



# Family-Based Psychological Treatment for Obsessive Compulsive Disorder in Children and Adolescents: A Meta-analysis and Systematic Review

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## Abstract

A significant number of children and adolescents with obsessive compulsive disorder (OCD) demonstrate poor response to the current gold standard treatment, cognitive behaviour therapy (CBT) with exposure and response prevention (ERP). Recent findings suggest that family variables affect treatment response highlighting the need for a meta-analytic review of the precise impact of family variables on OCD-related symptoms and processes. The current review and meta-analysis examined the effect of family-based interventions on OCD symptom and family factor outcomes for children and adolescents with OCD. The moderating effects of the degree of parental involvement and number of family factors targeted in treatment were investigated. An extensive literature search identified 37 eligible studies (1727 OCD participants). Large significant pooled mean effect sizes for OCD symptoms and Family Accommodation (FA), respectively, were obtained at posttest ( $g = 1.56$ ;  $g = 1.00$ ) and follow-up ( $g = 1.69$ ;  $g = 1.98$ ). Moderator analyses indicated that the number of family factors targeted in treatment significantly moderated outcomes on measures of FA ( $z = 2.21$ ,  $p = 0.03$ ), but not on Children's/Yale-Brown Obsessive Compulsive Scale (C/Y-BOCS) outcomes. FA has been significantly correlated with OCD symptom severity and poorer treatment outcomes, and there is data to suggest that FA may mediate OCD symptom outcomes (e.g., Piacentini et al. in *J Am Acad Child Adolesc Psychiatry* 50:1149–1161, 2011). Findings show that the greater the number of family factors targeted, the greater the reduction in FA at post, highlighting the importance of addressing a range of family factors in child OCD treatment to optimise outcomes.

**Keywords** Obsessive compulsive disorder · Treatment · Child · Family · Meta-analysis · Systematic review

## Introduction

Obsessive compulsive disorder (OCD) affects 1–4% of children and adolescents (Heyman et al. 2003; Rapoport et al. 2000; Valleni-Basile et al. 1995; Zohar 1999) and is characterised by obsessions and/or compulsions. Obsessions include recurrent and unwanted intrusive thoughts, images, or impulses that typically evoke significant anxiety or distress. Attempts are made to ignore or suppress these intrusions, or compulsions are performed to neutralise the obsessions and related distress (American Psychiatric Association 2013). Compulsions involve repetitive behaviours

(e.g., hand washing) or mental acts (e.g., counting) that an individual feels compelled to perform in response to obsessions or according to rigid rules (American Psychiatric Association 2013). Obsessions and compulsions are time-consuming and commonly cause significant impairment in functioning across areas of life, including social, familial, academic, and occupational domains (American Psychiatric Association 2013; Piacentini et al. 2003; Storch et al. 2010a). OCD in children and adolescents is thought to be similar to adult OCD in both prevalence and clinical presentation. However, diagnostic criteria specify that young persons are not required to have insight into their symptoms, such as their excessive or unreasonable nature (American Psychiatric Association 2013). In addition, young people with OCD may present with compulsions without distinct or clearly defined obsessions (Geller and March 2012). In youth, the disorder typically has a chronic, yet fluctuating, course and can significantly disrupt development that occurs

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during childhood and adolescence (Piacentini et al. 2003; Storch et al. 2010a).

Cognitive-behavioural therapy (CBT) that includes exposure and response prevention (ERP) has been established as the psychological treatment of choice for children and adolescents with OCD (Brauer et al. 2011; Freeman et al. 2018; Rosa-Alcázar et al. 2015). ERP involves prolonged and repeated exposure to feared obsessional stimuli (e.g., dirt; thoughts about death of a parent), while refraining from engaging in compulsions (e.g., hand washing; checking on a parent). As distress reduces with repeated exposure and by refraining from performing rituals, the individual learns that compulsions are not necessary to manage distress or to prevent the occurrence of feared events. However, a significant number of young people with OCD either fail to respond to ERP-based CBT or demonstrate only partial response. In the largest RCT to date examining treatment outcomes for children and adolescents with OCD, 60% of participants in the ERP-based CBT condition failed to demonstrate clinical remission (POTS Paediatric OCD Treatment Study (POTS) Team 2004). Recent findings have identified that the family environment can affect treatment response.

Family accommodation (FA) has been the focus of much of the recent research examining family environment factors in child/adolescent OCD. FA is the process whereby other family members participate in or assist with a child's OCD symptoms. FA can range from active participation in symptoms (e.g., answering a child's repetitive questions in an attempt to reduce their distress) to family members assisting with the avoidance of anxiety-provoking situations and/or modifying daily routines to assist with OCD (Lebowitz et al. 2012). The most common types of FA involve providing reassurance and waiting for the completion of rituals (Lebowitz et al. 2012). Rates of FA are remarkably high in families with a child with OCD, with the majority of families involved in frequent accommodation of OCD symptoms. Flessner et al. (2011) found that 99% of parents reported participating in at least one type of accommodation behaviour and 77.1% reported daily FA. FA can be negatively reinforcing for parents by temporarily reducing both child distress and parental distress associated with managing a child with OCD symptoms (Kagan et al. 2017; Lebowitz et al. 2014). Although generally well intentioned, FA reinforces child avoidance behaviours (contradictory to the goals of ERP) and maintains OCD symptoms and anxiety (Kagan et al. 2017; Wu et al. 2016). FA has been strongly associated with OCD symptom severity (e.g., Lebowitz et al. 2012; Strauss et al. 2015; Wu et al. 2016) and child functional impairment (e.g., Bipeta et al. 2013; Caporino et al. 2012). FA has also been linked to significantly reduced treatment outcomes (e.g., Garcia et al. 2010; Gorenstein et al. 2015; Peris et al. 2017; Piacentini et al. 2011). In a trial by Gorenstein et al. (2015), young people with higher FA scores

at pre-treatment showed poorer OCD symptom outcomes. Garcia et al. (2010) found that FA significantly predicted treatment outcomes for young people with OCD: Youth with lower levels of FA demonstrated greater symptom improvement across treatment conditions. FA accounted for changes in clinical symptoms in a study by Peris et al. (2017). Furthermore, in a trial by Piacentini et al. (2011), FA mediated OCD symptom outcomes and a reduction in FA was found to temporally precede OCD symptom change.

Recent findings suggest that other family variables may also affect treatment response. Peris et al. (2012a) found that families demonstrating higher levels of cohesion and lower levels of family conflict and blame of the young person prior to treatment were more likely to have a child who responded to CBT. Families that exhibited higher functioning in all three aforementioned domains had a 93% response to treatment compared to a 10% treatment response for families with poorer functioning in these three domains. High maternal expressed emotion (i.e., criticism and/or emotional overinvolvement) has been identified as a predictor of poor treatment response for young people with OCD (Peris et al. 2012b). Other family factors associated with the development and maintenance of child/adolescent OCD, and therefore relevant to treatment outcome, include over-responsibility placed on children (Farrell et al. 2013; Mathieu et al. 2015; Pietrefesa et al. 2010), poor family problem-solving skills (Barrett et al. 2002), and high parental control of child behaviour, such as overprotection (Haciomeroglu and Karanci 2013; Timpano et al. 2010).

Practise guidelines and reviews of the child and adolescent OCD literature commonly highlight the importance of involving family members in treatment to optimise treatment response (e.g., Brauer et al. 2011; Freeman et al. 2018; Geller and March 2012). However, the degree to which family members are included in treatment and the nature of their involvement vary significantly across studies and treatment programs. Interventions that involve family members typically fall into two broad categories: (1) Interventions where family member(s) attend treatment sessions to some extent, however family factors are not specifically targeted; and (2) Interventions that involve family member(s) and actively target family factors. Category 1 acts as a useful control with which to compare interventions that directly address family factors.

In the only RCT to date to compare Category 1 and Category 2 interventions for young people with OCD, Reynolds et al. (2013) investigated low and high levels of family involvement (FI) in a CBT intervention. Low FI was characterised by parents attending 3 of the 14 sessions and no family factors were directly addressed (Category 1), whereas high FI comprised parents attending all sessions and FA was targeted (Category 2). Low FI and High FI groups both demonstrated large positive effect sizes at posttest ( $d=1.45$ ;

$d=1.27$ ) and follow-up ( $d=1.53$ ;  $d=1.50$ ), with no significant differences found between groups. However, the authors acknowledged that the sample size was small and the study underpowered. Further investigation into Category 1 and 2 studies is necessary to better understand optimal family involvement.

Recent treatment programs have been extended to directly target some of the family factors identified to date in the child and adolescent OCD literature. Barrett et al. (2004) added 30 minutes of parent skills training (targeting problem-solving and FA) to one-hour child-focused CBT sessions. Similarly, Piacentini et al. (2011) supplemented child-focused sessions with 30-min family sessions addressing parental blame of the young person, FA, and unhelpful patterns of family interaction. One-h family sessions were added to child-focused sessions every second week in studies by Peris and Piacentini (2013) and Peris et al. (2017), targeting FA, family conflict, and blame, and enhancing cohesion and problem-solving skills. Excellent treatment outcomes have been reported where a number of family factors have been addressed, as evidenced by large effect sizes ( $d=2.65$ , Barrett et al. 2004;  $d=2.37$ , Piacentini et al. 2011;  $d=2.59$ , Peris and Piacentini 2013;  $d=2.07$ , Peris et al. 2017). A better understanding is required of the key family factors to target in OCD interventions for young people to enhance treatment response.

### Previous Meta-analyses and Systematic Reviews

The effectiveness of CBT in the treatment of child/adolescent OCD has been well established (Freeman et al. 2018). Some initial meta-analytic support has been provided for the effectiveness of family-based CBT for young people with OCD (Iniesta-Sepúlveda et al. 2017; Thompson-Hollands et al. 2014). Very few studies have considered the effectiveness of these interventions regarding family factor outcomes, such as FA. Importantly, meta-analytic studies have not yet identified many within-group treatment moderators systematically affecting response to treatment. Neither mode of treatment (e.g., individual vs group) nor therapeutic components (e.g., psychoeducation, cognitive training, contingency management) has been found to significantly and consistently affect treatment outcomes at a meta-analytic level (e.g., Olatunji et al. 2013). Very few studies have explored the effect of family-related within-group treatment moderators. Rosa-Alcázar et al. (2015) investigated the moderating effect of level of parental involvement on treatment outcomes and found that parental involvement (low, moderate, high) had a significant relationship ( $p=.002$ ) with OCD symptom outcomes, explaining 34% of variance. Iniesta-Sepúlveda et al. (2017) examined various techniques used in parenting components of interventions (i.e., FA; exposure assistance training; contingency management training;

and problem-solving) with results indicating nil significant treatment-moderating effects. Importantly, the number of family factors addressed in treatment has yet to be explored as a potential treatment moderator.

### Aims and Objectives

The current study aims to evaluate the effectiveness of family-based interventions for children and adolescents with OCD using both meta-analytic and systematic review techniques. A family-based intervention was broadly defined as an intervention that included a parent to some extent, regardless of whether or not family factors were specifically targeted. This study, therefore, includes two broad categories of family-based interventions: (1) parental involvement without addressing family factors directly and (2) parental involvement with the direct targeting of family factors. The effect of Category 1 and Category 2 family-based treatments on OCD symptoms as well as on FA, the primary family factor assessed by included studies, is investigated. In addition, the effect of family-based treatments on a range of other family variables, including blame, cohesion, conflict, and general family functioning, is calculated in the systematic review. This is the first study to consider the relative effectiveness of CBT with, versus without, the direct targeting of family factors.

The current meta-analysis examines the moderating effects of family-related treatment variables on OCD symptom and FA outcomes for young people with OCD. This is the first meta-analytic study to examine the number of family factors addressed in treatment as a potential treatment moderator. The effects on treatment outcomes of a large number of family factors is examined, including FA, problem-solving skills, conflict, blame/criticism, and communication. In addition, this study explores the moderating effects of the type and degree of parental/family involvement in treatment. Rather than code for parental/family involvement using broad categories or rating scales (e.g., low, moderate, high), a more precise method was used: The proportion of the total treatment time that parents were seen alone, as well as when participating in family sessions, was calculated. Quantifying parental/family involvement allows for a more objective examination of the number of family factors targeted in treatment. The current study aims to illuminate family-related treatment factors associated with improved outcomes.

The systematic review component of the current study analyses eligible studies according to seven main categories: Study design, participants, assessment, treatment, outcome measures, and symptom and family factor outcomes. Studies are also assessed for risk of bias according to domains defined by the Cochrane Handbook for Systematic Reviews of Interventions (Higgins and Green 2011). Thus, the quality

and reliability of the research and findings are evaluated for all included studies. The preferred reporting items for systematic reviews and Meta-Analyses (PRISMA) guidelines inform the summarising of information and reporting of results, and meta-analytic guidelines for conducting statistics are used (Liberati et al. 2009). The meta-analysis synthesises the results of eligible studies and examines the moderating effects of family-related treatment factors, including number of family factors targeted in treatment, total parent hours, and total family hours, on treatment outcomes (both OCD symptom and FA outcomes).

## Method

### Search Strategy

An extensive literature search was conducted using the databases: PsychInfo, Medline, Cochrane Central Register of Controlled Trials, and PubMed to identify published treatment studies that included family-based treatment interventions for children and adolescents with OCD. The key search terms employed included: (*obsessive compulsive disorder* or *OCD* or <sup>1</sup>*obsessive/compulsive neurosis*) AND (*intervention* or *therapy* or *trial* or *manual* or *treatment* or *cognitive behavio(u)r therapy*, or *CBT* or *exposure and response prevention* or *ERP* or *psychotherapy* or *program*) AND (*child* or *p(a)ediatric* or *adolescent* or *teen* or *schoolchild* or *boy* or *girl* or *preschool* or *youth* or *young person/people*) AND (*family* or *parent* or *mother* or *father* or *home* or *primary carer/caregiver* or *attachment* or *paternal* or *maternal*). Limits were set to include only peer-reviewed journal articles written in English. No limits were placed on publication date. Reference lists of relevant articles were also examined to identify any additional studies relevant to the review. The final search was conducted on 16 May 2018.

### Selection and Exclusion

Titles, abstracts, and full texts were systematically reviewed to eliminate studies that did not meet inclusion criteria for the review. Refer to Fig. 1 for a PRISMA flowchart of the selection process. Studies retained for further review were treatment trials involving children and adolescents (0–18 years old) with a principal diagnosis of OCD. Studies were only included if the OCD intervention involved a family member to some extent for all cases. Studies (e.g., case studies and case series) that did not include sufficient quantitative statistics to calculate effect sizes (e.g., overall

means and standard deviations) were excluded. In addition, studies with a very small sample size ( $n \leq 5$ ) were excluded. Where studies included several diagnostic groups, the results for OCD participants were required to be reported separately to be eligible for inclusion. Finally, studies with non-English-speaking populations were excluded. All abstracts were reviewed by a second rater and a 97% agreement rate was obtained. Differences in ratings were discussed and 100% agreement was reached on abstracts to be retained for the full text review. Full text articles were reviewed and discussed with the second rater prior to inclusion in the review and 100% agreement was reached on papers meeting the selection criteria. Out of the 872 articles screened, a total of 37 studies were eligible for inclusion in the current review. Publication dates ranged from 1994 to 2018.

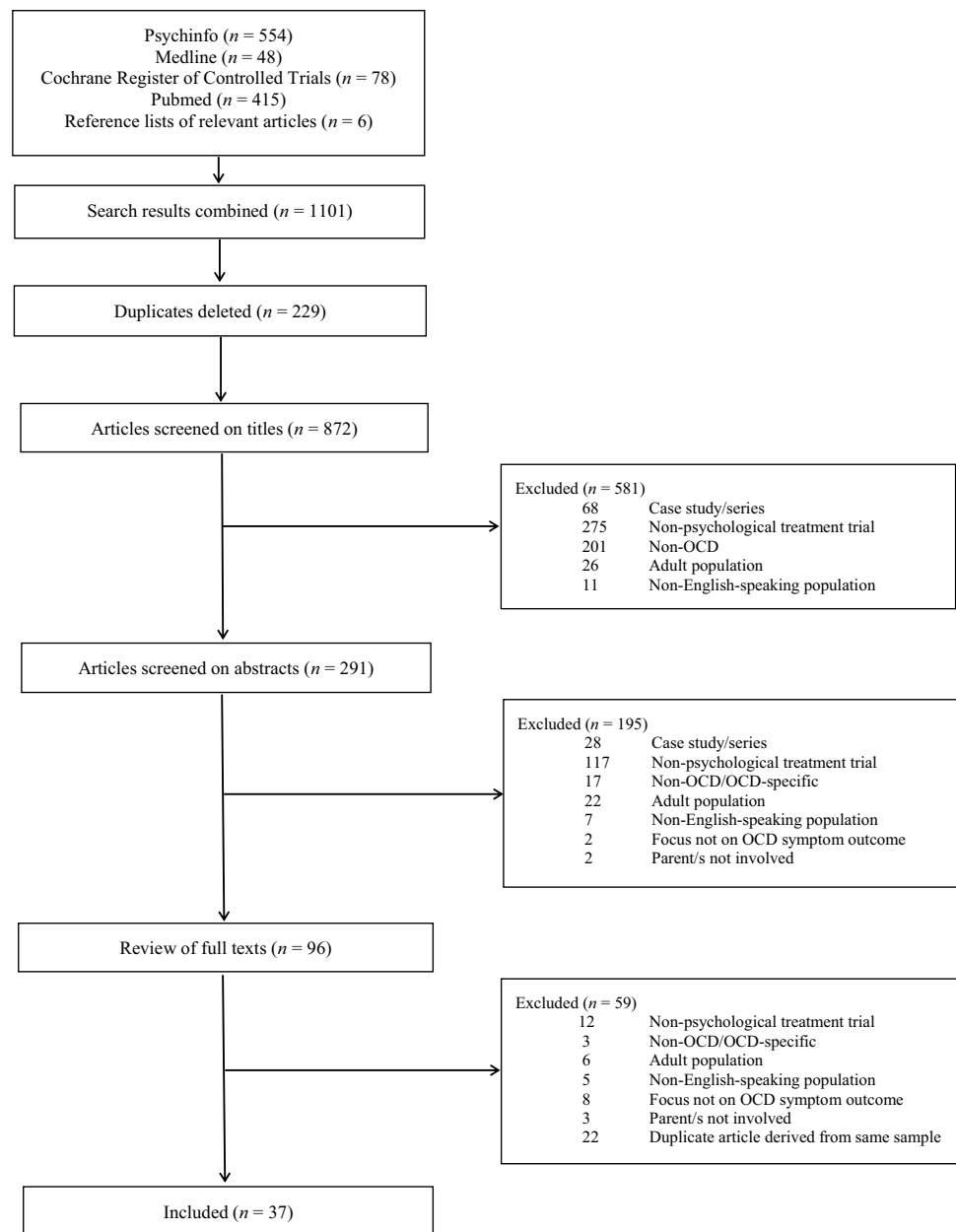
### Data Analysis

The 37 eligible research articles were reviewed to extract relevant data, including study design, participant characteristics, diagnostic and outcome measures, intervention type and characteristics (level of family involvement and whether or not family factors were targeted), and outcomes. Authors were contacted to request any relevant information not included in the published articles and this data were incorporated where provided. All corresponding authors were furnished with their study's respective calculations comprising Table 2 (and used in moderator analyses) and invited to provide any additional information not included in the published papers. A total of 64% of studies' authors responded, the majority to confirm the data presented to them. To evaluate the effectiveness of each study's intervention(s), in particular the degree of symptom and family factor change over time, within-group effect sizes (Cohen's  $d$ ) for pre-post and pre-follow-up treatment effects were calculated for all relevant outcome measures. Cohen's  $d$  within-group effect sizes for control conditions (e.g., waitlist) are available on request from authors.

Risk of bias was evaluated according to domains identified in the Cochrane Collaboration's Tool for Assessing Risk of Bias (Higgins et al. 2011): Selection bias (random allocation; allocation concealment), performance bias (blinding of participants and personnel), detection bias (blinding of outcome assessment—client report/externally rated), attrition bias (incomplete outcome data—post/follow-up), and reporting bias (selective reporting). Risk of bias ratings (low, high, or uncertain) were assigned to studies for each of the aforementioned categories, where relevant. Risk of bias was assessed by a second rater for 35% of the articles and a 96% agreement rate was obtained.

Meta-analytic statistics were employed using the program Comprehensive Meta-Analysis (CMA; Borenstein et al. 2005). The random effects model, rather than the fixed

<sup>1</sup> Current and past conceptualisations of OCD were used to broaden the literature search and enhance identification of all potentially relevant research articles.

**Fig. 1** PRISMA flowchart of study selection process

effects model, was used for all analyses as the included studies varied somewhat in design and participant population (Borenstein et al. 2009). Within-group effect sizes and variances were calculated using the Hedge's  $g$  statistic to assess pre-post and pre-follow-up treatment effects for the main OCD symptom and family factor measures. As recommended by Rosenthal (1993), a conservative estimate ( $r=0.7$ ) of the correlations between pre- and post-treatment measures was used as these were not typically reported in included articles. Within-group pooled mean effect sizes were also computed. Heterogeneity was assessed using Cochran's  $Q$  statistic and  $p$  value, and the  $I^2$  statistic. A significant  $p$  value for the  $Q$  statistic indicates heterogeneity

(i.e., the true effects vary from study to study). The  $I^2$  statistic indicates the degree to which variation between studies is due to heterogeneity and is reported in percentages, where 0% reflects no heterogeneity, 25% indicates low heterogeneity, and 50% and 75% reflect moderate and high heterogeneity, respectively. Risk of publication bias was determined using Duval and Tweedie's Trim and Fill Procedure (2000a, b). The pooled effect size is adjusted in order to yield an unbiased estimate of the effect size. In addition, the Classic fail-safe  $N$  (Rosenthal 1979) was calculated to identify the number of missing papers required to reduce a significant  $p$  value to less than alpha ( $<0.05$ ). Meta-regression analyses were employed to investigate moderators of treatment

effects. The variables investigated included Family Hours (the proportion of total treatment time that parents/family members attended treatment sessions with the young person), Parent Hours (the proportion of total treatment time that parents were seen on their own during treatment), and Number of Family Factors (the number of family factors directly targeted in treatment).

## Results

### Systematic Review

#### Demographics, Assessment and Outcome Measures

Refer to Table 1 for details.

#### Participant Characteristics

A total of 1727 participants comprised the 37 studies included in the review. Sample sizes of included studies ranged from 6 to 204 participants, with a median sample size of 31 participants. The mean age of OCD participants ranged from 5.8 to 14.5 years (*Mdn* = 12.8 years). The median percentage of female participants was 45%, with a range of 17–67%.

#### Study Design Characteristics

The majority of studies were designed as uncontrolled trials (UCT; 51%), followed by randomised controlled trials (RCT; 41%) and multiple baseline controlled trials (MBCT; 8%). A total of 65% of the studies included a follow-up time point for outcome measures, with a range of 1–18 months (*Mdn* = 6 months).

#### Assessment Measures

The majority of studies used semi-structured interviews to establish clinical diagnoses for participants: 65% of studies used the Anxiety Disorders Interview Schedule – Child/Parent versions (ADIS-C/P; Silverman et al. 1996) and 11% used the Kiddie Schedule for Affective Disorders and Schizophrenia—Present/Lifetime versions (K-SADS-P/L; Kaufman et al. 1997). Clinical interviews (CI) were employed in the remaining 24% of studies.

#### Outcome Measures

The primary outcome measure used to assess OCD symptoms in the majority (95%) of studies was the Children's

Yale-Brown Obsessive Compulsive Scale (CY-BOCS; Scapill et al. 1997; 33 studies) or the Y-BOCS (Goodman et al. 1989; 2 studies). A range of other symptom measures were also used, as reported in Table 1 and Table 3. A total of 32% of studies assessed family factors: All 12 studies assessed FA, predominately (75%; *n* = 9) using the Family Accommodation Scale (FAS; Calvocoressi et al. 1995, 1999; FAS-PR; Flessner et al. 2009). A second family variable was only assessed pre- and post-treatment in 11% of studies (*n* = 4) using a range of measures reported in Table 1 and Table 3. Behavioural tasks were rarely used (5% of studies; *n* = 2).

#### Treatment Program Characteristics

All studies used a CBT with ERP intervention. The majority of studies (76%; *n* = 28) delivered treatment in an individual face-to-face format (I), followed by group (G; 24%; *n* = 9) and individual remote (R; audio/video calls; 16%; *n* = 6) formats. A total of 32% of studies reported that treatment was based on March and Mullen's (1998) CBT manual. The remaining studies used a range of other CBT treatment programs outlined in Table 2.

The total number of treatment hours provided across studies ranged from 8 to 33 h over a range of 3–18 weeks (*Mdn* = 17 h/13 weeks). Programs included time with parents without the young person present in 49% of studies for a median of 5.5 h (range = 0.5–13.5 h). The number of family hours provided, where family members (usually parent/s) were included in sessions with the young person, ranged from 0 h to 21 h across studies, with a median of 6 h.

A total of 14% (*n* = 5) of studies did not directly address family factors during treatment, and therefore, comprise Category 1. The remaining 86% (*n* = 32) of studies met criteria for Category 2 by targeting at least one family factor in treatment. FA was the family factor most commonly targeted, in a total of 97% (*n* = 31) of Category 2 studies. The majority of Category 2 studies (47%; *n* = 15) reported addressing a total of one family factor during treatment, either FA (93%; *n* = 14), or conflict (7%; *n* = 1). Fewer studies addressed a second (28%; *n* = 9) or third (19%; *n* = 6) family factor. Only 2 studies (6%) reported targeting more than three family factors in treatment: Peris and Piacentini (2013) and Peris et al. (2017) addressed a total of five family factors in their interventions.

#### Overall Treatment Effects for Symptom and Family Factor Measures

As detailed in Table 3, all studies demonstrated a large positive within-group effect for pre- to post-treatment and pre-treatment to follow-up time points for the main OCD symptom measure used, the C/Y-BOCS. Cohen's *d* effect sizes for the C/Y-BOCS ranged from *d* = 0.79 to *d* = 3.61 (*Mdn d* = 1.88) for Pre-Post, and *d* = 1.31 to *d* = 3.34 (*Mdn d* = 2.01) for Pre-Follow-up. The

**Table 1** Demographics, assessment and outcome measures

No.	Study	N	% F	Age M (SD)	Design	Dx interview	F/up	Outcome measures								
								C/Y-BOCS	Other OCD	Anxiety	Depression	Behaviour	Parent	FA	Other fam.	Beh. task
1	Barrett et al. (2003)	24	42	11.21 (0.54)	RCT	ADIS-P	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓
2	Barrett et al. (2004, 2005)	77	51	11.87 (2.74)	RCT	ADIS-P	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓
3	Benazon et al. (2002)	16	50	NR	UCT	K-SADS-L (C/P)	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓
4	Comer et al. (2017)	22	41	6.65 (1.3)	RCT	ADIS-C/P	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓
5	Farrell et al. (2010)	35	46	12.29 (2.59)	UCT	ADIS-P	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓
6	Farrell et al. (2012)	43	30	11.09 (2.52)	UCT	ADIS-P	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓
7	Farrell et al. (2016)	10	40	13.6 (1.84)	MBCT	ADIS-P	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓
8	Fernandez de la Cruz et al. (2013)	153	48	14.22 (2.38)	UCT	CI	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓
9	Fernandez de la Cruz et al. (2015)	204	47	14.29 (2.29)	UCT	CI	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓
10	Fischer et al. (1998)	15	40	14.5 (1.73)	UCT	CI	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓
11	Franklin et al. (1998)	14	29	14.1 (2.2)	UCT	CI	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓
12	Freeman et al. (2008)	42	57	7.11 (1.26)	RCT	K-SADS-PL (C/P)	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓
13	Freeman et al. (2014)	127	53	7.2 (1.2)	RCT	K-SADS-PL (C/P)	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓
14	Hudson et al. (2015)	34	NR	NR	UCT	ADIS-C/P	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓
15	Lavell et al. (2016)	43	30	11.09 (2.52)	UCT	ADIS-P	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓
16	Lewin et al. (2014)	31	29	5.8 (1.6)	RCT	ADIS-P	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓
17	March et al. (1994)	15	67	14.3 (NR)	UCT	CI	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓
18	Martin and Thienemann (2005)	14	64	11.3 (NR)	UCT	CI	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓
19	Merlo et al. (2010)	16	38	13.3 (3.0)	RCT	ADIS-C/P	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓
20	Nakatani et al. (2011)	109	49	NR	UCT	CI	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓
21	Peris and Piacentini (2013)	20	45	12.35 (2.58)	RCT	ADIS-C/P	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓
22	Peris et al. (2017)	62	43	13.12 (2.68)	RCT	ADIS-C/P	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓
23	Piacentini et al. (2002)	42	60	11.8 (3.5)	UCT	CI/ADIS-C/P	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓
24	Piacentini et al. (2011)	71	63	12.2 (2.5)	RCT	ADIS-C/P	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓
25	POTS (2004)	112	50	11.78 (2.74)	RCT	ADIS-C	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓
26	Reynolds et al. (2013)	50	52	14.38 (1.50)	RCT	ADIS-C/P	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓
27	Scahill et al. (1996)	7	29	13 (2.02)	UCT	CI	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓
28	Selles et al. (2018)	85	54	13.9 (2.49)	UCT	ADIS-P	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓
29	Storch et al. (2007)	40	55	13.3 (2.7)	RCT	ADIS-P	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓
30	Storch et al. (2010)	30	50	13.4 (3.2)	UCT	ADIS-P	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓
31	Storch et al. (2011)	31	39	11.10 (2.59)	RCT	ADIS-C/P	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓
32	Sukhodolsky et al. (2013)	6	17	13 (2.38)	MBCT	K-SADS-PL	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓
33	Turner et al. (2009)	10	20	NR	UCT	CI	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓

Table 1 (continued)

No. Study	N	% F	Age M (SD)	Design	Dx interview	F/up	Outcome measures									
							C/Y-BOCS	Other OCD	Anxiety	Depression	Behaviour	Parent	FA	Other fam.	Beh. task	
34 Turner et al. (2014)	72	46	14.35 (2.12)	RCT	ADIS-C/P	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓
35 Waters et al. (2001)	7	40	NR	UCT	ADIS-C/P	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓
36 Whiteside and Jacobsen (2010)	16	44	13.13 (2.1)	UCT	ADIS-C	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓
37 Whiteside, McKay et al. (2014)	22	32	12.59 (3.1)	MBCT	ADIS-C	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓

% F Percentage female participants, Age M (SD) Age mean (standard deviation), NR Not reported, RCT Randomised controlled trial, UCT Uncontrolled trial, MCBT Multiple baseline controlled trial, Dx interview Diagnostic interview, C Child, P Parent, ADIS Anxiety disorders interview schedule, K-SADS-PL Kiddie schedule for affective disorders and schizophrenia—present and lifetime version, CI Clinical interview, F/up Follow-up, C/Y-BOCS Children's Yale-Brown obsessive compulsive scale, Other OCD Other OCD symptom measures, FA Family accommodation, Other fam. Other family factor measures, Behav. Task Behavioural tasks

first follow-up time point was used in calculations where studies reported multiple follow-up time points.

All studies indicated positive treatment effects on measures of FA: 10 studies demonstrated large effects and 1 study a medium effect at posttest, and at follow-up 5 studies showed a large effect and 1 study a small effect. Effect sizes for measures of FA ranged between  $d=0.52$  and  $d=2.04$  ( $Mdn d=1.04$ ) for Pre-Post and from  $d=0.32$  to  $d=1.77$  ( $Mdn d=1.29$ ) for Pre-Follow-up. An extended table with Cohen's  $d$  within-group effect sizes for anxiety, depression, behaviour, and parent symptom measures is available on request from authors.

**Risk of Bias**

Table 4 illustrates that overall a Low risk of bias status was identified for studies in the categories: Blinding of outcome assessment (client report), Incomplete outcome data—Post, and Follow-up (where relevant), and Selective reporting, with the majority of studies receiving a Low risk rating. The categories comprising Selection bias, namely Random sequence generation and Allocation concealment, were not relevant to the designs of a large percentage (51%) of included studies. Where relevant, the majority of studies (30%) received an Unclear risk of bias rating as insufficient information was provided in the aforementioned two categories to assist with making clear decisions regarding how well studies complied with best practise. The majority (86%) of the remaining seven studies were evaluated to be at Low risk of bias in both categories. The category of Blinding of participants/personal comprising Performance bias was not applicable to the design of 86% of the studies. Where applicable, most of the studies (80%) received an Unclear rating. The Blinding of outcome assessment (externally rated) category predominately received Low risk of bias ratings (57%), however, a large percentage of studies (38%) received Unclear ratings. Overall, the studies included in this review were evaluated as meeting criteria for low risk of bias, generally indicating research of sound quality. However, more detailed information provided in published papers related to randomisation (where relevant) and assessment procedures would enhance appraisal of the quality of research and reliability of findings.

**Meta-analysis**

**C/Y-BOCS Within-Group Effects**

Hedges's  $g$  within-group effect sizes were generated for the 30 studies ( $N=1227$ ) assessing OCD symptoms at pre- and post-treatment using the C/Y-BOCS (see Fig. 2a). All studies indicated a significant positive effect: two studies showed a medium effect according to Cohen's (1988) rule of thumb and the remaining 32 studies demonstrated a large effect. The pooled mean effect size was  $g=1.56$ , 95% CI [1.42,



Table 2 Treatment characteristics

No. Study	Treatment program	Family factors			Intervention time				
		Format	FA	Prob. S	Conf. B/C	Comm.	Total Hrs/weeks	Parent Hrs/total Hrs	Fam. Hrs/total Hrs
1	March and Mulle (1998); Waters et al. (2001) (parent)	I	✓	✓			21/14	7/21	3.5/21
2	Barrett et al. (2004, 2005)	I; G	✓	✓			21/14	7/21	2.3/21
3	March and Mulle (1998); Schwartz (1996)	I	✓				12/12-16	0/12	5.3+/12
4	Freeman and Garcia (2009)	I; R	✓				12/14	0/12	12/12
5	Barrett et al. (2004)	I; G	✓	✓			12/12 (D); 18/12 (G)	3-6/18 (D); 3-6/18 (G)	0/12 (D); 0/18 (G)
6	Farrell and Waters (unpublished)	G	✓	✓	✓		24.5/13	3/24.5	5.25/24.5
7	Farrell et al. (2016)	I+R	✓	✓			9.25/5	0/9.25	4.25-5.25/9.25
8	Fernandez de la Cruz et al. (2013)	I	✓				12-14/12-14	0/12-14	2-(12-14)/12-14
9	Fernandez de la Cruz et al. (2015)	I	✓				12-14/12-14	0/12-14	2-(12-14)/12-14
10	Fischer et al. (1998)	G	✓	✓			12/7	0/12	1.5/12
11	Franklin et al. (1998)	I					16/16 or 27/4	0/16 or 0/27	1+(e)/16 or 1.5+(e)/27
12	Freeman et al. (2008)	I	✓	✓	✓		13/14	3/13	10/13
13	Freeman et al. (2014)	I	✓	✓	✓		13/14	3/13	10/13
14	Hudson et al. (2015)	G					18-24(e)/11-12	6.75-12 (e)/18-24	4.5-6 (e)/18-24
15	Lavell et al. (2016)	G	✓	✓	✓		24.5/13	3/24.5	5.25/24.5
16	Lewin et al. (2014)	I	✓				12/6	0/12	12/12
17	March et al. (1994)	I	✓				16/16	0/16	6.25(e)/16
18	Martin and Thienemann (2005)	G	✓	✓			33/14	12/33	9/33
19	Merlo et al. (2010)	I					22-22.5/3	0/22-22.5	22-22.5/22-22.5
20	Nakatani et al. (2011)	I; G; R	✓	✓			8-12/8-12	0/8-12	1-(8-12)/8-12
21	Peris and Piacentini (2013)	I	✓	✓	✓		18/14	0.5/18	6/18
22	Peris et al. (2017)	I	✓	✓	✓		18/14	0.5/18	6/18
23	Piacentini et al. (2002)	I	✓	✓			12.5/12.5 (m)	3.1-4.13/12.5	0/12.5
24	Piacentini et al. (2011)	I	✓		✓		18/14	8/18	0/18
25	POTS (2004)	I	✓				14/12	0/14	3/14
26	Reynolds et al. (2013)	I	✓				11.7/14	0/11.7	11.7/11.7
27	Scahill et al. (1996)	I	✓				19/9-14	5+/19	3/19
28	Selles et al. (2018)	G	✓	✓			30-33/12	12-15/30-33	3-6/30-33
29	Storch et al. (2007)	I	✓		✓		21/3 or 21/14	0/21	21/21
30	Storch et al. (2010)	I	✓				21/3	0/21	21/21

Table 2 (continued)

No. Study	Treatment program	Family factors			Intervention time				
		Format	FA	Prob. S	Confl.	B/C	Comm.	Total Hrs/weeks	Parent Hrs/total Hrs
31	Storch et al. (2011)	I+R	✓				14-21/12	0/14-21	14-21/14-21
32	Sukhodolsky et al. (2013)	March and Mulle (1998); Barkley et al. (1997; 1999) (parent)	I	✓			18/18	6/18	3-5/18
33	Turner et al. (2009)	Turner et al. (unpublished)	R	✓			14/14	0/14	2.3/14
34	Turner et al. (2014)	Turner et al. (unpublished)	I; R	✓			14/17	0/14	2.3/14
35	Waters et al. (2001)	March et al. (1994; March and Mulle 1998); Authors' protocol (parent)	I	✓	✓		21/14	4.5/21	6/21
36	Whiteside and Jacobsen (2010)	Authors' manual (unpublished)	I				8.3-12.5/5	0/8.3-12.5	7.72-11.5/8.3-12.5
37	Whiteside, McKay et al. (2014)	Authors' manual (unpublished)	I				8.3-12.5/5	0/8.3-12.5	8.3-12.5/8.3-12.5

I Individual face-to-face treatment format, G Group, R Remote (audio/video calls), FA Family accommodation, Prob. S Problem solving, Confl. Conflict, B/C Blame/criticism, Comm. Communication, (e) Estimated, (m) Mean

1.7],  $p = 0.000$ , indicating a large, significant effect. Tests of heterogeneity demonstrated the presence of high-moderate significant heterogeneity,  $Q(33) = 125.28$ ,  $p = 0.000$ ,  $I^2 = 73.66\%$ . Hedges's  $g$  within-group effect sizes were generated for the 20 studies ( $n = 575$ ) reporting pre- and follow-up scores for the C/Y-BOCS (see Fig. 2b). The pooled mean effect size was  $g = 1.69$ , 95% CI [1.53, 1.85],  $p = 0.000$ , indicating a large, significant effect. Tests of heterogeneity showed significant moderate heterogeneity,  $Q(23) = 55.53$ ,  $p = 0.000$ ,  $I^2 = 58.58\%$ . The difference between the pooled mean effect size generated for pre-post and for pre-follow-up time points was not significant ( $p = 0.22$ ).

**FA Within-Group Effects**

Hedges's  $g$  within-group effect sizes were computed for the nine studies ( $n = 274$ ) assessing FA at pre- and post-treatment (the 11 values are detailed in Fig. 3a). All studies indicated a significant positive effect: eight values demonstrated a large effect, and two values a medium effect. A significant large pooled mean effect size of 1.00, 95% CI [0.8, 1.21],  $p = 0.000$ , was calculated. Significant moderate heterogeneity was indicated,  $Q(10) = 29.7$ ,  $p = 0.001$ ,  $I^2 = 66.33\%$ . Hedge's  $g$  within-group effect sizes were calculated for the six studies ( $n = 196$ ) assessing FA at pre-treatment and follow-up time points, outlined in Fig. 3b. A significant, large pooled mean effect size of  $g = 1.98$ , 95% CI [0.83, 1.53],  $p = 0.000$ , was computed. Significant, high heterogeneity was indicated,  $Q(7) = 36.38$ ,  $p = 0.000$ ,  $I^2 = 80.76\%$ . The difference between the pooled mean effect size for pre-post and for pre-follow-up time points was not significant ( $p = 0.75$ ).

**Publication Bias**

Duval and Tweedie's Trim and Fill Procedure (2000a, b) was employed to provide an adjusted pooled mean effect size for the C/Y-BOCS, taking into account any publication bias identified in the funnel plot (see Fig. 4). The mean effect size was reduced from  $g = 1.56$  to  $g = 1.42$  ( $n = 8$  values removed), to yield an estimate of the unbiased pooled effect size. An adjusted mean effect size for FA was also computed: The pooled mean effect size was reduced from  $g = 1.00$  to  $g = 0.9$  ( $n = 2$  values removed).

Publication bias was also assessed using the Classic fail-safe N calculation (Rosenthal 1979). The number of missing papers needed to reduce the  $p$  value to less than alpha ( $< 0.05$ ) was calculated as 6152 papers. As the total number of studies meeting inclusion criteria for this review was 37, it is highly unlikely that 6152 papers were missed. This suggests that publication bias does not affect the significant relationship found between family-based interventions for children and adolescents with OCD and OCD symptom outcome measured by the C/Y-BOCS.

**Table 3** Cohen's *d* within-group pre-post and pre-follow-up effect sizes for OCD symptom and family factor measures

No.	Dx <i>d</i>	C/Y-BOCS <i>d</i>	C/Y-BOCS F/up <i>d</i>	Other OCD <i>d</i>	Other OCD F/up <i>d</i>	FA <i>d</i>	FA F/up <i>d</i>	O. Fam. <i>d</i>	O. Fam. F/up <i>d</i>
1		NR*		NIMH-GOCS NR*					
2		(I) 2.65*	12 m (I) 2.64 <sup>b</sup>	NIMH-GOCS (I) 2.66*	12 m NIMH-GOCS (I) 3.11 <sup>b</sup>	SAS (I) 0.70*	NR <sup>b</sup>	Mo FAD (I) -0.24	12 m (I) Mo FAD 0.46 <sup>b</sup>
		(G) 2.01*	18 m (I) 2.20 <sup>b</sup>	NIMH-GOCS (G) 3.05*	18 m NIMH-GOCS (I) 2.71 <sup>b</sup>	SAS (G) 0.65*		Mo FAD (G) -0.23	18 m (I) Mo FAD 0.59 <sup>b</sup>
			12 m (G) 2.13 <sup>b</sup>	CGI-I (W6-P) (I) 1.21*	12 m NIMH-GOCS (G) 3.79 <sup>b</sup>			Fa FAD (I) -0.27	12 m (I) Fa FAD -0.13 <sup>b</sup>
			18 m (G) 2.28 <sup>b</sup>	CGI-I (W6-P) (G) 2.47*	18 m NIMH-GOCS (G) 3.97 <sup>b</sup>			Fa FAD (G) 0.00	18 m (I) Fa FAD 0.28 <sup>b</sup>
3		1.65*		NIMH-GOCS NR*					12 m (G) Mo FAD 0.37 <sup>b</sup>
4	(R) 1.67*	(R) 1.35*	6 m (R) 1.52*	CGI-S (R) 1.45*	6 m CGI-S (R) 1.25*	FAS (R) 1.14*	6 m FAS (R) 1.21*		18 m (G) Mo FAD 0.67 <sup>b</sup>
	(I) 1.61*	(I) 1.51*	6 m (I) 1.99*	CGI-S (I) 1.0*	6 m CGI-S (I) 1.39*	FAS (I) 0.93*	6 m FAS (I) 1.55*		12 m (G) Fa FAD 0.75 <sup>b</sup>
5		2.13*		CGAS (R) 1.31*	6 m CGAS (R) 1.48*				18 m (G) Fa FAD 0.99 <sup>b</sup>
				CGAS (I) 0.61*	6 m CGAS (I) 0.86*				
				NIMH-COGS 2.01*					
				CGI-S 2.13*					
				COIS-P 0.51*					
				COIS-C 0.54*					
6	1.61*	0.92*	6 m 1.43*	NIMH-COGS 1.65*	6 m NIMH-COGS 2.03*				
				COIS-P 0.90*					
				COIS-C 0.69*					
7	2.11*	(C) 2.31*	(C) 6 m 2.49*	CGI-S 2.30*	6 m CGI-S 2.48*				
		(P) 1.97*	(P) 6 m 1.77*	NIMH GOCS 1.74*	6 m NIMH GOCS 1.72*				
8		1.7*							
9		1.77*							
10		0.84*	6 m 1.31*						
11		(Int) 2.48*	9 m (Int) 2.03*						
		(wkly) 3.57*	9 m (wkly) 3.10*						
12		1.85 <sup>b</sup>							

**Table 3** (continued)

No.	Dx <i>d</i>	Dx F/up <i>d</i>	C/Y-BOCS <i>d</i>	C/Y-BOCS F/up <i>d</i>	Other OCD <i>d</i>	Other OCD F/up <i>d</i>	FA <i>d</i>	FA F/up <i>d</i>	O. Fam. <i>d</i>	O. Fam. F/up <i>d</i>
13		2.05*			COIS-P 0.96* CGI-S 1.86*					
14					SCAS-P-OC 1.37 <sup>b</sup> SCAS-C-OC 0.59 <sup>b</sup> NIMH GOCS NR*	3-12 m SCAS-P-OC 1.38 <sup>b</sup> 3-12 m SCAS-C-OC 0.53 <sup>b</sup> 6 m NIMH GOCS NR*				
15	NR	6 m NR* 12 m NR*	1.21 <sup>b</sup>	6 m 1.56* 12 m 1.49*	NIMH GOCS NR*	12 m NIMH GOCS NR*				
16	1.43 <sup>b</sup>	1 m 1.86 <sup>b</sup> 3 m 2.20 <sup>b</sup>	1.66 <sup>b</sup>	1 m 2.34 <sup>b</sup> 3 m 2.31 <sup>b</sup>	NIMH GOCS 1.40 <sup>b</sup> CGI-S 1.20 <sup>b</sup> CSDS 1.55 <sup>b</sup>	1 m NIMH GOCS 2.23 <sup>b</sup> 3 m NIMH GOCS 2.30 <sup>b</sup> 1 m CGI-S 2.22 <sup>b</sup> 3 m CGI-S 2.17 <sup>b</sup> 1 m CSDS 1.98 <sup>b</sup> 3 m CSDS 2.15 <sup>b</sup>	FAI 1.19 <sup>b</sup>	1 m FAI 1.77 <sup>b</sup> 3 m FAI 2.11 <sup>b</sup>		
17		1.57 <sup>*,a</sup>		NR*	NIMH GOCS NR*	NIMH GOCS NR*				
18		0.79*			NIMH GOCS 0.98* COIS-P 1.12* COIS-C 0.23 CGI-S NR <sup>b</sup>					
19		3.24 <sup>b</sup>								
20		2.01*								
21		2.59 <sup>b</sup>		3 m 1.94 <sup>b</sup>	CGAS 2.26 <sup>b</sup>	3 m CGAS 2.08 <sup>b</sup>	FAS 2.04 <sup>b</sup>	3 m FAS 1.72 <sup>b</sup>	PABS-Blame Mo 0.7 <sup>b</sup> PABS-Blame Fa 1.15 <sup>b</sup> FES-Cohes. Mo 0.10 <sup>b</sup> FES-Cohes. Fa 0.62 <sup>b</sup> FES-Confl. Mo 0.08 <sup>b</sup> FES-Confl. Fa 0.70 <sup>b</sup>	3 m PABS-Blame Mo 0.92 <sup>b</sup> 3 m PABS-Blame Fa 1.19 <sup>b</sup> 3 m FES-Cohes. Mo 0.03 <sup>b</sup> 3 m FES-Cohes. Fa 0.38 <sup>b</sup> 3 m FES-Confl. Mo 0.51 <sup>b</sup> 3 m FES-Confl. Fa 0.70 <sup>b</sup>
22		2.07*		2.35*	COIS-P 0.98*		FAS 1.65*		PABS-Blame 0.73* FES-Cohesion 0.39* FES-Conflict 0.65*	
23					NIMH-GOCS 1.75*					
24		2.37 <sup>b</sup>		1 m NR <sup>b</sup> 6 m NR <sup>b</sup>	COIS-P 1.01 <sup>b</sup> COIS-C 0.81 <sup>b</sup>	1 m; 6 m COIS-P NR <sup>b</sup> 1 m; 6 m COIS-C NR <sup>b</sup>	FAS 0.78 <sup>b</sup>	FAS NR <sup>b</sup>		

Table 3 (continued)

No.	Dx <i>d</i>	C/Y-F/up <i>d</i>	C/Y-BOCS <i>d</i>	C/Y-BOCS F/up <i>d</i>	Other OCD <i>d</i>	Other OCD F/up <i>d</i>	FA <i>d</i>	FA F/up <i>d</i>	O. Fam. <i>d</i>	O. Fam. F/up <i>d</i>
25		1.61								
26		1.27*	6 m 1.51*							
27		2.04*	1 m 1.80*							
			3 m 2.16*							
28		(C) 1.47*	(C) 1.46*	COIS-P 0.67*	COIS-P 1.01*	COIS-P 1.01*	FAS 1.02*	FAS 1.31*	OFF-C 1.05*	OFF-C 1.29*
		(P) 1.32*	(P) 1.61*	COIS-C 0.87*	COIS-C 0.98*	COIS-C 0.98*			OFF-P Fa 0.88*	OFF-P Fa 0.79*
29		(Int) 2.62*	3 m (Int) 2.20*	CGI-S (int) 3.29*	3 m CGI-S (Int) 3.11*	3 m CGI-S (Int) 3.11*	FAS (Int) 1.41*	3 m FAS (int) 1.24 <sup>b</sup>	OFF-P Mo 0.5*	OFF-P Mo 1.04*
		(wkly) 1.73*	3 m (wkly) 2.33*	CGI-S (wkly) 1.68*	3 m CGI-S (wkly) 2.44*	3 m CGI-S (wkly) 2.44*	FAS (wkly) 0.52*	3 m FAS (wkly) 0.32 <sup>b</sup>		
30		2.37*	3 m 2.24*	COIS-P (Int) 1.3*	3 m (int) COIS-P 1.89*	3 m (int) COIS-P 1.89*	FAS 0.79*			
				COIS-P (wkly) 0.45*	3 m (wkly) COIS-P 0.57*	3 m (wkly) COIS-P 0.57*				
				CGI-S 2.91*	3 m CGI-S 2.85*	3 m CGI-S 2.85*				
				COIS-P 0.75*	COIS-P 0.75*	COIS-P 0.75*				
				COIS-C 0.72*	COIS-C 0.72*	COIS-C 0.72*				
31		1.81*	3 m 1.98*	COIS-P 1.08*	3 m CGI-S 2.01*	3 m CGI-S 2.01*	FAS 0.83			
				COIS-C 1.05*	COIS-C 1.05*	COIS-C 1.05*				
				CGI-S 1.56*	CGI-S 1.56*	CGI-S 1.56*				
32	NR <sup>b</sup>									
33		1.8*	6 m 1.70*	ChOCI-P 0.71*	6 m ChOCI-P 1.15*	6 m ChOCI-P 1.15*				
			12 m 1.77*	ChOCI-C 0.86*	12 m ChOCI-P 1.30*	12 m ChOCI-P 1.30*				
					6 m ChOCI-C 0.98*	6 m ChOCI-C 0.98*				
					12 m ChOCI-C 0.96*	12 m ChOCI-C 0.96*				
34		2.41 (I) <sup>b</sup>	3 m (I) 2.2 <sup>b</sup>	ChOCI-C (I) 1.05 <sup>b</sup>	3-12 m ChOCI-C (I) 1.07-1.21 <sup>b</sup>	3-12 m ChOCI-C (I) 1.07-1.21 <sup>b</sup>				
		1.91 (R) <sup>b</sup>	6 m (I) 2.56 <sup>b</sup>	ChOCI-C (R) 1.21 <sup>b</sup>	3-12 m ChOCI-C (R) 1.16-1.19 <sup>b</sup>	3-12 m ChOCI-C (R) 1.16-1.19 <sup>b</sup>				
			12 m (I) 2.70 <sup>b</sup>	ChOCI-P (I) 1.14 <sup>b</sup>	3-12 m ChOCI-P (I) 1.07-1.20 <sup>b</sup>	3-12 m ChOCI-P (I) 1.07-1.20 <sup>b</sup>				
			3 m (R) 1.84 <sup>b</sup>	ChOCI-P (R) 1.11 <sup>b</sup>	3-12 m ChOCI-P (R) 1.21-1.27 <sup>b</sup>	3-12 m ChOCI-P (R) 1.21-1.27 <sup>b</sup>				
			6 m (R) 1.97 <sup>b</sup>	CGAS (I) 2.11 <sup>b</sup>	3-12 m CGAS (I) 1.84-2.25 <sup>b</sup>	3-12 m CGAS (I) 1.84-2.25 <sup>b</sup>				
			12 m (R) 2.09 <sup>b</sup>	CGAS (R) 1.58 <sup>b</sup>	3-12 m CGAS (R) 1.56-2.15 <sup>b</sup>	3-12 m CGAS (R) 1.56-2.15 <sup>b</sup>				
				CGI-I (I) 3.61 <sup>b</sup>	3-12 m CGI-I (I) 2.39-2.86 <sup>b</sup>	3-12 m CGI-I (I) 2.39-2.86 <sup>b</sup>				
				CGI-I (R) 2.63 <sup>b</sup>	3-12 m CGI-I (R) 3.05-3.17 <sup>b</sup>	3-12 m CGI-I (R) 3.05-3.17 <sup>b</sup>				
35		3.61*	3 m 3.34*	CGAS 1.35	3 m CGAS 1.88	3 m CGAS 1.88	FAS NR*		FAD-General NR <sup>b</sup>	
				NIMH GOCS NR*	3 m NIMH GOCS NR*	3 m NIMH GOCS NR*				
36		(C+P) 2.07*	5 m (C) 2.77*							

**Table 3** (continued)

No.	Dx <i>d</i>	Dx F/up <i>d</i>	C/Y-BOCS <i>d</i>	C/Y-BOCS F/up <i>d</i>	Other OCD <i>d</i>	Other OCD F/up <i>d</i>	FA <i>d</i>	FA F/up <i>d</i>	O. Fam. <i>d</i>	O. Fam. F/up <i>d</i>
37	0.86*	3 m 1.34*	1.37*	5 m (P) 2.88* 3 m 1.98*	COIS-P 0.55	3 m COIS-P 0.90*	FAI 1.04*	3 m FAI 1.27*		
					COIS-C 0.09	3 m COIS-C 0.70*				

Dx Diagnostic assessment, *d* Cohen's *d* effect size, *F/up* Follow-up, *I* Individual face-to-face treatment format, *G* Group treatment format, *R* Remote treatment format (telephone/web), *m* Months, *C* Child, *P* Parent, *NR* Not reported, *FA* Family accommodation, *Mo* Mother, *Fa* Father, *Cohes* Cohesion, *Confl* Conflict, *(W6-P)* Week 6—Post-treatment, *Int* Intensive, *Wkly* Weekly measures, *C/Y-BOCS* Children's Yale-Brown obsessive compulsive scale, *NIMH GOCS* NIMH global obsessive compulsive scale, *CGI-I/S* Clinical global impression—improvement/severity, *CGAS* Children's global assessment scale, *COIS-C/P* Child obsessive compulsive impact scale, *ChOCI* Children's obsessional compulsive inventory, *SCAS-OC* Spence children's anxiety scale—obsessive compulsive subscale, *SAS* Sibling accommodation scale, *FAS* Family accommodation scale, *FAI* Family accommodation interview, *FAD* Family assessment device, *PABS* Parental attitudes and behaviors scale, *FES* Family environment scale, *OFF* OCD family functioning scale

<sup>a</sup>ES from Barrett et al. (2008)

<sup>b</sup>Significance not reported

\*Significant differences (<.05) between pre- and post-treatment score and gains maintained or continued at follow-up. A minus before an ES indicates worsening score (no improvement). An ES of .2 is small, .5 is medium, and .8 is large

### Exploration of Within-Group Effects Moderators

The continuous variables of family hours, parent hours, and number of family factors were employed in meta-regression analyses for OCD symptom and FA outcomes using the random effects model. The number of family factors targeted in treatment was found to significantly moderate outcome on measures of FA ( $z = 2.21, p = 0.03$ ). The greater the number of family factors targeted in treatment, the larger the effect size for FA and therefore the greater the reduction in FA from pre- to post-treatment. Considering the variables of Parent Hours and Family Hours for the nine studies that assessed FA at pre- and post-treatment, 67% yielded an identical score for both Parent Hours ( $Mdn = 0$ ) and Family Hours ( $Mdn = 1$ ). Due to the low variability in scores evident for parent hours and family hours for the small number of studies assessing FA, no further analyses for FA were undertaken using these two variables. Neither family hours ( $z = 0.07, p = 0.94$ ), parent hours ( $z = -0.17, p = 0.86$ ), nor number of family factors ( $z = -0.72, p = 0.47$ ) yielded significant point estimates of the slope for the symptom outcome measure, the C/Y-BOCS, including when combined. The categorical variable of Category 1/Category 2 studies was investigated as a potential moderator of OCD symptom and FA outcomes. Category of study was not found to be a significant moderator of OCD ( $z = -1.3, p = 0.19$ ) nor FA ( $z = -0.19, p = 0.85$ ) outcomes.

### Discussion

The current meta-analysis and systematic review examined the effect of family-based interventions on OCD symptom outcomes as well as on a range of family factor outcomes (including FA, blame, cohesion, conflict, and general family functioning) for children and adolescents with OCD. The broad inclusion criteria encompassed controlled and uncontrolled studies with a wide range of parental involvement in treatment, including interventions that directly sought to address family factors (Category 2) as well as those that did not target family factors (Category 1). The current meta-analysis aimed to illuminate family-related treatment factors associated with improved outcomes and uniquely considered the relative impact on treatment outcomes of CBT with, versus without, the direct targeting of family factors for young people with OCD. This is the first meta-analytic study to consider the moderating effects of the number of family factors targeted in treatment on OCD symptoms and FA outcomes, including precise calculations of the proportion of total treatment time that parents were seen alone, and when involved in family sessions.

Overall, family-based interventions were found to be effective for children and adolescents with OCD. All studies

**Table 4** Risk of bias

No.	Study	Type of bias							
		Selection		Performance	Detection		Attrition		Reporting
		Random sequence generation	Allocation concealment	Blinding of participants/ personnel	Blinding of outcome ax (client report)	Blinding of outcome ax (externally rated)	Incomplete outcome data (post)	Incomplete outcome data follow-up	Selective reporting
1	Barrett et al. (2003)	?	?	n/a	+	+	+	n/a	+
2	Barrett et al. (2004, 2005)	?	?	n/a	+	+	+	?	+
3	Benazon et al. (2002)	n/a	n/a	n/a	+	?	+	n/a	–
4	Comer et al. (2017)	+	+	n/a	+	+	+	+	+
5	Farrell et al. (2010)	n/a	n/a	n/a	+	?	+	+	+
6	Farrell et al. (2012)	n/a	n/a	n/a	+	?	+	?	+
7	Farrell et al. (2016)	+	+	n/a	+	+	+	+	+
8	Fernandez de la Cruz et al. (2013)	n/a	n/a	n/a	+	?	+	n/a	+
9	Fernandez de la Cruz et al. (2015)	n/a	n/a	n/a	+	?	+	n/a	+
10	Fischer et al. (1998)	n/a	n/a	n/a	+	+	+	+	+
11	Franklin et al. (1998)	–	–	n/a	+	+	+	+	+
12	Freeman et al. (2008)	?	?	n/a	+	+	+	n/a	+
13	Freeman et al. (2014)	+	+	n/a	+	+	+	n/a	+
14	Hudson et al. (2015)	n/a	n/a	n/a	+	+	+	+	+
15	Lavell et al. (2016)	n/a	n/a	n/a	+	?	+	+	+
16	Lewin et al. (2014)	?	?	–	+	+	+	+	+
17	March et al. (1994)	n/a	n/a	n/a	+	?	?	+	+
18	Martin and Thienemann (2005)	n/a	n/a	n/a	+	?	+	n/a	+
19	Merlo et al. (2010)	?	?	?	+	+	+	n/a	+
20	Nakatani et al. (2011)	n/a	n/a	n/a	+	?	+	n/a	+
21	Peris and Piacentini (2013)	?	?	?	+	+	+	+	+
22	Peris et al. (2017)	?	?	n/a	+	+	+	+	+
23	Piacentini et al. (2002)	n/a	n/a	n/a	+	–	+	n/a	+

**Table 4** (continued)

No.	Study	Type of bias							
		Selection	Allocation concealment	Performance	Detection		Attrition		Reporting
		Random sequence generation		Blinding of participants/personnel	Blinding of outcome ax (client report)	Blinding of outcome ax (externally rated)	Incomplete outcome data (post)	Incomplete outcome data follow-up	Selective reporting
24	Piacentini et al. (2011)	?	?	n/a	+	+	+	+	+
25	POTS (2004)	+	+	?	+	+	+	n/a	+
26	Reynolds et al. (2013)	?	?	?	+	+	+	+	+
27	Scahill et al. (1996)	n/a	n/a	n/a	+	?	+	+	+
28	Selles et al. (2018)	n/a	n/a	n/a	+	–	–	–	+
29	Storch et al. (2007)	?	?	n/a	+	+	+	?	+
30	Storch et al. (2010)	n/a	n/a	n/a	+	+	+	+	+
31	Storch et al. (2011)	+	+	n/a	+	+	+	+	+
32	Sukhodolsky et al. (2013)	?	?	n/a	+	?	+	n/a	+
33	Turner et al. (2009)	n/a	n/a	n/a	+	?	+	+	+
34	Turner et al. (2014)	+	+	n/a	+	+	+	+	+
35	Waters et al. (2001)	n/a	n/a	n/a	+	?	+	+	+
36	Whiteside and Jacobsen (2010)	n/a	n/a	n/a	+	?	+	+	+
37	Whiteside, McKay et al. (2014)	n/a	n/a	n/a	+	+	–	–	+

+ Low risk of bias, - High risk of bias, ? Unclear risk of bias, n/a Not applicable, ax Assessment

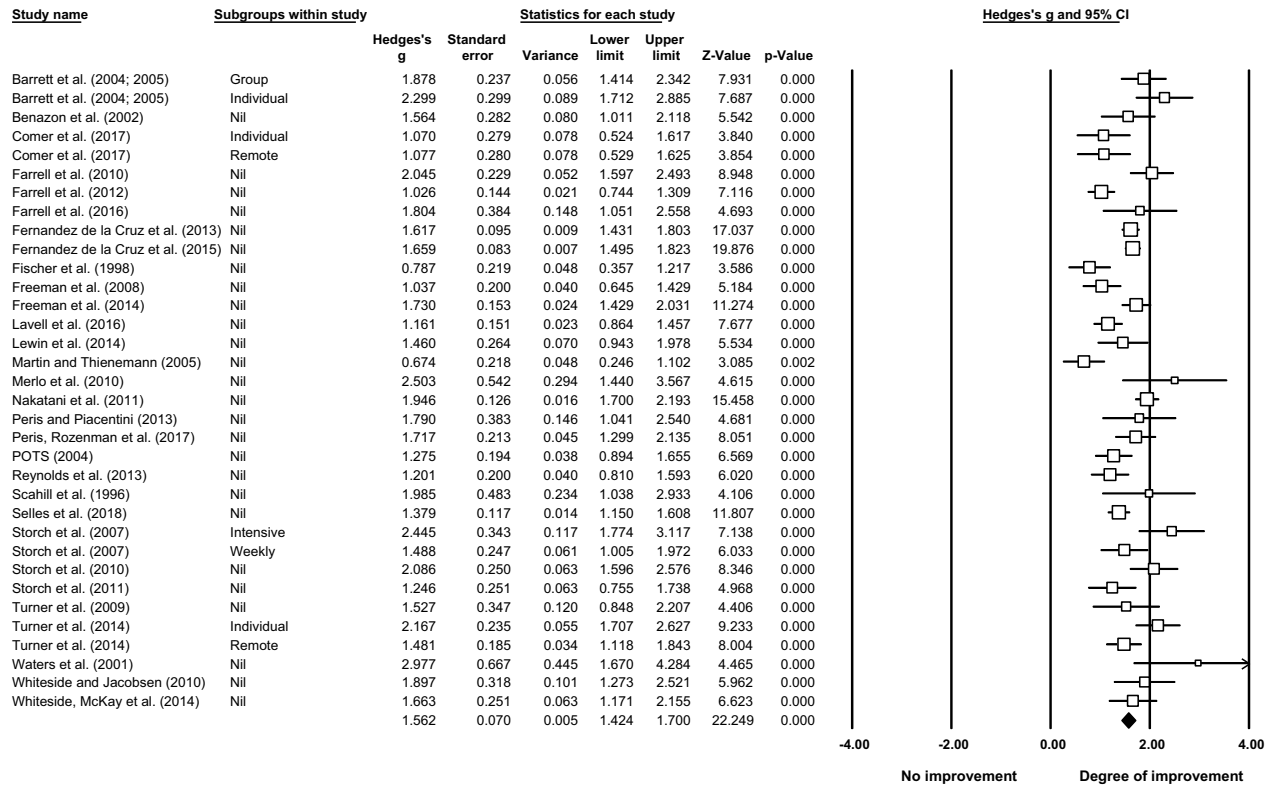
evaluated demonstrated a significant positive treatment effect for both OCD symptoms and FA, at posttest and follow-up, regardless of whether family factors were directly targeted (i.e., Category 1 and Category 2 interventions). The pooled mean effect size for the C/Y-BOCS was large and significant for both pre-post and pre-follow-up comparisons. Large, significant treatment effects were also demonstrated for FA at both time points. The large treatment effects obtained for OCD symptoms and FA are consistent with findings of previous meta-analyses (e.g., Iniesta-Sepúlveda et al. 2017), lending further support for the effectiveness of family-based interventions in reducing OCD symptoms, as well as FA, in young people with OCD.

One of the primary findings of this study was that the number of family factors targeted in treatment significantly

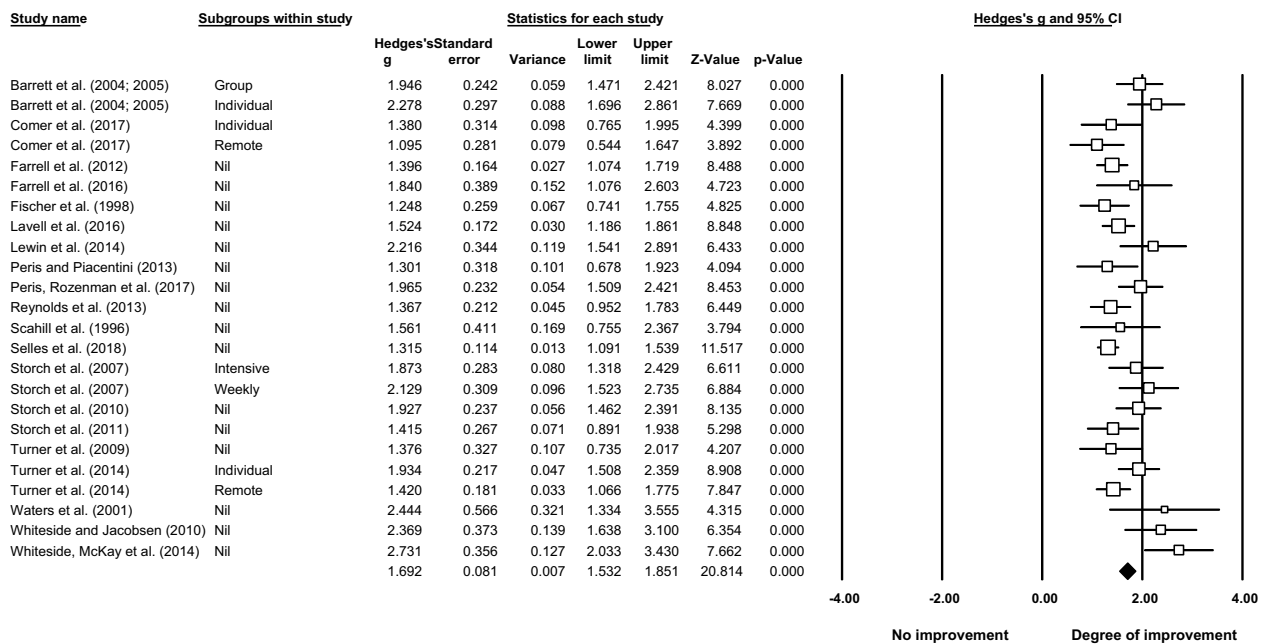
moderated treatment outcomes on measures of FA. Thus, the greater the number of family factors targeted in treatment, the greater the reduction in FA, an unhelpful family response, from pre- to post-treatment. FA scores have been significantly correlated with OCD symptom severity in previous meta-analyses (e.g., Strauss et al. 2015; Wu et al. 2016). Treatment trials have demonstrated the association between FA and poorer treatment outcomes (e.g., Garcia et al. 2010; Merlo et al. 2009; Storch et al. 2008). Peris et al. (2017) found that changes in FA accounted for changes in clinical symptoms, and therefore clinical improvement, for young people with OCD. The authors identified FA as a potential mechanism for change in the treatment of youth with OCD, contributing to previous findings by Piacentini et al. (2011). Piacentini et al. (2011)



**A**

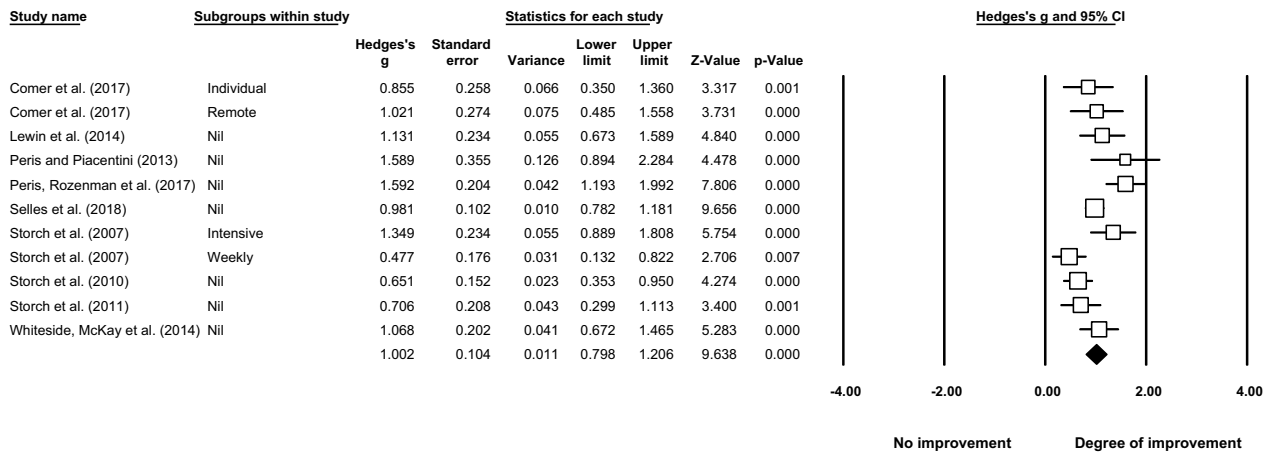


**B**

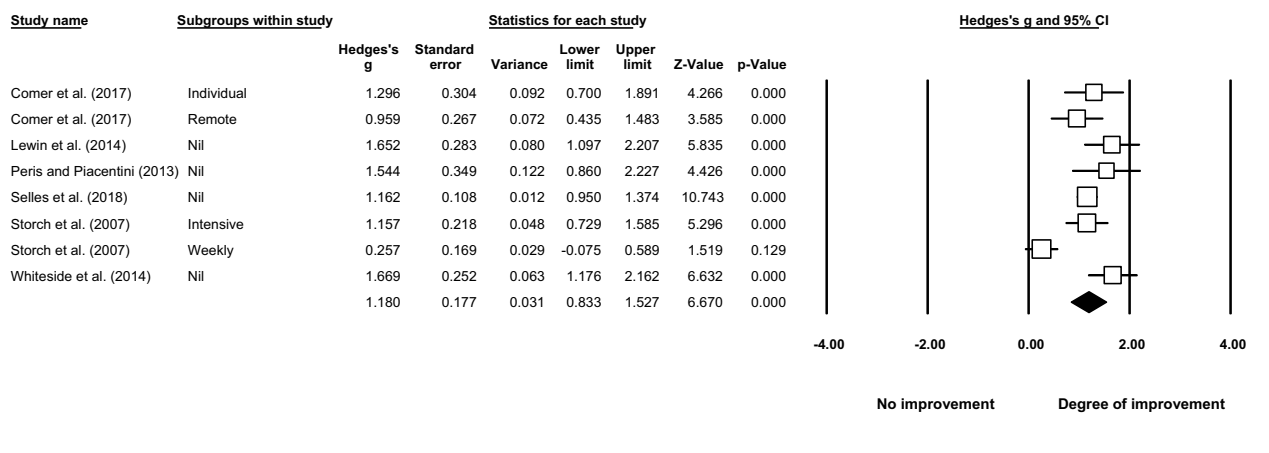


**Fig. 2 a** Forest plot of Hedges's g indices for OCD symptoms (C/Y-BOCS) pre-post treatment. **b** Forest plot of Hedges's g indices for OCD symptoms (C/Y-BOCS) pre-follow-up

**A**



**B**

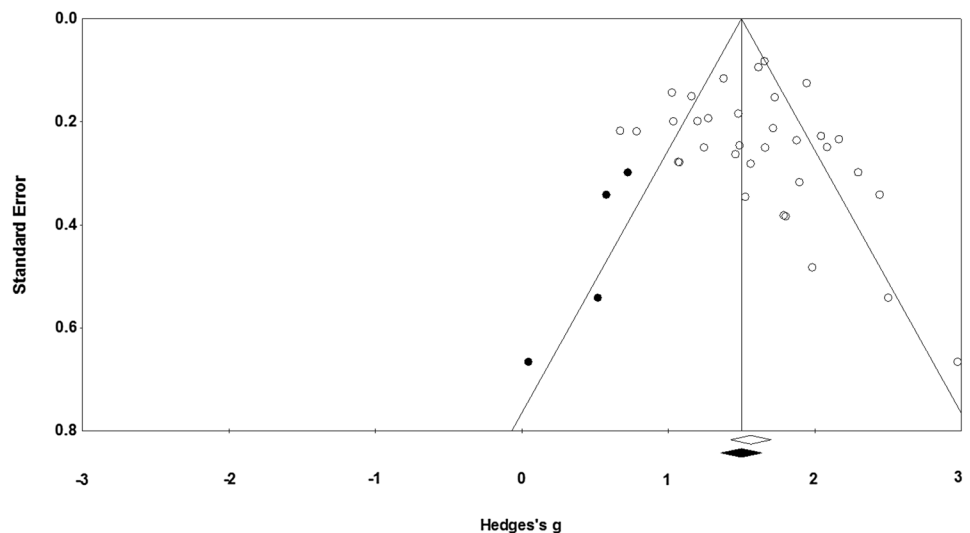


**Fig. 3** **a** Forest plot of Hedges's g indices for family accommodation measures pre-post treatment. **b** Forest plot of Hedges's g indices for family accommodation measures pre-follow-up

demonstrated that FA-mediated OCD symptom outcomes and found that changes in FA preceded OCD symptom change. The findings of the current study highlight the importance of reducing family-related maintaining factors, such as FA, by specifically targeting these family factors in OCD interventions for young people to optimise treatment response. The current findings indicate that FA is not the only critical family variable to target in treatment to enhance outcomes. In fact, the more family factors targeted, the greater are these outcomes. This study's findings add further support to preliminary results presented in the treatment literature that specific family factors, such as family cohesion, conflict, and parental blame of the young person, can affect response to treatment for young people with OCD (Peris et al. 2012, Peris et al. 2017).

The number of family factors targeted in treatment did not significantly moderate OCD symptom outcomes as measured by the C/Y-BOCS. It may be that measures of FA better assess behaviour associated with unhelpful family factors than a measure assessing OCD symptom severity. Mounting evidence suggests that changes in FA may moderate or even mediate OCD symptom change (Peris et al. 2017; Piacentini et al. 2011). Although this relationship was not significant in the current study, this may be due to the small number of studies assessing FA at pre- and post-treatment. Despite the limited measurement of FA outcomes, a small correlation of  $r = 0.35$  was still identified in the current study between change in FA scores and change in OCD symptom scores (C/Y-BOCS) scores pre- to post-treatment. Nil significant moderating

**Fig. 4** Funnel plot of standard error by Hedges's  $g$  indices for observed and imputed comparisons of OCD symptoms (C/Y-BOCS) pre-post treatment



effects were found for Category 1 versus Category 2 studies on OCD symptom and FA outcomes. As the continuous variable, Number of Family Factors, is a more precise operationalisation of the family factors targeted in treatment compared to the aforementioned categorical variable that uses only two broad categories (family factors vs no family factors), the former variable likely better identified the moderating effects of family factors on FA outcomes.

The current study found that the amount of time parents spent in family sessions and/or alone with the therapist did not significantly moderate OCD symptom outcomes. The impact of Parent Hours and Family Hours on FA outcome was not analysed due to low variability in scores for the few eligible studies that measured FA. Nonetheless, the current findings possibly suggest that the amount of family/parent time may be of less importance than the number of family factors addressed during this time. A previous meta-analysis by Rosa-Alcázar and colleagues (Rosa-Alcázar et al. 2015) found that parents' active participation in treatment, such as when parents were trained to assist their children as ERP coaches, had a significant positive association with the effect size for OCD symptoms. This may be the case for family factors, such that time spent by parents actively participating in treatment to address family maintaining factors may have more of an impact on OCD symptoms than merely the amount of time spent by parents attending treatment sessions.

### Limitations and Future Directions

Limitations of the current study include that family factors may have been indirectly targeted in Category 1 studies when parents were present in treatment sessions, even when not part of the treatment protocol, particularly if these factors were interfering with treatment progress. Alternatively,

family factors may have been targeted directly as part of a study's standard treatment protocol without authors including this information in their written description of treatment. In an effort to overcome the latter limitation, authors of all studies were contacted to confirm the specific family factors addressed in each treatment trial. The majority of authors responded and verified the family factors collated by the current study.

Only 32% of included studies assessed at least one family factor both pre- and post-treatment. All 12 studies measured FA and only four studies assessed a family factor other than FA. Due to the small number of studies assessing FA and the limited variability in scores for Parent Hours and Family Hours for these studies, moderator analyses for FA could not be performed for the two aforementioned variables. The impact of potential moderators on other family factors, such as blame, criticism, cohesion, could not be explored due to the extremely small number of studies measuring these additional family factors.

While all Category 2 studies addressed family factors, predominately FA, there is likely to have been variation across studies in how family factors were addressed. Differences in therapist level of training, therapist style, and content of the treatment programs addressing family factors could have affected treatment outcomes. In particular, some programs may have addressed family factors in the context of psychoeducation, whereas other programs may have included a more practical skills-training approach. In addition, more experienced therapists may be adept at experientially addressing a range of family factors, and understanding the function of these factors for individual families in the maintenance of OCD symptoms. The quality of the approach employed to address family factors and the nature of the potential differences between studies would only be quantifiable through observation of sessions, such

as the coding of treatment session recordings. As such, this remains a focus area for future research, however, relies on the availability of recordings (or transcripts thereof) and/or publicly available data.

Moving forward, relatively little is still known about the components of family-based interventions that enhance treatment response for young people with OCD, including optimal family-related treatment content as well as the dose and nature of family involvement. Future meta-analyses would advance the literature and build on current findings by focusing further on family-related treatment moderators for young people with OCD, particularly as additional data becomes available. Findings from the current study's examination of treatment moderators emphasise the importance of addressing a number of family factors (e.g., FA, conflict, blame/criticism, problem-solving, and communication) in future interventions for young people with OCD. Further research is needed to better understand the most effective way of addressing these family factors. Questions for further exploration include whether family factors need to be extensively addressed and the relevant skills practised by families or whether merely educating families about the unhelpful nature of these factors and the role they play in maintaining OCD is sufficient. In addition, whether particular family factors play more of a maintaining role in the disorder and, therefore, need greater attention in treatment to bolster symptom improvement remains unclear.

Improved reporting of the specific family factors addressed in treatment will assist in identifying those associated with enhanced outcomes: Authors are encouraged to provide this information in the method section of forthcoming research papers. More frequent assessment of family variables both pre- and post-treatment using standardised measures is necessary to assist with identification of these core family maintaining factors. It is suggested that the effects on FA outcomes of parent and family time (in addition to other potential family-related treatment moderators) be re-examined once a larger number of studies have measured FA. Future research would further assist by focusing on the development and validation of measures assessing a broader range of family factors, as current measures are limited. Measures created for multiple responders, such as parent, child, and clinician (where possible), would further enrich findings. A range of assessment modes would assist to capture family factor change from pre to post treatment, including behavioural tasks such as parent–child discussion or interaction tasks.

Empirical research comparing interventions that include nil/minimal family involvement with those with high levels of involvement may provide a better understanding of the relative impact of degree of family participation. This could involve further comparing interventions where family factors are directly targeted with those where family factors are

not addressed (Category 1 vs. Category 2). Studies comparing interventions that target different types of family factors would significantly assist to identify key family factors affecting treatment response. The relationship between family factors addressed in treatment, FA change, and OCD symptom change pre- to post-treatment needs further investigation, including the mediatory relationship between FA and OCD symptom change proposed by previous research (e.g., Piacentini et al. 2011).

This systematic review and meta-analysis highlights the importance of addressing a range of family factors in the treatment of child/adolescent OCD in order to enhance outcomes for young people and their families. Further research is warranted to improve theoretical models and explain the impact of parental involvement in treatment and the direct targeting of family factors on family factor outcomes implicated in the maintenance of OCD.

## Compliance with Ethical Standards

**Conflict of interest** The authors declare that they have no conflict of interest.

**Ethical Approval** This article does not contain any studies with human participants or animals performed by any of the authors.

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