayas et al., 2014; Martin et al., 2013; Orive et al., 2013; Quiles et al., 2016; Sepúlveda et al., 2012a; Sepulveda, Kyriacou, & Treasure, 2009	10	F
Depression, Anxiety and Stress Scale	DASS	Lovibond & Lovibond, 1993.	SR	Distress	Subscales Depression, Stress and Anxiety. Higher scores indicate higher distress. Good psychometric properties, well validated in clinical samples.	Goddard et al., 2013; Hibbs et al., 2015; Raenker et al., 2013; Rhind et al., 2016; Salerno et al., 2016; Sepulveda et al., 2010	6	F
General Severity Index (Symptom Checklist 90-Revised)	GSI	Derogatis, 1994; Spanish: González de Rivera et al., 1989	SR	Psycho- pathology	Well-validated measure of psychopathological distress; 90 items across 9 subscales of the Symptom Checklist Revised (Somatization, Obsessive-compulsive, Interpersonbal sensitivity, Depression, Anxiety, hostility, Phobic anxiety, Paranoid ideation, Psychoticism) with likert scoring (1-4) are divided by 90 to give GSI. Clinical cut off is 8 in men and ≥12 in women.	Rienecke, Richmond & Lebow, 2016; Sepulveda et al., 2014a, 2014b	3	F

Measures continued

Construct Name of Measure	Abbrev-iation	Authors & Date	Admin	Construct measured	Subscales, Scoring & Comments	Studies Using Measure	# Used	Validation
Eating Disorder								
Anoretic Behaviour Observation Scale	ABOS	Vandereycken , 1992; Spanish: Institutio Nacional de la Salud, 1995; Japanese: Uehara et al.,	C re: SU	Service-user's eating behaviour	Subscales: Concern about diet, Bullimic behaviour, Hyperactivity. Higher scores indicate more carer observations of AN symptoms and behaviour. Good reliability reported, also validated in Spanish and Japanese.	Ohara et al. 2016; Kyriacou, Treasure & Schmidt, 2008;	2	F
Short evaluation of eating disorders	SEED	Bauer, Winn, Schmidt, & Kordy, 2005	Clin/ SU SR	Presence and severity of Anorexia Nervosa and Bulimia Nervosa symptoms	Weighted scoring. Higher score indicates higher ED severity. Validated in clinical sample.	Rhind et al., 2016	1	F
Eating Disorders Examination	EDE-Q	Fairburn & Beglin, 1994	SU SR	Presence and severity of ED symptoms	Higher scores indicate greater symptom severity. Validated in clinical sample with good psychometric properties and diagnostic validity.	Goddard et al., 2013	1	F
Carer's needs and coping								
Carers' Needs Assessment	CNA	Wancata et al. 2006	C SR	Carers needs	Subscales: Information about caring for somoene with an ED; Support from other people/organizations; Support for self. 3-point scale, higher scores indicate greater needs. Developed for carers of people diagnosed with Schizophrenia. Adapted for study by Graap et al. 2008, maintained good internal reliability.	Graap et al., 2008	1	P
Carers Needs Assessment Measure	CaNAM	Haigh & Treasure, 2003	C SR	Carers' met and unmet needs in caring for someone with an eating disorder	Designed for carers of people with eating disorders, but validated in one sample only. Assesses carers' perceptions about the amount of support and information about the illness they have received. Binomial (yes/no) scoring system. Higher scores indicate greater needs.	Coomber & King 2012, 2013; Haigh & Treasure, 2003	2	P

Measures continued

Construct Name of Measure	Abbrev-iation	Authors & Date	Admin	Construct measured	Subscales, Scoring & Comments	Studies Using Measure	# Used	Validation
Carer's needs and coping								
Brief COPE	Brief COPE	Carver, 1997	C SR	The way people cope with specified situations	Maladaptive coping and Adaptive coping (problem-focussed; emotion-focussed). 4-point Likert scale. Higher score on maladaptive coping indicates less adaptive coping, and vice versa. Validated, good reliability.	Coomber & King 2012, 2013	2	P
Coping Inventory for Stressful Situations - Japanese version	CISS	Endler & Parker, 1990; Japanese: Furukawa, Suzuki-Moor, Saito, & Hamanaka, 1993	C SR	coping styles in stressful situations	Subscales: Task-oriented coping, Emotion-oriented coping (negative, e.g. self-blame and venting) Avoidance-oriented coping. 48 items on a 5-point likert scale. Higher scores indicate the corresponding coping styles are more frequently used. Japanese version validated.	Ohara et al., 2016	1	P
Burden								
Eating Disorders Symptom Impact Scale	EDSIS	Sepulveda et al., 2008; Spanish: Carral-Fernández et al.,	C SR	Burden specific to impact of ED symptoms	24 items with 1-4 likert scale: Nutrition, Dysregulated behaviour, Guilt, and Social isolation. Good psychometric properties. Higher scores indicate high impact of symptoms.	Raenker et al. 2013; Sepulveda et al., 2008, 2012a, 2014b	4	F
Involvement Evaluation Questionnaire	IEQ	van Wijngaarden et al., 2000	C SR	consequences or burden of caregiving	Subscales: Tension, supervision, worrying, urging; 31-items, 5-point likert scales. Evaluates burden of caring in psychosis and mood disorders, good psychometric properties. Validated in Spanish.	Martin et al. 2013; Las Hayas et al. 2014; Martin et al., 2013	3	F

Measures continued

Construct Name of Measure	Abbreviation	Authors & Date	Admin	Construct measured	Subscales, Scoring & Comments	Studies Using Measure	# Used	Validation
Burden								
Care-ED	Care-ED	Raenker et al., 2013	Res	Time spent dealing with specific aspects of ED caregiving role in last month.	Semi-structured telephone inteview developed for carers of people with eating disorders. Subscales: Medical, Food, Practical related Care, Emotional support. Score indicates time spent on providing support. Retrospective measure vulnerable to memory bias.	Rhind et al. 2016; Salerno et al., 2016	2	N
Burden Assessment Scale	BAS	Reinhard, Gubman, Horwitz, & Minsky, 1994	C SR	Subjective and objective caregiving burden	Objective and Subjective burden subscales. 19 items rated on a likert scale (1-4). High scores indicate higher burden. Good validity and reliability in samples of carers of severe mental illness.	Dimitropoulos et al., 2008	1	P
Experience of Caregiving Inventory	ECI	Szmukler, Burgess, Herrman, Benson, Colusa, & Bloch, 1996	C SR	Experiences of caregiving	Eight negative (negative behaviours, negative symptoms, stigma, problems with services, effects on the family, loss, dependency, need for back-up) and 2 positive (reverse-scored) subscales. 66-items on a 5-point (0-4) likert scale. Higher scores indicate more negative experience.	Sepulveda et al., 2010; Sepúlveda et al. 2012a; Whitney et al. 2007; Winn et al., 2007	1	F
Caregiving Stress Scale	CSS	Pearlin, Mullan, Semple, & Skaff, 1990	C SR	Caregiver stress	Subscales: Family conflict, expressive support, relational deprivation, overload, role captivity, loss of self, self-esteem, personal gain, coping, competence, and mastery. Some items reworded by Kyriacou, Treasure, & Schmidt, (2008) to be more relevant to ED, retaining good internal consistency.	Kyriacou, Treasure & Schmidt, 2008	1	F/N
Zarit Caregiver Burden Interview	J-ZBI_8	Zarit, Reever, & Bach-Peterso, 1980. Japanese 8-item: Arai, Tamiya, & Yano, 2003	C SR	Caregiving burden	5-point likert scale, higher scores indicate higher burden. Designed for use with carers of elderly relatives. Short Japanese version validated.	Ohara et al., 2016	1	F

Measures continued

Construct Name of Measure	Abbrev-iation	Authors & Date	Admin	Construct measured	Subscales, Scoring & Comments	Studies Using Measure	# Used	Validation
Carer's skills								
The Revised Scale for Caregiving Self-Efficacy	CSE	Steffen, McKibbin, Zeiss, Gallagher-Thompson, Bandura, 2002	C SR	caregiver self-efficacy	15 items measure carer self-percieved self-efficacy. Scores are marked on a Likert scale ranging from 1 through to 100 in increments of 10. Good internal reliability.	Goddard et al., 2011	1	P
The Caregiver Skills	CASK	Hibbs et al., 2015c	C SR	Carer skills specific to the support of a person with an ED	Developed measure change following carers skills interventions. Subscales: Bigger picture, Self-care, Biting-your-tongue, Insight and acceptance, Emotional intelligence, Frustration tolerance. 27 items on a 10-point likert scale. Higher score indicates greater skills.	Hibbs et al. 2015; Rhind et al., 2016; Salerno et al., 2013	3	F
Quality of Life								
The Short-Form 12 - Spanish	SF12	Ware et al., 1996; Spanish: Gandek et al., 1998	SR	Individual's percieved health-related quality of life	12 items, scores range from 0-100. A mental component scale (MCS) and physical component scale (HCS). Higher scores indicate better quality of life. Spanish version validated.	Las Hayas et al. 2014; Martin et al., 2013	2	F
World Health Organisation Quality of Life questionnaire - short version	WHO-QOL	Harper, 1998	SR	Individual's quality of life	Quality of life measured across 4 domains: Physical health, psychological health, social relationships, environment. 5-point Likert scale; good psychometric properties. Raenker et al. 2013 used only the 'social wellbeing' scale.	Raenker et al., 2013	1	P
Health Related Quality of Life ED Short Form	HeRQoLED	Engel et al., 2010	SU SR	Health related quality of life in ED	Measures social maladjustment and mental and functional health. Higher scores indicate a better quality of life. Validated for ED population in Spanish.	Las Hayas et al., 2014	1	F

Measures continued

Construct Name of Measure	Abbrev-iation	Authors & Date	Admin	Construct measured	Subscales, Scoring & Comments	Studies Using Measure	# Used	Validation
Social support								
Social Support Questionnaire-Short Form	SSQ6	Sarason, Sarason, Shearan, & Pierce, 1987	SR	Emotional (rather than practical) support from social network	Higher scores indicate more social support. Does not assess objective, practical support. Good internal consistency.	Coomber & King 2012, 2013	2	P
OSLO three-item social support scale	OSLO-3-SS	Meltzer, 2003	SR	Social support	Measures perception of level of support from family, friends and neighbourhood. Higher scores indicate greater perception of support recieved. Good predictive validity in relation to psychological distress.	Rhind et al., 2016	1	P
Social Provisions Scale	SPS	Cutrona & Russell, 1987	SR	Social support	24 items on a 4-point likert scale. Higher scores indicate greater support. Instructions changed for study to assess support outside the family.	Dimitropoulos et al., 2008	1	N
Social Network Questionnaire	SNQ	Magliano et al., 1998	SR	social support	14 items, subscales; Social contacts, Practical support, Affective support, Partner support. Higher scores indicate greater social support. For Ohara et al.'s (2016) study the 'Partner' subscale (2 items) was excluded due to differing concepts between Europe and Japan. Developed for carers of schizophrenia and not validated in ED sample, or in Japanese.	Ohara et al., 2016	1	N
Expressed								
Family Questionnaire	FQ	Weidemann, Rayki, Feinstein, & Hahlweg, 2002; Spanish: Sepulveda et al., 2014b	SR	expressed emotion	Subscales; Criticism and Emotional over-involvement. 4-point likert scale. Frequently used measure with excellent psychometric properties and high convergent validity with more indepth clinician-administered measures. Validated in Spanish for ED population. Higher scores indicate more expressed emotion.	Coomber & King, 2012, 2013; Goddard et al. 2011, 2013; Hibbs et al., 2015; Rhind et al. 2016. Salerno et al., 2013. Spanish: Sepulveda et al., 2014b	6	F

Measures continued

Construct Name of Measure	Abbreviation	Authors & Date	Admin	Construct measured	Subscales, Scoring & Comments	Studies Using Measure	# Used	Validation
Expressed								
Levels of Expressed Emotion	LEE	Cole & Kazarian, 1988; Spanish: Sepulveda et al. 2012b	C SR	Expressed emotion	Subscales: Attitude towards the illness, Hostility, Tolerance/ Coping with illness, Intrusiveness. True/false scoring of 60 items, good psychometric properties. Versions for completion by patient or carer. Valdated in Spanish for ED sample.	Grover et al. 2011 Sepulveda et al., 2012b, 2014a	2	F
Five-Minute Speech Sample	FMSS	Magana, Goldstein, Karno, Miklowitze, Jenkins, Falloon,	Clin	expressed emotion	Subscales: Criticism (CC) and emotional overinvolvement (EOI). Clinicians rate 5 minutes of speach about service-user. Coded as high, borderline or low. Good convergent and discriminative validity.	Sepulveda et al., 2010	1	F
Brief Dyadic Scale of Expressed Emotion	BDSEE	Medina Pradas, Navarro, Lopez, Grau, & Obiols, 2011. Adolescent validation: Schmidt, Tetzlaff, & Hilbert, 2016.	SU re: C	Patient's perception of their carer's expressed emotion	10-point likert-scale, high score means high percieved expressed emotion: higher criticism and emotional involvement and lower warmth. Completed seperately for each carer. Validated, including for adolescents.	Rhind et al. 2016; Salerno et al., 2016	2	F

Measures continued

Construct Name of Measure	Abbrev-iation	Authors & Date	Admin	Construct measured	Subscales, Scoring & Comments	Studies Using Measure	# Used	Validation
Expressed								
- Levels of Expressed Emotion	LEE	Cole & Kazarian, 1988; Spanish: Sepulveda et al. 2012b	C SR	Expressed emotion	Subscales: Attitude towards the illness, Hostility, Tolerance/ Coping with illness, Intrusiveness. True/false scoring of 60 items, good psychometric properties. Versions for completion by patient or carer. Valdated in Spanish for ED sample.	Grover et al. 2011 Sepulveda et al., 2012b, 2014a	2	F
- Five-Minute Speech Sample	FMSS	Magana, Goldstein, Karno, Miklowitze, Jenkins, Falloon, 1986.	Clin	expressed emotion	Subscales: Criticism (CC) and emotional overinvolvement (EOI). Clinicians rate 5 minutes of speach about service-user. Coded as high, borderline or low. Good convergent and discriminative validity.	Sepulveda et al., 2010	1	F
- Brief Dyadic Scale of Expressed Emotion	BDSEE	Medina Pradas, Navarro, Lopez, Grau, & Obiols, 2011. Adolescent validation: Schmidt, Tetzlaff, & Hilbert, 2016.	SU re: C	Patient's perception of their carer's expressed emotion	10-point likert-scale, high score means high percieved expressed emotion: higher criticism and emotional involvement and lower warmth. Completed seperately for each carer. Validated, including for adolescents.	Rhind et al. 2016; Salerno et al., 2016	2	F

Appendix E: Multivariate Analysis

Overview of Multivariate Models of Carer Distress

Sample	Study	Non-significant variables tested in model	Time	Significant predictors in multivariate model	Time	Distress measure	Time	Variance % accounted for by final model	# Sample comparisons
c	Coomber & King, 2013	Satisfaction with social support; expressed emotion; maladaptive coping; total carer needs	0, 4.5m	-	-	GHQ-12	4.5m, 9m	-	-
d	Kyriacou, Treasure, & Schmidt, 2008	SU rejection of having ED; rejection of carer's help; AN symptoms; difficult behaviours	0	Parental gender, interpersonal strains, self-related strains	0	HADS	0	41*	2
d	Whitney, Haigh, Weinman, & Treasure, 2007	Difficult behaviours, problems with services, effect on family, need for back-up, loss	0	Dependancy, stigma	0	GHQ-12	0	53*	2
	Goddard et al., 2013	C marital status, contact time, C accomodating and enabling	0	SU age, C history of eating difficulties, expressed emotion, C psychological control	0	DASS	0	-*	-
b	Coomber & King, 2012	C satisfaction with social support, expressed emotion, total needs, contact hours, SU importance and confidence in change (estmiated	0	C maladaptive coping	0	GHQ-12	0	32*	-
	Winn et al., 2007	Positive experiences of caregiving,	0	Negative experiences of caregiving	0	GHQ-12	0	24*	7
a	Sepulveda et al., 2014a - Total sample	ED psychopathology, SU dysfunctional behaviors, C social isolation, hostility toward patient, tolerance or coping with illness	0	SU Purging; Impact of SU poor nutrition; C Guilt; C attitude towards the illness; C intrusiveness	0	GSI	0	39*	7
	- Carers of people who purge	C intrusiveness, ED psychopathology, SU dysfunctional behaviors, C social isolation, hostility toward patient, tolerance or coping with illness		Nutritional impact, guilt, C attitude towards illness	0	GSI	0	25*	7
	- Carers of people who do not purge	C Guilt; C attitude towards the illness; ED psychopathology, SU dysfunctional behaviors, C social isolation, hostility toward patient, tolerance or coping with illness		Nutritional impact, carer intrusiveness	0	GSI	0	44*	7

Overview of Multivariate Models of Carer Distress continued

Sample	Study	Non-significant variables tested in model	Time	Significant predictors in multivariate model	Time	Distress measure	Time	Variance % accounted for by final model	# Sample comparisons
a	Sepulveda et al., 2012a - Primary Carers	Illness duration; problems with services; SU dependency; Positive experiencec; C guilt; C social isolation	0	Parent's educational level; Loss; good relationship; Impact of nutritional problems	0	HADS-A	0	44*	7
	- Secondary Carers	Loss; good relationship; Impact of nutritional problems; Illness duration; problems with services; SU dependency; Positive experience	0	Parent's educational level; C guilt; C social isolation	0	HADS-A	0	47*	7
	- Primary Carers	Loss; problems with services; SU dependency; Positive experience; Parent's educational level; C guilt;	0	Illness duration; good relationship; Impact of nutritional problems; C social isolation	0	HADS-D	0	42*	7
	- Secondary Carers	Loss; problems with services; Parent's educational level; C guilt; Illness duration; good relationship;	0	SU dependency; Positive experience; Impact of nutritional problems; C social isolation	0	HADS-D	0	42*	7
	Dimotropolous	Burden, family conflict	0	none	0	GHQ-12	0	N/A	-
f	Las Hayas et al.,	-	B	C anxiety, burden	B	HADS-A	Ch. B-1y	-*	6
				Change in Burden, SU depression	B-1y				
	Salerno et al,. 2016	Patient BMI, duration of illness, patient age (all contributed to variance accounted for in final model)	B	Carer distress, patient distress	B	DASS	Ch. B-1y	43*	-
	Ohara et al., 2016	Social contact; C observations of AN; General family functioning	0	Emotion-oriented coping; Affective social support; contact time	0	GHQ-28	Ch. B-1y	60.2*	-

Overview of Multivariate Models of Carer Distress continued

Sample	Study	Non-significant variables tested in model	Time	Significant predictors in multivariate model	Time	Distress measure	Time	Variance % accounted for by final model	# Sample comparisons
f	Orive et al., 2013	Relationship: partner/sibling/child (vs. mother); C depression	B	Relationship: father (vs.mother); SU drug addiction	B	HADS-A	B	-*	6
		-	-	C anxiety; SU Restrictive ED only (compared to purging-type)	B	HADS-A	Ch. B-1y	-*	-
		Relationship: father/partner (vs. mother); Education:secondary (vs. primary/none)	B	Relationship: sibling/child (vs. mother); Education: univeristy (vs. primary/none); C anxiety; SU suidical intent	B	HADS-D	Ch. B-1y	-*	-
		SU clinical depression	B	SU Restrictive ED only (vs. purging-type); SU poss. depression; C depression; C education: secondary/univeristy (vs. primary/none).	B	HADS-D	Ch. B-1y	-*	-

Note: Analysis of predictors of distress in participants recieving intervention (Sepulveda et al., 2010; Salerno et al., 2016) have not been included in this table, due to the confounding effect of intervention. Data from Salerno et al.'s (2016) treatment as usual group is presented. Sample only given where sample is analysed by > 1 study. Number of comparisons only given where >1. *Model significant to at least p<0.5. 0: Cross-sectional study. B: Baseline. EoT: End of treatment. C: Carer. SU: Service-user. - : Not given. PC/SC: Analysis relates to primary/secondary carer.

Appendix F: Proposed Predictors of Carer Distress by Type

Predictors of Carer Distress

Category	*Sample*	*Author*	*Type*	*Lv. Evidence*	*Predictor*	*Rater*	*Measure*	*Time*	*Distress measure*	*Time*	*Test*	*Statistic*	*p*	*Power*	# study comparisons	# sample comparisons
Service-user Demographic & Clinical																
Service-user age																
	c	Goddard et al., 2013	UP	4	Service-user age	SU	SD-Q	0	DASS	0	r	0.16	<.05		7	
	c	Goddard et al., 2013	MP	2	Service-user age	SU	SD-Q	0	DASS	0	β	0.14	<.05	M		
	e	Salerno et al., 2016	MP	1	Service-user age	SU	SD-Q	B	DASS	Ch. B-1y	β	0.12	0.488	L		
Illness duration																
	a	Sepúlveda et al., 2012a	MP	2	Illness duration	PC	clinical	0	HADS-A	0	β	-	>.05	S-M	4	7
	a	Sepúlveda et al., 2012a	MP	2	Illness duration	PC	clinical	0	HADS-D	0	β	-	>.05	S-M	4	7
	a	Sepúlveda et al., 2012a	MP	2	Illness duration	SC	clinical	0	HADS-A	0	β	-0.19	<.001	S-M	4	7
	a	Sepúlveda et al., 2012a	MP	2	Illness duration	SC	clinical	0	HADS-D	0	β	-	>.05	S-M	4	7
	f	Las Hayas et al., 2014	UP	3	Duration of illness	Clin	Clinical	B	HADS-A	Ch. B-1yr	β	-0.09	0.042		2	6
	e	Salerno et al., 2016	MP	1	Duration of illness	Clin	Clinical	B	DASS	Ch. B-1yr	β	-0.17	0.352	L		
Co-morbidities																
	e	Rhind et al., 2016	UP	4	OCD Severtiy	SU	CYBOCS	0	DASS	0	r_s	-0.04	>.05		17	36
	e	Rhind et al., 2016	UP	4	P Social apptitude	C	SAS	0	DASS	0	r_s	-0.26	<.05		17	36
	f	Orive et al., 2013	UP	4	SU suicidal intent	Clin	clinical	B	HADS-A	B	β	1.83	0.08		30	78
	f	Orive et al., 2013	UP	4	SU suicidal intent	Clin	clinical	B	HADS-D	B	β	2.85	0.005		30	78
	f	Orive et al., 2013	UP	4	SU drug addiction	Clin	clinical	B	HADS-A	B	β	5.96	0.06		30	78
	f	Orive et al., 2013	UP	4	SU drug addiction	Clin	clinical	B	HADS-D	B	β	6.99	0.02		30	78
	f	Orive et al., 2013	UP	4	SU psychosis	Clin	clinical	B	HADS-A, D	B	β	4.63, 1.88	.16, .58		30	78
	f	Orive et al., 2013	MP	2	SU Suicidal intent	Clin	clinical	B	HADS-D	B	β	2.65	0.002	M	4	6
	f	Orive et al., 2013	MP	2	SU Drug addiction	Clin	clinical	B	HADS-A	B	β	4.71	0.04	S-M	4	6
	f	Orive et al., 2013	UP	3	SU suicidal intent	Clin	clinical	B	HADS-A, D	Ch. B-1y	β	-1.23,	.16, .59		30	78
	f	Orive et al., 2013	UP	3	SU drug addiction	Clin	clinical	B	HADS-A, D	Ch. B-1y	β	2.20, 1.32	.35, .48		30	78
	f	Orive et al., 2013	UP	3	SU psychosis	Clin	clinical	B	HADS-A, D	Ch. B-1y	β	0.36, 0.29	.88, .89		30	78

Predictors of Carer Distress continued

Category	*Sample*	*Author*	*Type*	*Lv. Evidence*	*Predictor*	*Rater*	*Measure*	*Time*	*Distress measure*	*Time*	*Test*	*Statistic*	*p*	*Power*	# study comparisons	# sample comparisons
ED symptoms																
	c	Goddard et al. 2013	UP	4	ED symptoms	SU	EDE-Q	0	DASS	0	*r*	0.1	>.05		7	
		Ohara et al., 2016	UP	4	C Observations of AN behaviour	SU	ABOS	0	GHQ-28	0	*r*	0.41	<.01		27	
		Ohara et al., 2016	UP	4	C Observations of AN behaviour	SU	ABOS	0	GHQ-SD	0	*r*	0.31	<.01		27	
		Ohara et al., 2016	UP	4	C Observations of AN behaviour	SU	ABOS	0	GHQ-A/ Insomnia	0	*r*	0.39	<.001		27	
	e	Rhind et al., 2016	UP	4	AN severity	SU	SEED	0	DASS	0	r_s	-0.06	>.05		17	36
	a	Sepulveda et al., 2014a	MP	2	Severity of ED (by trmt required)	Clin	Day/in-Service-user vs not	0	GSI	0		-	0.53	M	3	7
		Ohara et al., 2016	MP	2	C Observations of AN behaviour	C	ABOS	0	GHQ-28	0	β	-	>.05	L		
	b	Coomber & King, 2012	MP	2	SU's importance of Change	C	Likert scale 0-11	0	GHQ-12	0	β	0.01	>.05	L		
	b	Coomber & King, 2012	MP	2	SU's confidence to Change	C	Likert scale 0-11	0	GHQ-12	0	β	-0.06	>.05	L		
	d	Kyriacou, Treasure & Schmidt, 2008	MP	2	Sufferer rejection of having ED; sufferer rejection of carer's help ; AN symptoms	C	ABOS	0	HADS	0	β	-	>.05	M		2
	e	Salerno et al., 2016	MP	1	Service-user BMI	Clin	Clinical	B	DASS	Ch. B-1y	β	0.2	0.11	L		
Treatment																
	f	Orive et al., 2013	UP	4	SU time in clinical treatment (by 1 m	Clin	clinical	B	HADS-A, D	B	β	0.004, -	.55, .10		30	78
	f	Orive et al., 2013	UP	3	SU time in clinical treatment (by 1 m	Clin	clinical	B	HADS-A, D	Ch. B-1y	β	-0.008,	.11, .65		30	78
	f	Orive et al., 2013	UP	3	SU time in clinical treatment (by 1 m	Clin	clinical	B	HADS-A, D	Ch. B-1y	β	-0.01, -	.20, .99		30	78
Compensatory behaviour																
	f	Orive et al., 2013	UP	4	SU Restrictive ED only (compared to	Clin	clinical	B	HADS-A, D	B		-0.62, -	.43, .25		30	78
	f	Orive et al., 2013	UP	3	SU Restrictive ED only (compared to	Clin	clinical	B	HADS-A, D	Ch. B-1y	β	1.07, 0.62	.12, .24		30	78
	a	Sepulveda et al., 2014a	MP	2	SU purging	C	Y/N	0	GSI	0	β	0.14	<.05	M	3	7

Predictors of Carer Distress Continued

Category	Sample	Author	Type	Lv. Evidence	Predictor	Rater	Measure	Time	Distress measure	Time	Test	Statistic	p	Power	# study comparisons	# sample comparisons
Service-user's Distress																
	c	Goddard et al., 2013	UP	4	Service-user distress	SU	DASS	B	DASS	B	r	0.24	<.001		7	
	e	Rhind et al., 2016	UP	4	Service-user distress	SU	DASS	B	DASS	B	r_s	-0.45	<.001		17	36
	f	Orive et al., 2013	UP	4	SU Depression (possible, clinical)	SU	HADS-D (8-10, ≥11)	B	HADS-A	B	β	1.33, 1.38	.18, .14		30	78
	f	Orive et al., 2013	UP	3	SU baseline depression (possible, clinical)	SU	HADS-D (8-10, ≥11)	B	HADS-A	Ch. B-1y	β	1.17, -.31	.17, .69		30	78
	f	Orive et al., 2013	UP	3	SU baseline depression (possible)	SU	HADS-D (8-10)	B	HADS-D	Ch. B-1y	β	1.42	0.03		30	78
	f	Orive et al., 2013	UP	3	SU baseline depression (clinical)	SU	HADS-D (≥11)	B	HADS-D	Ch. B-1y	β	-0.41	0.49		30	78
	e	Salerno et al., 2016	MP	1	SU distress	SU	DASS	B	DASS	Ch. B-1yr	β	0.34	0.01	L		
	f	Las Hayas et al., 2014	MP	1	Change in SU depression	SU	HADS-D	Ch. B-1yr	HADS-A	Ch. B-1yr	β	1.53	0.015	S-M	2	6
Service-users quality of life																
	f	Las Hayas et al., 2014	UP	3	Service-user mental health and functionality	SU	HeRQoL ED-ss	B	HADS-A	Ch. B-1y	β	-0.02	0.21		16	78
	f	Las Hayas et al., 2014	UP	3	Service-user social maladjustment	SU	HeRQoL ED-ss	B	HADS-A	Ch. B-1y	β	-0.01	0.36		16	78
	f	Las Hayas et al., 2014	UP	3	Service-user mental quality of life	SU	SF12-MCS	B	HADS-A	Ch. B-1y	β	0.008	0.8		16	78
	f	Las Hayas et al., 2014	UP	3	Service-user physical quality of life	SU	SF12-PCS	B	HADS-A	Ch. B-1y	β	0.1	<.001		16	78
	f	Las Hayas et al., 2014	UP	3	Service-user mental health and functionality	SU	HeRQoL ED-ss	Ch. B-1yr	HADS-A	Ch. B-1y	β	0.04	0.02		16	78

Predictors of Carer Distress Continued

Category	*Sample*	*Author*	*Type*	*Lv. Evidence*	*Predictor*	*Rater*	*Measure*	*Time*	*Distress measure*	*Time*	*Test*	*Statistic*	*p*	*Power*	*# study comparisons*	*# sample comparisons*
Carer Gender and relationship to service-user																
	f	Las Hayas et al., 2014	UP	3	Relationship: Sibling/child (vs. mother)	C	SD-Q	B	HADS-A	Ch. B-1yr	β	0.46	0.65		16	78
	f	Las Hayas et al., 2014	UP	3	Female (vs. male) gender	C	SD-Q	B	HADS-A	Ch. B-1yr	β	-0.16	0.77		16	78
Contact & Time Spent Caregiving																
	a	Sepúlveda et al., 2012a	UP	4	Contact time	PC, SC	SD-Q	0	HADS -A/D	0	*r*	-	>.05		15	22
	c	Goddard et al., 2013	UP	4	Contact time	C	SD-Q h/w	0	DASS	0	*r*	0.26	<.001		7	
		Dimitropoulos et al., 2008	UP	4	Contact hours	C	SD-Q h/w	0	GHQ-12	0	*r*	0.05	>.05		10	
	e	Rhind et al., 2016	UP	4	Time spent caregiving	C	Care-ED h/w	0	DASS	0	r_s	0.24	>.05		17	36
	e	Rhind et al., 2016	UP	4	Time spent caregiving	M	Care-ED h/w	0	DASS	0	β	0.24	<.01		17	36
	e	Rhind et al., 2016	UP	4	Time spent caregiving	F	Care-ED h/w	0	DASS	0	β	0.07	>.05		17	36
	a	Sepúlveda et al., 2012a	UP	4	Living with SU	PC, SC	SD-Q y/n	0	HADS -A, D	0	*r*	-	>.05		15	22
	b	Coomber & King, 2012	MP	2	Contact hours	C	SD-Q h/w	0	GHQ-12	0	β	0.23	>.05	L		
	c	Goddard et al., 2013	MP	2	Contact time	C	SD-Q h/w	0	DASS	0	β	0.06	>.05	M		
		Ohara et al., 2016	MP	2	Contact time	C	SD-Q h/d	0	GHQ-28	0	β	2.0	0.042		27	
Carer Clinical																
Carer Distress																
	e	Hibbs et al., 2015	UP	4	General/psychological health	C	GHQ-12	0	DASS-21	0	r_s	0.68	<.01		19	36
	f	Orive et al., 2013	MP	2	Depression (possible, clinical)	C	HADS-D (8-10, ≥11)	0	HADS-A	0	β	5.08, 6.26	<.001	S-M		

Predictors of Carer Distress Continued

Category	Sample	Author	Type	Lv. Evidence	*Predictor*	*Rater*	Measure	Time	*Distress measure*	*Time*	Test	*Statistic*	*p*	Power	# study comparisons	# sample comparisons
Carer Clinical																
Carer Distress continued																
	f	Orive et al., 2013	MP	2	Possible anxiety	C	HADS-A (8-10)	0	HADS-D	0	β	2.01	0.01	M	4	6
	f	Orive et al., 2013	MP	2	Clinical anxiety	C	HADS-A ≥11	0	HADS-D	0	β	4.67	<.001	M	4	6
	f	Las Hayas et al., 2014	UP	3	Change in depression	C	HADS-D	Ch. B-1yr	HADS-A	Ch. b-1yr	β	1.68	0.018		16	78
	f	Las Hayas et al., 2014	MP	1	Carer anxiety	C	HADS-A	B	HADS-A	Ch. b-1yr	β	0.42	<.001	S-M	2	6
	e	Salerno et al., 2016	MP	1	Carer distress	C	DASS	B	DASS	Ch. b-1yr	β	0.47	<.000	L		
Carers' Quality of Life																
	f	Martin et al., 2013	UP	4	Quality of life (physical)	M	SF-12 - PCSS	0	HADS-A	0	*r*	-0.41, -0.42	<.0001		12	78
	f	Martin et al., 2013	UP	4	Quality of life (physical)	F	SF-12 - PCSS	0	HADS-A, D	0	*r*	-0.14, -0.1	.28, .41		12	78
	f	Martin et al., 2013	UP	4	Quality of life (mental)	M	SF-12 - MCSS	0	HADS-A, D	0	*r*	-0.67, -0.75	<.0001		12	78
	f	Martin et al., 2013	UP	4	Quality of life (mental)	F	SF-12 - MCSS	0	HADS-A	0	*r*	-0.46, -0.45	.0001, .0002		12	78
Carer History of Dating Difficulties																
	c	Goddard et al., 2013	MP	2	Carer history of eating difficulties	C	self-report	0	DASS	0	β	0.17	<.05	M		
Hypothesised reactions to the ED																
Expressed Emotion																
	e	Hibbs et al., 2015	UP	4	Expressed emotion	C	FQ	0	GHQ-12	0	r_s	0.57	<.01		19	36
	e	Hibbs et al., 2015	UP	4	Expressed emotion	C	FQ	0	DASS-21	0	r_s	0.53	<.01		19	36
	a	Sepulveda et al., 2012b	UP	4	Expressed emotion	C	LEE-S	0	GHQ-12	0	r_s	0.34	<0.01		7	22
	c	Goddard et al., 2013	UP	4	Expressed Emotion	C	FQ	0	DASS	0	*r*	0.46	<.001		7	
	e	Rhind et al., 2016	UP	4	Expressed Emotion	M	FQ,	0	DASS	0	β	0.51	<.001		17	36
	e	Rhind et al., 2016	UP	4	Expressed Emotion	F	FQ, BDSEE	0	DASS	0	β	0.47	<.001		17	36

Predictors of Carer Distress Continued

Category	*Sample*	*Author*	*Type*	*Lv. Evidence*	*Predictor*	*Rater*	*Measure*	*Time*	*Distress measure*	*Time*	*Test*	*Statistic*	*p*	*Power*	*# study comparisons*	*# sample comparisons*
Hypothesised reactions to the ED																
Expressed Emotion continued																
		Sepulveda et al., 2014b	UP	4	Criticism	M	FQ-Spanish	0	GSI	0	r_s	0.36	<.001		4	
		Sepulveda et al., 2014b	UP	4	Criticism	F	FQ-Spanish	0	GSI	0	r_s	0.32	<.01		4	
	b	Coomber & King, 2012	MP	2	Expressed emotion	C	FQ	0	GHQ-12	0	β	0.21	>.05	L		
	c	Goddard et al., 2013	MP	2	Expressed emotion	C	FQ	0	DASS	0	β	0.25	<.05	M		
		Sepulveda et al., 2014b	UP	4	Emotional over-involvement	M	FQ-Spanish	0	GSI	0	r_s	0.24	<.01		4	
Hypothesised reactions to the ED																
Expressed Emotion continued																
		Sepulveda et al., 2014b	UP	4	Emotional over-involvement	F	FQ-Spanish	0	GSI	0	r_s	0.24	<.01		4	
	a	Sepulveda et al., 2012b	UP	4	Attitude toward illness (LEE subscale)	C	LEE-S-ss	0	GHQ-12	0	r_s	0.09	>.05		7	22
	a	Sepulveda et al., 2012b	UP	4	Intrusiveness; Hostility toward Service-user; Lack of Tolerance/coping with illness	C	LEE-S-ss	0	GHQ-12	0	r_s	0.30 - 0.29	<0.01		7	22
	a	Sepulveda et al., 2014a	MP	2	Attitude towards illness	C	LEE-S-ss	0	GSI	0	β	0.13	<.05	M	3	7
	a	Sepulveda et al., 2014a	MP	2	Intrusiveness	C	LEE-S-ss	0	GSI	0	β	0.15	<.05	M	3	7
	a	Sepulveda et al., 2014a	MP	2	Hostility towards Service-user; Lack of tolerance/coping with illness	C	LEE-S-ss	0	GSI	0		-	0.43	M	3	7
	a	Sepulveda et al., 2014a	MP	2	Tolerance or coping with illness	C	LEE-S-ss	0	GSI	0		-	0.98	M	3	7
	b	Coomber & King, 2013	UP	3	Expressed Emotion	C	FQ	B, 4.5m	GHQ-12	4.5m, 9m	*r*	0.7 - 0.28	>.05		4	12
	b	Coomber & King, 2013	MP	1	Expressed emotion	C	FQ;	B, 4.5m	GHQ-12	4.5m, 9m	β	-	>.05	L		
Family functioning and conflict																
		Dimitropoulos et al., 2008	UP	4	Family Conflict	C	FCS	0	GHQ-12	0	*r*	0.28	<.05		10	
		Dimitropoulos et al., 2008	UP	4	Family conflict regarding person with AN	C	FCS-ss	0	GHQ-12	0	*r*	0.24	>.05		10	

Predictors of Carer Distress Continued

Category	Sample	Author	Type	Lv. Evidence	Predictor	Rater	Measure	Time	Distress measure	Time	Test	Statistic	p	Power	# study comparisons	# sample comparisons
Family functioning and conflict continued																
		Dimitropoulos et al., 2008	UP	4	Family conflict regarding other family member	C	FCS-ss	0	GHQ-12	0	*r*	0.12	>.05		10	
		Dimitropoulos et al., 2008	UP	4	Family functioning	C	FAD-	0	GHQ-12	0	*r*	-0.32	<.05		10	
		Ohara et al., 2016	UP	4	General Family functioning	C	FAD-	0	GHQ-28	0	*r*	0.25	<.05		27	
		Ohara et al., 2016	UP	4	General Family functioning	C	FAD-	0	GHQ-A/I	0	*r*	0.19	>.05		27	
		Ohara et al., 2016	UP	4	General Family functioning	C	FAD-	0	GHQ-SD	0	*r*	0.19	>.05		27	
		Dimitropoulos et al., 2008	MP	2	Family Conflict	C	FCS	0	GHQ-12	0	β	0.23	0.8			
		Ohara et al., 2016	MP	2	General Family functioning	C	FAD-	0	GHQ-28	0	β		>.05			
Carer efforts to control the eating disorder																
	c	Goddard et al., 2013	UP	4	Psychological control	C	PCS	0	DASS	0	*r*	0.38	<.001		7	
	c	Goddard et al., 2013	MP	2	Psychological control	C	PCS	0	DASS	0	β	0.17	<.05	M		
Accommodating and Enabling																
	c	Goddard et al., 2013	UP	4	Accommodation and Enabling	C	AESED	0	DASS	0	*r*	0.38	<.001		7	
		Quiles et al., 2016	UP	4	Avoidance and Modifying routines	C	AESEDs	0	HADS-D	0	*r*	0.46	<.01		15	
		Quiles et al., 2016	UP	4	Reassurance Seeking	C	AESEDs	0	HADS-D	0	*r*	0.35	<.01		15	
		Quiles et al., 2016	UP	4	Meal rituals	C	AESEDs	0	HADS-D	0	*r*	0.27	<.05		15	
		Quiles et al., 2016	UP	4	Control of family	C	AESEDs	0	HADS-D	0	*r*	0.36	<.01		15	
		Quiles et al., 2016	UP	4	Turning a blind eye	C	AESEDs	0	HADS-D	0	*r*	0.18	>.05		15	
		Quiles et al., 2016	UP	4	Avoidance and Modifying routines	C	AESEDs	0	HADS-A	0	*r*	0.47	<.01		15	
		Quiles et al., 2016	UP	4	Reassurance Seeking	C	AESEDs	0	HADS-A	0	*r*	0.38	<.01		15	
		Quiles et al., 2016	UP	4	Meal rituals	C	AESEDs	0	HADS-A	0	*r*	0.2	>.05		15	
		Quiles et al., 2016	UP	4	Control of family	C	AESEDs	0	HADS-A	0	*r*	0.35	<.01		15	
		Quiles et al., 2016	UP	4	Turning a blind eye	C	AESEDs	0	HADS-A	0	*r*	0.18	>.05		15	
		Quiles et al., 2016	UP	4	Avoidance and Modifying routines	C	AESEDs	0	GHQ-12	0	*r*	0.37	<.01		15	
		Quiles et al., 2016	UP	4	Reassurance Seeking	C	AESEDs	0	GHQ-12	0	*r*	0.36	<.01		15	
		Quiles et al., 2016	UP	4	Meal rituals	C	AESEDs	0	GHQ-12	0	*r*	0.19	>.05		15	

Predictors of Carer Distress Continued

Category	Sample	Author	Type	Lv. Evidence	*Predictor*	*Rater*	Measure	Time	*Distress measure*	*Time*	Test	*Statistic*	*p*	Power	# study comparisons	# sample comparisons
Caregiving Burden																
Overall burden continued																
	a	Sepúlveda et al., 2012a	UP	4	Negative caregiving experience [1]	PC, SC	ECI-ss	0	HADS-A, D	0	*r*	0.45 -	<.01		15	22
	f	Gonzalez et al., 2012	UP	4	Total caregiving burden[1]	C	IEQ	0	HADS -A, D	0	*rs*	0.49, 0.43	<0.05		10	78
		Dimitropoulos et al., 2008	UP	4	Burden	C	BAS	0	GHQ-12	0	*r*	0.26	<.05		10	
		Ohara et al., 2016	UP	4	Caregiver burden	C	J-ZBI_8	0	GHQ-28 total & ss; A/I, SD	0	*r*	0.55 - 0.61	<.001		27	
	a	Sepúlveda et al., 2012a	UP	4	ED-specific caregiving burden [1]	PC, SC	EDSIS	0	HADS-A, D	0	*r*	0.54 -	<.01		15	22
	g	Sepulveda et al., 2008	UP	4	ED-specific caregiving burden	C	EDSIS	0	GHQ-12	0	*r*	0.32	<.01		5	17
		Dimitropoulos et al., 2008	MP	2	Burden	C	BAS	0	GHQ-12	0	β: R	0.24: 0.06	0.6	M-L	63	
		Winn et al., 2007	MP	2	Negative experiences of caregiving	C	ECI-ss	0	GHQ-12	0	*	0.09	<.001	M		
	d	Kyriacou, Treasure & Schmidt, 2008	MP	2	Total of combined interpersonal strains scales	C	CSS items 26–104	0	HADS	0	R^2	0.065	0.008	M		2
	d	Kyriacou, Treasure & Schmidt, 2008	MP	2	Total of combined self-related strains scales	C	CSS items 57–228	0	HADS	0	R^2	0.31	<.001	M		2
	f	Las Hayas et al., 2014	UP	3	caregiving burden	C	IEQ	B	HADS-A	Ch. B-1yr	β	-0.06	0.024		16	78
	f	Las Hayas et al., 2014	UP	3	caregiving burden	C	IEQ	Ch. B-1yr	HADS-A	Ch. B-1yr	β	0.1	0.001		16	78
	f	Las Hayas et al., 2014	MP	1	caregiving burden	C	IEQ	B	HADS-A	Ch. B-1yr	β	-0.1	<.001	S-M	2	6
	f	Las Hayas et al., 2014	MP	1	caregiving burden	C	IEQ	Ch. B-1yr	HADS-A	Ch. B-1yr	β	0.11	<.001	S-M	2	6
Components of burden																
	g	Sepulveda et al., 2008	UP	4	Dysregulated behaviour	C	EDSIS-	B	GHQ-12	B	*r*	0.23	<.05		5	17
	a	Sepulveda et al., 2014a	MP	2	Dysregulated behaviour	C	EDSIS-	0	GSI	0	β	-	0.11	M	3	7
	g	Sepulveda et al., 2008	UP	4	Impact of nutritional problems	C	EDSIS-	B	GHQ-12	B	*r*	0.18	>.05		5	17
	a	Sepulveda et al., 2014a	MP	2	Impact of nutritional problems	C	EDSIS-	0	GSI	0	β	0.44	<.001	M	3	7

Predictors of Carer Distress Continued

Category	Sample	Author	Type	Lv. Evidence	*Predictor*	*Rater*	Measure	Time	*Distress measure*	*Time*	Test	*Statistic*	*p*	Power	# study comparisons	# sample comparisons
Components of burden continued																
	a	Sepúlveda et al., 2012a	MP	2	Impact of nutritional problems	PC	EDSIS-	0	HADS-A	0	β	0.5	<.001	S-M	4	7
	a	Sepúlveda et al., 2012a	MP	2	Impact of nutritional problems	SC	EDSIS-	0	HADS-D	0	β	0.3	<.001	S-M	4	7
	a	Sepúlveda et al., 2012a	MP	2	Impact of nutritional problems	SC	EDSIS-	0	HADS-A	0	β	0.39	<.001	S-M	4	7
	a	Sepúlveda et al., 2012a	MP	2	Impact of nutritional problems	PC	EDSIS-	0	HADS-D	0	β	-	>.05	S-M	4	7
	g	Sepulveda et al., 2008	UP	4	Guilt	C	EDSIS-	0	GHQ-12	0	*r*	0.24	<.05		5	17
	a	Sepúlveda et al., 2012a	MP	2	Guilt	PC	EDSIS-	0	HADS-A	0	β	-	>.05	S-M	4	7
	a	Sepúlveda et al., 2012a	MP	2	Guilt	PC	EDSIS-	0	HADS-D	0	β	0.38	<.001	S-M	4	7
	a	Sepúlveda et al., 2012a	MP	2	Guilt	SC	EDSIS-	0	HADS-A	0	β	-	>.05	S-M	4	7
	a	Sepúlveda et al., 2012a	MP	2	Guilt	SC	EDSIS-	0	HADS-D	0	β	-	>.05	S-M	4	7
	g	Sepulveda et al., 2008	UP	4	Social isolation	C	EDSIS-	0	GHQ-12	0	*r*	0.33	<.01		5	17
	a	Sepúlveda et al., 2012a	MP	2	Social isolation	PC	EDSIS-	0	HADS-A	0	β	-	>.05	S-M	4	7
	a	Sepúlveda et al., 2012a	MP	2	Social isolation	PC	EDSIS-	0	HADS-D	0	β	0.38	<.001	S-M	4	7
	a	Sepúlveda et al., 2012a	MP	2	Social isolation	SC	EDSIS-	0	HADS-A	0	β	0.24	<.001	S-M	4	7
	a	Sepúlveda et al., 2012a	MP	2	Social isolation	SC	EDSIS-	0	HADS-D	0	β	0.29	<.001	S-M	4	7
	d	Whitney et al., 2007	MP	2	Dependency	C	ECI-ssc	0	GHQ-12	0	β	0.39	<.001	S-M		2
	a	Sepúlveda et al., 2012a	MP	2	Dependency	PC	ECI-ssc	0	HADS-A	0	β	-	>.05	S-M	4	7
	a	Sepúlveda et al., 2012a	MP	2	Dependency	PC	ECI-ssc	0	HADS-D	0	β	-	>.05	S-M	4	7
	a	Sepúlveda et al., 2012a	MP	2	Dependency	SC	ECI-ssc	0	HADS-A	0	β	-	>.05	S-M	4	7
	a	Sepúlveda et al., 2012a	MP	2	Dependency	SC	ECI-ssc	0	HADS-D	0	β	0.27	<.01	S-M	4	7
	d	Whitney et al., 2007	MP	2	Problems with services	C	ECI-ssc	0	GHQ-12	0	β	-	>.05	S-M		2
	a	Sepúlveda et al., 2012a	MP	2	Problems with services	PC	ECI-ssc	0	HADS-A	0	β	-	>.05	S-M	4	7
	a	Sepúlveda et al., 2012a	MP	2	Problems with services	PC	ECI-ssc	0	HADS-D	0	β	-	>.05	S-M	4	7
	a	Sepúlveda et al., 2012a	MP	2	Problems with services	SC	ECI-ssc	0	HADS-A	0	β	0.13	<.05	S-M	4	7
	a	Sepúlveda et al., 2012a	MP	2	Problems with services	SC	ECI-ssc	0	HADS-D	0	β	-	>.05	S-M	4	7
	d	Whitney et al., 2007	MP	2	Loss	C	ECI-ssc	0	GHQ-12	0	β	-	>.05	S-M		2
	a	Sepúlveda et al., 2012a	MP	2	Loss	PC	ECI-ssc	0	HADS-A	0	β	0.22	<.001	S-M	4	7

Predictors of Carer Distress Continued

Category	*Sample*	*Author*	*Type*	*Lv. Evidence*	*Predictor*	*Rater*	*Measure*	*Time*	*Distress measure*	*Time*	*Test*	*Statistic*	*p*	*Power*	# study comparisons	# sample comparisons
Components of burden continued																
	a	Sepúlveda et al., 2012a	MP	2	Loss	PC	ECI-ssc	0	HADS-D	0	β	-	>.05	S-M	4	7
	a	Sepúlveda et al., 2012a	MP	2	Loss	SC	ECI-ssc	0	HADS-A	0	β	-	>.05	S-M	4	7
	a	Sepúlveda et al., 2012a	MP	2	Loss	SC	ECI-ssc	0	HADS-D	0	β	-	>.05	S-M	4	7
	d	Whitney et al., 2007	MP	2	Difficult behaviours; Effect on family; Need for backngup	C	ECI-ssc	0	GHQ-12	0	β	-	>.05	S-M		2
Stigma																
		Dimitropoulos et al., 2008	UP	4	Percieved stigma towards SU	C	DCCF-ss	0	GHQ-12	0	*r*	-0.06	>.05		10	
		Dimitropoulos et al., 2008	UP	4	Percieved stigma towards family	C	DCCF-ss	0	GHQ-12	0	*r*	-0.02	>.05		10	
	d	Whitney et al., 2007	MP	2	Stigma	C	ECI - stigma subscal	0	GHQ-12	0	β	0.26	0.01	S-M		2
Carer Needs and coping																
Carer needs																
		Graap et al., 2008	UP	4	Number of Service-user problem areas	C	CNA	0	GHQ-12	0	*r*	0.615	0.001		2	
		Graap et al., 2008	UP	4	Number of needed ED interventions	C	CNA	0	GHQ-12	0	*r*	0.364	0.001		2	
	d	Haigh & Treasure, 2003	UP	4	Carer's need for information about eating	C	CaNAM	0	GHQ-12	0	r_s	-	>.05		3	
	d	Haigh & Treasure, 2003	UP	4	Carer's need for support for self	C	CaNAM	0	GHQ-12	0	r_s	-	>.05		3	
	d	Haigh & Treasure, 2003	UP	4	Carer's need for support from other	C	CaNAM	0	GHQ-12	0	r_s	-	>.05		3	
	b	Coomber & King, 2012	MP	2	Total needs	C	CaNAM	0	GHQ-12	0	β	0.03	>.05	L		
	b	Coomber & King, 2013	UP	3	Total needs	C	CaNAM	B	GHQ-12	4.5m	*r*	0.01	>.05		4	
	b	Coomber & King, 2013	UP	3	Total needs	C	CaNAM	B	GHQ-12	9m	*r*	0.06	>.05		4	
	b	Coomber & King, 2013	UP	3	Total needs	C	CaNAM	4.5m	GHQ-12	9m	*r*	-0.08	>.05		4	
	b	Coomber & King, 2013	MP	1	total carer needs	C	CaNAM	B	GHQ-12	4.5m, 9m	β	-	>.05	L		
	b	Coomber & King, 2013	MP	1	total carer needs	C	CaNAM	4.5m	GHQ-12	9m	β	-	>.05	L		

Predictors of Carer Distress Continued

Category	*Sample*	*Author*	*Type*	*Lv. Evidence*	*Predictor*	*Rater*	*Measure*	*Time*	*Distress measure*	*Time*	*Test*	*Statistic*	*p*	*Power*	# study comparisons	# sample comparisons
Carer coping																
		Ohara et al., 2016	UP	4	Task-oriented coping	C	CISS	0	GHQ-28 total & ss (A/I, SD)	0	*r*	-0.09 - -0.14	>.05		27	
		Ohara et al., 2016	UP	4	Avoidance-oriented coping	C	CISS	0	GHQ-28 total & ss (A/I, SD)	0	*r*	0.2 - 0.18	>.05		27	
		Ohara et al., 2016	UP	4	Emotion-oriented coping	C	CISS	0	GHQ-28 total & ss (A/I, SD)	0	*r*	0.55 - 0.59	<.001		27	
	b	Coomber & King, 2012	MP	2	Maladaptive coping	C	Brief COPE	0	GHQ-12	0	β: R	0.32 0.20	<.05	L		
		Ohara et al., 2016	MP	2	Emotion-oriented coping	C	CISS	0	GHQ-28	0	β	0.52	<.001	L		
	b	Coomber & King, 2013	UP	3	Maladaptive coping	C	Brief	B	GHQ-12	4.5m	*r*	0.34	<.05		4	12
	b	Coomber & King, 2013	UP	3	Maladaptive coping	C	Brief	B, 4.5m	GHQ-12	9m	*r*	0.23	>.05		4	12
	b	Coomber & King, 2013	MP	1	maladaptive coping;	C	Brief COPE;	B, 4.5m	GHQ-12	4.5m, 9m	β	-	>.05	L		
Protective factors																
Carer employment and education																
	a	Sepúlveda et al., 2012a	UP	4	Employment	C	SD-Q	0	HADS -A,D	0	*r*		>.05		15	22
	a	Sepúlveda et al., 2012a	UP	4	Highest educational level	SC	SD-Q	0	HADS-D	0	*r*	-0.24	0.01		15	22
	a	Sepúlveda et al., 2012a	MP	2	Highest educational level	PC	SD-Q	0	HADS-A	0	β	-0.14	<.05	S-M	4	7
	a	Sepúlveda et al., 2012a	MP	2	Highest educational level	PC	SD-Q	0	HADS-D	0	β	-0.18	<.05	S-M	4	7
	a	Sepúlveda et al., 2012a	MP	2	Highest educational level	SC	SD-Q	0	HADS-A	0	β		>.05	S-M	4	7
	a	Sepúlveda et al., 2012a	MP	2	Highest educational level	SC	SD-Q	0	HADS-D	0	β		>.05	S-M	4	7
	f	Orive et al., 2013	MP	2	Highest education: Secondary (vs none/primary)	C	SD-Q	B	HADS-D	B	β	0.28	0.7	M	4	6

Predictors of Carer Distress Continued

Category	*Sample*	*Author*	*Type*	*Lv. Evidence*	*Predictor*	*Rater*	*Measure*	*Time*	*Distress measure*	*Time*	*Test*	*Statistic*	*p*	*Power*	# study comparisons	# sample comparisons
Positive experiences																
	f	Orive et al., 2013	MP	2	Highest education: Univeristy (vs none/primary)	C	SD-Q	B	HADS-D	B	β	-1.41	0.03	M	4	6
	a	Sepúlveda et al., 2012a	UP	4	Positive experiences in caregiving [1]	PC, SC	ECI-ss	0	HADS-A, D	0	r	-0.09 - 0.09	>.05		15	22
		Winn et al., 2007	MP	2	Positive experiences of caregiving	C	ECI-ss	0	GHQ-12	0	*	0.11	0.133	M		
	a	Sepúlveda et al., 2012a	MP	2	Good relationship	PC, SC	ECI-ss	0	HADS-A	0	β	-0.18, -0.22	<.001	S-M	4	7
	a	Sepúlveda et al., 2012a	MP	2	Good relationship	PC, SC	ECI-ss	0	HADS-D	0	β	-	>.05	S-M	4	7
	a	Sepúlveda et al., 2012a	MP	2	Positive experience	PC	ECI-ss	0	HADS-A, D	0	β	-	>.05	S-M	4	7
	a	Sepúlveda et al., 2012a	MP	2	Positive experience	SC	ECI-ss	0	Secondary carer HADS-A	0	β	-	>.05	S-M	4	7
	a	Sepúlveda et al., 2012a	MP	2	Positive experience	SC	ECI-ss	0	Secondary carer HADS-D	0	β	-0.3	<.001	S-M	4	7
Support recieved																
		Rienecke, Richmond &	UP	4	Therapeutic alliance	C	WAI-SF	0	GSI	0	r_s	-	>.05		1	
		Dimitropoulos et al., 2008	UP	4	Professional support	C	PSI	0	GHQ-12	0	r	-0.04	>.05		10	
		Dimitropoulos et al., 2008	UP	4	Social support	C	SPS	0	GHQ-12	0	r	-0.12	>.05		10	
		Ohara et al., 2016	UP	4	Social contacts	C	SNQ	0	GHQ-28	0	r	-0.33	<.01		27	
		Ohara et al., 2016	UP	4	Social contacts	C	SNQ	0	GHQ-A/I	0	r	-0.28	<.05		27	
		Ohara et al., 2016	UP	4	Social contacts	C	SNQ	0	GHQ-SD	0	r	-0.25	<.05		27	
	e	Rhind et al., 2016	UP	4	Social Support	C	OSLO-3-	0	DASS	0	r_s	-0.29	<.05		17	36
		Ohara et al., 2016	UP	4	Practical social support recieved	C	SNQ	0	GHQ-28	0	r	-0.24	>.05		27	
		Ohara et al., 2016	UP	4	Practical social support recieved	C	SNQ-ss	0	GHQ-A/I	0	r	-0.17	>.05		27	
		Ohara et al., 2016	UP	4	Practical social support recieved	C	SNQ-ss	0	GHQ-SD	0	r	-0.23	>.05		27	
		Ohara et al., 2016	UP	4	Affective social support recieved	C	SNQ-ss	0	GHQ-28	0	r	-0.48	<.001		27	
		Ohara et al., 2016	UP	4	Affective social support recieved	C	SNQ-ss	0	GHQ-A/I	0	r	-0.33	<.01		27	

Predictors of Carer Distress Continued

Category	*Sample*	*Author*	*Type*	*Lv. Evidence*	*Predictor*	*Rater*	*Measure*	*Time*	*Distress measure*	*Time*	*Test*	*Statistic*	*p*	*Power*	*# study comparisons*	*# sample comparisons*
Support received continued																
		Ohara et al., 2016	UP	4	Affective social support recieved	C	SNQ-ss	0	GHQ-SD	0	*r*	-0.37	<.01		27	
	b	Coomber & King, 2012	MP	2	Satisfaction with social support	C	SSQ6	0	GHQ-12	0	β	-0.01	>.05	L		
		Ohara et al., 2016	MP	2	Affective social support recieved	C	SNQ-ss	0	GHQ-28	0	β	-0.42	<.001	L		
		Ohara et al., 2016	MP	2	Social contact	C	SNQ-ss	0	GHQ-28	0	β	-	>.05	L		
	b	Coomber & King, 2013	UP	3	Satisfaction with social support	C	SSQ6	B	GHQ-12	4.5m	*r*	-0.14	>.05		4	12
	b	Coomber & King, 2013	UP	3	Satisfaction with social support	C	SSQ6	B	GHQ-12	9m	*r*	-0.11	>.05		4	12
	b	Coomber & King, 2013	UP	3	Satisfaction with social support	C	SSQ6	4.5m	GHQ-12	9m	*r*	-0.12	>.05		4	12
	b	Coomber & King, 2013	MP	1	Satisfaction with social support;	C	SSQ6;	B	GHQ-12	4.5m, 9m	β	-	>.05	L		
	b	Coomber & King, 2013	MP	1	Satisfaction with social support;	C	SSQ6;	4.5m	GHQ-12	9m	β	-	>.05	L		
Carer Skills																
	e	Hibbs et al., 2015	UP	4	Carers' skills[1]	C	CASK	B	DASS-21	B	r_s	-0.38	<.01		19	36
	e	Hibbs et al., 2015	UP	4	Carers' skills[1]	C	CASK	B	GHQ-12	B	r_s	-0.51	<.01		19	36
	e	Rhind et al., 2016	UP	4	Carers' skills	C	CASK	B	DASS	B	r_s	-0.45	<.001		17	36
	e	Rhind et al., 2016	UP	4	Carers' skills	M	CASK	B	DASS	B	β	-0.42	<.001		17	36
	e	Rhind et al., 2016	UP	4	Carers' skills	F	CASK	B	DASS	B	β	-0.46	<.01		17	36

Note: Sample a-g; please see Table 6. Power: For multi-variate analysis, size of effect the study is powered to find is given. # study/sample comparisons: Number of univariate comparisons made within the same study or sample without correction of significance level, see Table 6. As cross-sectional community samples may present with meaningfully different experiences to those sampled at baseline of an intervention trial, timepoints of the latter are marked with 'B', and the former with '0'. Multivariate analysis of burden and expressed emotion as predictors of distress split by purging/non-purging subgroup by Sepulveda et al. (2014a) are not included in the table for clarity but are discussed below, analysis of these variables for the total sample are included. SDQ: Socio-demographic questionnaire. GHQ-A/I: GHQ Anxiety & Insomnia subscale. GHQ-SD: GHQ Severe Depression subscale.

[1] All subscales also tested with same significance and similar relationship. *: Coefficient from Huberwhite sandwich estimators of variance. PC: Primary carer. SC: Secondary carer.

Type: UP: Univariate predictor; MP: Multivariate predictor. Lv. Evidence: Level of evidence, please see text.

Appendix G: Ethical Approval

The original ethics form is presented in Appendix H. Multiple updates were made to approve the study for additional years, and minor changes to participant materials. The approval letters for these updates can be provided on request. I was granted Permission to use the archival data by the lead researcher Janet Treasure, and informed I did not need to be a named researcher. I made the Research and Development department aware of my involvement (by telephone, they did not require me to submit any written information).

Confirmation of this agreement was provided by email by Prof. Treasure:

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Appendix H: Approved Ethical Application for the Trial from which This Study Analysed Archival Data

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Appendix I: Letter of Ethical Approval for the Trial from which This Study Analysed Archival Data

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Appendix J: Mediation Analysis Tested with Variables in Alternative Positions

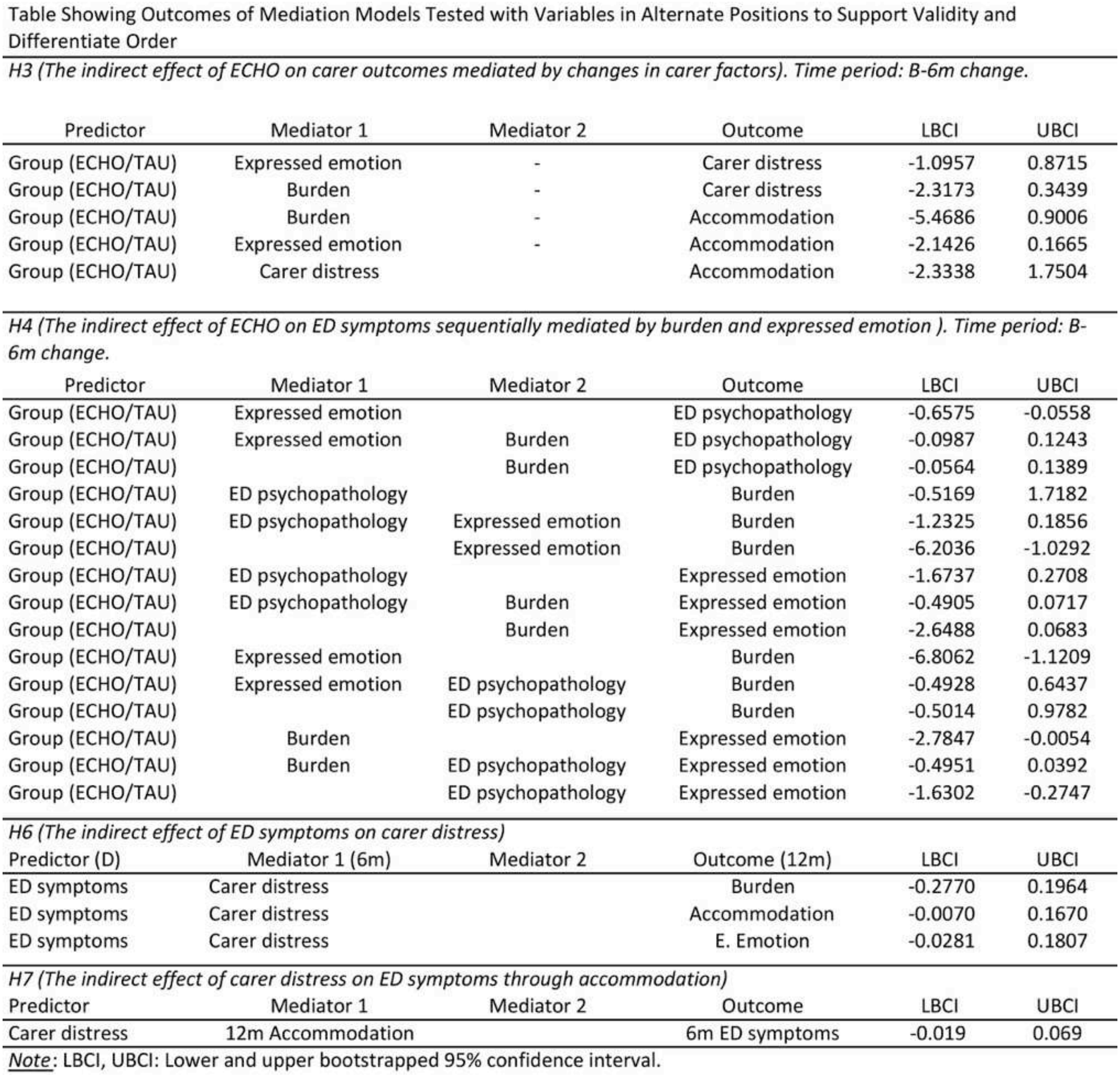


Table Showing Outcomes of Mediation Models Tested with Variables in Alternate Positions to Support Validity and Differentiate Order

H3 (The indirect effect of ECHO on carer outcomes mediated by changes in carer factors). Time period: B-6m change.

Predictor	Mediator 1	Mediator 2	Outcome	LBCI	UBCI
Group (ECHO/TAU)	Expressed emotion	-	Carer distress	-1.0957	0.8715
Group (ECHO/TAU)	Burden	-	Carer distress	-2.3173	0.3439
Group (ECHO/TAU)	Burden	-	Accommodation	-5.4686	0.9006
Group (ECHO/TAU)	Expressed emotion	-	Accommodation	-2.1426	0.1665
Group (ECHO/TAU)	Carer distress		Accommodation	-2.3338	1.7504

H4 (The indirect effect of ECHO on ED symptoms sequentially mediated by burden and expressed emotion). Time period: B-6m change.

Predictor	Mediator 1	Mediator 2	Outcome	LBCI	UBCI
Group (ECHO/TAU)	Expressed emotion		ED psychopathology	-0.6575	-0.0558
Group (ECHO/TAU)	Expressed emotion	Burden	ED psychopathology	-0.0987	0.1243
Group (ECHO/TAU)		Burden	ED psychopathology	-0.0564	0.1389
Group (ECHO/TAU)	ED psychopathology		Burden	-0.5169	1.7182
Group (ECHO/TAU)	ED psychopathology	Expressed emotion	Burden	-1.2325	0.1856
Group (ECHO/TAU)		Expressed emotion	Burden	-6.2036	-1.0292
Group (ECHO/TAU)	ED psychopathology		Expressed emotion	-1.6737	0.2708
Group (ECHO/TAU)	ED psychopathology	Burden	Expressed emotion	-0.4905	0.0717
Group (ECHO/TAU)		Burden	Expressed emotion	-2.6488	0.0683
Group (ECHO/TAU)	Expressed emotion		Burden	-6.8062	-1.1209
Group (ECHO/TAU)	Expressed emotion	ED psychopathology	Burden	-0.4928	0.6437
Group (ECHO/TAU)		ED psychopathology	Burden	-0.5014	0.9782
Group (ECHO/TAU)	Burden		Expressed emotion	-2.7847	-0.0054
Group (ECHO/TAU)	Burden	ED psychopathology	Expressed emotion	-0.4951	0.0392
Group (ECHO/TAU)		ED psychopathology	Expressed emotion	-1.6302	-0.2747

H6 (The indirect effect of ED symptoms on carer distress)

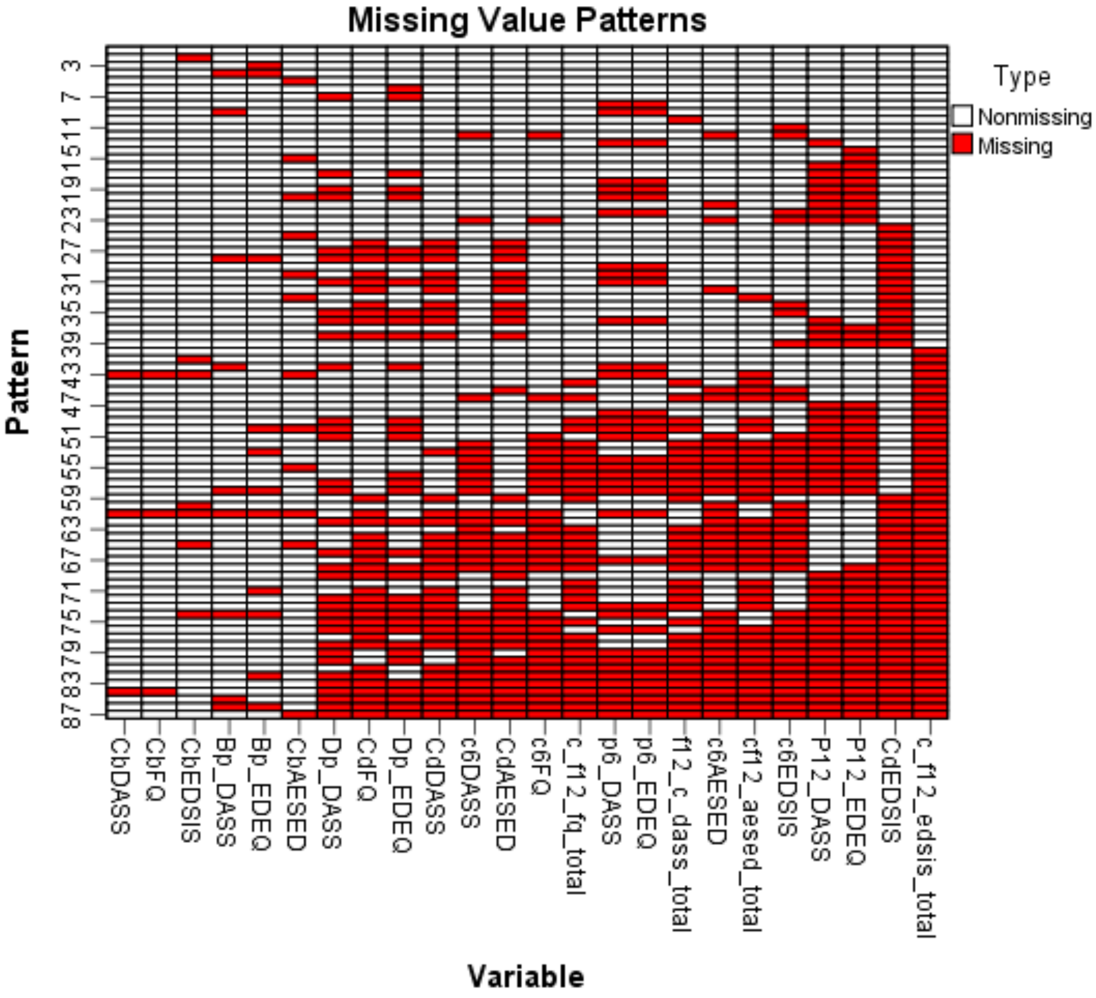
Predictor (D)	Mediator 1 (6m)	Mediator 2	Outcome (12m)	LBCI	UBCI
ED symptoms	Carer distress		Burden	-0.2770	0.1964
ED symptoms	Carer distress		Accommodation	-0.0070	0.1670
ED symptoms	Carer distress		E. Emotion	-0.0281	0.1807

H7 (The indirect effect of carer distress on ED symptoms through accommodation)

Predictor	Mediator 1	Mediator 2	Outcome	LBCI	UBCI
Carer distress	12m Accommodation		6m ED symptoms	-0.019	0.069

Note: LBCI, UBCI: Lower and upper bootstrapped 95% confidence interval.

Appendix K: SPSS Missing Value Analysis Showing Evidence of Questionnaire Data Not Missing At Random (MAR)



Appendix L: Proportion of Missing Data by Questionnaire and Time-Point

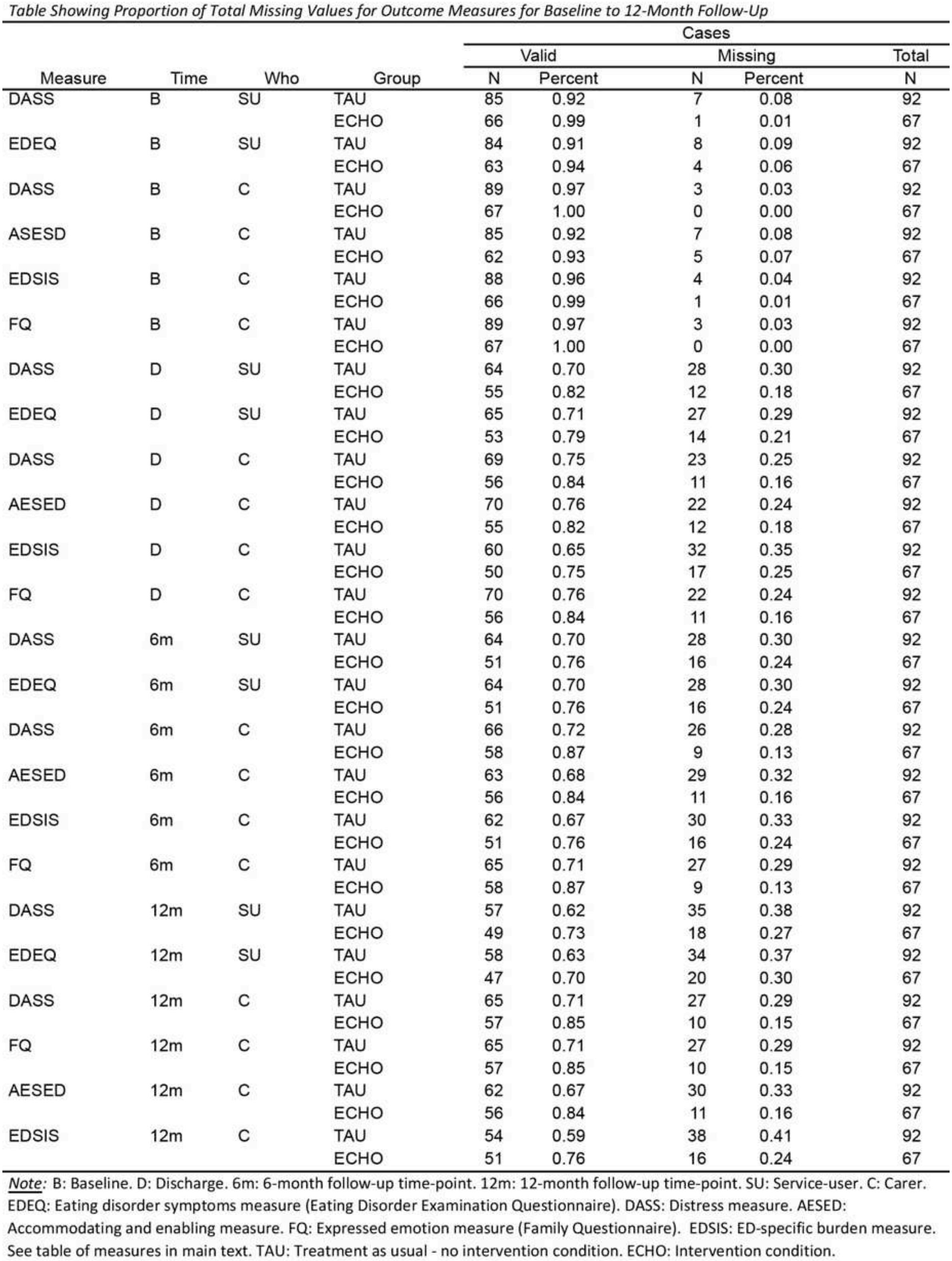


Table Showing Proportion of Total Missing Values for Outcome Measures for Baseline to 12-Month Follow-Up

				Cases				
				Valid		Missing		Total
Measure	Time	Who	Group	N	Percent	N	Percent	N
DASS	B	SU	TAU	85	0.92	7	0.08	92
			ECHO	66	0.99	1	0.01	67
EDEQ	B	SU	TAU	84	0.91	8	0.09	92
			ECHO	63	0.94	4	0.06	67
DASS	B	C	TAU	89	0.97	3	0.03	92
			ECHO	67	1.00	0	0.00	67
ASESD	B	C	TAU	85	0.92	7	0.08	92
			ECHO	62	0.93	5	0.07	67
EDSIS	B	C	TAU	88	0.96	4	0.04	92
			ECHO	66	0.99	1	0.01	67
FQ	B	C	TAU	89	0.97	3	0.03	92
			ECHO	67	1.00	0	0.00	67
DASS	D	SU	TAU	64	0.70	28	0.30	92
			ECHO	55	0.82	12	0.18	67
EDEQ	D	SU	TAU	65	0.71	27	0.29	92
			ECHO	53	0.79	14	0.21	67
DASS	D	C	TAU	69	0.75	23	0.25	92
			ECHO	56	0.84	11	0.16	67
AESED	D	C	TAU	70	0.76	22	0.24	92
			ECHO	55	0.82	12	0.18	67
EDSIS	D	C	TAU	60	0.65	32	0.35	92
			ECHO	50	0.75	17	0.25	67
FQ	D	C	TAU	70	0.76	22	0.24	92
			ECHO	56	0.84	11	0.16	67
DASS	6m	SU	TAU	64	0.70	28	0.30	92
			ECHO	51	0.76	16	0.24	67
EDEQ	6m	SU	TAU	64	0.70	28	0.30	92
			ECHO	51	0.76	16	0.24	67
DASS	6m	C	TAU	66	0.72	26	0.28	92
			ECHO	58	0.87	9	0.13	67
AESED	6m	C	TAU	63	0.68	29	0.32	92
			ECHO	56	0.84	11	0.16	67
EDSIS	6m	C	TAU	62	0.67	30	0.33	92
			ECHO	51	0.76	16	0.24	67
FQ	6m	C	TAU	65	0.71	27	0.29	92
			ECHO	58	0.87	9	0.13	67
DASS	12m	SU	TAU	57	0.62	35	0.38	92
			ECHO	49	0.73	18	0.27	67
EDEQ	12m	SU	TAU	58	0.63	34	0.37	92
			ECHO	47	0.70	20	0.30	67
DASS	12m	C	TAU	65	0.71	27	0.29	92
			ECHO	57	0.85	10	0.15	67
FQ	12m	C	TAU	65	0.71	27	0.29	92
			ECHO	57	0.85	10	0.15	67
AESED	12m	C	TAU	62	0.67	30	0.33	92
			ECHO	56	0.84	11	0.16	67
EDSIS	12m	C	TAU	54	0.59	38	0.41	92
			ECHO	51	0.76	16	0.24	67

Note: B: Baseline. D: Discharge. 6m: 6-month follow-up time-point. 12m: 12-month follow-up time-point. SU: Service-user. C: Carer. EDEQ: Eating disorder symptoms measure (Eating Disorder Examination Questionnaire). DASS: Distress measure. AESED: Accommodating and enabling measure. FQ: Expressed emotion measure (Family Questionnaire). EDSIS: ED-specific burden measure. See table of measures in main text. TAU: Treatment as usual - no intervention condition. ECHO: Intervention condition.

Appendix M: Socio-Demographics and Clinical Information for Excluded Vs. ECHO Included Group, with Tests of Difference

Table Showing Sociodemographics and Clinical Characteristics of Patients and Their Carers at Baseline, Continued

	Group				Statistics	
	ECHO 0: Non-compliant/msg (*n* = 19)		ECHO 1: Trmt. Compliant (*n* = 67)		ECHO 0 vs. 1	
	Carer	Patient	Carer	Patient	C	P
Relationship						
Relationship: n (%)					F = 7.42, *p* = .08	
Mother	14 (73.68)		57 (85.07)			
Father	1 (5.26)		0 (0.00)			
Partner/spouse	4 (21.05)		7 (10.45)			
Sibling/friend	0 (0.00)		3 (4.48)			
Living together: y/n (% yes)		13/5 (72.22)		43/23 (65.15)	C^2 = 0.32, *p* = .78	
Contact h/w: n (%)					*F* = 6.02, *p* = .26	
0-7		5 (26.32)		20 (29.85)		
8–14		2 (10.53)		13 (19.40)		
15–21		3 (15.79)		2 (2.99)		
>21		8 (42.11)		31 (46.27)		
Other		1 (5.26)		1 (1.49)		
Clinical characteristics						
Duration of illness, months: mean (SD)		133.27 (121.73)		81.87 (84.74)		U = 546.0, *p* = .13
BMI: mean (SD)		15.57 (2.84)		15.43 (2.35)		U = 574.0, *p*
Length of admission, days: mean (SD)		171.73 (120.41)		179.36 (135.83)		U = 618.5, p = .85
Compensatory vomiting: y/n (% yes)		6/12 (33.33)		19/46 (29.23)		C^2 = 0.72, *p* = .57
Comorbidity: n (%)						*F* = 0.64, *p* = .76
None		4 (25.00)		20 (3.33)		
Depression		11 (68.75)		34 (56.67)		
Anxiety		0 (0.00)		0 (0.00)		
OCD		1 (6.25)		6 (10.00)		
Borderline		0 (0.00)		0 (0.00)		
Carer ED Hx y/n (%yes)	5/14 (26.32)		20/47 (29.85)		C^2 = 0.09, *p* = .77	

Note: (%) Indicates percentage of respondents by group. Contact hours is face to face. E0: ECHO 0: Participants assigned to ECHO intervention who did not recieve any intervention, or for whom this information was missing. E1: ECHO 1: Participants who recieved some intervention. T: TAU: Treatment as usual. aUnemployed/sick/student/retired/homemaker/other. OCD: Obsessive Compulsive Disorder. Carer ED Hx: Carer has history of eating difficulties. SPSS only gives Fishers's Exact Test p-value for 2x2 tables.

Appendix N: Pre-Hoc Analysis of Group as a Moderator of Relationships Tested by Mediation Analysis

Table showing results of pre-hoc analysis of whether experimental group moderates any of the relationships involved in planned mediation analyses

H6: Results of 3 Pre-Hoc Regression Analyses using PROCESS Model 59. Evidence of Non-Significant Moderation of the Direct and Indirect Relationships Between ED Symptoms and Carer Distress by Group.

Mediator in model tested	Predictor	Outcome	LBCI	UBCI
Burden	ED symptoms	Burden	-3.325	5.621
	Burden	Carer distress	-0.849	0.836
	ED symptoms	Carer distress	-8.138	9.121
Moderation of indirect effect through mediator Burden			-3.842	7.942
AESED	ED symptoms	Accommodation	-13.002	1.368
	Accommodation	Carer distress	-0.554	0.570
	ED symptoms	Carer distress	-6.020	10.737
Moderation of indirect effect through mediator accommodation			-10.462	1.566
EE	ED symptoms	E. Emotion	-4.208	1.188
	E. Emotion	Carer distress	-1.687	0.964
	ED symptoms	Carer distress	-6.880	9.450
Moderation of indirect effect through mediator E. Emotion			-7.075	0.861

H7: Results of 3 Pre-Hoc Regression Analyses using PROCESS Model 59. Evidence of Group as a Non-Significant Moderator of the Relationships Between Carer Distress and Service-User ED symptoms

Mediator in model tested	Predictor	Outcome	LBCI	UBCI
E.Emotion & SU distress	Carer distress	Expressed emotion	-0.110	0.089
(sequential mediation)	Carer distress	SU distress	-0.248	0.530
	Expressed emotion	ED symptoms	-0.120	0.069
	SU distress	ED symptoms	-0.021	0.039
	Carer distress	ED symptoms	-0.020	0.015
Moderation of indirect effect through mediator E. Emotion			-0.005	0.013
Moderation of indirect effect through mediator P distress			-0.005	0.011

H8: Results of a Pre-Hoc Regression Analysis using PROCESS Model 59. Evidence of Group as a Non-Significant Moderator of the Relationship Between Carer Distress and Service-User ED symptoms

Mediator in model tested	Predictor	Outcome	LBCI	UBCI
Accommodation	Carer distress	Accommodation	-0.428	0.625
	Accommodation	ED symptoms	-0.054	0.036
	Carer distress	ED symptoms	-0.029	0.044
Moderation of indirect effect through mediator Accommodation			-0.025	0.021

Note: E. Emotion: Expressed emotion. LBCI, UBCI: Lower and upper bootstrapped 95% confidence interval. SU: Service-user.

Appendix O: Examples of PROCESS Statistical Readouts

Simple Moderation Analysis

```
Run MATRIX procedure:

************** PROCESS Procedure for SPSS Release 2.15 ******************

          Written by Andrew F. Hayes, Ph.D.       www.afhayes.com
    Documentation available in Hayes (2013). www.guilford.com/p/hayes3

**************************************************************************
Model = 1
    Y = cB6FQ
    X = GPCYN9
    M = N_C_0or1

Sample size
        122

**************************************************************************
Outcome: cB6FQ

Model Summary
          R       R-sq        MSE          F        df1        df2
p
      .3454      .1193    49.1035     4.8742     3.0000   118.0000
.0031

Model
              coeff         se          t          p       LLCI       ULCI
constant    -2.0466     1.1999    -1.7056      .0907    -4.4228      .3296
N_C_0or1     3.8273     1.8493     2.0695      .0407      .1651     7.4894
GPCYN9       -.4829     1.8061     -.2674      .7897    -4.0595     3.0937
int_1       -6.6251     2.5950    -2.5530      .0120   -11.7639    -1.4862

Product terms key:

 int_1    GPCYN9      X     N_C_0or1

R-square increase due to interaction(s):
         R2-chng          F        df1        df2          p
int_1      .0500     6.5177     1.0000   118.0000      .0120

*************************************************************************

Conditional effect of X on Y at values of the moderator(s):
   N_C_0or1      Effect           se            t            p        LLCI
ULCI
      .0000       -.4829       1.8061       -.2674        .7897      -4.0595
3.0937
     1.0000     -7.1079      1.8634     -3.8146       .0002    -10.7979     -
3.4180
```

```
***************************************************************************

Data for visualizing conditional effect of X on Y
Paste text below into a SPSS syntax window and execute to produce plot.

DATA LIST FREE/GPCYN9 N_C_0or1 cB6FQ.
BEGIN DATA.

      .0000      .0000    -2.0466
     1.0000      .0000    -2.5295
      .0000     1.0000     1.7806
     1.0000     1.0000    -5.3273

END DATA.
GRAPH/SCATTERPLOT=N_C_0or1 WITH cB6FQ BY GPCYN9.

******************** ANALYSIS NOTES AND WARNINGS *************************

Level of confidence for all confidence intervals in output:
    95.00

NOTE: Some cases were deleted due to missing data.  The number of such
cases was:
  37

NOTE: All standard errors for continuous outcome models are based on the
HC3 estimator

------ END MATRIX -----
```

Simple Mediation Analysis

```
Run MATRIX procedure:

************** PROCESS Procedure for SPSS Release 2.15 ******************

          Written by Andrew F. Hayes, Ph.D.       www.afhayes.com
    Documentation available in Hayes (2013). www.guilford.com/p/hayes3

***************************************************************************
Model = 4
    Y = B6EDEQ
    X = GPCYN9
    M = cB6FQ

Sample size
         90

***************************************************************************
Outcome: cB6FQ

Model Summary
          R       R-sq        MSE          F        df1        df2
p
      .3046      .0928    53.6936     8.8162     1.0000    88.0000
.0038

Model
              coeff         se          t          p       LLCI       ULCI
```

```
constant      .0270     1.0919      .0247      .9803    -2.1429     2.1970
GPCYN9      -4.6387     1.5623    -2.9692      .0038    -7.7434    -1.5340

Covariance matrix of regression parameter estimates
           constant     GPCYN9
constant     1.1923    -1.1923
GPCYN9      -1.1923     2.4407

**************************************************************************
Outcome: B6EDEQ

Model Summary
          R         R-sq          MSE            F          df1          df2
p
      .2417        .0584       1.8037       2.7547       2.0000      87.0000
.0692

Model
              coeff         se          t          p       LLCI       ULCI
constant     -.5006      .1818    -2.7534      .0072     -.8620     -.1392
cB6FQ         .0412      .0184     2.2375      .0278      .0046      .0777
GPCYN9       -.0862      .2930     -.2941      .7694     -.6685      .4962

Covariance matrix of regression parameter estimates
           constant      cB6FQ     GPCYN9
constant      .0331      .0001     -.0324
cB6FQ         .0001      .0003      .0010
GPCYN9       -.0324      .0010      .0858

************************** TOTAL EFFECT MODEL ****************************
Outcome: B6EDEQ

Model Summary
          R         R-sq          MSE            F          df1          df2
p
      .1017        .0104       1.8742        .8786       1.0000      88.0000
.3511

Model
              coeff         se          t          p       LLCI       ULCI
constant     -.4995      .1744    -2.8635      .0052     -.8462     -.1528
GPCYN9       -.2772      .2957     -.9374      .3511     -.8648      .3104

Covariance matrix of regression parameter estimates
           constant     GPCYN9
constant      .0304     -.0304
GPCYN9       -.0304      .0874

***************** TOTAL, DIRECT, AND INDIRECT EFFECTS ********************

Total effect of X on Y
     Effect         SE          t          p       LLCI       ULCI
     -.2772      .2957     -.9374      .3511     -.8648      .3104

Direct effect of X on Y
     Effect         SE          t          p       LLCI       ULCI
     -.0862      .2930     -.2941      .7694     -.6685      .4962

Indirect effect of X on Y
          Effect    Boot SE   BootLLCI   BootULCI
cB6FQ     -.1910      .1050     -.4502     -.0471
```

```
Partially standardized indirect effect of X on Y
          Effect    Boot SE   BootLLCI   BootULCI
cB6FQ     -.1396      .0746     -.3229     -.0335

Completely standardized indirect effect of X on Y
          Effect    Boot SE   BootLLCI   BootULCI
cB6FQ     -.0701      .0373     -.1623     -.0170

Ratio of indirect to total effect of X on Y
          Effect    Boot SE   BootLLCI   BootULCI
cB6FQ      .6891     6.4334     -.2288    50.7464

Ratio of indirect to direct effect of X on Y
          Effect    Boot SE   BootLLCI   BootULCI
cB6FQ     2.2163    22.2416      .7899   201.1036

R-squared mediation effect size (R-sq_med)
          Effect    Boot SE   BootLLCI   BootULCI
cB6FQ      .0094      .0166     -.0123      .0593

Preacher and Kelley (2011) Kappa-squared
          Effect    Boot SE   BootLLCI   BootULCI
cB6FQ      .0678      .0354      .0170      .1544

Normal theory tests for indirect effect
     Effect         se          Z          p
     -.1910      .1107    -1.7256      .0844

******************** ANALYSIS NOTES AND WARNINGS *************************

Number  of  bootstrap  samples  for  bias  corrected  bootstrap  confidence
intervals:
     1000

Level of confidence for all confidence intervals in output:
    95.00

NOTE: Some cases were deleted due to missing data.  The number of such
cases was:
  69

NOTE: All standard errors for continuous outcome models are based on the
HC3 estimator

------ END MATRIX -----
```

Moderated Mediation (Pre-Hoc)

```
Run MATRIX procedure:

************** PROCESS Procedure for SPSS Release 2.15 *******************

          Written by Andrew F. Hayes, Ph.D.       www.afhayes.com
    Documentation available in Hayes (2013). www.guilford.com/p/hayes3

**************************************************************************
Model = 59
    Y = f12_c_da
```

```
    X = Dp_EDEQ
    M = c6EDSIS
    W = GPCYN9

Sample size
         80

**************************************************************************
Outcome: c6EDSIS

Model Summary
          R         R-sq          MSE            F          df1          df2
p
      .3843        .1476     199.7372       5.3325       3.0000      76.0000
.0022

Model
              coeff         se          t          p       LLCI       ULCI
constant    25.6748     5.9251     4.3332      .0000    13.8739    37.4757
Dp_EDEQ      2.7561     1.6138     1.7079      .0917     -.4580     5.9702
GPCYN9     -11.7066     8.1477    -1.4368      .1549   -27.9342     4.5211
int_1        1.1482     2.2457      .5113      .6106    -3.3245     5.6208

Product terms key:

 int_1    Dp_EDEQ     X     GPCYN9

**************************************************************************
Outcome: f12_c_da

Model Summary
          R         R-sq          MSE            F          df1          df2
p
      .5826        .3394     514.0921       6.3542       5.0000      74.0000
.0001

Model
              coeff         se          t          p       LLCI       ULCI
constant    -1.3778    10.0199     -.1375      .8910   -21.3430    18.5873
c6EDSIS      1.1258      .2797     4.0257      .0001      .5686     1.6830
Dp_EDEQ     -3.1160     2.9195    -1.0673      .2893    -8.9332     2.7011
int_2        -.0066      .4226     -.0156      .9876     -.8487      .8355
GPCYN9      10.0082    18.4767      .5417      .5897   -26.8076    46.8241
int_3         .4918     4.3309      .1135      .9099    -8.1379     9.1214

Product terms key:

 int_2    c6EDSIS     X     GPCYN9
 int_3    Dp_EDEQ     X     GPCYN9

******************** DIRECT AND INDIRECT EFFECTS *************************

Conditional direct effect(s) of X on Y at values of the moderator(s):
     GPCYN9      Effect            SE            t            p         LLCI
ULCI
      .0000      -3.1160       2.9195      -1.0673        .2893      -8.9332
2.7011
     1.0000      -2.6243       3.1990       -.8203        .4147      -8.9985
3.7499

Conditional indirect effect(s) of X on Y at values of the moderator(s):
```

```
Mediator
            GPCYN9     Effect    Boot SE   BootLLCI   BootULCI
c6EDSIS      .0000     3.1029     1.8643      .2866     8.4504
c6EDSIS     1.0000     4.3697     2.2509     1.1403    10.5919

Values for quantitative moderators are the mean and plus/minus one SD from
mean.
Values for dichotomous moderators are the two values of the moderator.

******************* INDEX OF MODERATED MEDIATION ***********************

Mediator
             Index   SE(Boot)   BootLLCI   BootULCI
c6EDSIS     1.2669     2.9286    -3.8416     7.9415

When the moderator is dichotomous, this is a test of equality of the
conditional indirect effects in the two groups.

******************* ANALYSIS NOTES AND WARNINGS ************************

Number  of  bootstrap  samples  for  bias  corrected  bootstrap  confidence
intervals:
     1000

Level of confidence for all confidence intervals in output:
    95.00

NOTE: Some cases were deleted due to missing data.  The number of such
cases was:
  79

NOTE: All standard errors for continuous outcome models are based on the
HC3 estimator

------ END MATRIX -----
```

Sequential Mediation Analysis with Co-Variates

```
Run MATRIX procedure:

************** PROCESS Procedure for SPSS Release 2.15 ******************

          Written by Andrew F. Hayes, Ph.D.       www.afhayes.com
    Documentation available in Hayes (2013). www.guilford.com/p/hayes3

**************************************************************************
Model = 6
    Y = P12_EDEQ
    X = CbDASS
   M1 = CdFQ
   M2 = p6_DASS

Statistical Controls:
CONTROL= GPCYN9   Bp_EDEQ  Ceat_Hx

Sample size
         73

**************************************************************************
Outcome: CdFQ
```

```
Model Summary
          R         R-sq          MSE            F          df1          df2
p
      .4605        .2120      75.3609       5.0151       4.0000      68.0000
.0013

Model
              coeff         se          t          p       LLCI       ULCI
constant    32.1887     6.8291     4.7135      .0000    18.5614    45.8159
CbDASS        .1185      .0292     4.0597      .0001      .0603      .1768
GPCYN9        .7228     2.1308      .3392      .7355    -3.5293     4.9748
Bp_EDEQ      2.0225     1.0079     2.0067      .0488      .0113     4.0338
Ceat_Hx      1.1105     2.5377      .4376      .6631    -3.9534     6.1743

**************************************************************************
Outcome: p6_DASS

Model Summary
          R         R-sq          MSE            F          df1          df2
p
      .5114        .2615     785.2562       5.0932       5.0000      67.0000
.0005

Model
              coeff         se          t          p       LLCI       ULCI
constant    15.0430    26.0933      .5765      .5662   -37.0396    67.1257
CdFQ          .9011      .4725     1.9072      .0608     -.0420     1.8442
CbDASS        .1183      .1643      .7202      .4739     -.2097      .4464
GPCYN9      -2.3103     6.9499     -.3324      .7406   -16.1825    11.5618
Bp_EDEQ      6.3619     3.2338     1.9673      .0533     -.0928    12.8166
Ceat_Hx    -11.6510     9.4930    -1.2273      .2240   -30.5991     7.2972

**************************************************************************
Outcome: P12_EDEQ

Model Summary
          R         R-sq          MSE            F          df1          df2
p
      .6652        .4425       1.7037      17.3263       6.0000      66.0000
.0000

Model
              coeff         se          t          p       LLCI       ULCI
constant     1.4573     1.2468     1.1689      .2467    -1.0320     3.9466
CdFQ         -.0114      .0188     -.6044      .5477     -.0490      .0262
p6_DASS       .0189      .0061     3.1086      .0028      .0067      .0310
CbDASS        .0078      .0065     1.1889      .2387     -.0053      .0208
GPCYN9       -.1640      .3286     -.4992      .6193     -.8201      .4920
Bp_EDEQ       .4961      .1318     3.7627      .0004      .2329      .7593
Ceat_Hx      -.5694      .4706    -1.2099      .2306    -1.5091      .3702

************************** TOTAL EFFECT MODEL ****************************
Outcome: P12_EDEQ

Model Summary
          R         R-sq          MSE            F          df1          df2
p
      .5907        .3489       1.9313      22.8364       4.0000      68.0000
.0000
```

```
Model
              coeff         se          t          p       LLCI       ULCI
constant     1.9216     1.0997     1.7474      .0851     -.2728     4.1160
CbDASS        .0107      .0078     1.3588      .1787     -.0050      .0263
GPCYN9       -.2036      .3405     -.5978      .5519     -.8830      .4759
Bp_EDEQ       .6274      .1249     5.0236      .0000      .3782      .8767
Ceat_Hx      -.7830      .4590    -1.7060      .0926    -1.6989      .1329

***************** TOTAL, DIRECT, AND INDIRECT EFFECTS ********************

Total effect of X on Y
     Effect         SE          t          p       LLCI       ULCI
      .0107      .0078     1.3588      .1787     -.0050      .0263

Direct effect of X on Y
     Effect         SE          t          p       LLCI       ULCI
      .0078      .0065     1.1889      .2387     -.0053      .0208

Indirect effect(s) of X on Y
           Effect    Boot SE   BootLLCI   BootULCI
Total:      .0029      .0040     -.0047      .0114
Ind1 :     -.0014      .0024     -.0067      .0029
Ind2 :      .0020      .0013      .0004      .0060
Ind3 :      .0022      .0031     -.0026      .0100
(C1)       -.0034      .0029     -.0105      .0011
(C2)       -.0036      .0037     -.0123      .0027
(C3)       -.0002      .0035     -.0076      .0066

Partially standardized indirect effect of X on Y
           Effect    Boot SE   BootLLCI   BootULCI
Total:      .0021      .0028     -.0038      .0078
Ind1 :     -.0010      .0017     -.0049      .0022
Ind2 :      .0014      .0009      .0003      .0039
Ind3 :      .0016      .0021     -.0023      .0066

Completely standardized indirect effect of X on Y
           Effect    Boot SE   BootLLCI   BootULCI
Total:      .0583      .0777     -.0865      .2230
Ind1 :     -.0272      .0470     -.1430      .0551
Ind2 :      .0405      .0234      .0093      .1064
Ind3 :      .0449      .0594     -.0526      .1955

Ratio of indirect to total effect of X on Y
           Effect    Boot SE   BootLLCI   BootULCI
Total:      .2719    12.4007    -2.0091     2.4812
Ind1 :     -.1267    17.8091    -4.7442      .5605
Ind2 :      .1891     9.0972     -.4880     3.2449
Ind3 :      .2095     3.4981     -.8769     2.2700

Ratio of indirect to direct effect of X on Y
           Effect    Boot SE   BootLLCI   BootULCI
Total:      .3734    12.6594    -1.8410    15.4617
Ind1 :     -.1740     5.2400    -2.9971     1.5788
Ind2 :      .2597     5.4094     -.8301     8.5684
Ind3 :      .2877     7.1101    -1.2360     7.5406

Indirect effect key
 Ind1 :   CbDASS   ->       CdFQ     ->       P12_EDEQ
 Ind2 :   CbDASS   ->       CdFQ     ->       p6_DASS  ->       P12_EDEQ
 Ind3 :   CbDASS   ->       p6_DASS  ->       P12_EDEQ
```

```
Specific indirect effect contrast definitions
(C1)   Ind1       minus      Ind2
(C2)   Ind1       minus      Ind3
(C3)   Ind2       minus      Ind3

******************* ANALYSIS NOTES AND WARNINGS ************************

Number  of  bootstrap  samples  for  bias  corrected  bootstrap  confidence
intervals:
     1000

Level of confidence for all confidence intervals in output:
    95.00

NOTE: Some  cases  were  deleted  due  to  missing  data.   The  number  of  such
cases was:
  86

NOTE: All  standard  errors  for  continuous  outcome  models  are  based  on  the
HC3 estimator

------ END MATRIX -----
```

Appendix P: Information for Submission to International Journal of Eating Disorders

Author Guidelines

ORIGINALITY

The journal accepts for review manuscripts that have not been published or are not currently elsewhere under review.

CONTENT TYPES

Manuscripts published in IJED include: (1) Original Articles; (2) Brief Reports; (3) Reviews (systematic reviews and meta-analyses); (4) Commentaries; (5) Clinical Case Reports; and (6) “An Idea Worth Researching". All word limits relate to the body of the text (i.e., not including abstract, references, tables and figures) and represent maximum lengths. Authors are encouraged to keep their manuscript as short as possible while communicating clearly.

When uploading their manuscript, authors will be asked to complete a checklist indicating that they have followed the Author Guidelines pertaining to the appropriate article type.

To summarize, the article types are:

(1) Original Articles report substantive research that is novel, definitive or complex enough to require a longer communication. Only a subset of research papers are expected to warrant full length format.

- Word Limit: 4,000 (excluding abstract, references, tables or figures)
- Abstract: 250 words.
- References: 60 are recommended; more are permissible, for cause.
- Figures/Tables: a maximum of 8 essential tables/figures, overall.

The methods section should include a statement about sample selection, response rate, and other factors that would impact selection or response bias and, in turn, representativeness of the sample. Inclusion of small samples requires justification and authors should be mindful of the recommendations concerning minimal sample sizes in subfields (e.g., genetic research, instrument development, etc., where adequate samples may number in the hundreds). Authors also are asked to provide information about reliability and validity of study measures as applicable to their sample.

If the study involves qualitative data, authors need to include a statement about sample size in relation to theme saturation. We recommend that authors review the BMJ checklist for studies involving qualitative methods and conduct and report their analyses accordingly.

If the work involves cross-cultural assessment or assessment in a new language or study population, authors should provide information about local literacy in the language of assessment, the validity of (or process for validating) a translation of an assessment, and for inclusion of regional samples, a statement about the representativeness of the regional sample (or distinction from) the national sample. If statistical analyses are employed, effect size estimates should be reported in the results section.

(2) Brief Research Reports. This contribution type is intended for manuscripts describing studies with straightforward research designs, pilot or "proof of concept" studies, and replications. Authors are advised that the instructions regarding sample description and, if applicable, description of qualitative methods or cultural assessments provided for Original Articles (see above) also apply to Brief Reports.

- Word Limit: 1,500 (excluding abstract, references, tables or figures).
- Abstract: 200 words.
- References: 20 are recommended; more are permissible, for cause.
- Figures/Tables: a maximum of 2 essential tables/figures, overall.

If statistical analyses are employed, effect size estimates should be reported in the results section.

(3) Review articles critically review the status of a given research area and propose new directions for research and/or practice. Both systematic and meta-analytic review papers are welcomed if they review a literature that is advanced and/or developed to the point of warranting a review and synthesis of existing studies. Reviews of topics with a limited number of studies are unlikely to be deemed as substantive enough for a Review paper. The journal does not accept papers that merely describe or compile a list of previous studies without a critical synthesis of the literature that moves the field the forward.

- Word Limit: 7,000 (excluding abstract, references, tables or figures).
- Abstract: 250 words.
- References: 100.
- Figures/Tables: no maximum, but should be appropriate to the material covered.

All Review articles must follow the PRISMA Guidelines (www.prisma-statement.org), summarized in a 2009 *J. Clin. Epidemiol.* article by Moher et al. entitled "*Preferred Reporting Items for Systematic Reviews and Meta-Analyses: The PRISMA Statement*" (DOI: 10.1016/j.jclinepi.2009.06.005), freely available for download in both English and Spanish.

Authors who choose this contribution type must complete the Review Checklist upon submission of the manuscript, an example of which can be found here. This example is for informational purposes only. During the submission process, Authors will be prompted to complete the Review Checklist directly in ScholarOne. The rationale for any unchecked items on the Review Checklist must be explicitly described in the accompanying Cover Letter.

(4) Commentaries are solicited by the Editors when multiple perspectives on or critical appraisal of an article would assist in placing that article in context. Unsolicited commentaries are not accepted.

- Word Limit: 1,500 (excluding abstract, references, tables or figures).
- Abstract: no abstract.
- References: 5, using the footnote format rather than the journal's standard format.
- Figures/Tables: none.

(5) Clinical Case Reports detail key elements of cases where there is novelty in the presentation, pathology or treatment, and where that novelty will inform clinicians and researchers about rare presentations or novel ideas. This category will often be appropriate to rare biological or psychological presentations. Reports of rigorously conducted studies employing single-case experimental designs are especially welcome.

Every effort should be taken to ensure the anonymity of the patient concerned, and any clinicians not involved as authors. If there is any potentially identifiable information, then it is the responsibility of the authors to obtain approval from the local Institutional Review Board (IRB) (or equivalent) for the case to be reported, and a copy of that approval should be made available to the Editor on request.

- Word Limit: 1,500 (excluding abstract, references, tables or figures).
- Abstract: 150 words.
- References: 20.
- Figures/Tables: a maximum of 2 essential tables/figures, overall.

(6) "An idea Worth Researching" is a contribution type where authors propose an idea that may not yet have adequate empirical support or be ready for full empirical testing, but holds great promise for advancing research of eating disorders. Authors are encouraged to write a piece that is bold, forward looking, and suggestive of new and exciting avenues for research and/or practice in the field.

- Word Limit: 1,500 (excluding abstract, references, tables or figures).
- Abstract: no abstract.
- References: 5 maximum, in footnote format.
- Figures/Tables: a maximum of 2 essential tables/figures, overall

MANUSCRIPT PREPARATION & FORMAT

Speaking of That: Terms to Avoid or Reconsider

Authors should refrain from using terms that are stigmatizing or terms that are ambiguous. For further explanation and examples, see the 2016 IJED article by Weissman et al. entitled "*Speaking of that: Terms to avoid or reconsider in the eating disorders field*" (DOI: 10.1002/eat.22528).

General Format

Manuscripts must be typed in English and double-spaced throughout, with margins of at least one inch at the top, bottom, and both sides of each page. Please use line numbers, restarting the numbering of lines on each page. All manuscripts are subject to copyediting; however, it is the primary responsibility of the authors to proofread thoroughly and ensure correct spelling and punctuation, completeness and accuracy of references, clarity of expression, thoughtful construction of sentences, and legible appearance prior to the manuscript's submission. Preferred spelling follows Webster's New Collegiate Dictionary or Webster's Third New International Dictionary. The manuscript should conform to accepted English usage and syntax. Use headings to indicate the manuscript's general organization. Do not use a heading for the introduction. In general, manuscripts will contain one of several levels of headings. Centered upper case headings are reserved for Methods, Results, and Discussion sections of the manuscript. Subordinate headings (e.g., the Participants or Procedure subsection of Methods) are typed flush left, underlined, in upper case and lower case letters. The text begins a new paragraph. Number all pages of the manuscript except the figures (including title page and abstract) consecutively. Manuscripts that do not conform to the Author Guidelines stated here will not be considered further. Number all pages of the manuscript except the figures (including title page and abstract) consecutively.

Parts of the manuscripts should be arranged in the following sequence:

(1) Title page. (numbered 1). Titles should be short and specific, conveying the main point of the article. When developing the title (and abstract), authors are encouraged to review tips for improving search engine optimization (SEO) to ensure that their articles are highly visible to potential readers. Tips on SEO are given here; visit www.wileyauthors.com for more helpful hints for authors. The title page should include the full names, titles, and affiliations of all authors, and an abbreviated title (Running Head) that should not exceed 50 characters, counting letters, spacing, and punctuation. The Running Head should be typed in upper case letters centered at the bottom of the title page. Each page of the manuscript (excluding figures) should be identified by typing the first two or three words of the full title in the upper right-hand corner above the page number. No running head is required for letters to the editor. Indicate the word count for the abstract and the word count for the manuscript (excluding figures, tables, and references).

(2) Abstract. The word maximum and abstract format varies by contribution type (see above). When an abstract is required, the abstract should be typed as a single paragraph on a separate page, numbered 2. Type the word "Abstract" in upper and lower case letters, centered at the top of page 2. Provide the following information in the form of a structured abstract, using these headings: **Objective**: briefly indicate the primary purpose of the article, or major question addressed in the study. **Method**: indicate the sources of data, give brief overview of methodology, or, if review article, how the literature was searched and articles selected for discussion. For research based articles, this section should briefly note study design, how participants were selected, and major study measures. **Results**: summarize the key findings. **Discussion**: indicate main clinical, theoretical, or research applications/implications. The journal requires structured abstracts with two exceptions: the journal will continue to use unstructured abstracts for Clinical Case Reports, and no abstract is required for "An Idea Worth Researching".

(3) Text. Begin the text on page 3 and be sure to identify each page with the short title typed in the upper right-hand corner above the page number. Type the full title of the manuscript centered at the top, and then begin the text. The full title appears on page 3 only. Indent all paragraphs. The maximum length for article submissions is specified for each manuscript type. Authors are advised that content be conveyed as concisely as possible.

(4) References. Begin on separate page, with the word "References" typed in upper and lower case letters, centered at the top of the page. References must be double spaced.

(5) Appendices. Type each appendix on a separate page labeled "Appendix A, B", etc., in the order in which they are mentioned in the text.

(6) Footnotes. Start on separate page.

(7) Tables. Tables should be double-spaced, including all headings, and should have a descriptive title. If a table extends to another page, so should all titles and headings. Each table should be numbered sequentially in Arabic numerals and begin on a new page. Be sure to explain abbreviations in tables even if they have already been explained in-text. Consider the tables and figures to be self-contained and independent of the text. They should be interpretable as stand-alone entities.

(8) Figure captions. Start on separate page. Each figure caption should have a brief title that describes the entire figure without citing specific panels, followed by a description of each panel. Figure captions should be included in the submitted manuscript as a separate section. Be sure to explain abbreviations in figures even if they have already been explained in-text. Consider the tables and figures to be self-contained and independent of the text. They should be interpretable as stand-alone entities. Axes for figures must be labeled with appropriate units of measurement and description.

(9) Acknowledgements/Disclosure of Conflicts. Start on a separate page. Any possible conflict of interest, financial or otherwise, related to the submitted work must be clearly indicated in the manuscript. Acknowledge significant contributions that do not warrant authorship; list sources of support (e.g., federal, industry, or other funding).

Informed Consent

The Methods section should include a statement that the research was reviewed and approved by an institutional review board, and that participation involved informed consent.

Every effort should be taken to ensure the anonymity of the patient concerned, and any clinicians not involved as authors. If there is any potentially identifiable information, then it is the responsibility of the authors to seek and obtain approval from the local Institutional Review Board (IRB) (or equivalent) for the case to be reported, and a copy of that approval should be made available to the Editor on request.

Presenting Statistical Data in Text

For additional detail regarding statistical requirements for the manuscript, see IJED Statistical Formatting Requirements. For more detailed background information on statistical analyses and their rationale authors are referred to IJED Statistical Reporting Guidelines.

Manuscripts reporting statistical tests without effect size estimates may be rejected without review.

References

Wiley's Journal Styles Are Now in EndNote (Wiley's Journal Styles and EndNote) . EndNote is a software product that we recommend to our journal authors to help simplify and streamline the research process. Using EndNote's bibliographic management tools, you can search bibliographic databases, build and organize your reference collection, and then instantly output your bibliography in any Wiley journal style. If you already use EndNote, you can download the reference style for this journal. To learn more about EndNote, or to purchase your own copy, click here . If you need assistance using EndNote, contact endnote@isiresearchsoft.com , or visit www.endnote.com/support.

Except as noted for Commentaries and "Ideas Worth Researching", referencing follows the Vancouver method of reference citation. In this system, references are numbered consecutively in the order in which they are first mentioned in the text. Identify each reference in text, tables, and legends by Arabic numbers. All references cited should be listed numerically at the end of the paper. Prepare citations according to the style used in Index Medicus and the International list of periodical title word abbreviations (ISO 833).

All reference citations in the text should appear in the reference list. When there are less than seven authors, each must be listed in the citation. When seven or more authors, list the first six followed by et al. after the name of the sixth author. Representative examples are as follows:

Journal Article: 1. Endicott J, Spitzer RL. A diagnostic interview: The schedule for affective disorders and schizophrenia. Arch Gen Psychiatry 1978;35:837-844.

Book Chapter: 2. Fairburn CG, Cooper Z. The eating disorders examination (12th ed). In: Fairburn CG, Wilson GT, editors. Binge eating: nature, assessment, and treatment. New York: The Guilford Press, 1993, p. 317-331.

Book: 3. Tudor I. Learner-centeredness as language education. Cambridge: Cambridge University Press; 1996.

Preparation of Figures

To ensure the highest quality print production, your figures must be submitted in TIFF format according to the following minimum resolutions:

- 1200 dpi (dots per inch) for black and white line art (simple bar graphs, charts, etc.)
- 300 dpi for halftones (black and white photographs)
- 600 dpi for combination halftones (photographs that also contain line art such as labeling or thin lines)

Vector-based figures (usually created in Adobe Illustrator) should be submitted as EPS. Do not submit figures in the following formats: JPEG, GIF, Word, Excel, Lotus1-2-3, PowerPoint, PDF.

Graphs must show an appropriate grid scale. Each axis must be labeled with both the quantity measured and the unit of measurement. Color figures must be submitted in a CMYK colorspace. Do not submit files as RGB. All color figures will be reproduced in full color in the online edition of the journal at no cost to authors. Authors are requested to pay the cost of reproducing color figures in print. Authors are encouraged to submit color illustrations that highlight the text and convey essential scientific information. For best reproduction, bright, clear colors should be used.

Supplementary Materials

Supplementary materials will be made available to readers as a link to the corresponding articles on the journal's website. Supplemental materials should be placed at the very end of the manuscript and clearly marked with a centered title "Supplemental Materials: For Online Publication Only."

ADDITIONAL MANUSCRIPT PREPARATION GUIDELINES

1. Some authors use terms such as “anorexics” or “bulimics” as personal pronouns, referring to groups of individuals by their common diagnosis. Language of this type should be replaced with such terms as “individuals with anorexia nervosa”, “people with bulimia nervosa”, or “participants with eating disorders”.

2. The term “participants” should be used thought the article instead of “subjects”.

3. Standard rules will continue to govern the use of capitalization in Headings and Subheadings. However, when a minor word in a Heading or Subheading actually has special or unique meaning, the rule should be overridden.

4. When referring to gender, “males" and “females” should be used in cases where the study samples include both children (below age 18) and adults; when the participants comprise adults only, the terms “men” and “women” should be used. In articles that refer to children (i.e., below the age of 13), “boys” and “girls” should be used.

5. In articles that refer to genetic material, the names of genes should be spelled out in full the first time they appear in the text, after which an italicized abbreviation can be substituted.

6. The word “data” is plural; therefore, text should follow accordingly (for example, “The data show…the data are … the data were…”).

7. For information on how to present *p* values and other standard measurements see IJED Statistical Formatting Requirements.

VIDEO ABSTRACTS

A video abstract can be a quick way to make the message of your research accessible to a much larger audience. Wiley and its partner Research Square offer a service of professionally produced video abstracts, available to authors of articles accepted in this journal. You can learn more about it at www.wileyauthors.com/videoabstracts . If you have any questions, please direct them to videoabstracts@wiley.com .

SUBMISSION

Prepare your manuscript and illustrations in appropriate format, according to the instructions given here.

If you have not already done so, create an account for yourself in the system at the submission site, manuscriptcentral.com/ijed by clicking on the "Create an Account" button. To monitor the progress of your manuscript throughout the review process, just log in periodically and check your Author Center.

Please be sure to study the Instructions and Forms given at the site carefully, and then let the system guide you through the submission process. Online help is available to you at all times during the process. You are also able to exit/re-enter at any stage before finally "submitting" your work. All submissions are kept strictly confidential. If you have any questions, do not hesitate to contact us at support@scholarone.com.

PEER REVIEW

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Appendix Q: Participant Consent Form

Gerald Russell Eating Disorders Unit &

Larkbarrow Daypatients Unit

Bethlem Royal Hospital

South London & Maudsley NHS Trust

Institute of Psychiatry

KING'S College LONDON Founded 1829

University of London

COMPARING THE COST EFFECTIVENESS OF SUPPLEMENTING STANDARD CARE WITH AN INTERVENTION FOR CARERS (CARERS ASSESSMENT, SKILLS AND INFORMATION SHARING, CASIS) OF PEOPLE WITH EATING DISORDERS

Carer Information Sheet

You are being invited to take part in an evaluation of a new development in the type of service offered to families of people with eating disorders. It is based on work that has shown that family members have difficulties in knowing what to do to help the individual with an eating disorder and do not feel that their needs are addressed with standard services. It involves an educational intervention for family members to supplement standard care. However, in order for this intervention to be provided as part of standard practice it necessary to demonstrate that it has a positive impact on family members and individuals with an eating disorder and is cost effective.

Before you decide whether you are able to contribute to this project we will explain why it is considered to be important. We will explain the question we are addressing and what your participation will involve. Please take time to read the following information carefully and discuss it with others if you wish. Ask us if there is anything that is not clear or you would like more information about. Take time to decide whether you are able to contribute and commit to this evaluation process.

Thank you for reading this.

Who should be involved?

All family members are invited to be involved. If at all possible it is extremely helpful to have more than one perspective from a family and a joint collaborative approach. Therefore, if possible, please can more than one family member comment and evaluate the project over time (i.e. can we have two sets of questionnaires from each family- if you need more packs please ask the research co-ordinator involved. We are very happy to have siblings and other family members to also contribute).

What is the purpose of this study?

Many people are affected by an eating disorder in the UK and we aim to do everything we can to ensure they receive treatments that are based on a sound evidence base. Research to date has shown that involving carers (family members and close others) in the management and treatment of people with eating disorders can improve the results of the treatment. Research has also shown that by providing carers with information and training, carers are likely to feel more confident, feel less distress and anxiety relating to the eating disorder, and are motivated to acquire new skills that will assist them in more effectively helping their relative. Also, importantly, the earlier someone with an eating disorder engages in treatment the better the outcome. Carers can play an important role in the engagement process.

The purpose of this study is to examine whether our training and information package for carers is useful and effective. Another issue we want to address is whether this training package will have a subsidiary effect for the person with an eating disorder. Lastly, we will look at whether this intervention has benefits in terms of long-term cost effectiveness to you, your family and the public health care system.

Why have I been chosen to take part?

We have invited you, as a carer for someone currently receiving treatment in our service, to participate in this study. All carers of people being treated in our service are given this information form and are invited to take part in the study. We are collaborating with a number of services and hope to recruit a total of about 350 families.

Do I have to take part?

It is up to you to decide whether or not to take part. If you do decide to take part you will be given this information sheet to keep and be asked to sign a consent form. If you decide to take part you are still free to withdraw at any time and without giving a reason without prejudicing your present care or the care of the patient. All the data you have provided would also be removed from our database and files unless it has been assigned an anonymous numeric code after which time we will not be able to remove the data. If you choose not to take part your present care and the care of the patient will not be affected.

What are the benefits of participating in the study?

We hope that participating in the study will be helpful to you and your relative with an eating disorder. However, this cannot be guaranteed as this intervention has only recently been developed and needs more evaluation to properly understand its effects. The information we will obtain from this study may help us to provide better treatment for future patients with an eating disorder.

What are the possible disadvantages and risks of taking part?

The main disadvantage to you in taking part is that we will ask for your time and continued help in reporting on the well being of you and your family during the phase of intensive treatment and for a period of up to a year afterwards. We have tried to make this process as short and simple as possible. However we do have to repeat the same measures over time in order to be able to describe the longer-term impact (eating disorders have a protracted time course). Therefore we will ask you to donate your time and attention in order to provide reliable evidence upon which to base future services.

Our preliminary work suggests that there are no major risks. In some cases we do find that because the individual with an eating disorder has mixed feelings about change they can attempt to sabotage treatment by taking the educational materials away (we will happily replace items lost in this way). Also they can denigrate any effort made by you as carers to help them.

What will I have to do if I take part?

There will be two groups in the study. The core difference between the groups will be the amount of education offered to you as a carer. The treatment process for your relative with an eating disorder will be identical, i.e. the treatment offered as part of the inpatient service. A computer that has no information about the individuals, that is, by chance, selects the groups. Carers chosen by chance to be in the CASIS group will be offered educational materials.

Irrespective of the group you are allocated to, we will ask you to help us audit this process by filling out questionnaires and taking part in (short, mainly phone) interviews throughout the process. We would ask you to complete a series of questionnaires. These should take approximately 30 minutes to complete and need to be returned to Liz Goddard or Simone Raenker (CASIS trial co-ordinators). If you are part of the group receiving the educational materials you will be sent them once we have received the initial questionnaires.

In order for us to assess whether the effect of the educational intervention is effective over time (an important aspect if NICE were to judge whether this should be something added to services throughout the UK) we need you to be willing and able to fill in the same questionnaires again at discharge, 6 months and 12 months after patient discharge whether you receive the educational materials or not. We thank you in advance for this essential contribution.

Carers who are not allocated to the group receiving the material can request to have it after the 12 month follow up is completed.

To assess whether or not these educational materials for families have a subsidiary effect for the person with an eating disorder, we will follow the progress of your family member with an eating disorder during the period of intensive care and over the follow up. This will involve short telephone interviews and questionnaires. These will be administered by the co-ordinators of the study: Liz Goddard and Simone Raenker.

If you agree to help please complete and sign the Consent Form.

Reimbursement

In recognition for the time and effort that you will make we will be able to reimburse you £10 for each set of questionnaires completed and an additional £20 if you have been able to contribute data at all time points of the study. Therefore you can receive £60 as reimbursement for the time and effort you have given by participating in this study. We need more information from the individuals with an eating disorder themselves and so they are given a separate reimbursement.

Confidentiality

All information that you provide during the course of the research will be kept strictly confidential. The information will be made anonymous (any identifiable details such as name and address removed) and only then will it be entered onto the computer. Confidential information will only be accessible to authorised people (i.e. members of staff employed on the project). Interviews may be audio recorded. Any recordings that are collected will be securely stored on a protected file on a computer. Only key researchers will have access to this file. All information you provide will be identified by a numeric code. If any publication results from this research, you will not be identified by name.

Your relative's GP or health professional will be informed that you are taking part in this study and will receive an information pack describing the research. They will not, however, have access to any of your responses or information you give us, as this information will remain confidential.

Alternative contact

At first contact with the study co-ordinator, once you have consented to taking part in the study, we will ask you to name two people (family or friends) for the research team to contact in case you move or we cannot contact you. We would only contact these people in the event that you cannot be reached. In this instance the study co-ordinator would ask why you could not be found or are choosing not to participate. This will help us to ensure we are informed if your changing circumstances affect your ability to continue to participate in the study.

What if new information becomes available?

Sometimes during the course of a research project, new information becomes available about the treatment that is being studied. If this happens, your consultant will tell you about it and discuss with you whether you want to continue with the study. Here it is your choice whether you continue with the study. If you decide to continue in the study you will be asked to sign an updated consent form. If you choose to withdraw it will have no impact on the standard of the care your relative receives. Also, on receiving new information your consultant might consider it to be in your best interests to withdraw you from the study. He/she will explain the reasons.

What if something goes wrong?

If you wish to discuss or complain about any aspect of the way you have been approached or treated during the course of this study, the normal National Health Service complaints mechanisms are available to you. King's College London No Fault Compensation scheme includes payment damages or compensations in respect of any claim made by research participants for bodily injury arising out of participation in any clinical trial. In the very unlikely event that taking part in this research project harms you in any way, there are no special compensation arrangements. However, if you are harmed due to someone's negligence, then you may have grounds for legal action, but you may not be eligible for financial support.

Results of the study

The results of the study will be submitted for publishing to public journals and to the newsletter produced by the Eating Disorders Unit, Guy's Hospital, King's College London. Results may also be presented at conferences. Following publication we would expect that this study will contribute evidence for the next NICE guideline review. In all instances, no names appear and there is no identification.

Who is organising the research?

The research is being organized by the Eating Disorder Research Unit (Institute of Psychiatry) and the Eating Disorder Clinical Team (South London & Maudsley NHS Trust).

This work is in part supported by an NIH-R National Institute of Health Research programme grant (Ref number RP-PG-0606-1043) Treatment of Anorexia nervosa: Translating experimental neuroscience into clinical practice". 2007-2011. ARIADNE to U. Schmidt, J.Treasure, K. Tchanturia, H. Startup, S. Ringwood, S. Landau, M. Grover, I. Eisler, I. Campbell, J. Beecham, M. Allen and G. Wolff.

ARIADNE Carers Project

Ulrike Schmidt, Janet Treasure, Miriam Grover, Liz Goddard, Simone Raenker

The consultants in charge of this study at your site are Professor Janet Treasure and Professor Ulrike Schmidt.

Further Information

If you would like more information about this research please feel free to contact the clinical researcher on the ward, Liz Goddard (02071880190) or Simone Raenker (0203 228 4526) or leave a message (0203 228 4402) or email us at Elizabeth.Goddard@iop.kcl.ac.uk or Simone.Raenker@iop.kcl.ac.uk.

If you would like to take part in this study please keep this Information Sheet and a signed copy of the Consent Form for your own records. Please return the first signed copy of the Consent Form to Liz Goddard or Simone Raenker.

Gerald Russell Eating Disorders Unit &
Larkbarrow Daypatients Unit

Bethlem Royal Hospital
South London & Maudsley NHS Trust

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Larkbarrow Daypatients Unit

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CARER'S CONSENT FORM

COMPARING THE COST EFFECTIVENESS OF SUPPLEMENTING STANDARD CARE WITH AN INTERVENTION (CASIS) FOR CARERS OF PEOPLE WITH EATING DISORDERS

Please initial box

1 I confirm that I have read and understand the information sheet dated 28/04/08 (version 2) for the above study and have had the opportunity to ask questions. ☐

2 I understand that my participation is voluntary and that I am free to withdraw at any time, without giving any reason, without my medical care or legal rights being affected. ☐

3 I agree to having the interviews audiotaped and understand that excerpts of the transcript might be used, albeit anonymously, in the reporting of the findings of this research. ☐

4 I understand that workshop discussion and discussion with the mentor are confidential. ☐

5 I agree to take part in the above study. ☐

____________________	____________________	____________________
Name of Carer	Date	Signature
____________________	____________________	____________________
Name of person taking consent (If different from the researcher)	Date	Signature
____________________	____________________	____________________
Researcher	Date	Signature

Appendix R: Summary of Part A for Health Professionals

Introduction

It is widely recognised that caring for someone with an eating disorder is often difficult and distressing, with carers (family, partners and friends providing care) of people with eating disorders more likely to experience clinically relevant depression and anxiety than non-carers[1]. In addition to the rationale for supporting carers generally, research and theory[2,3] suggests that the effect of the eating disorder (ED) on carers can result in caregiving responses (e.g. expressed emotion[4]) which may not only lead to carer distress but also perpetuate a vicious cycle of worsening ED symptoms[3]. Studies have shown that supporting carers can improve ED recovery[5], likely through a virtuous cycle[2].

However, it is not clear what predicts the amount of distress carers of people with eating disorders will experience. There is even less clarity around the processes leading to these heightened levels of distress, or why it is more distressing for some carers than others. Interventions for carers of people with ED are recently becoming more widely available, and typically take the form of peer support, skills-sharing and psycho-education. It is also not clear what predicts reductions in distress as an outcome of such interventions, the processes by which these reductions occur, or which carers they are most effective for.

Methods

To address these questions, we conducted a review of the literature into predictors of carer distress in eating disorders covering papers published from inception to October 2017.

Included within this was a review of moderators (factors that affect the size of impact of the predictor on the outcome) and mediators (factors that explain or account for the affect the predictor has on the outcome) of both the amount of distress carers experience and the reduction in distress carers experience following intervention. Searches through electronic databases, and by hand, retrieved 424 studies that were screened for quality and relevance, resulting in 30 included papers (details available on request).

Results, Discussion and Implications

Findings showed that, while a multitude of potential predictors were investigated, robust evidence was lacking overall. However, there was reasonable evidence for several aspects of the caregiving experience being associated with greater distress, which are described in turn.

Understandably, the person with an eating disorder expressing suicidal intent, or abusing substances was associated with carers being more distressed, as was carers feeling they had inadequate skills for their caring role, and finding it difficult to cope. In practice, these findings may be useful in identifying carers potentially in need of additional support.

Additionally, three areas related to carers' experiences of the eating disorder were found to predict greater carer distress; expressed emotion, accommodation, and burden. Expressed emotion refers to caregivers' criticism and emotional over-involvement. However, rather than being a one-way behaviour, it is proposed to describe the relational interaction between carer and cared-for factors, as carers attempt to care for their unwell loved one[6]. Accommodating and enabling behaviours ('accommodation') refers to carers' attempts to accommodate family life to the demands of the ED, and to enable ED behaviours in an effort to reduce the negative impact of the illness[7]. The most robustly evidenced finding was that that greater caregiving burden (more demands, strains and negative experiences related to caregiving) is likely to lead to higher levels of distress. This suggests that it's not the eating disorder symptoms themselves that cause distress for carers, but the way in which they become personally relevant for the carer/s.

These findings indicate that addressing how carers respond to the eating disorder symptoms may provide an avenue for reducing carer distress. This may provide professionals and carers with some optimism regarding the opportunity for improvement in carer distress, even in cases where the cared-for is unwilling to engage in treatment or recovery has plateaued. Intervention focused on supporting carers to reduce levels of expressed emotion, accommodation and burden are currently running at the Maudsley hospital in London, and have been manualised for health professional use[8,9].

Unsurprisingly, there was also good evidence that carers either having higher levels of distress themselves, or caring for someone who is very distressed, is a good predictor of feeling more distressed in the future. This echoes the chronicity of eating disorders and reinforces the importance of providing services for carers addressing distress, which seems otherwise likely to persist.

Only five of the studies investigated what predicts how much carer distress will reduce after intervention. The methodology employed by the majority of these studies, and inconsistencies in results, meant it was not possible to draw clear conclusions. This was also the case for moderators and mediators of both carer distress in EDs, and changes to carer distress as an intervention outcome. Therefore, further research is strongly indicated in these areas.

Limitations

Limitations included generalisability of the sample, which was predominantly mothers (58%) of people with Anorexia Nervosa (72%), who lived together (84%). While processes involved in carer distress are hypothesised to be comparable across eating disorder diagnoses[2], differences have been noted between the caring roles (e.g. fathers, partners)[10]. Many of the questionnaires used by the reviewed studies were of questionable validity for the sample, and methodology, especially for moderators and mediators, was sometimes insufficient to draw clear conclusions. However, there was agreement amongst all studies as to whether associations between predictors and carer distress were positive or negative, affording some confidence.

Conclusion

The findings of this review support understandings of carer distress that focus on the ways the eating disorder becomes personally relevant for the carer, including the carers' emotional and behavioural responses. Overall, evidence illuminating the probably complex processes underlying carer distress in eating disorders is lacking, and deserves further investigation.

References

1. Anastasiadou, D., Medina-Pradas, C., Sepulveda, A. R., & Treasure, J. (2014). A systematic review of family caregiving in eating disorders. Eating behaviors, 15(3), 464-477. doi:10.1016/j.eatbeh.2014.06.001

2. Treasure, J., & Nazar, B. P. (2016). Interventions for the Carers of Patients With Eating Disorders, *Current Psychiatry Reports, 18*(16), 1–7. doi: 10.1007/s11920-015-0652-3
3. Treasure, J., & Schmidt, U. (2013). The cognitive-interpersonal maintenance model of Anorexia Nervosa revisited: a summary of the evidence for cognitive, socio-emotional and interpersonal predisposing and perpetuating factors. Journal of Eating Disorders, 1(1), 13. doi:10.1186/2050-2974-1-13
4. Butzlaff, R. L., & Hooley, J. M. (1998). Expressed emotion and psychiatric relapse: A meta-analysis. *Archives of General Psychiatry*, *55*(6), 547-552. doi:10.1001/archpsyc.55.6.547
5. Hibbs, R., Magill, N., Goddard, E., Rhind, C., Raenker, S., Macdonald, P., … Treasure, J. (2015). Clinical effectiveness of a skills training intervention for caregivers in improving patient and caregiver health following in- patient treatment for severe Anorexia Nervosa : pragmatic randomised controlled trial. British Journal of Psychiatry, 1, 56–66. doi:10.1192/bjpo.bp.115.000273
6. Hooley, J. M. (2007). Expressed emotion and relapse of psychopathology. *Annual Review of Clinical Psychology, 3,* 329-352. doi:10.1146/annurev.clinpsy.2.022305.095236
7. Sepulveda, A. R., Kyriacou, O., & Treasure, J. (2009). Development and validation of the accommodation and enabling scale for eating disorders (AESED) for caregivers in eating disorders. *BMC Health Services Research, 9*, 171. doi:10.1186/1472-6963-9-171

8. Treasure, Sepulveda, Whitaker, Todd, Lopez, & Whitney, 2007; Treasure, J., Schmidt, U., & Macdonald, P. (Eds.). (2009). *The Clinician's Guide to Collaborative Caring in Eating Disorders: The New Maudsley Method*. London and New York: Routledge.
9. Treasure, J., Sepulveda, A. R., Whitaker, W., Todd, G., Lopez, C., & Whitney, J. (2007). Collaborative Care between Professionals and Non-Professionals in the Management of Eating Disorders: A Description of Workshops Focussed on Interpersonal Maintaining Factors. *European Eating Disorders Review*, *15*, 24–34. doi: 10.1002/erv
10. Kyriacou, O., Treasure, J., & Schmidt, U. (2008). Understanding how parents cope with living with someone with Anorexia Nervosa: Modelling the factors that are associated with carer distress. *International Journal of Eating Disorders*, *41*(3), 233-242. doi:10.1002/eat.20488

Appendix S: Summary of Part B for Health Professionals

Introduction

Eating disorders are serious psychiatric illnesses with high mortality rates[1]. The NICE[2] guidelines recommend outpatient management as the first form of treatment for all types of eating disorders. This places a burden of care onto parents and close others who often experience their role as distressing and burdensome[3,4], with a large proportion experiencing clinically relevant levels of anxiety and depression[5]. In addition to the negative impact on quality of life for carers[6], their distress and related[7] behavioural responses to the eating disorder (ED), specifically *expressed emotion* (the relational interaction between carer and cared-for factors, resulting in criticism and over-involvement) and *accommodation* (going along with the cared-for's symptomatic behaviours in an effort to reduce the ED's impact) may unintentionally maintain the ED[8].

Recent trials of skill-sharing interventions for carers generally report small-to-moderate sized reductions in carer distress, expressed emotion and burden[9] (demands, strains and negative caregiving experiences). However, the processes involved in achieving these outcomes are unclear. Additionally, the hypothesis that more severe cared-for ED symptoms are associated with greater carer distress is central to leading models of ED caregiving[7], but this is poorly supported empirically[5].

Aims

- To determine the processes involved in the beneficial outcomes from a recent randomised controlled trial (RCT) of an intervention for carers of people with eating disorders[10].
  - To determine why or how the intervention was effective, and when, or for who, it was more effective.
- To investigate the relationships between ED symptoms and carer distress.

Methodology

Carers of people presenting for hospital treatment of Anorexia Nervosa were randomly allocated to either treatment as usual or a skills-sharing intervention. The intervention, based on the New Maudsley Method[11], consisted of a book, five DVDs and telephone coaching. Detailed guides for providing similar interventions are available[12,13]. Consenting service-users (n=159) and their primary caregivers (non-professional most actively involved in their care, n=159) completed questionnaires at baseline and discharge of the cared-for's hospital admission, and at six and 12-month follow-up. Previous exploration[10] found the intervention to be associated with reductions in expressed emotion, burden and ED symptoms, but not carer distress or accommodation.

Moderation and mediation analyses aim to clarify processes involved in the effect of one variable (e.g. intervention) on another (e.g. reduction in distress). They suggest how or why an effect occurs (moderation), and when, or for whom, it is more likely to occur (mediation)[14]. To investigate why or how the intervention was effective, and who it was more effective for, several variables were entered as potential moderators (including number of carers per service-user, service-user BMI and age) and mediators (including expressed emotion and burden) of the relationship between receiving the intervention and size of reduction in the outcomes of interest (reduction in ED severity; carer distress; accommodation; expressed emotion; burden).

Burden, accommodation, expressed emotion and service-user distress were explored as potential mediators assessing whether they accounted for a relationship between ED symptoms and carer distress. The longitudinal design of the RCT enabled the proposed mediators to be entered in sequence to investigate how ED symptoms may lead to carer distress, and how carer distress may lead to ED symptoms.

Results, Discussion and Implications

Moderation of intervention outcomes: When, or for whom, the intervention was most effective

Greater reduction in expressed emotion following intervention only occurred for the group of carers who took part with another of the service users' carers, not for carers who took part alone. It may therefore be useful for carers to attend with another carer of the same service-user, where possible. This was the only significant[15] moderator found.

Mediation of intervention outcomes: Why, or how, the intervention was effective

Findings indicated that intervention predicted greater reductions in both carer distress and accommodation, but that this was indirectly, through reductions in burden and expressed emotion. Therefore, the current findings increase the known efficacy of the intervention compared to previous exploration of only direct effects[10]. This is potentially valuable for securing future funding.

The greater reductions in burden and expressed emotion following intervention also significantly mediated the greater reduction in ED symptoms; it appears that the intervention reduced burden, which reduced expressed emotion, which in turn reduced ED symptoms. This highlights the importance of interventions targeting carer burden and expressed emotion as these appear to be potentially key processes in the amelioration of clinically relevant problems for both service user (ED symptoms) and carer (distress).

Longitudinal investigation of relationships between ED symptoms and carer distress

Findings indicated that the cared-for having more severe ED symptoms led to carers showing higher accommodation, burden and expressed emotion, which in turn led to greater carer distress over time. It appears therefore that the ED symptoms increase carer distress through the ways in which the ED becomes personally relevant for the carer; having to organise family life around the ED, a difficult relationship with the cared-for, and increased demands, strains and negative experiences. This finding of indirect associations offers support and clarification to leading models[7,8] that have struggled to explain the lack of empirical support for a direct link between ED symptoms and carer distress[5,86].

Finally, results suggested that greater carer distress led to greater expressed emotion, which in turn led to greater service user distress, which in turn led to more severe ED symptoms over time. This is the first time that carer distress has been statistically implicated as a predictor of eating disorder symptoms, suggesting a possible mechanism by which this happens and supporting the case for improved services aimed at reducing carer distress.

Strengths and Limitations

This study used a robust longitudinal RCT design, although missing data may have introduced bias. While mediation analysis is arguably placed to imply causality under certain conditions, the design of this study means cause and effect cannot be confidently stated.

Conclusion

Carer distress, often clinically relevant for carers, can be both a factor in eating disorder maintenance and ameliorated with skills-sharing interventions for carers. It is therefore imperative that carers are offered the support they frequently ask for[17].

References

1. Arcelus, J., Mitchell, A. J., Wales, J., & Nielsen, S. (2011). Mortality rates in patients with Anorexia Nervosa and other eating disorders: a meta-analysis of 36 studies.

Archives of General Psychiatry, *68*(7), 724-731. doi:10.1001/archgenpsychiatry.2011.74

2. National Institute of Clinical Excellence (2004). Eating disorders: Core interventions in the treatment and management of Anorexia Nervosa, Bulimia Nervosa and related eating disorders. CG9. Retrieved from http://guidance.nice.org.uk/CG9
3. Whitney, J., Murray, J., Gavan, K., Todd, G., Whitaker, W., & Treasure, J. (2005). Experience of caring for someone with Anorexia Nervosa: A qualitative study. *The British Journal of Psychiatry*, *187*(5), 444-449. doi:10.1192/bjp.187.5.444
4. Zabala, M. J., Macdonald, P., & Treasure, J. (2009). Appraisal of caregiving burden, expressed emotion and psychological distress in families of people with eating disorders: A systematic review. *European Eating Disorders Review, 17*(5), 338–349. doi:10.1002/erv.925
5. Kyriacou, O., Treasure, J., & Schmidt, U. (2008). Understanding how parents cope with living with someone with Anorexia Nervosa: Modelling the factors that are associated with carer distress. *International Journal of Eating Disorders*, *41*(3), 233-242. doi:10.1002/eat.20488
6. Martin, J., Padierna, A., Aguirre, U., Quintana, J. M., Las Hayas, C., & Munoz, P. (2013). Quality of life among caregivers of patients with eating disorders. *Quality of Life Research,20* (9) 1359-1369. doi: 10.1007/s11136-011-9873-z
7. Treasure, J., & Nazar, B. P. (2016). Interventions for the Carers of Patients With Eating Disorders, *Current Psychiatry Reports, 18*(16), 1–7. doi: 10.1007/s11920-015-0652-3
8. Treasure, J., & Schmidt, U. (2013). The cognitive-interpersonal maintenance model of Anorexia Nervosa revisited: a summary of the evidence for cognitive, socio-

emotional and interpersonal predisposing and perpetuating factors. Journal of Eating Disorders, 1(1), 13. doi:10.1186/2050-2974-1-13

9. Hibbs, R., Rhind, C., Leppanen, J., & Treasure, J. (2015). Interventions for caregivers of someone with an eating disorder: A meta-analysis. *International Journal of Eating Disorders,* 48(4), 349-361. doi: 10.1002/eat.22298
10. Hibbs, R., Magill, N., Goddard, E., Rhind, C., Raenker, S., Macdonald, P., … Treasure, J. (2015). Clinical effectiveness of a skills training intervention for caregivers in improving patient and caregiver health following in- patient treatment for severe Anorexia Nervosa : pragmatic randomised controlled trial. British Journal of Psychiatry, 1, 56–66. doi:10.1192/bjpo.bp.115.000273
11. Treasure, Sepulveda, Whitaker, Todd, Lopez, & Whitney, 2007; Treasure, J., Schmidt, U., & Macdonald, P. (Eds.). (2009). *The Clinician's Guide to Collaborative Caring in Eating Disorders: The New Maudsley Method*. London and New York: Routledge.
12. Goddard, E., Raenker, S., Macdonald, P., Todd, G., Beecham, J., Naumann, U., … Treasure, J. (2013b). Carers’ assessment, skills and information sharing: Theoretical framework and trial protocol for a randomised controlled trial evaluating the efficacy of a complex intervention for carers of inpatients with Anorexia Nervosa. *European Eating Disorders Review : The Journal of the Eating Disorders Association, 21*(1), 60–71. doi:10.1002/erv.2193
13. Treasure, J., Sepulveda, A. R., Whitaker, W., Todd, G., Lopez, C., & Whitney, J. (2007). Collaborative Care between Professionals and Non-Professionals in the Management of Eating Disorders: A Description of Workshops Focussed on Interpersonal Maintaining Factors. *European Eating Disorders Review*, *15*, 24–34. doi: 10.1002/erv

14. Hayes, A. F. (2013). *Introduction to mediation, moderation, and conditional process analysis: A regression-based approach*. New York, NY: Guilford Press
15. All significance values are p<.05
16. Goddard, E., Salerno, L., Hibbs, R., Raenker, S., Naumann, U., Arcelus, J., … Treasure, J. (2013c). Empirical examination of the interpersonal maintenance model of Anorexia Nervosa. *The International Journal of Eating Disorders, 46*(8), 867–74. doi:10.1002/eat.22172
17. Haigh, R., & Treasure, J. (2003). Investigating the needs of carers in the area of eating disorders: Development of the Carers' Needs Assessment Measure (CaNAM). *European Eating Disorders Review, 11*(2), 125–141. doi:10.1002/erv.487

Appendix T: Summary of Part A for Lay Carers

It is widely recognised that caring for someone with an eating disorder is often difficult and distressing. Research has supported this and found that carers (family, partners and friends providing care) of people with eating disorders are more likely to experience anxiety and depression than non-carers.

However, it is less clear why this is the case, or why it is more distressing for some carers than others. In the hope of understanding this better, we reviewed the relevant research into what issues, experiences or circumstances may be more likely to lead to higher levels of distress being experienced by carers of people with eating disorders.

A thorough search of published research was conducted through electronic databases, as well as searching journals by hand. Over 400 journal articles were screened for quality and relevance. This resulted in finding 30 research articles that were relevant to our questions.

The methods and results of these 30 studies were analysed to understand the strength of evidence for each issue, experience or circumstance that was tested. The way these had been tested by different studies meant that some were only able to say whether these occurred at the same time as distress, while others could say that they occurred before distress; a stronger position to suggest they caused distress, rather than the other way around. However, it would not be possible for this type of (correlational) research to *prove* causes of distress, so we use the term ‘associates’ (things that are statistically related) rather than ‘causes’.

Findings showed that, while many different possible associates of distress were investigated, overall there was not strong evidence for the majority of them. However, there was reasonable evidence for several aspects of the caregiving experience being associated with greater carer distress, which will be described in turn.

Understandably, the person with an eating disorder expressing suicidal intent, or abusing substances was associated with carers being more distressed, as was carers feeling they had inadequate skills for their caring role, and finding it difficult to cope.

Additionally, three areas to do with carers’ experiences of the eating disorder were found to predict greater distress; expressed emotion, accommodation, and burden. Expressed emotion describes the emotional atmosphere and interactions between carer and cared-for. Commonly in eating disorders, difficulties in these interactions can arise as a result of trying to care for someone who is very unwell but who may reject the help carers try to provide. Accommodation refers to refers to carers' attempts to accommodate family life to the demands of the eating disorder, and help enable the person they care for to carry out the eating disordered behaviour (e.g. special cooking rituals) in an effort to reduce the cared-for's distress and the negative impact of the illness. The most well evidenced finding was that that greater caregiving burden (more demands, strains and negative experiences related to caregiving) is likely to lead to higher levels of distress. This suggests that it's not the eating disorder symptoms themselves that cause distress for carers, but the way that they become personally relevant for the carer/s.

This is a helpful finding because it indicates areas that carers have control over as targets for change, rather than relying on the person with the eating disorder to make changes (something many carers will recognise as problematic). Interventions for carers are already running based on this model, with some success. Examples are carers groups and support projects at the Maudsley hospital in London.

Unsurprisingly, there was also good evidence that carers either having higher levels of distress themselves, or caring for someone who is very distressed, was a good predictor of the carer feeling more distressed in the future. This reinforces the importance of providing services for carers as, alongside the typically long course of eating disorders, carers' distress may persist.

While support for carers is often lacking, interventions specifically for carers of people with eating disorders are becoming more widely available. The majority of these interventions aim to reduce carer distress, and often take the form of peer support and sharing professional skills with carers. As well as the above, we looked at which carers are likely to benefit most from these kinds of interventions, and whether there is anything that leads to distress being more or less reduced by the intervention (e.g. how effective the intervention is), and whether the interventions were more useful for some carers than others (e.g. people with their own histories of eating distress). However, we found there is not yet enough evidence to draw clear conclusions, so we argue that there needs to be more research into this area.

Like all research, this study had some limitations. Firstly, many of the studies reviewed only looked at mothers of people with Anorexia Nervosa. Therefore, it may not be reasonable to suggest that our findings can or should be applied to other types of carers, for example partners of people with Binge Eating Disorder. We also had some concerns about how appropriate some of the questionnaires used by the studies were. However, there were not serious questions about the conclusions that can be drawn from this study, and it is likely to be broadly relevant to caregivers’ experiences across eating disorder diagnoses and types of relationships.

The findings of this study support understandings of carer distress that focus on the ways the eating disorder impacts on the carer, and suggests that interventions should continue to focus on these areas in order to reduce carer distress. It will also be important to continue researching this area in order to better understanding the processes contributing to carers’ distress, so interventions can become better at reducing it more effectively in the future.

Appendix U: Summary of Part B for Lay Carers

As a teenager, my best friend was diagnosed with Anorexia Nervosa. In an effort to relieve her devastating distress we tried accommodating life and meals to her requirements, and enabling strange food rituals and rules, hoping it would help her eat *something*. We bargained with the eating disordered part of her, and criticised the madness of the eating disorder logic. There was little support available for us as her carers, and we felt excluded from her treatment. She became frighteningly thin, and was finally admitted to general hospital, where we were told she may die.

Then one day, some weeks later, she decided to get better; and just like that, she did.

This began my fascination with eating disorders; what changed for my friend that day? Could it have changed sooner? How?! We asked her and each other these questions many times, with no answer. With these questions in my mind I started studying psychology and working at Beat running carers' groups. I saw how my own experience had not been unique, and understood that a vicious cycle can occur where the often frustrating and anxiety-provoking experience of caring for someone with an eating disorder can cause carers to try to help in ways that can ultimately have unintended effects. I realised that the eating disorder can affect not only the sufferer, but those all around them, insidiously corrupting our well-intentioned interactions to support its own maintenance.

Working with carers (family, partners and friends with a caring role), I was inspired by their hope, tenacity and determination. I saw how carers can, and do, have a hugely positive impact on their cared-fors' recovery, especially when they have access to adequate information and support. Having gained enough experience to progress from the magic question of ‘what will make people spontaneously recover from eating disorders?’, I began a Psychology doctorate and turned my attention to something more answerable.

Research shows that carers of people with eating disorders experience high levels of distress, anxiety, and depression, but how the eating disorder might lead to carers’ distress is less clear. I wanted to understand this, as well as whether (and if so how) an intervention for carers based on skills-sharing reduced carers’ distress. I also asked whether the intervention was more useful if more than one carer per sufferer attended.

Working with Janet Treasure and the research team at the Maudsley Hospital, London, we designed a study using data from their recent trial of a programme of support for carers (a book, DVDs and telephone coaching). This was a Randomised Controlled Trial (RCT), the gold standard in research into treatments. Carers (159) of people with Anorexia Nervosa admitted for in-patient treatment took part. The carers and the people they cared for completed questionnaires at several time-points over the course of hospital treatment and a year after discharge.

Commonly in eating disorders, carers report noticing themselves becoming more critical of the person they care for and/or taking on responsibility for all aspects of the suffers life in an attempt to protect them from further difficulties. This, and the generally difficult emotional atmosphere that eating disorders often create, is termed expressed emotion. Findings showed that the intervention reduced expressed emotion only when more than one carer per sufferer took part, not for carers who took part alone. This might be because taking part with someone else who knows the person they care for helped carers to think about how the skills could be applied to their personal situation, and they were able to support each other. Therefore, there are plans to provide a moderated online support group for carers as an additional part of the intervention in future.

Findings also showed that carer distress was reduced by the intervention, but indirectly: Carers who took part in the intervention reported they felt less burdened. It appears that carers who felt less burdened had more time to look after their own needs, increasing their capacity to care and finding it easier not to get drawn into unhelpful interactions (reduced expressed emotion). Having more positive emotional experiences with the person they cared for may then have led to carers feeling less distressed.

Similar processes appeared to be at work when looking at how the cared-fors' eating disorder symptoms led to carers' distress. Caring for someone with worse symptoms was linked to understandably feeling more burdened, accommodating and enabling more, and there being more expressed emotion. In turn, these difficulties were linked with higher distress for carers. So it seems that it's the way that these symptoms become personally relevant for the carer that lead to distress, rather than the symptoms themselves. This is encouraging as these areas are often easier to change than eating disorder symptoms, indicating carers can be supported to reduce their levels of distress, without needing the sufferer to recover first.

Further to this was the important finding that carers who were less distressed were likely to have more positive interactions with the person they cared for (e.g. less expressed emotion). This appears to have led to less distress for the person with the eating disorder, in turn leading to enhanced improvement in their eating disorder symptoms following treatment. This is exciting because it's the first study to show this, backing the case for more funding for better services and skills-sharing for carers, to support carers ensure this become a virtuous cycle rather than a vicious one. It also provides evidence backing the adage ‘put on your own oxygen mask first’; carers looking after their own needs really does help their loved one’s recovery.

Today, my friend is married, with three beautiful children to whom I am a very proud godmother. I thank her for inspiring my career and reminding me to always hold onto the hope, as we had to in those dark days that now seem like a lifetime ago. Knowing how difficult it can be to get people with eating disorders into treatment, and how passionate many carers are about helping the person they care for, the findings of this study give me increased hope that through increased collaboration including better services for carers, together we can beat eating disorders.

Appendix V: Feedback to R&D

Following completion of the MRP, I emailed R&D a summary of Part B, presented below. Further analysis of the archival data is being undertaken by the IoP research team, so the on-going wider study has not closed.

---------- Forwarded message ----------
From: **Hannah King** <kinghannah99@googlemail.com>
Date: 5 March 2017 at 21:08
Subject: Feedback R&D2008/029
To: tempr&d@kcl.ac.uk, janet.treasure@kcl.ac.uk

Dear Hannah,

Please be advised that I have now completed my analysis of data from this study:

Study title: A randomised pragmatic trial comparing the cost effectiveness of supplementing standard care with an intervention for carers (Carers assessment, skills and information sharing, CASIS) of people with eating disorders
REC Ref: 08/H0720/41
SLaM R&D Ref: R&D2008/029

I have attached a summary for your information. Please let me know if you would like me to send any further details.

Although my use of the data from this trial has ended, I understand that the team has not completed analysis. I have copied in Janet Treasure as an ongoing contact.

With best wishes
Hannah

Hannah King
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Salomons Centre for Applied Psychology
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TN3 0TF

Moderators and Mediators of Relationships between Eating Disorders and Related Carer Difficulties, in the Context of a Skills Sharing Intervention for Carers of People with Anorexia Nervosa

Summary

Background: Carers of people with eating disorders (EDs) have heightened levels of distress, expressed emotion, burden and accommodation to the ED; factors implicated in maintenance of the ED. Although carers' skills interventions are helpful, how they effect change, and the processes involved in hypothesised relationships between ED symptoms and carer distress, is unclear.

Aims: To determine the processes involved in the beneficial carer and service-user outcomes from a carers' skills RCT, including by longitudinally examining relationships between ED symptoms and carer distress.

Method: This study utilised data from a multi-site large-scale carers' skills RCT**.** Primary carers (159) of people presenting for Anorexia Nervosa hospital treatment were randomly allocated to the intervention or treatment as usual. Moderators and mediators of intervention outcomes, and of longitudinal relationships between ED symptoms and carer distress, were examined.

Results, Discussion and Implications

Moderation of intervention outcomes: Greater reduction in expressed emotion following intervention only occurred for the group of carers who took part with another of the service users' carers, not for carers who took part alone. It may therefore be useful for

carers to attend with another carer of the same service-user, where possible. This was the only significant moderator found (p<.05).

Mediation of intervention outcomes: Findings indicated that intervention predicted greater reductions in both carer distress and accommodation, but that this was indirectly, through reductions in burden and expressed emotion. Therefore, the current findings increase the known efficacy of the intervention compared to previous exploration of only direct effects (Hibbs et al., 2015). The greater reductions in burden and expressed emotion following intervention also significantly mediated the greater reduction in ED symptoms; it appears that the intervention reduced burden, which reduced expressed emotion, which in turn reduced ED symptoms. This highlights the importance of interventions targeting carer burden and expressed emotion as these appear to be potentially key processes in the amelioration of clinically relevant problems for both service user (ED symptoms) and carer (distress).

Longitudinal investigation of relationships between ED symptoms and carer distress: Findings indicated that the cared-for having more severe ED symptoms led to carers showing higher accommodation, burden and expressed emotion, which in turn led to greater carer distress over time. It appears therefore that the ED symptoms increase carer distress through the ways in which the ED becomes personally relevant for the carer; having to organise family life around the ED (accommodating and enabling), a difficult relationship with the cared-for (expressed emotion) and increased demands, strains and negative experiences (burden). Finally, results were consistent with the hypothesis that greater carer distress led to greater expressed emotion, which in turn led to greater service user distress, which in turn led to more severe ED symptoms over time.

Limitations & Implications: Missing data may have introduced bias. Accessing only primary carers of severely unwell service-users limited generalisability. Results support the importance of carers' skills interventions in addressing ED-related service-user and carer difficulties.

Appendix W: Questionnaires Completed by Participants

The initial demographic questionnaire, purpose-made for the intervention, is shown below. Other (standardized) questionnaires have been removed from the final version as per the handbook's instruction, as they are either copyright or widely available.

PARTICIPANT'S CODE _______________ *COREC ref. no* 08/H0720/41/ *Version 1 20/02/08*

CASIS Carer Questionnaire - please return the questionnaire to the research team

1. Section – Code and Date

1.1 Code (filled in by the research team) ☐

1.2 Today's Date

_ _ / _ _ / _ _
DD MM YY

2. Section – Introduction and Instructions

Thank you very much for taking part in our study. Please read the information sheet to get all the information about the project. Here are some notes that may guide you through the process. The headers on each sheet let you know whether the sheet is one that you keep or whether it is one that we need.

If you have any questions please contact either:

Liz Goddard
02071880190/07828975513
Elizabeth.Goddard@iop.kcl.ac.uk

Simone Raenker
02071880169 / 07917852497
Simone.Raenker@iop.kcl.ac.uk

Eating Disorder Unit - Academic Medicine
5th Floor Thomas Guy House
Guys Hospital, London SE1 9RT

CASIS Carer Questionnaire → Please return the questionnaire to the research team after you have filled in every question. Please read the instructions given carefully and answer every question.

a. Your Details
b. Demographic Questions
c. Questions about the person you are caring for
d. Other Issues
e. Psychological Questionnaires

We appreciate you taking the time and spending the effort to support us in our research project. Therefore, if you and the person you care for with an eating disorder are able to complete the set we will be able to reimburse you as carers with £60 and the person you care for with an eating disorder with £120 (smaller tokens are available for each set) .

PARTICIPANT'S CODE ______________ *COREC ref. no* 08/H0720/41/ *Version 1 20/02/08*

CASIS Carer Questionnaire - please return the questionnaire to the research team

3. Section – Site

Please choose the Hospital your relative is admitted to. (Tick one box)

○ Bethlem Hospital, London
○ Vincent Square Clinic, London
○ St. Georges Hospital, London
○ Highfield Unit, Oxford
○ Cotswold House, Oxford
○ Brandon Mental Health Unit, Leicester
○ Kinver Centre, South Staffordshire
○ Darwin Centre, North Staffordshire
○ Affinity Healthcare, Cheadle Royal Hospital, Manchester
○ Seacroft Hospital, Leeds
○ Coventry and Warwickshire Trust
○ STEPS Eating Disorder Unit, Bristol
○ Cotswold House, Marlborough
○ Kimmeridge Court, Dorset
○ Birmingham Eating Disorder Service
○ Diana Princess of Wales Hospital, Grimsby
○ Maidstone
○ Haldon Unit, Exeter

4. Section "Demographic Questions"

4.1 Date of birth: _ _ / _ _ / _ _
DD MM YY

4.2 Gender: ○ Female ○ Male

4.3 What is your current employment status? (Tick one box)

○ Full time employed
○ Part time employed
○ Unemployed
○ Student
○ Retired
○ Sick leave
○ Housewife / -husband
○ Other
If other, please specify

____

PARTICIPANT'S CODE _______________ *COREC ref. no* 08/H0720/41/ *Version 1 20/02/08*

CASIS Carer Questionnaire - please return the questionnaire to the research team

4.4 What is the highest level of education you have completed? (Tick one box)

○ No qualifications
○ O Level / GCSE
○ A Level / NVQ
○ University degree
○ Postgraduate degree
○ Diploma / BTEC
○ Other
    If other, please specify

____

____

____

4.5 How many years of education have you received?

___________ years and ___________ months

4.6 Is your first language English?

○ Yes
○ No

4.7 What is your ethnicity?
○ White British
○ White Irish
○ Other White
○ Mixed White and Black Caribbean
○ Mixed White and Black African
○ Mixed White and Asian
○ Other Mixed
○ Asian or Asian British – Indian
○ Asian or Asian British – Pakistani
○ Asian or Asian British – Bangladeshi
○ Other Asian
○ Black or Black British – Caribbean
○ Black or Black British – African
○ Other Black
○ Chinese
○ Other ethic group

4.8 What is your marital status?
○ Married
○ Living together
○ Single
○ Divorced
○ Separated
○ Widowed

PARTICIPANT'S CODE _______________ *COREC ref. no* 08/H0720/41/ *Version 1 20/02/08*

CASIS Carer Questionnaire - please return the questionnaire to the research team

4.9 Do you have a daughter(s)?

○ Yes
○ No

4.10 Do you have a son(s)?

○ Yes
○ No

4.11 If you have answered yes to question 4.9 and 4.10, please type in the ages of your children in the table.

Children	***1. child - age***	***2. child - age***	***3. child - age***	***4. child - age***	***5. child - age***
Daughter(s)					
Son(s)					

Section 5 "Details about the person you are caring for"

5.1 Please describe your relationship to the person you are caring for with the following options: (Tick one box)
She / He is my…

○ Spouse
○ Partner
○ Child
○ Sibling
○ Parent
○ Other relative

If other, please specify

____

____

○ Friend
○ Other

If other, please specify

____

____

Carer Assessment, Skills, & Information Sharing

PARTICIPANT'S CODE _______________ *COREC ref. no* 08/H0720/41/ *Version 1 20/02/08*

CASIS Carer Questionnaire - please return the questionnaire to the research team

5.2 What is the name of the person you are caring for :

____

5.3 What is her / his date of birth?

_ _ / _ _ / _ _
DD MM YY

5.4 What is her /his age?

__ __ years

5.5 What is her / his gender?

○ Female
○ Male

5.6 What is her / his marital status? (Tick one box)
○ Married
○ Living together
○ Single
○ Divorced
○ Separated
○ Widowed

5.7 We are interested to know what sort of an eating disorder the person you care for has/had. Please answer to all the behaviours and symptoms below that describe their eating difficulties:

Severely underweight?
○ Yes ○ No

Restricting food intake?
○ Yes ○ No

Exercising excessively?
○ Yes ○ No

Vomiting after meals?
○ Yes ○ No

Missing menstrual periods for 3 months or more?
○ Yes ○ No

Eating unusually large amounts of food in one sitting (binge eating)?
○ Yes ○ No

Eating in secret?
○ Yes ○ No

Stealing food/money in order to binge?
○ Yes ○ No

Severely overweight?
○ Yes ○ No

Others? Please specify

____

____

____

____

5.8 Has the person that you care for ever been diagnosed with an eating disorder by a health professional?
○ Yes
○ No

If Yes, what was the diagnosis? (You may tick more than one if the eating disorder has changed over time)

☐ Anorexia nervosa
☐ Bulimia nervosa
☐ Binge eating disorder
☐ I don't know
☐ Other
If other, please specify

____

____

5.9 At what age was the person you care for first diagnosed with an eating disorder?

____

____

5.10 At what age did the person you care for start to experience these eating difficulties?

____

____

PARTICIPANT'S CODE _____________ *COREC ref. no* 08/H0720/41/ *Version 1 20/02/08*

CASIS Carer Questionnaire - please return the questionnaire to the research team

5.11 While the person you care for is suffering from their eating disorder do she / he has any additional problems? Please answer to all the behaviours below. E.g. does she / he:

Drink a lot of alcohol
○ Yes ○ No

Use illegal drugs
○ Yes ○ No

Use legal drugs (e.g. sleeping tablets) in excess of the recommended amount
○ Yes ○ No

Deliberately hurt themselves (e.g. by biting, cutting burning themselves)
○ Yes ○ No

Steal (including food)
○ Yes ○ No

Gamble
○ Yes ○ No

Others? Please specify

____

____

____

____

5. 12 On average, how many hours of face to face contact do you have with the person you care for each week? (Tick one box)

○ 0 – 7 hours / week
○ 8 – 14 hours / week
○ 15 – 21 hours / week
○ More than 21 hours / week
○ Other

If other, please specify

____

____

5. 13 On average, how many hours of other contact do you have with the person you care for each week? (e.g. telephone, text, email etc)

○ 0 – 7 hours / week
○ 8 – 14 hours / week
○ 15 – 21 hours / week
○ More than 21 hours / week
○ Other

If other, please specify

____

____

Section 6 "Other Issues"

6.1 Have you ever had a problem with eating or weight?

○ Yes
○ No

If Yes, please give brief details of your eating problem (e.g. anorexia, over-eating); and whether you have ever been diagnosed with an eating disorder:

____

____

6.2 Has anyone else in your family ever had a problem with eating or weight?

○ Yes
○ No

If Yes, please detail their relationship to you (e.g. mother or cousin); their eating problem (e.g. anorexia, over-eating); and whether they have ever been diagnosed with an eating disorder:

____

____

6.3 Is there anything else that you would like to add?

____

____