

Adherence to Family-Based Treatment (FBT) in Studies of Youth with Anorexia Nervosa: A Critical Review of the Literature

Afeeq Shaszwan Zambre^{1*}, Lesley O'Hara², Sara McDevitt³, Kristen Michelle Maunder^{4,5} and Fiona McNicholas^{1,2,5,6}

¹Department of Medical Science, University College Dublin, Belfield, Dublin, Republic of Ireland

²Lucena Clinic Services, Rathgar, Rathfarnham, Dublin, Republic of Ireland

³Department of Psychiatry, University College Cork, Cork, Republic of Ireland

⁴Department of Psychiatry, The University of Dublin, College Green, Dublin, Republic of Ireland

⁵Department of Child Psychiatry, Our Lady's Hospital for Sick Children, Crumlin, Dublin, Republic of Ireland

⁶Geary Institute, University College Dublin, Belfield, Dublin, Republic of Ireland

* **Corresponding author:** Afeeq Shaszwan Zambre, Department of Medical Science, University College Dublin, Belfield, Dublin, Republic of Ireland, Tel: +60176307371; E-mail: afeeqshaszwan@gmail.com

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Abstract

Objectives: Family-Based Treatment (FBT) is a manualised out-patient approach delivered by trained clinicians. FBT is recommended as first-line treatment for anorexia nervosa (AN) in youth. As FBT moves from specialist centers to the community setting, clinician drift may contribute to variations in delivery of the treatment and subsequently outcome. The aim of this research, in the context of the development of national FBT training in Ireland, was to systematically review FBT-effectiveness literature, and to determine based on details in articles published if the reader can ascertain the extent of FBT treatment adherence and key components of treatment success. For the purpose of this paper, reporting on FBT adherence rather than fidelity is reviewed.

Methods: 320 papers were screened and 15 papers reporting on FBT outcome in AN were included in the final analysis.

Results: Evidence such as shorter duration of illness, fewer hospital admissions and better cost effectiveness compared to some other therapies support FBT as an evidenced based and effective intervention in youth with AN. General principles of FBT were given in most papers as were important details on therapist expertise and supervision. However, specific details measuring therapist adherence to the core aspects of FBT treatment were absent from most of the written articles, making it impossible to establish the impact of manual deviation on outcome.

Conclusion: Examining associations between treatment adherence and therapeutic outcome is essential to ensure optimal training and delivery of FBT for youth with AN, especially as treatment becomes disseminated to varied settings.

Keyword: Fidelity; Adherence; Anorexia nervosa; Family-Based treatment; Adolescence

Introduction

Anorexia Nervosa (AN) is a severe eating disorder characterized by body image disturbance, pursuit of thinness, and disordered eating behaviour. AN is estimated to occur in 1-4% of women and 0.2% of men. Although incidence rates have been stable over the last decade overall, the rate among adolescent females aged 15-19 has increased. AN continues to pose considerable risk by virtue of its often-chronic course, high mortality rate and adverse effects on family. Recent meta-analysis suggests that the standardised mortality ratio (SMR) for AN is 5.86 (95% CI;4.17-8.26) with a mean follow up period of 14 years [1].

Given the impact on the young person and family, it is fitting that strenuous efforts have been made in identifying effective treatments. There is a growing body of evidence supporting the essential role played by parents for treatment success. Family-Based Treatment (FBT) is an out-patient based treatment, delivered over three phases, commencing with weekly sessions for about 10 weeks and focussing almost entirely on seeing the family as a resource and empowering parents to refeed their child, with the support of siblings as all family members are expected to attend the sessions. The second phase gradually shifts the responsibility of eating back to the adolescent, and it is only in the 3rd phase with monthly appointments, that broader adolescent issues become a focus of attention and are addressed. Some features, considered key to treatment success, such as the setting in the first session of 'a grave scene' by highlighting the severity and high mortality rate of the illness, 'the family meal' in the second session, and a delayed focus on non-related eating disorder difficulties to the last phase,

separate FBT from other therapies (**Figure 1**). Other components also considered essential by the authors of FBT include a deliberate focus on early refeeding, regular weighing of the child, and utilizing families as a resource and empowering parents to interrupt the child's symptoms [2,3]

FBT has now been recommended as first line treatment in youths with AN by the 2014 Royal Australian and New Zealand College of Psychiatry Guidelines and by the American Academy of Child & Adolescent Psychiatry Guidelines. Although not specifically named as first line by 2017 NICE guidelines (Eating disorders: recognition and treatment, the recommended 'AN-focussed family therapy' share many of the guiding principles of the FBT model [4]. A treatment manual facilitating standardized delivery of FBT has been developed by Lock and Le Grange, modelled on a previously delivered family-based intervention originating at the Maudsley Hospital in London. Alongside the establishment of a National Clinical Programme for Eating Disorders in Ireland, the Health Service Executive has recommended FBT as the treatment of choice, and established a national FBT training program [5-8].

Despite the existence of evidence-based approaches and training in specific treatment modalities within mental health and psychology services, it is recognised that clinicians regularly diverge from, or are non-adherent to, core components of treatments. This is true even in the absence of clinical rational and especially as treatment moves from research and specialist centres to real world community mental health settings [9].

Substantial divergence in adherence limits treatment efficacy, and if unrecognised, may also limit conclusions drawn from research studies. Assessment of treatment adherence is therefore essential when evaluating treatment effectiveness. Manualisation of treatment goes some way towards treatment standardization but treatment deviations still need to be measured and reported. Guidelines for both the conduct and the systematic reporting on effectiveness studies have been established, with the aim to maximize the likelihood that researchers conduct and subsequently report in a standardised way on key elements of studies. Guidelines for development and conduct of clinical trials, Good Clinical Practice (GCP), are regularly evaluated and updated, ensuring most current standards of rigor and practice are applied. These shape the manual of procedure (MOP) which details the operating procedures for any study, and can be made available to the reader. The most familiar reporting guidelines are for randomised controlled trials (RCT) studies, developed by the Consolidated Standards of Reporting Trials (CONSORT) group. Less frequently used is the TREND (Transparent Reporting of Evaluations with Nonrandomized Designs). However, it is recognised that often papers published in peer-reviewed journals fall short of reported items as set out in CONSORT [10-12].

Applying these principles to the case of studies reporting on FBT, a clear description of the intervention given, and both treatment fidelity and adherence is required in published manuscripts allowing for study replication, and minimisation of Type I or II errors, and to give readers security on data credibility and validity. If treatment adherence is not measured, ad-hoc practice may ensue, ineffective treatments may be disseminated, and effective treatments erroneously discarded at significant personal and service cost.

There is a subtle but important difference between adherence and fidelity. In this paper we consider adherence to be concerned with therapist delivery of the therapy and participants' behaviour in terms of following advice offered. For example did the therapist set a grave scene, request siblings to attend sessions, and focus on weight restoration? Equally adherence would concern itself with the degree to which parents and youth took on board the advice offered, for example did the parents prepare and plate the food, or ensure meal completion, and did they avoid giving responsibility for meal supervision to a sibling. Treatment non-adherence may also occur when parents don't attend scheduled appointments, or when therapists give more frequent or longer sessions. On the other hand FBT fidelity, as outlined in the fidelity literature, is the degree to which treatment is 'delivered in a comparable manner to all participants and is true to the theory and goals underlying the research'. It also incorporates therapist competence, the degree to how well procedures were carried out with sophisticated clinical skills that promote behavioural change, and how effective it was, as well as whether the therapist avoided interventions not specified in FBT manual, as set out by Lock & Le Grange. These attributes are fundamental to discerning the efficacy of FBT studies because non-adherence may cause an underestimation of the treatment effect or a failure to detect a treatment effect when one actually exists (i.e., Type II errors). Despite being a key variable in outcome research, outside of medication RCTs, evaluation of treatment adherence remains limited [13-17].

Thus, the aim of this research, in anticipation of a roll out and evaluation of the national FBT training program in Ireland, was (i) to systematically review and report on the FBT-effectiveness literature for youth with AN and (ii) to establish whether any intervention-specific mediators are identified in past studies of FBT to help inform a future Irish FBT evaluation study. As referred to above, treatment adherence is defined as the extent to which the published manuscript provides sufficient detail so that the reader can ascertain whether the treatment offered adhered to the treatment components as outlined in the FBT manual [3]. A skills study specific adherence tool was designed by the authors to assist in their evaluation of each reviewed paper.

Methods

A literature review was conducted to ascertain all outcome studies in AN using FBT. The Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guidelines were used for reporting our findings.

Search strategy

The search strategy was conducted using the following databases PubMed, Web of Science, PsycINFO and PsycARTICLES (ASZ) and updated in December 2018 (KM).

Search terms

A literature search was carried out using 3 headings. The first search term heading was "Family-based treatment" OR "Family based therapy" OR "family therapy" OR "FBT" OR "family-based treatment" OR "Maudsley model" OR "Maudsley method" OR "Maudsley family therapy" OR "Maudsley approach", the second term was "Eating disorder" OR "Eating disorders" OR "anorexia nervosa" OR "anorexia" OR "anorexic", and the third term was "Outcome".

Inclusion/exclusion criteria

Given that the FBT treatment manual was published in 2001, the search included empirical studies published between the time frame of 01/01/2001 and 17/12/2018. In addition, only publications in English were included. Articles reporting on FBT, but restricted to bulimia nervosa were excluded.

Paper extracon

Search results were reviewed by ASZ and KM to ensure they fulfilled inclusion criteria and duplicates were removed. All papers included in the review were evaluated by all research team members. In cases of uncertainty, all authors reviewed the full article and reached an agreement following team discussion. **Figure 1** shows the PRISMA flow diagram detailing the number of articles excluded after a title, abstract review and full-text review.

A list of all eligible studies, along with clinical and study details including patient demographics of those assigned to FBT, treatment duration, study outcomes and support for efficacy of FBT, are included. Some papers were follow-up papers based on previously described outcomes and these have been highlighted. If the main focus of the paper was not on outcome, it was excluded. However, as the intention of this paper was to consider each published article and the degree to which a reader can evaluate treatment adherence, it was not omitted from overall scoring.

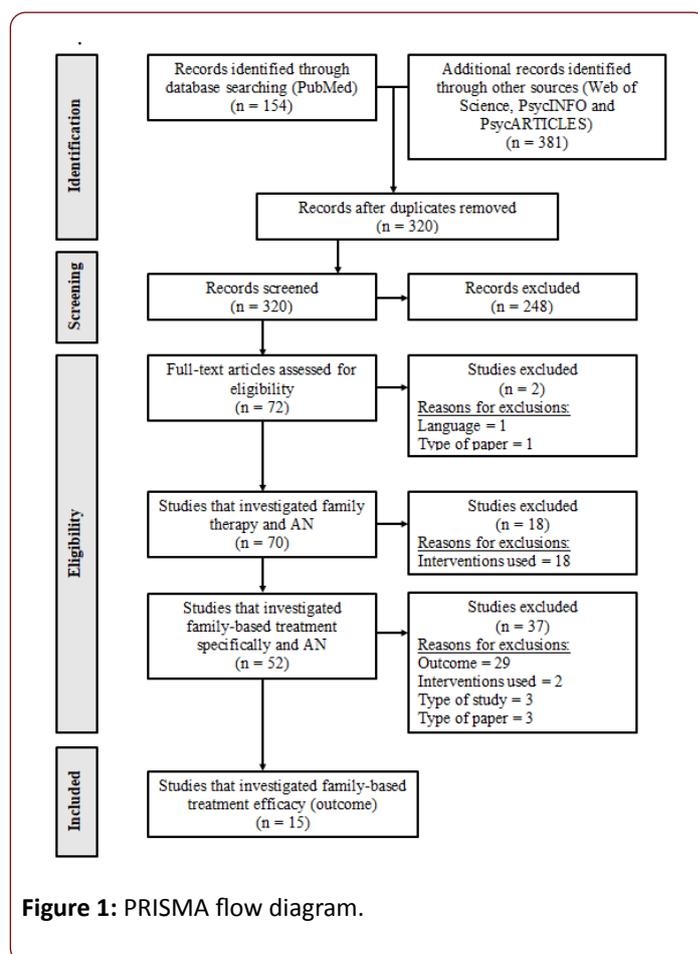


Figure 1: PRISMA flow diagram.

Adherence testing

A study specific FBT adherence checklist was constructed incorporating the fundamental guiding principles outlined by the FBT authors [18]. The first 4 items pertain to FBT structure (salience of family involvement, session frequency of approximately 20, utilising a family meal, taking and sharing weight at outset) and the subsequent 7 pertain to FBT principles and prescribed interventions (setting a grave scene, taking an agnostic approach, being non-authoritarian, encouraging externalisation of the disorder, empowering parents to take control of the renourishment process, nutrition and weight gain, and encouraging sibling support). These are listed in **Table 1** (Treatment manual for anorexia nervosa: a family-based approach) and reflect key goals of treatment.

All papers were rated by the lead author (ASZ) and at least one other team member (LOH, SMD). All 4 authors (ASZ, LOH, SMD, FMN) rated one paper at the start to assist with rating calibration.

Each on the 11 adherence items using the following criteria: 'no mention in paper' (0) 'some mention but not explicit' (1 point) and a 'detailed, clear mention' (2 points). This paper does not report on whether the therapist was in fact adhering to the protocol, but rather whether, and if so to what extent, there was degree of adherence referenced in the manuscript. It is to be made clear that we are attempting to measure adherence which is the extent to which the authors document on whether the

therapists providing treatment follow the manualised FBT approach. We are not referring to the study participants (i.e. families and youth) adherence to recommendations made. All raters reviewed each paper independently, keeping narrative

notes to justify adherence scores given. Discrepancies between scores were discussed as a group and if a consensus was not reached, the scores on sub sections were averaged and an overall paper agreement established by consensus and senior author opinion (FMN).

Table 1: FBT key structural elements.

No.	Adherence Item	Item description with reference to FBT Manual	Adherence Rating 0-no mention 1-some mention 2-detailed mention
1	Family involvement	FBT manual recommend that all family members attend.	
2	Session frequency	Frequency of attendance.	
3	The family meal	Parents are invited to prepare and bring a family meal that will adequately commence the process of re-nourishing their child. This session is considered essential as it allows insight into family meal planning, decision making and ends with the encouragement to have the parent direct their child to 'eat one more mouthful'.	
4	Taking and sharing weight at outset	Weekly weights should be taken and shared with the family.	
	FBT Methodology		Total 0-8 range
FBT Principles and Prescribed Interventions			
5	Setting a grave scene by conveying the seriousness of AN	The therapeutic aim is to engage the family's concern about the seriousness of the patient's condition in a manner that is described as simultaneously grave and portentous yet sympathetic and warm. The aim is to calibrate parental anxiety and concern (heightened anxiety when low and contain when high) so that parents can take appropriate action.	
6	An agnostic view about the cause of AN	Therapists hold an agnostic view as to the cause of illness, removing guilt from the family or others and focuses on commencing treatment.	
7	Non-Authoritarian stance as a therapist	Therapists need to adopt an active consultative and collaborative stance in relationship with the parents. The therapist is not responsible for weight restoration or directly interceding with patients to address other necessary behavioural changes. The therapist does not direct or prescribe interventions for parents, but rather provides information about strategies that might work, assists parents to think through options and to evaluate success of parental efforts.	
8	Encouragement to externalise disorder (externalisation)	In stressing that the patient has little control over their illness, the therapist tries to enable the parents to take drastic action against the illness and not against their child. By stressing that the eating disorder is not identical to the patient themselves, parents can provide support for their child while at the same time distinguishing the illness from the patient as a person. A metaphor can be used aimed at the parent's level of anxiety and understanding of the illness.	
9	Empowering parents to take control of the re-nourishment process and interrupting eating disorder behaviours	Empowering parents as competent agents for ensuring weight gain in the starved offspring. Again and again, attention is brought back to the parents need to take charge in sessions and make decisions necessary to refeed their child. The family should be the major resource for recovery. The therapist should alert the parents not to become engaged in discussions about 'diet' food and emphasize that they should nourish the patient according to the state of malnutrition, not according to the wishes of the illness, AN.	
10	Salience of eating, nutrition and weight gain (directing family to food and eating)	FBT treatment focuses of these three aspects. There is directing and redirecting of conversations back to food, feeding and weight gain.	
11	Sibling support	The focus on sibling involvement is one of support (outside of mealtimes), reinforcing healthy intergenerational boundaries and preventing siblings from interfering with the parents' task of restoring weight to their starving child. Sibling support in not to assist in weight restoration, as this is exclusively within the parents' domain. The support will vary depending on the relationship that existed between the siblings prior to the onset of illness.	
	FBT Principles and Prescribed Interventions		Total 0-14
	Overall FBT Adherence		Total score 0-22

Data Collection

Individual item scores were assigned by team members for each paper allowing calculation of a subtotal FBT process adherence score (items 1 through 4, score range 0-8), subtotal FBT principles and prescribed interventions (items 5 through 11 range 0-14), and total adherence score (range 0-22). A higher score corresponded to higher reported adherence to the FBT treatment manual.

Raters also reported on the overall description provided in the manuscript on key structural elements of FBT, reference to the use of the FBT manual, description of therapist expertise and supervision provided. Inter-rater percentage agreement was calculated for the rater pairs: rater 1/2 68%, rater 1/3 78%, and for all 4 70%.

Result

Search results

320 articles were screened, and 72 full-text articles were read fully for suitability from which 15 were included in the quantitative synthesis. **Figure 1** shows the PRISMA flow diagram and documents the number of articles excluded at each stage: after a title review, abstract review, and full-text review of the studies.

Study data

Clinical summary data for the studies are displayed in **Table 2**. All of the 15 papers involved the use of FBT treatment for youth (age range 9-18), diagnosed with AN, with a duration of follow up from 6-12 months. Most participants were females.

Of the 15 studies, 8 referenced the 2002 edition of the FBT treatment manual, studies referenced the second edition in

2013, while 3 studies did not specify which treatment manual was used. Some papers were drawn from previous study populations [19-29].

FBT outcomes

Outcome measures varied between studies, from weight restoration, return of menses and/or reduction in eating disorder symptomatology. Some of the outcomes were also described in some of the studies as a combination of these variables with remission including both physical and psychological measurements. FBT was compared with active treatment in six studies, focussed individual therapy (3), Parent Focused Treatment (2) and systemic family therapy (1). 9 studies reported on outcomes of FBT treatment alone [30,31]

All 15 studies supported the efficacy of FBT. This was seen in the various studies showing evidence of FBT being acceptable and effective for treating AN via effective weight restoration, improvement of psychological symptoms (e.g. negative affect), return of menses and having positive impacts on family functioning. FBT was also successful at facilitating full remission at follow-up with outcomes being stable post treatment. When compared directly with Systemic Family Therapy, although there was no difference in BMI outcomes at follow up, FBT was found to result in faster recovery, resulted in fewer hospital admissions and was more cost effective.

FBT training and supervision

8 out of the 15 studies mentioned that FBT supervision was provided by the authors of FBT (James Lock and/or Daniel Le Grange) directly, online, or by telephone. All the studies we identified (N=15) had at least one of the authors of the FBT manual involved as authors in the paper. 10 of the 15 studies identified in this systematic review gave details on professional clinical background and training received.

Table 2: Study descriptive data.

Study		Study Info					
		N	Age (range years)	Female (%)	FBT Length (months)	Description of Study Outcome Variable(s)	Support for Efficacy of FBT
Lock J et al., 2005 (Lock et al., 2005)	Short term	44	Dec-18	39 (89%)	6	Comparison of short and long term FBT treatment in youths diagnosed with AN.	Positive. No difference between short or long term treatments.
	Long term	42	Dec-18	38 (91%)	12		
Le Grange D et al., 2005 (Le Grange et al., 2005)		45	Sep-18	39 (87%)	12	Description of a case series of children and adolescents with AN receiving manualised FBT	Positive. Preliminary support for FBT found.
Lock J et al., 2006 (Lock et al., 2006)		29	9-12.9	26 (81.2%)	9	Examining usefulness of FBT with younger children	Positive. FBT was acceptable and effective for treating AN in children.
Loeb KL et al., 2007 (Loeb et al., 2007)		20	Dec-17	19 (95%)	12	Investigating the feasibility and effectiveness of delivering FBT	Positive. FBT can be successfully disseminated.

					at a site beyond the treatment's origin and manualisation	
Couturier J et al., 2010 (Couturier et al., 2010)	14	Dec-17	14 (100%)	12	Assessing the dissemination of FBT, including effectiveness, fidelity, and acceptability	Positive. FBT was effective in weight restoration and improving psychological symptoms.
Turkiewicz G et al., 2010 (Turkiewicz et al., 2010)	9	Dec-17	9 (100%)	6	Examining the feasibility, acceptability, and effectiveness of FBT in Brazil	Positive. FBT was effective for Brazilian families.
Lock J et al., 2010 (Lock et al., 2010)	61	Dec-18	54 (89%)	12	Evaluating the relative efficacy of FBT and AFT for adolescents with AN in full remission	Positive. Both treatments led to improvements. FBT was more effective in facilitating full remission at follow-up.
Faust JP et al., 2013 (Faust et al., 2013)	84	NS	84 (100%)	12	Examining ROM in relation to EBW and psychosocial markers in adolescents with AN	Positive. FBT results in ROM with ROM occurring at weights close to reference norms for percent EBW.
Agras WS et al., 2014 (Agras et al., 2014)	78	Dec-18	67 (86%)	9	Comparison of FBT with SyFT for the treatment of adolescent-onset AN.	Positive. FBT is the preferred treatment with lower costs than SyFT.
Le Grange D et al., 2014 (Le Grange et al., 2014)	The sample participant data was taken from Lock J et al., 2010.				Reporting on relapse from full remission and attainment of remission during a four-year open follow-up period	Positive. Outcomes were generally stable post treatment.
Accurso EC et al 2015 (Accurso et al., 2015)	84	Sep-18	75 (88%)	No fixed dose.	Comparing outcomes that youths with AN achieved after receiving FBT in a randomized trial versus a fee-for-service clinic	Positive. FBT is effective and efficacious except for youths with lower initial body weights.
Ciao AC et al., 2015 (Ciao et al., 2015)	The sample participant data was taken from Lock J et al., 2010.				Examining family functioning in two treatments for adolescent AN (FBT and AFT) from multiple family members' perspectives	Positive. FBT had a more positive impact on family functioning compared to AFT.
Lock J et al., 2015 (Lock et al., 2015)	10	NS	9 (90%)	6	Examining the feasibility and effects of a novel adaptive treatment (IPC) aimed at enhancing parental self-efficacy in poor early responders to FBT	Positive. Adaptive designs of FBT are feasible.
Le Grange et al., 2016 (Le Grange et al., 2015)	107	Dec-18	93 (88%)	6	Randomized Clinical Trial of FBT versus Parent Focused Treatment (PFT) comparing remission rates for adolescents with AN	Positive. FBT is an efficacious intervention to mobilize parents to restore weight in this patient population.

Adherence Scoring

Papers were rated as to whether they referred to the use of the FBT manual, and included a brief overview of the principles guiding treatment in the paper introduction (as opposed to providing actual study details of therapist adherence to this method), a simple yes/no rating was used, without qualifying the level of detailed description required or given. All 15 studies had referred to the manual as part of setting the context for their study, the expectation being that the guidelines in the manual were followed. 14 studies also gave some degree of information regarding therapist level of expertise and supervision provided in the methodology.

Two out of the 15 studies specified the use of a fidelity or adherence measurement tool /In one very large RCT designed study, fidelity to either FBT or systematic family therapy was

described as being assessed. Trainer raters reviewed a selection of randomly chosen therapy sessions, and although reference is made to use of an appropriate and psychometrically tested fidelity instrument, the instrument itself was not described or referenced. The study reported good overall mean scores for fidelity; FBT 4.15 (0.94) and SyFT 4.38 (0.48), on a 0 to 6 scale.

The second study, conducted with a small sample of 14 youth with AN in Canada, was the first study to examine the dissemination of Family-Based Treatment, and possibly the first and only study to examine in detail the aspect of therapist fidelity to treatment. The treating fidelity scale is described in detail, and raters viewed a random series of FBT sessions, examining for the presence of 25 main therapeutic goals of FBT sessions. These are rated as being 'not at all' or 'very much' present, resulting in a score per item from 0 to 7. Acceptable treatment fidelity was set by the authors as a score of 5 or

greater, and this was achieved 72% of the time in phase I , 47% of the time in phase II, and 54% of the time in phase III of FBT treatment.

None of the other 13 studies reported on the degree of therapist adherence to FBT. However, general comments were made about the use of methods to enhance treatment adherence, for example using videotape monitoring, but further information about the outcome of such monitoring was not given.

In any attempt to quantify nonspecific reference to adherence, the adherence checklist was used to rate specific components of FBT delivery, both the FBT process (4 items, checklist score range 0-8) and FBT principles and prescribed interventions (7 items, checklist score range 0-14), the sum of both giving a total adherence score range of 0-22 (**Table 3**).

All 15 studies had at least some mention of the FBT process, with a mean of 2.86 (range of 1-8) Three papers had a rating of

just 1, suggesting only the briefest of description of specific FBT methodology. The mean adherence rating score for FBT principles and prescribed interventions was 4.00, (range of 0-8). The mean total score combining both FBT key structural elements and FBT principles and prescribed interventions was 6.73 which correspond to an average FBT manualised adherence rate of only 30.59% for all studies included in our review.

A final rating was assigned by consensus for each paper, based on adherence rating scores, setting the context, and group discussion, as to whether a reader could ascertain an adequate or inadequate degree of FBT treatment adherence based on information presented or inferences made in each paper. Overall, an adequate degree of FBT treatment adherence was depicted in 10 out of the 15 studies with 5 studies deemed inadequate.

Table 3: Study score.

Study	Adherence checklist mean score	Mention in paper				
	Methods (0-8)	Therapist style (0-14)	Total (0-22)	Specifics of FBT	Therapist clinical training	Overall FBT Treatment Fidelity Reporting
Couturier J et al., 2010 (Couturier et al., 2010)	2	8	10	Yes	Yes	Adequate
Le Grange D et al., 2005 (Le Grange et al., 2005)	4.75	6	10.5	Yes	Yes	Adequate
Loeb KL et al., 2007 (Loeb et al., 2007)	3.5	6	9.5	Yes	Yes	Adequate
Turkiewicz G et al., 2010 (Turkiewicz et al., 2010)	3	4	7	Yes	Yes	Adequate
Lock J et al., 2010 (Lock et al., 2010)	2.5	3.5	6	Yes	Yes	Adequate
Agras WS et al., 2014 (Agras et al., 2014)	1	3	4	Yes	Yes	Adequate
Lock J et al., 2005 (Lock et al., 2005)	2.5	7.5	10	Yes	Yes	Adequate
Accurso EC et al 2015 (Accurso et al., 2015)	1	0	1	Yes	Yes	Adequate
Faust JP et al., 2013 (Faust et al., 2013)	2	2.5	4.5	Yes	Yes	Adequate
Lock J et al., 2006 (Lock et al., 2006)	2	3	5	Yes	Yes	Inadequate
Lock J et al., 2015 (Lock et al., 2015)	3.5	4.5	8	Yes	No	Inadequate
Le Grange D et al., 2014 (Le Grange et al., 2014)	1	1	2	Yes	No	Inadequate
Ciao AC et al., 2015 (Ciao et al., 2015)	2.5	4.5	6.5	Yes	No	Inadequate
Le Grange et al., 2016 (Le Grange et al., 2015)	8	3	11	Yes	Yes	Adequate

Murray et al., 2017 (Murray et al., 2017)	4	2	6	Yes	No	Inadequate
Total mean (range)	2.86(1-8)	4.6 (0-8)	6.73(1-10.5)			

Discussion

Given the development of a National Eating Disorder Clinical Program in Ireland, and the roll out of FBT training across Child and Adolescent Mental Health teams, the authors endeavoured to examine efficacy studies on FBT in adolescent AN and to examine their internal validity (the extent to which treatment is delivered as intended) to help predict external validity (that FBT benefits may be replicated in real world settings, such as amongst FBT trained clinicians in Ireland).

Although many papers referencing family treatment of AN were identified, given our specific interest in efficacy studies, only 15 met inclusion criteria and eligible for inclusion in the systematic review. All 15 studies attested to the benefits of FBT in the treatment of AN, with results equivalent to or better than other family or individual approaches. Most papers reviewed referred in general principles to 'adherence to the manual', and the benefits of a 'manualised treatment' aiding a standardised approach, however, this reference to fidelity and adherence, apart from one study was not-detailed and vague. This leaves the reader without enough information to decide to what degree the FBT model was adhered to, and by association, whether the benefit seen was specific to FBT per se or might be linked to other factors. Examples of this include vague statements such as 'use of manualised treatments' and 'the treatment was manual based' which require more detail to understand the level of adherence by the therapists.

The mean total score from the fidelity checklist was well below the median 50%. The low mean total score of the adherence checklist highlights the low frequency of mention of adherence to core features. Some papers were found to lack information which would allow a reader to understand the degree of treatment adherence. Thus, it is difficult for the reader to draw conclusions about the extent to which the outcomes can be linked to specific FBT-interventions. This might lead a therapist, whether experienced or not, to loosely apply FBT. Negative finding in an independent study might erroneously cast doubt over treatment effectiveness. Given the challenge of trying to establish adherence post intervention, and from published papers, where a standard format is lacking, the routine use of an adherence checklist would assist in the systematic capturing and reporting of treatment adherence. This was examined in the dissemination study from Courtier and colleagues, but the suggested scale used was not formally psychometrically tested. This has now been at least partially addressed and a psychometrically robust fidelity checklist, The Family Therapy Fidelity and Adherence Check or FBT-FACT has been developed. This is a welcome development to the field, and incorporates both therapist behavior or adherence and competence in the early phases of treatment. Principal component analysis of the questionnaire has identified certain components which are more robust than others, perhaps due to

the multifaceted nature of main items, and further work is being conducted to examine this further and extend fidelity check to the overall treatment. However, the authors conclude that the therapist 'overall fidelity', despite variability in single-item agreement, can be measured with good validity. Therapist knowledge and skill vary, and unexpected gaps may exist. As highlighted in a national audit of psychological therapies in England and Wales [32] more than 30% of therapists providing a range of psychological therapies were delivering them without formal training. New therapies, such as FBT, therefore need both training and ongoing skill maintenance to promote efficacy.

In the studies we reviewed, papers reported good details on the level of training and expertise of therapists providing the FBT, along with the study's degree of supervision, allowing clarity as to their level of proficiency. Eight out of fifteen studies mentioned that FBT supervision was provided by the authors of FBT directly, online or by telephone. (Many studies mentioned some level of supervision by the authors of FBT (James Lock and/or Daniel Le Grange). Only two out of the 15 studies specified use of a fidelity or adherence measurement tool.

In moving from 'ivory tower' settings to real world settings, the therapist's experience and training and supervision in the specific treatment model can be important predictive factors for treatment success [33], All the studies reviewed involved authors of the FBT manual as co-authors, contributors or supervisors, and hence methodological rigour and skilled supervision and training was likely to be high. Independent replication of FBT studies will be necessary to test generalizability and it will be imperative that therapist adherence, expertise and training will also be collected and presented in reports to allow rigorous replications. This will be especially important if results deviate from previously reported positive results such as those reported in all 15 papers reviewed in this study.

The FBT treatment manual recommends several clinical disciplines suited to deliver FBT including psychiatry, psychology, mental health nursing and family therapy. There was some detail of professional background and training detailed in most of the studies. The degree of supervision required to support more junior trainees and to maintain treatment effectiveness is also important as FBT is rolled out in community settings and training programmes need to emphasise these components as doing so may enhance treatment efficacy. It is recognised that supervision can reduce 'therapist drift' over time [34] even to the point of compensating for any skills deficits. Studies have shown that treatment by clinically inexperienced student therapists, when carried out under close supervision by experienced supervisors; achieve equivalent clinical outcomes as if delivered by experienced therapists [35].

The reporting of treatment adherence and fidelity is negligible throughout eating disorder treatment literature and literature

more broadly [36]. Difficulty in adherence to FBT has been recognised by therapists, and has been attributed to factors within the therapist, the family and the treatment itself [37]. A study exploring therapists' perceptions regarding the uptake of FBT for adolescents with AN found a number of barriers: time commitment for therapists and families, inadequate attention to comorbid symptoms, need for parental consistency, lack of family meals in the real world context, difficulty weighing the child each time, and the need for sibling involvement. Such reported barriers highlight the risk for therapist drift and the importance of addressing these at the outset of training. In the presence of many perceived adherence barriers, whilst perhaps not acceptable, therapist drift becomes more understandable. It is also recognised that the measurement of either fidelity or adherence poses its own difficulties, with both quantitative and qualitative aspects to be considered, and might account for the scant presentation in the literature.

This has been recognised and prioritized by authors of the FBT manual, who regard knowledge and research into FBT fidelity to be 'in its infancy'. To establish core aspects of FBT predictive of success, good fidelity measures are required and preliminary measures such as the Fidelity of Implementation Rating System has been developed.

To ensure good clinical outcomes, then adequate adherence is required. To provide focussed and essential training and supervision, knowledge of areas known to have consistently low fidelity need to be targeted. Such research and attention to adherence will allow for further refinement of the manual, and improve the outcomes for youth with eating disorders.

Four months after the FBT introductory training in Ireland, clinicians completed a 15 item self-report adherence rating scale suggesting that only 54% 'always' completed a family meal and 69% 'always' charged the family with refeeding. When asked about their subjective impression of treatment adherence to FBT, 33% reported 'always' adhering and a further 1/3 as 'frequently' [38]. In that cohort, adherence to FBT was perceived to be enhanced by facilitated FBT peer supervision group attendance, and in Ireland, it is intended that this method be included in the ongoing training programme. Differences between recommended FBT and reported practice have previously been reported with over a third (34%) of clinicians deviating from the standard FBT approach. Therapists reported to also collect individual therapy food diaries, offer mindfulness techniques, motivational work and reflective team family therapy. These additional modalities were associated with clinician anxiety and complex caseload patterns.

Treatment adherence issues can also be found in other eating disorder treatments. In a paper reporting on adherence to Cognitive Behavioural Therapy (CBT), no single core CBT technique was routinely used by half of the participating again with clinician anxiety less adherent [39].

Assessment of and reporting on treatment adherence gives the reader greater confidence in study results and facilitates replication, essential for service delivery [40]. It protects against potentially effective treatments being prematurely discarded and unsuccessful treatments being implemented in clinical and

public health settings with negative repercussions to patients, providers and organizations [41].

In our review, the majority of the studies did not report specifically on the degree of therapist adherence although some general comments were made about methods used to enhance treatment adherence, for example using videotape monitoring. The outcome of this, and any deviance from expected delivery was not detailed given. The absence of reporting on adherence of specific FBT techniques does not mean that FBT was not strictly adhered to, or that studies did not establish and monitor treatment adherence, but that there was a lack of presentation of this data in the published manuscript, leaving the reader uncertain. Indeed as the original authors of FBT were involved directly or indirectly in almost all studies reviewed, the familiarity and thus adherence to the model is likely to have been strong, however it is not specifically reported in the articles. Low adherence reporting has been found in a systematic review examining health behaviour change and found that only 27% of the 342 studies critiqued checked adherence to protocols [42].

This study extends a previous report measuring clinical adherence to FBT, by taking the readers' perspective and by systematically examining whether adherence to the treatment manual can be ascertained from the data provided in the FBT literature. Having examined the literature, this study concludes that the existing literature on FBT effectiveness does not provide enough information on adherence to the model to allow the reader determine whether FBT was delivered as intended, or whether certain threshold therapist skills are necessary.

Limitations

Coding and rating of narrative information is open to subjective interpretation. In order to enhance the reliability of our coding, operational criteria were set in advance and the 5 raters (3 clinical; 2 research) reviewed the papers separately before discussion and consensus formation. A checklist was designed incorporating core features of the treatment and with reference to a published clinical adherence analysis of FBT. Care has been taken in the presentation of the findings to stress that the absence of written information in published papers, and thus the finding of 5 papers providing inadequate detail, is not evidence of the absence of rigorous methodological standards of any the studies, but rather lack of adequate detail in the published manuscript.

Despite these limitations, this study provides some preliminary evidence supporting the need for the inclusion of standard methods of reporting of adherence and fidelity in studies on psychological treatments. The creation of the Core Treatment Objectives Clinician Rating Scale by Ellison and colleagues is an example of standard recording of salient therapeutic objectives [43]. A similar adherence rating scale of actual therapist practice would complement this. Preliminary results from the FBT FACT are very promising and wait further testing. Scientific journals may consider offering author guidelines on how to report on treatment adherence rates when submitting manuscripts.

Finding the essential ingredients of any efficacious treatment is crucial to ensuring its effectiveness in real world settings. Standardized and routine reporting on treatment adherence can help in the essential process of establishing the reliability of treatments offered and of key ingredients. From a training perspective, adherence measures can also be regularly monitored and included in clinical supervision sessions and can re-enforce mastery of key components. Reporting on these measures in publications will help ensure that, as treatment moves from the 'ivory tower' setting to the community, efficacious treatments may retain their effectiveness, and be acceptable for roll out, as in the proposed case in Ireland. There also needs to be a standardization within publications on the reporting of fidelity and therapist adherence. The routine use of an adherence checklist would improve the confidence in study results and help in ascertaining essential ingredients.

This study examining adherence to treatment comes at an important time whereby there is a formal drive for rolling out manualized psychosocial treatments in mental health and for eating disorders internationally. In Ireland, this has included an enhanced form of CBT (CBT-E) and FBT to date, as part of the Health Service Executive [44-47] National Clinical Programme for Eating Disorders. Use of checklists, such as the one used in this study and incorporation into supervision and training would help ensure high standards of care.

Conclusion

Treatment adherence is an integral part of mental healthcare delivery and is associated with earlier recovery and better clinical outcomes. It should be closely measured and monitored to ensure that treatment is properly carried out. We recommend that studies reporting psychosocial interventions measure adherence to treatment manuals and clearly state this data in the study results section. Developing and reporting on a MOP, and following recommended reporting guidelines such as CONSORT or TREND, will assist in the identification of degree of any deviations from intended treatment delivery by therapist. Such checklists and tools should also be included in treatment manuals, and ideally be independently validated.

Developing and reporting on a MOP will assist in the identification of degree of any deviations from intended treatment delivery by therapist.

Conflicts Of Interest

All authors have no conflicts of interest.

Ethical Standards

The authors assert that all procedures contributing to this work comply with the ethical standards of the relevant national and institutional committee on human experimentation with the Helsinki Declaration of 1975, as revised in 2008. The authors assert that ethical approval for publication of this paper was not required by their local Ethics Committee.

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