A longitudinal examination of dyadic distress patterns following a skills intervention for carers of adolescents with anorexia nervosa

Dr Laura Salerno^{1**}, Dr Charlotte Rhind^{2*}, Dr Rebecca Hibbs², Dr Nadia Micali^{3,4}, Prof. Ulrike Schmidt², Prof. Simon Gowers⁵, Dr Pamela Macdonald², Dr Elizabeth Goddard², Ms Gillian Todd⁶, Dr Kate Tchanturia^{2,7}, Prof Gianluca Lo Coco¹ and Prof Janet Treasure²

¹ University of Palermo, Department of Psychological and Educational Sciences, Palermo, Italy

² King's College London, Institute of Psychiatry, Psychology & Neuroscience, Department of Psychological

Medicine, Section of Eating Disorders, London, UK

³University College London, Institute of Child Health, Behavioural and Brain Sciences Unit, London, UK

⁴Icahn School of Medicine at Mount Sinai, Department of Psychiatry, New York, NY, US

⁵University of Liverpool, Adolescent Psychiatry, Chester, UK

⁶South London and Maudsley NHS Foundation Trust, London, UK

⁷Ilia State University, Tbilisi, Georgia

* L.S. and C.R. contributed equally to this work

**Corresponding author: Laura Salerno | Address: Department of Psychology and Educational Sciences, University of Palermo, Viale delle Scienze, Edificio 15 – 90128 Palermo, Italy | Email: laurasalerno81@hotmail.com | Ph +39 091 23897725 | fax +39 091 6513825

Acknowledgements: This report/article presents independent research commissioned by the National Institute for Health Research (NIHR) under RfPB programme. PB –PG-0609-19025. Research Title: Expert Carers Helping Others (ECHO) (IRAS Code: 55754 CSP: 55754). The views expressed in this publication

are those of the author(s) and not necessarily those of the NHS, the NIHR or the Department of Health. JT and US receive salary support from the National Institute for Health Research (NIHR) Mental Health Biomedical Research Centre at South London and Maudsley NHS Foundation Trust and King's College London and C.R by the Psychiatry Research Trust. We also thank the following principal investigators and site involved in the ECHO project: S. Clark-Stone, 2gether NHS Foundation Trust; W. Clarke, Anuerin Bevan Local Health Board; D. Robertson, Birmingham and Solihull Mental Health NHS Foundation Trust; N. Dawson, Bradford District Care Trust; C. Schrieber-Kounine, Avon and Wiltshire Mental Health Partnership NHS Trust; J. Shapleske, Cambridgeshire and Peterborough NHS Foundation Trust; J. Whittaker, Central Manchester University Hospitals NHS Foundation Trust; A. Jones, Cornwall Partnership NHS Trust; K. Goss, Coventry & Warwickshire Partnership NHS Trust; H. Crowson, Derbyshire Mental Health Services NHS Trust; K. Higgins, Dudley and Walsall Mental Health NHS Trust; N. Green, Humber Mental Health Teaching NHS Trust; A. Lodwick, Hywel dda Health Board; N. Jacobs, Kent and Medway NHS and Social Care Partnership Trust; C. Newell, Kimmeridge Court, Dorset Healthcare University NHS Foundation Trust; J. Morgan, Leeds Partnership NHS Foundation Trust and St George's University of London; J. Arcelus, Leicestershire Partnership NHS Trust; H. Birchall, Lincolnshire Partnership NHS Foundation Trust; R. Thompson, Norfolk and Waveney Mental Health Foundation Trust; H. Stephens, North Bristol NHS Trust; I. Lea, North Essex Partnership Foundation NHS Trust; L. Addicott, Nottinghamshire Healthcare NHS Trust; S. Sankar, Northamptonshire Healthcare NHS Trust; J. Holliday, Oxford Health NHS Foundation Trust; B. Waites, Powys Local Health Board; H. Strachan, Royal Bolton Hospital; A. Fennell, Black Country Partnership NHS Foundation Trust; A. Wolton, Somerset Partnership NHS Foundation Trust; H. Gahan, South Essex Partnership University NHS Foundation Trust; G. Moss, Sheffield Children's Hospital; J. Orme, Sheffield Health and Social Care NHS Foundation Trust; K. Moore, South Staffordshire and Shropshire Healthcare NHS Foundation Trust; G. Burgoyne, Suffolk Mental Health Partnership NHS Trust; I. Eisler, South London and Maudsley NHS Foundation Trust; B. Bamford, South-West London and St George's Mental Health Trust; I. Yi, Surrey and Borders Partnership NHS Foundation Trust; and P. Parker, Worcestershire Mental Health Partnership NHS Trust. Our appreciation extends also to the Mental Health Research Network who have been instrumental in the successful setup of this project.

Abstract

Family interventions in anorexia nervosa (AN) have been developed to ameliorate maladaptive patterns of patient-carer interaction that can play a role in illness maintenance. The primary aim of this study is to examine the inter-relationship between baseline and post-treatment distress in dyads of carers and patients with AN in order to examine the interdependence between carers and patients. A secondary aim is to examine whether a carer skills intervention (Experienced Carer Helping Others; ECHO) impacts on this inter-relationship. Dyads consisting of treatment-seeking adolescents with AN and their primary carer (n=149; mostly mothers) were randomised to receive a carer skills intervention (ECHO) in addition to treatment as usual (TAU), or TAU alone, as part of a larger clinical trial. Carers and patients completed a standardised measure of psychological distress (The Depression, Anxiety, and Stress Scale) at baseline and 12-months post-treatment. The Actor Partner Interdependence Model was used to examine longitudinal changes in interdependence by treatment group. As expected, post-treatment levels of distress were related to baseline levels in both groups (actor effects). Moreover, carer distress at 12 months was related to patient distress at baseline for the TAU (partner effects), but not for the ECHO group. Finally, carers' distress change was not a significant predictor of patients' Body Mass Index (BMI) change in the two treatment conditions. These findings are limited to predominantly mother-offspring dyads and may not generalise to other relationships. The ECHO intervention which is designed to teach carers skills in illness management and emotion regulation may be an effective addition to TAU for ameliorating interdependence of distress in patients and their primary carers over time.

Keywords: Anorexia Nervosa, Caregiving, Actor-Partner Interdependence Model (APIM), Longitudinal study, Distress, Clinical trial.

Introduction

Adolescent anorexia nervosa (AN) is a serious psychiatric disorder [1] characterized by high morbidity and mortality rates, and the prognosis is improved with early intervention [2]. The evidence base concerning treatment for adolescents with AN indicates that involving the family is a key factor [3]. Family based therapy has been studied most extensively [3]; it focuses on engaging the parents to manage the eating behaviour [4]. Most research using family based treatment has only measured patient outcomes and the impact on carers is unknown. However there has been a call for considering the needs of carers alongside the needs of patients, so called "carer proofing" [5].

Caring for an individual with an eating disorder is burdensome [6] and is associated with distress and a reduction in quality of life [7]. The caregiving role can be "helpful" as exemplified in family based therapy but less helpful, "vicious" circles of interaction can develop whereby high expressed emotion (criticism, hostility, and over protection) and the tendency to collude with the illness by accommodating to and/or enabling eating disorders symptoms and division within the family serve to maintain the illness, as described in the cognitive interpersonal model [8]. The core features of this model are that there are predisposing factors such as obsessive compulsive features and anxious avoidance (particularly of close relationships) which increase the vulnerability to AN and that these contribute also to the maintenance of the disorder because they foster pro anorexia nervosa beliefs and behaviours [9]. Moreover, set shifting and social communication difficulties may be familial vulnerability traits and so have a wider effect by causing a maladaptive response by family members. These less helpful forms of behaviours are often associated with high levels of distress and anxiety in both the patient and the parent [10]. A recent systematic review indicated that parent distress and burden can be reduced by interventions for parents [11]. However, not all of these interventions have targeted these less helpful "vicious" circles of interaction.

The carer skills intervention (Experienced Carers Helping Others; ECHO) was specifically developed [12] to target the interpersonal relationship patterns developed in the cognitive interpersonal model. One facet of the model is based on the premise of emotional contagion within families whereby observing the emotional expressions of another person causes an automatic tendency to share the person's experiences [13]. Carers are therefore taught emotional regulation strategies so that they do not mirror the distress in their

offspring (a potentially "less helpful" interaction which escalates distress in the family). Carers are also taught how to reduce their own emotionally-driven behaviours such as overprotection and/or criticism and accommodation. There have, as yet, been no longitudinal studies that have examined whether this intervention does impact on the inter-relationship between parents' (or primary caregivers) and their offspring's distress and Body Mass Index (BMI).

Most studies about family involvement in the management of people with eating disorders have assessed changes in psychological distress of parents and/or patients. These approaches evaluate how the members of the dyads (patients and parents) change individually, but they do not capture if and how the dyads' interrelationship may have changed. To date, no studies have examined changes in mutual interdependence. This study uses the Actor–Partner Interdependence Model (APIM) [14] as a method for identifying relational changes. The APIM is ideal for examining a relational phenomenon between two interdependent individuals because it treats the dyad (instead of the individual carer or patient) as the unit of analysis [15]. This model has been used to examine mutual collaboration or influence by modeling the impact of one dyad member's ratings on the other member's ratings (partner effects) while accounting for the interdependent nature of their relationship. Furthermore, it has been used in a variety of settings, including those where there is chronic illness in one family member, for example, spouses with chronic heart failure [16, 17], children with asthma [18], cystic fibrosis [19], and elderly parents [20].

The aim of this study was to examine the relationship over time between distress levels in both primary carer and the eating disorder patient, and also to examine the impact of baseline patients' Body Mass Index (BMI), duration of illness and age on carer and patients distress over time. These variables were included as covariates in the APIM, as previous research suggests that they are related to the distress experienced by patients with eating disorders and their carers [11, 21]. A supplementary aim was to examine whether the addition ECHO intervention impacted on this interaction. Finally, we examined whether carers' and patients' change on distress levels over the course of treatment impacts on patients' outcome in terms of BMI. Hypothesis 1: Actor effects. We hypothesized that carers' initial distress would be positively related to their distress at the end of treatment in both the two treatment conditions; and that patients' initial distress will be positively related to their distress at the end of treatment in both the two treatment conditions.

Hypothesis 2: Partner effects. We predicted that, in the treatment as usual (TAU) condition, patients' distress at baseline would predict carers' distress after 12 months whereas, for the carers given the ECHO intervention, no such relationship would be found. This is because one of the skills taught in the ECHO intervention was emotional regulation and a calm caregiving style; and therefore the partner effect would be ameliorated. We expected an association between the carers' distress and the patients' distress after 12 months in the ECHO condition, given the preliminary knowledge of the influence of carer's pre-treatment distress on AN patient's distress [22].

Hypothesis 3: Finally, regarding patients' outcome in terms of BMI change, we expected an association between both patient's and carer's distress change over the course of ECHO intervention and patient's outcome. This is because the collaborative care model focuses on reducing carer's distress and anxiety as they can trigger expressed emotion and accommodating behaviors, which in turn maintain patient's symptoms. On the other hand, patients who improve on their distress level are more likely to report better outcome in terms of BMI.

Methods

Design and Participants

This longitudinal study was a part of a multi-site randomized controlled trial examining the effect of a carer skills intervention (Experienced Carers Helping Others; ECHO) on the outcome of adolescents newly referred for specialist outpatient treatment for AN (Trial registration: ISRCTN83003225) [22]. Thirty-eight eating disorder outpatient services across the UK recruited patients (n=149) and up to three of their carers (n=225) for the study. Carers were identified as by the patient and needed to be currently living with the patient with the intention of living together for the following year. "Carers" are defined by criteria of the Princess Royal Trust (www.carers.org) as someone who provides unpaid help and support to a parent, child partner, relative, friend or neighbour. Carers also described themselves as primary or secondary. Definition of primary and secondary carer in the self-report questionnaire is referred to the level of dependence by the patient and number of hours of contact (i.e. primary carer might be the person who spends the most time with the patient or is the person who is called upon first; a secondary carer may be someone who is still very involved but maybe does not spend quite as much time with the patient or perhaps sees them less frequently).

The collaborating sites included 17 Child and Adolescent Mental Health Services (CAMHS), 13 Adult Mental Health Services and 8 offering both CAMHS and Adult teams. All sites were managed within the National Health Service (NHS) and are listed in the acknowledgements section. Ethics approval was granted by the Northwick Park Hospitals Ethics Committee (11/H0725/4). Site specific ethics and governance approval was granted on all participating sites and this study adopted by the Clinical Research Network.

Consenting carers of patients aged 12-21 years with a primary diagnosis of AN or atypical AN, according to ICD-10 criteria [24] were randomly allocated to receive TAU or TAU in addition to ECHO. For the purposes of the present study, all patients randomized to the clinical trial and their primary carers only (*n* = 149 carer/patient dyads, typically mother/offspring dyads), were included in the analysis. Participants were grouped according to the type of treatment they received: 49 dyads received the ECHO intervention, 50 dyads also received carer coaching (ECHOc; see treatment section), whereas 50 dyads received TAU alone.

Procedure

Following consent families were randomised to ECHO treatment or TAU. The ECHO materials and coaching were delivered from the research hub. There was no further interaction between the research hub and the contributing clinical sites where usual care was delivered. Follow up assessments included computerised self-report instruments and structured, blinded to allocation, interviews for both carer and patients.

Intervention

Treatment as usual

The National Institute of Health and Clinical Excellence (NICE) guidelines have one grade B and several Grade C recommendations for the treatment of AN in adolescents and young adults [25]. Accordingly, most people with AN and atypical presentations should be managed on an outpatient basis with psychological treatment and physical monitoring provided by a health care professional competent to administer care and assess physical risk. NICE recommends this combination of treatment for at least six months' duration, with more intensive forms of treatment to be considered in cases of significant deterioration, non-improvement or in cases of physical risk. Weight restoration in an outpatient setting should aim for an average weekly weight gain of 0.5 kg and for adolescents, family interventions that directly address the eating disorder should be offered (grade B).

As would be expected for a pragmatic trial in EDs, TAU varied to some degree across the 38 collaborating sites included in the present study. However, TAU accessed by patients in the present study sample was comparable between the two treatment groups (TAU alone vs TAU+ECHO intervention) as would be expected as the patients were randomized by site. During the 6-month period from ED outpatient referral, 80.59% of patients received some form of individual therapy (TAU=82%, ECHO=80%), 23.5% of the sample received family therapy (TAU=24%; ECHO=23%), and 33% received dietician counselling (TAU=22%; ECHO=38%). A further 44% of patients also reported visiting their General Practitioner during this time for reasons related to their ED (TAU=48%; ECHO=41%), 11% reported using self-help or telephone helplines (TAU=14%; ECHO=9%) and 34% of the patient sample also reported use of other services, such as the community psychiatric nurse, crisis intervention team, or occupational therapy (TAU=34%; ECHO=41%). During this 6-month period following outpatient referral, 13% of patients (TAU=16%; ECHO=12%) were admitted to inpatient ED, 9% entered a daypatient ED programme (TAU=12%; ECHO=7%), and 15% accessed General Hospital (e.g. Emergency) services for medical reasons related to the ED (TAU=20%; ECHO=13%). Moreover, carers were asked about their use of services for individual reason(s) besides to accompany their relative (i.e. because of direct or indirect consequence of caring activities ED related). During the 6-month period from ED outpatient referral, 13.4% of carers reported visiting their General Practitioner during this time for individual reasons ED related (TAU=16%; ECHO=12.1%), 10.1% reported using self-help or telephone helplines (TAU=10%; ECHO=10.1%) and 3.4% of the carers also reported use of other services, such as the community psychiatric nurse, group therapy, or dietician (TAU=2%; ECHO=4%).

Carer skills intervention (Experienced Carers Helping Others; ECHO).

ECHO uses a skills training approach and consists of a book [12] and five DVDs (three theoretical, two practical). The DVDs complement the information presented in the book with role plays and practical examples. The information on the DVDs is presented visually with audio voiceover. Moreover, 50.5% (n=50) of the ECHO sample (n=99) also received carer coaching (ECHOc). The ECHOc package included 5 telephone coaching sessions per individual. Participants were contacted by the coach within two weeks of receiving the material by post. Calls were made on a regular basis with time in between (e.g. two weeks) for carers to practice the skills. The time taken to complete calls varied between families depending on

individual circumstances. The expectation was that telephone calls lasted up to 40 minutes with a minimum of five calls (per family) to have completed the intervention [26]. The content of the intervention has been summarised [27] and includes teaching carers emotional self-management, how to restrain their own emotionally driven behaviours (high expressed emotion, including emotional over-involvement, and accommodating and enabling behaviours) and communication skills (the basic principles of motivational interviewing).

Measures

Carers and patients provided standard demographic (age, ethnicity, marital/employment status, years in education) and clinical information (illness duration, diagnosis, height and weight) by self-report as part of the baseline assessment of the larger trial. Patient diagnosis and height and weight information were validated by clinicians at the treating site. Both Body Mass Index (BMI) and age standardised weight-for-height percentage, using Great Ormond Street Hospital for Children criteria, version 4.22 UK, were calculated for each patient. Furthermore, the AN-severity index (AN-TSI) of the Short Evaluation of Eating Disorders (SEED) [28] measure was used for the assessment of the severity of AN symptoms.

The Depression, Anxiety, and Stress Scale (DASS-21) [29, 30] was used as an overall assessment measure of psychological distress in patients and carers at baseline (pre-treatment) and 12-month follow-up (post-treatment). The DASS is a 21-item measure assessing depression, anxiety, and stress over the past 7 days using a 4-point Likert Scale. Only the total score was considered in this study. This instrument has good psychometric qualities (Cronbach's alpha = .87 - .94) [31] and has good internal consistency in the present study (DASS at T0: Cronbach's = .94 and .94 for patients and carers, respectively; DASS at T1: Cronbach's = .96 and .95, for patients and carers, respectively).

Statistical Analyses

Data were acquired from questionnaires collected from both patients and carers at two time periods: TO (baseline assessment, prior to treatment) and T1 (following one year of treatment). Descriptive statistics (mean and standard deviations) were examined. Assumptions for parametric data were evaluated and no positive skewed distributions were found. Paired samples t-tests were conducted on patients and carers' DASS to examine differences between T0 and T1 assessments.

The APIM model tested in this study is displayed in Figure 1. Mplus software version 6.12 was used to test the model. Missing values (1.3% for carers' DASS at T0; 0.7% for patients' DASS at T0; 20%, 26.2%, 18.8% for carers and patients' DASS at T1 and patients' BMI at T1, respectively) were handled with FIML, a direct model-based method for estimating parameters in the presence of missing data. Because the path model examined is identified (saturated) there is a perfect fit, so no model fitting statistics are showed. There are four variables in this longitudinal APIM model: two outcome variables (carers' and patients' distress at T1) and two independent (carers' and patients' distress at T0) that are expected to predict the outcome variables. Moreover, three additional patient variables (patients' BMI, duration of illness and age at T0) are also included in the model as independent variables. Carers' and patients' distress at T0 are based on the same measurement instruments as the outcome variables, but measured at an earlier point in time (baseline assessment, prior to treatment).

Using regression language to describe the relationships depicted in the figure there are five actor effects (a measure of a person's own characteristics predicts his or her outcome): (a) the effect of the carers' distress at T0 on their own distress at T1, (b) the effect of the patients' distress at T0 on their own distress at T1, (c) the effect of the patients' BMI at T0 on their own distress at T1, (d) the effect of the patients' duration of illness at T0 on their own distress at T1, and e) the effect of the patients' age at T0 on their own distress at T1. Actor effects are represented in Figure 1 by the five paths labelled *a*. There are also five partner effects (a measure of interdependence, i.e. the extent to which a person's outcome is predicted by his or her partner's earlier characteristics): (a) the effect of the carers' distress at T0 on their children's distress at T1, (b) the effect of the patients' distress at T0 on their carers' distress at T1, (c) the effect of the patients' BMI at T0 on their carers' distress at T1, (d) the effect of the patients' duration of illness at T0 on their carers' distress at T1. Partner effects are represented in Figure 1 by the paths labelled *b*. The APIM also models eleven within dyad correlations (specified by the double-headed arrow in figure 1): the relationships between carers' and patients' distress, BMI, duration of illness and age at baseline and the relationship between carers' and patients' scores after 12 months.

Finally, changes in patients' BMI and distress and carers' distress over the course of treatment were calculated by regressing baseline variables over 12 months variables and a second model was tested. There

are three variables in this longitudinal model: an outcome variable (patients' BMI change during the course of treatment) and two independent variables (carers' and patients' distress change during the course of treatment) that are expected to predict the outcome. Moreover, two additional patient's variables (patients' duration of illness and age at T0) were also included in the model as independent variables. In this model, there are three actor effects: (a) the effect of the patients' distress change on their own BMI change, (b) the effect of the patients' duration of illness at T0 on their own BMI change, and (c) the effect of the patients' age at T0 on their own BMI change. There is also one partner effect: (a) the effect of the carers' distress change on their children's BMI change. The model also tested six correlations: the relationships between carers' and patients' distress change, duration of illness and age.

Results

Baseline Characteristics of Patient-carer Dyads

In the total sample, the patients were adolescents (average age 16.9 years) with a short duration of illness (average duration 22 months). The average BMI was 16.8 kg/m² (age-adjusted weight-for-height 83%) and 75% fulfilled the criteria for ICD-10 AN and 25% for atypical AN (n = 21 with restrictive food intake and n = 16 with binge eating and/or purging behaviors). The majority (93%) of the carers were mothers, of whom 74% were married and 30% were in full time employment. Other carers included fathers (5%), siblings (1%), and grandparent (1%). The clinical and socio-demographic characteristics, differentiated between ECHO and TAU groups are shown in Table 1.

Carer and Patient Distress at baseline and 12 months follow up

No differences were found between ECHO and ECHOc groups on BMI (t = .031, p = .975), duration of illness (t = .062, p = .951) and levels of distress at baseline (for carers, ECHO: M (ds) = 34.62 (24.26), ECHOc: M (ds) = 34.54 (29.51), t = .016, p = .987; for patients, ECHO: M (ds) = 62.98 (29.35), ECHOc: M (ds) = 62.84 (31.74), t = .021, p = .983) and 12 months follow up (for carers, ECHO: M (ds) = 32.73 (26.85), ECHOc: M (ds) = 31.78 (22.68), t = .169, p = .866; for patients, ECHO: M (ds) = 51.85 (33.43), ECHOc: M (ds) = 47.79 (29.81), t = .564, p = .575), so in the current study we compared the ECHO (N = 99) with the TAU condition (N = 50).

The means and standard deviations for all of the variables examined in this study, separated for ECHO and TAU groups, are displayed in Table 2. The ECHO and TAU groups did not significantly differ (data not shown in the table) on level of distress (t = 1.83, p = .07 and t = -1.41, p = .16, for patients and carers, respectively), BMI (t = .481, p = .63) and duration of illness (t = .159, t = .87) at baseline. Patients had extremely severe levels of distress at baseline (mean 67 (30)) and carers had moderate levels (mean 32 (28)). Patients' distress decreased over time in both the ECHO and TAU groups.

Carers in the ECHO group had a small decrease in DASS scores at T1 whereas in the TAU group there was a small increase in DASS.

Actor-Partner Interdependence Model (APIM) Analyses

Actor Effects

The results of APIM approach are displayed in Table 3. For the TAU group, patient and carer's distress, patients' BMI, duration of illness and age at baseline accounted for 43% and 32% of the variance in carer and patient's distress at T1, respectively. For the TAU group, there were significant actor effects for both patients and carers. Specifically, carers' distress at baseline was a significant predictor of their own distress after 12 months and patients' distress at baseline was a significant predictor of their own distress after 12 months. Neither patients' BMI nor duration of illness and age were significant predictors of their own distress at 12 months.

For the ECHO group, patient and carer's distress, BMI, duration of illness and age at baseline accounted for 24% and 30% of the variance in carer and patient's distress after 12 months, respectively. For the ECHO group, there were significant actor effects for both patients and carers. Specifically, carers' distress at baseline was a significant predictor of their own distress after 12 months and patient's distress at baseline was a significant predictor of their own distress after 12 months. Moreover, duration of illness was a nearly significant predictor of patients' distress at 12 months, whereas no relationship was found between patients' BMI and age and their own distress at 12 months.

Partner Effects

The results of partner effects from APIM approach are displayed in Table 3. In the TAU group there was a significant partner effect between patient's distress at baseline and carer's distress at 12 months.

Neither patients' BMI nor duration of illness and age were significant predictor of carers' distress at 12 months. In the ECHO group there were no partner effects for carers and patients.

Actor and Partner Effects on patients' BMI change over the course of treatment

Carer's and patient's change variables accounted for 17% and 19% of the variance in patient's BMI change for the TAU and ECHO groups, respectively. For the TAU group, neither patients' nor carers' distress change over the course of treatment, duration of illness and patients' age were significant predictors of patients' BMI change. For the ECHO group, only patients' distress change was a significant predictor of their own BMI change during the course of treatment (b(β) = -4.380 (-.353), SE = 1.587, t = -2.760, p < .01). Carers' distress change, and patients' duration of illness and age were not significant predictors of patients' BMI change.

Discussion

The aim of this study was to examine the relationship over time between distress levels in both primary carer and the eating disorder patient, and also to examine the impact of baseline patients' BMI, duration of illness and age on carer and patients distress over time. A secondary aim was to examine whether this interaction could be improved (i.e. reduce distress inter-dependence) with treatment augmented with ECHO; and whether patient's and carer's distress change over the course of ECHO intervention would be associated with patient's outcome, in terms of BMI change.

Both the carers' and patients' actor effect hypotheses were supported, in that we found that carers' initial distress was positively related to their distress at the end of treatment in both the treatment conditions; and patients' initial distress was positively related to their distress at the end of treatment in both the treatment conditions. We also confirmed our second hypothesis in that we found that carers who received the ECHO treatment were less impacted by patients' initial levels of distress (i.e. no partner effects were found), whereas there were partner effects for the TAU group, as carers' levels of distress at one year were related to the patient's initial level of distress. Neither patients' baseline BMI nor duration of illness and age were significant predictors of carers' and patients' distress after 12 months.

The findings on actor effects seem consistent with previous studies which showed the relationship between pre and post treatment carers' distress [11] as well as the negative relationship between symptomatic distress at baseline and AN patients' mental health after 6-12 month or at the end of treatment [32]. Interestingly, when we also examined the patients' outcome in terms of BMI change, we found that patients who improved on their distress level were more likely to report a better outcome after 12 months in terms of BMI change, in the ECHO condition. Our findings are also in line with previous research which showed that carers of AN patients reported moderate levels of distress as measured with the DASS, especially for mothers [33].

In the current study, the results on the partner effects suggest that the ECHO intervention, delivered in addition to TAU, might be effective in ameliorating interdependence of distress over time. These findings are in line with previous evidence of interdependence between family members in chronic illness. In one of the few studies which investigated the interdependence between mothers and fathers on unhelpful caregiving behaviors, and its impact on the outcome of AN patients [34], it was showed that patients' symptomatic distress was worse when both mother and father were high in accommodation. Also in a study of families with elderly relatives a partner effect was found, whereby caregiver's spirituality significantly influenced the elder's psychological well-being [20]. Interestingly, the hypothesis regarding the impact of carers' distress change on patients' outcome (in terms of BMI) was not supported, suggesting that the dyad interdependence seems to play a role more for patients' distress than for their BMI. However, consistently with the collaborative care model, patients' change on distress level predicted their BMI change in the ECHO condition. Further research is needed to establish the complex interactions between carer's pretreatment distress and patient's outcome, as suggested by the cognitive interpersonal model of AN.

To our knowledge, this has been the first study which has examined the impact of an intervention targeting caregiving behaviours on these partner effects. Future research needs to collect more dyadic data on the effects of patients interventions on carers and consider carers' health outcomes as well as patients' [5].

It is of note that the majority of studies in the literature regarding the efficacy of family-based treatment with adolescent AN patients have only focused on the patient's outcome, without reporting data relating to the carers [21, 31, 35]. Thus, we have very little evidence about the influence of the patient's distress on carer's outcome. Moreover, no previous study on the efficacy of family-based treatment modeled

couple-related data. The findings of the current study are promising and seem to support the complex interactions between AN patients and carers which are described in the cognitive-interpersonal maintenance model of AN [9], but further evidence is needed on how interpersonal relationships become entangled with the eating disorder and what therapeutic strategy can be effective in reducing dysfunctional relationship patterns. Carers usually find their caregiving role as burdensome and distressing [33]. Our findings suggest that when carers received the ECHO intervention, which utilized a skills training approach with several strategies that have been identified as important for behaviour change [36], they were less influenced by their children's depression, anxiety and stress.

The strengths of the study described in this paper include the use of a sample with high external validity and the use of the APIM model, which is specifically designed to test associations between correlated constructs from two members of a dyad. Despite such strengths, various limitations must be considered when interpreting the results of this study. We have only considered the interaction effect between the primary carer (predominantly mothers) and patient (typically offspring). Although up to three carers were invited to participate in the overall clinical trial, including fathers for 47% of patients, there were insufficient data to analyse other dyadic relationships. However, it would also be interesting to examine partner effects for fathers as they have an important impact. Furthermore, the patients included in this present study predominantly have a short illness course and a moderate severity. Therefore, these findings may be limited in the extent to which they can be generalized to patients with duration of illness longer than two years. Moreover, as inclusion criteria for the larger clinical trial required at least one carer to also participate, the present sample might be biased towards a sample with parents and other carers more actively involved in their care. It is also worth note that the measure of baseline and post-treatment distress is a short 21-items screening tool with a Likert scale and a global evaluation of distress is considered for this study. Limitations of the Likert scale are well known and future studies should use different diagnostic measures to deeper understand which specific aspects of patients' and carers' distress are more affected by the ECHO intervention. A limitation of the study design is the heterogeneity in TAU provision across treatment centres, given that patients and families can vary in their level of engagement in treatment programmes. It is interesting that only 24% of participants in the TAU reported having received family therapy as family therapy/involvement was recommended for adolescents in since the NICE guidelines. The use of TAU as a

comparison condition in a controlled trial presents several strengths as well as challenges and limitations [37], including the huge variability of what TAU encompasses. Further research is needed to compare the findings of the ECHO condition with a more structured family-based intervention and to evaluate possible treatment type (e.g. individual or family therapy or hospitalization rate) and distress interaction. Finally, in the current study we adopted the APIM approach to study the effect of predictors on individual's distress in both members of the couple (patient and caregivers). Further research should address how the couple as a whole copes with stressors and illness and the Common fate model could be an appropriate analytic model for this goal, as suggested by Lederman & Kenny [38].

Clinical implications

The findings from the current study have clinical implications. Despite previous research highlighting the impact that living with anorexia nervosa has on multiple aspects of family functioning [11, 39], only a few studies examined how the dyads' inter-relationship have changed and looked for "carer proof' effects. These findings suggest that the emotional regulation skills taught in the ECHO intervention had a beneficial effect in terms of breaking the unhelpful cycle of maintenance in which carers mirror the distress in their affected relative, which in turn leads to more distress in the patient, and other maladaptive caregiver responses (e.g. high levels of expressed emotion). Our findings showed that carers of AN patients report moderate/high levels of distress. This highlights the need to develop specific interventions to address their suffering. The current results are promising in validating family treatments based on the cognitive interpersonal maintenance model of AN, as it suggests that a specific intervention targeting one of the maintaining factors (i.e., accommodating and enabling behaviors, high expressed emotions) can reduce carer's distress [9]. Other family interventions that share skills in emotion regulation within an interpersonal context may also provide similar benefit. Finally, the finding that neither the patient's BMI nor the duration of illness was a predictor of carer's distress after one year, highlighted the need to assess patient's distress alongside BMI when working with family caregivers, given its potential negative influence on carer's distress.

Conclusion

Our findings suggest that Actor–Partner Interdependence Model of Analyses can be a useful method of examining the processes involved in family interventions. In the TAU group we found interdependence of

distress between patients and primary carers (mostly mothers) over time. However, this mutual influence was not seen in the ECHO group. This suggests that skills taught in the ECHO intervention were having a positive effect in terms of breaking a positive feedback effect, whereby carers mirror the distress in their offspring which, in turn, leads to more distress in the patient, as theorized in the interpersonal component of the cognitive interpersonal maintenance model of AN.

Conflict of Interest

JT is a co-author of the book used in the carers skills training intervention ECHO and receives royalties.

References

- Micali N, Hagberg KW, Petersen I, Treasure JL (2013) The incidence of eating disorders in the UK in 2000-2009: Findings from the General Practice Research Database. BMJ Open 3.
- Treasure J, Stein D, Maguire S (2015) Has the time come for a staging model to map the course of
 eating disorders from high risk to severe enduring illness? An examination of the evidence. Early Interv
 Psychiatry 9:173-184. doi: 10.1111/eip.12170
- Lock J (2015) An Update on Evidence-Based Psychosocial Treatments for Eating Disorders in Children and Adolescents. J Clin Child Adolesc Psychol 44: 702-721. doi: 10.1080/15374416.2014.971458
- 4. Lock J, Le Grange D (2013) Treatment manual for anorexia nervosa: A family-based approach (2nd ed.). New York, NY: Guilford.
- 5. Al-Janabi H, Nicholls J, Oyebode J (2016). The need to "carer proof" healthcare decisions. BMJ, 352:i1651. Doi: 10.1136/bmj.i1651
- 6. Raenker S, Hibbs R, Goddard E, Naumann U, Arcelus J, Ayton A, Bamford B, Boughton N, Connan F, Goss K, Lazlo B, Morgan J, Moore K, Robertson D, Schreiber-Kounine C, Sharma S, Whitehead L, Beecham J, Schmidt U, Treasure J (2013) Caregiving and coping in carers of people with anorexia nervosa admitted for intensive hospital care. Int J Eat Disord 46: 346-354. doi: 10.1002/eat.22068
- 7. Anastasiadou D, Medina-Pradas C, Sepulveda AR, Treasure J (2014) A systematic review of family caregiving in eating disorders. Eat Behav 15:464-477. doi: 10.1016/j.eatbeh.2014.06.001
- 8. Schmidt U, Treasure J (2006) Anorexia nervosa: valued and visible. A cognitive-interpersonal maintenance model and its implications for research and practice. Br J Clin Psychol 45:343-366.
- 9. 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. J Eat Disord 15:1-13. doi: 10.1186/2050-2974-1-13
- 10. Goddard E, Salerno L, Hibbs R, Raenker S, Naumann U, Arcelus J, Ayton A, Boughton N, Connan F, Goss K, Lacey H, Laszlo B, Morgan J, Moore K, Robertson D, Schreiber-Kounine C, Sharma S, Whitehead L, Schmidt U, Treasure J (2013) Empirical examination of the interpersonal maintenance model of anorexia nervosa. Int J Eat Disord 46:867-874. doi: 10.1002/eat.22172

- 11. Hibbs R, Rhind C, Leppanen J, Treasure J (2015) Interventions for caregivers of someone with an eating disorder: A meta-analysis. Int J Eat Disord 48:349-361. doi: 10.1002/eat.22298
- 12. Treasure J, Smith G, Crane A (2007) Skills-based learning for caring for a loved one with an eating disorder: The new Maudsley method. London, Routledge.
- 13. Frith CD, Frith U (2006) The Neural Basis of Mentalizing. Neuron *5*0:531-534. doi: 10.1016/j.neuron.2006.05.001
- Kenny DA (1996) Models of non-independence in dyadic research. J Soc Pers Relat 13:279-294. doi: 10.1177/0265407596132007
- 15. Kenny DA, Kashy DA, Cook WL (2006) Dyadic data analysis. New York, NY: Guilford Press.
- 16. Chung ML, Moser DK, Lennie TA, Rayens MK (2009) The effects of depressive symptoms and anxiety on quality of life in patients with heart failure and their spouses: testing dyadic dynamics using Actor-Partner Interdependence Model. J Psychosom Res 67:29-35. doi: 10.1016/j.jpsychores.2009.01.009
- 17. Vellone E, Chung ML, Cocchieri A, Rocco G, Alvaro R, Riegel B (2014) Effects of self-care on quality of life in adults with heart failure and their spousal caregivers: testing dyadic dynamics using the actorpartner interdependence model. J Fam Nurs 20:120-141. doi: 10.1177/1074840713510205
- Rayens MK, Svavarsdottir EK (2003) A new methodological approach in nursing research: an actor, partner, and interaction effect model for family outcomes. Res Nurs Health 26:409-419. doi: 10.1002/nur.10100
- Driscoll KA, Schatschneider C, McGinnity K, Modi AC (2012) Application of dyadic data analysis in pediatric psychology: cystic fibrosis health-related quality of life and anxiety in child-caregiver dyads. J Pediatr Psychol 37:605-611. doi: 10.1093/jpepsy/jss063
- Kim SS, Reed PG, Hayward RD, Kang Y, Koenig HG (2011) Spirituality and psychological well-being: testing a theory of family interdependence among family caregivers and their elders. Res Nurs Health 34:103-115. doi: 10.1002/nur.20425
- La Grange D, Lock J, Agras WS, Moye A, Bryson SW, Jo B, Kraemer HC (2012) Moderators and mediators of remission in family-based treatment and adolescent focused therapy for anorexia nervosa.
 Behav Res Ther 50:85-92. doi: 10.1016/j.brat.2011.11.003

- 22. 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. Int J Eat Disord 41:233-242. doi: 10.1002/eat.20488
- 23. Rhind C, Hibbs R, Goddard E, Schmidt U, Micali N, Gowers S et al (2014) Experienced Carers Helping Others (ECHO): Protocol for a pilot randomised controlled trial to examine a psycho-educational intervention for adolescents with anorexia nervosa and their carers. Eur Eat Disord Rev 22:267-277. doi: 10.1002/erv.2298
- 24. WHO (1992) The International Classification of Disease Version 10 (ICD-10): Classification of Mental and Behavioural Disorders. Geneva, World Health Organisation.
- 25. NICE (2004) National Collaborating Centre for Mental Health. Eating Disorders: Core Interventions in the treatment and management of anorexia nervosa, bulimia nervosa and related eating disorders. Number CG9 London: The British Psychological Society.
- 26. Goddard, E., Raenker, S., Macdonald, P., Todd, G., Beecham, J., Naumann, U., Bonin, E. M., Schmidt, U., Landau, S. & Treasure, J. (2013). 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. Eur Eat Disord Rev 21, 60-71. doi: 10.1002/erv.2193
- Treasure J, Rhind C, Macdonald P, Todd G (2015) Collaborative Care: The New Maudsley Model. Eat
 Disord 23:366-376. doi: 10.1080/10640266.2015.1044351
- 28. Bauer S, Winn S, Schmidt U, Kordy H (2005) Construction, scoring and validation of the short evaluation of eating disorders (SEED). Eur Eat Disord Rev 13:191-200. doi: 10.1002/erv.637
- Lovibond PF, Lovibond SH (1995) The structure of negative emotional states: Comparison of the depression anxiety stress scales (DASS) with the Beck Depression and Anxiety Inventories. Behav Res Ther 33:335-343. doi:10.1016/0005-7967(94)00075-U
- Lovibond, S.H. & Lovibond, P.F. (1995). Manual for the Depression Anxiety Stress Scales. (2nd.
 Ed.) Sydney: Psychology Foundation. ISBN 7334-1423-0.

- 31. Antony MM, Cox BJ, Enns MW, Bieling PJ, Swinson RP (1998) Psychometric properties of the 42-item and 21-item versions of the Depression Anxiety Stress Scales in clinical groups and a community sample. Psychol Assessment, 10:176-181. doi: 10.1037/1040-3590.10.2.176
- 32. Vall E, Wade TD (2015) Predictors of treatment outcome in individuals with eating disorders: A systematic review and meta-analysis. Int J Eat Disord 7:946–971. doi: 10.1002/eat.22411
- 33. Anastasiadou, D., Medina-Pradas, c., Sepulveda, A., & Treasure, J. (2014). A systematic review of family caregiving in eating disorders. Eat Behav, 15(3):464-77. doi: 10.1016/j.eatbeh.2014.06.001
- 34. Salerno L., Rhind C., Hibbs R., Micali N., Schmidt U., Gowers S., Macdonald P., Goddard E., Todd G., Lo Coco G., 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. J Affect Disorders, 191: 230-236. doi: 10.1016/j.jad.2015.11.016
- 35. Agras WS, Lock J, Brandt H, Bryson SW, Dodge E, Halmi KA, Jo B, Johnson C, Kaye W, Wilfley D, Woodside B (2014). Comparison of 2 family therapies for adolescent anorexia nervosa: a randomized parallel trial. JAMA Psychiatry 71:1279-1286. doi: 10.1001/jamapsychiatry.2014.1025
- 36. Michie S, van Stralen MM, West R (2011). The behaviour change wheel: a new method for characterising and designing behaviour change interventions. Implement Sci. 6:42. doi: 10.1186/1748-5908-6-42
- 37. Kazdin AE (2015). Treatment as usual and routine care in research and clinical practice. Clin Psychol Rev. 42: 168-178. doi: 10.1016/j.cpr.2015.08.006
- 38. Ledermann, T., & Kenny, D. A. (2012). The common fate model for dyadic data: Variations of a theoretically important but underutilized model. J Fam Psychol, 26, 140–148. doi: 10.1037/a0026624
- 39. Zabala MJ, Macdonald P, Treasure J (2009). Appraisal of caregiving burden, expressed emotion and psychological distress in families of people with eating disorders: A systematic review. Eur Eat Disord Rev, 17, 338-349. doi: 10.1002/erv.925