Efficacy of metacognitive therapy in improving mental health: A meta-analysis of single-case studies

Abstract

Context. Metacognitive therapy and one of its treatment components, the attention training technique are increasingly being delivered to improve mental health. **Objective.** To examine the efficacy of metacognitive therapy and/or attention training technique on mental health outcomes from single-case studies. **Methods.** Fourteen studies (53 patients) were included. The *d*-statistic for multiple baseline data and the percentage change index were used to compute the effect sizes. **Results**. Metacognitive therapy has a large effect on depression, anxiety, other psychopathological symptoms, and all outcomes together. Effect sizes were significantly moderated by the number of sessions, the severity and duration of symptoms, and patient gender, but not by study quality or attention training technique when used as a stand-alone treatment. At the follow-up, 77.36% of the individuals were considered recovered or had maintained improvement. **Conclusion**. Metacognitive therapy and attention training technique strongly contribute to improving mental health outcomes. This study effectively informs evidence-based practice in the clinical milieu.

Keywords: Metacognitive therapy; attention training technique; single-case; mental health; anxiety; depression; meta-analysis.

Introduction

Metacognitive therapy was developed 2 decades ago to treat anxiety-related disorders, but has since been expanded as a general and transdiagnostic treatment approach (Wells, 2009). This therapy is based on the Self-Regulatory Executive Function model, a transdiagnostic and metacognitive model of emotional disorders. The model posits that the cause of many psychological disorders is rooted in the activation of a dysfunctional pattern of thinking and self-regulation called the "cognitive attentional syndrome". More specifically, the cognitive attentional syndrome includes several features such as perseverative thinking (e.g., worry and rumination), threat monitoring, and counterproductive cognitive-behavioral coping strategies such as thought control and suppression, reassurance seeking, emotional avoidance, or substance use (Wells, 2009). These dysfunctional coping strategies in particular contribute to dysregulation and the maintenance of negative beliefs and emotional distress. Although the cognitive attentional syndrome aims to deal with negative thoughts, beliefs, or emotions, it prevents these experiences from being efficiently regulated. In addition, the excessive conceptual processing underlying the cognitive attentional syndrome is fueled by diminished attentional control and, in particular, marked difficulty in disengaging from rumination, worry, and other forms of self-focused attention.

According to the Self-Regulatory Executive Function model, the cognitive attentional syndrome results from positive and negative dysfunctional metacognitive beliefs (Wells, 2009). On the one hand, positive metacognitive beliefs refer to the usefulness of rumination, worry, rituals, threat monitoring, and other similar strategies (e.g., "I must worry in order to face daily life problems"; "I must control my thoughts or something bad will happen"). On the other hand, negative metacognitive beliefs refer to

the negative implications and meaning attributed to mental processes such as thoughts or mental images (e.g., “Some thoughts are dangerous or harmful”; “Intrusive images are a sign of folly”). Although it is predominantly the case that negative metacognitive beliefs are the most significant causal influence on pathology, the co-occurrence of positive and negative metacognitive beliefs can be detrimental as well (Wells, 2009).

In recent years, accumulating evidence corroborates the Self-Regulatory Executive Function model by showing that dysfunctional metacognitive beliefs play a pivotal role in the etiology of numerous mental disorders (Sun, Zhu, & So, 2017).

Rather than challenging the validity of thoughts and beliefs about the self, others, and the world, as in traditional cognitive-behavioral therapy, metacognitive therapy fosters the development of a detached awareness vis-à-vis one’s thoughts and aims at optimizing the control of dysfunctional cognitive emotion regulation strategies (e.g., worry, rumination) and maladaptive attentional strategies. Crucially, in contrast to cognitive-behavioral therapy, metacognitive therapy focuses on the process rather than the content of thinking. Indeed, it aims at interrupting the cognitive attentional syndrome triggered by negative thoughts by favoring a detached mindful state, improving attentional flexibility, and strengthening executive control. Ultimately, such a shift in the process of treating internal and external information is supposed to challenge both the positive and the negative metacognitive beliefs that perpetuate emotional distress. Typically, metacognitive therapy lasts for approximately 12 sessions and begins with the identification of the cognitive attentional syndrome through an idiosyncratic case formulation derived from both evidence-based, disorder-specific models and transdiagnostic models such as the Self-Regulatory Executive Function (Wells, 2009).

Specific intervention techniques have been developed within the framework of metacognitive therapy to optimize and facilitate effective executive control and to mitigate the cognitive attentional syndrome ; such techniques include attention training technique, detached mindfulness, and worry and rumination postponement (Wells, 2009). In particular, the efficacy of attention training technique as a stand-alone intervention has been investigated in a growing number of studies (for a review, see Fergus & Bardeen, 2016). During attention training technique sessions, participants are first provided with a rationale explaining that this technique has to be considered a strategy that aims to decrease self-focused attention. Regarding logistics, a series of close and distant environmental sounds (e.g., the voice of the therapist, a tapping on a table, the sound of water drops falling on a washbasin, the distant sound of traffic) are typically used in the framework of guided exercises that aim to recruit various attentional process involved in the improvement of cognitive control. The number of sounds used can vary providing the task is sufficiently difficult to tax attentional resources. In general, the clinician gives verbal instructions as to how patients should attend to auditory stimuli. However, an automated version (audio recording) has also been developed (Fergus & Bardeen, 2016).

According to Wells (2009), a typical attention training technique session comprises three types of exercises that successively require (1) selective attention (i.e., focusing on one specific sound after another), (2) rapid attention switching (i.e., rapidly switching from one sound to another), and (3) divided attention (i.e., deploying attention to all of the sounds simultaneously). The entire procedure lasts approximately 12 min. In accordance with Wells' (2009) procedure, patients are told that attention training technique must not be viewed and used as a coping strategy deployed to become distracted or to avoid negative thoughts or emotions, nor to control their negative

thoughts and emotions. From such a perspective, all intrusive thoughts or feelings that occur while performing the exercises may be noticed incidentally, and if so they should be treated as "background noise". Consequently, participants are clearly instructed not to deal with them, but rather to consider them as opportunities to improve attentional control. Each session ends with a collaborative review of the exercises, and then participants are instructed to practice attention training technique at home daily.

The reason for why the attention-training technique has been singled-out as a stand-alone intervention lies in the fact that it specifically targets excessive and inflexible self-focused attention, which constitutes a key process underlying many psychopathological states. Indeed, the rationale behind the technique is that the auditory monitoring exercises performed in the attention training technique are cognitively demanding enough to recruit and improve attentional control, interrupting the cognitive attentional syndrome (and the focus on self-focused repetitive thinking or threat monitoring) via the processing of external non-self-relevant stimuli (Knowles, Foden, El-Deredy, & Wells, 2016).

Although metacognitive therapy share common features with other psychological process-centered interventions, such as mindfulness-based interventions, it also differs from them. For instance, both attention training technique and mindfulness-based interventions aim at regulating the focus of attention, detached mindfulness in metacognitive therapy does not involve meditation or body-focused exercises and rather aims at developing meta-awareness of thoughts than present-to-moment awareness (Wells, 2009).

The efficacy of metacognitive therapy has been examined in a range of randomized controlled trials conducted in clinical samples. More specifically, a recent meta-analysis performed by Normann, van Emmerik, and Morina (2014) that

incorporated 16 studies conducted on 384 outpatients with mental disorders (including OCD, PTSD, GAD, major depressive disorder, postpartum depression, or comorbid anxious and mood disorders) showed a large effect size on the primary outcome measures (Hedges' *g* of 2.00 for within group pre- to posttreatment and 1.65 for within group pretreatment to follow-up). Of note, nine studies included in this meta-analysis were controlled trials, and seven were uncontrolled trials (with four open trials and three single-case designs). An important finding from this meta-analysis was that it supported the conclusion that metacognitive therapy is superior to both a control condition (waiting list) and a traditional cognitive-behavior treatment. The latter result must, however, be taken cautiously, given the small number of studies included (Normann et al., 2014). Notably, the meta-analysis showed not only that metacognitive therapy decreased symptoms related to patients' primary diagnosis, but also that it mitigated transdiagnostic processes, such as the dysfunctional metacognitive beliefs related to the cognitive attentional syndrome. This result in particular strengthens the feature of the Self-Regulatory Executive Function model that considers a common underlying harmful style of thinking to contribute to psychological distress across disorders (Wells, 2009). Overall, it appears that a convincing corpus of data supports metacognitive therapy as an effective approach to treat emotional disorders.

While this recent meta-analytic review of metacognitive therapy demonstrated large effect sizes across anxiety disorders and depression (Normann et al., 2014), the efficacy of the attention training technique as a stand-alone intervention has also been systematically assessed, with numerous studies demonstrating that it is efficacious across a number of psychopathological conditions (Fergus & Bardeen, 2016). Attention training technique might be relevant as a stand-alone intervention because it particularly targets excessive and inflexible self-focused attention underlying worry, rumination,

and threat monitoring that characterizes the cognitive attentional syndrome. More specifically, Knowles et al. (2016) recently performed a meta-analysis of both single-case and group studies on eight articles (four single cases and four randomized controlled trials) comprising 293 participants. Single-case outcomes indicated that attention training technique yields large effect size estimates, expressed as improvement rate difference, with a pooled effect size ranging from 0.74 to 1.00 for anxiety and depressive disorders. In addition, standardized effect size across the four randomized controlled trials indicated that attention training technique is more efficacious (with a small-to-large effect size depending on the outcome) than control groups in relation to various outcomes (e.g., intrusive thoughts, self-focused attention, attention flexibility, pain threshold, hypervigilance). Long-term effects (from 6 to 12 months) were also reported in most of the studies considered for the analyses.

Numerous studies have provided empirical evidence for the efficacy of metacognitive therapy or attention training technique as a stand-alone intervention, but no meta-analysis has to date been performed to examine the efficacy of metacognitive therapy in single-case studies, although an important number of single-case design studies on metacognitive therapy have been published in the last 2 decades. Single-case methodology has unique strengths for assessing the efficacy of a treatment and thus constitutes a clinically relevant and scientifically well-established alternative to traditional group comparison designs (e.g., Dattilio, 2006). The scientific rigor of single-case methodology has also been acknowledged by influential international groups in the evidence-based movement (OCEBM Levels of Evidence Working Group, 2011). In addition, encouraged by the evidenced-based practice movement, researchers have adapted (or developed) methodology and statistical analyses to single-case design

and these techniques have progressed in recent years (e.g., Manolov, Gast, Perdices, & Evans, 2014).

The primary aim of this meta-analysis was to estimate the effect of metacognitive therapy on mental health in single-case studies exclusively. The analyses focused on various mental health outcomes considered together or separately so that the results of the present study could be directly compared to those of previous systematic reviews of group designs on this topic (Normann et al., 2014; Sadeghi, Mokhber, Mahmoudi, Asgharipour, & Seyfi, 2015). For this purpose, we used a data analysis procedure specifically developed on the basis of statistical theory for single-case design outcomes so that they would be comparable to traditional group-design outcomes (Shadish, Hedges, & Pustejovsky, 2014). The second objective was to compare the efficacy of one specific component of metacognitive therapy, the attention training technique, as a stand-alone treatment, to the efficacy of metacognitive therapy as a whole package treatment program by including a moderator variable specifying whether attention training technique was or was not used as a stand-alone treatment. This question is particularly relevant because attention training technique is considered a low-cost, easy-to-administer technique that provides an economic advantage compared with the full-package therapeutic program (Fergus & Bardeen, 2016). The third objective was to examine the effect of specific moderators (e.g., sociodemographic variables, number of sessions, duration of symptoms, presence of comorbidity, psychotropic medication use, severity of the target behavior at baseline) on the outcomes to better understand for whom metacognitive therapy produced the greatest effect, thereby reinforcing evidence-based practice in the therapeutic milieu.

Methods

This study was conducted in accordance with the preferred reporting items for systematic reviews and meta-analyses that evaluate health care interventions (PRISMA; Moher et al., 2009).

Search strategy

A systematic literature search was conducted in three electronic databases: PsycINFO, PubMed, and Web of Science. Each database was initially searched for English language journal articles from the first available date until January 2, 2017, using the following search terms: (metacognitive therapy OR attention training technique) AND (single-case OR case series OR multiple baseline OR case study).

The search was also supplemented with the following steps: (a) authors who had published substantially in the area were contacted to obtain information about published or unpublished articles germane to the review, (b) Google Scholar was used to search for articles that may have been unidentified, and (c) reference sections of identified articles, in particular systematic reviews and meta-analyses on the topic (e.g., Fergus & Bardeen, 2016; Knowles et al., 2016; Normann et al., 2014; Sadeghi et al., 2015), were examined and a citation search was conducted to identify further studies.

Selection of studies

After the removal of duplicates, the remaining titles were reviewed and the abstracts of the potentially relevant articles screened. The full texts of the selected articles were then obtained and assessed for eligibility. The screening of titles, abstracts, and full-text articles was independently conducted by two authors (LR, JB). Disagreements between the authors were discussed until consensus was reached.

Figure 1 shows the search process in detail. The initial results identified 70 citations after de-duplication, which were then examined against the following inclusion criteria:

- Studies had to evaluate the effectiveness of metacognitive therapy or attention training technique as a stand-alone intervention.
- Studies had to be published in English only and in peer-reviewed journals.
- Studies had to use a single-case methodology in which the design had to include a series of discrete phases wherein an intervention was manipulated in an experimentally controlled manner (i.e., was systematically applied and withdrawn) and the target behavior was continuously and frequently measured (Tate et al., 2015). Single-case studies that used appropriate methodology to rule out threats to internal validity such as a single-case experimental design (e.g., reversal, multiple baseline, alternating treatments) were acceptable for inclusion. Furthermore, quasi-experimental designs (e.g., AB with maintenance phase) were included if they relied on validated psychopathology measures.
- Studies had to examine clinical adult samples with a formal diagnosis and/or who met criteria on a validated psychopathology measure.
- Studies had to use health-related outcome measures of psychopathology or symptom severity, or measures of cognitive modification related to the targeted psychopathology.

We excluded studies that (a) did not include any comparisons with at least three measurement points per phase of the intervention, because this is both a design requirement (Kratochwill et al., 2010) and a necessity for the statistical analyses; (b) were only descriptive; or (c) used a pre/post design, as this is not considered *stricto sensu* single-case methodology (Tate et al., 2015).

A total of 14 articles met full inclusion criteria, for which full paper copies were retrieved to assess further eligibility.

Data extraction and quality assessment

For each included study, the following data were extracted by the first author (LR) and then checked by the third author (JB): year of publication, number of patients included, type of diagnosis or targeted psychopathology, type of intervention and number of sessions, outcomes considered, and type of single-case design adopted (Table 1). For each participant of each included study, moderators were then extracted by the first author (LR) and further checked by the third author (JB): age, gender, number of sessions, presence versus absence of comorbid psychopathology, presence versus absence of psychotropic medication, duration of symptoms, and severity of symptoms at baseline. In addition, studies that examined the efficacy of attention training technique as a stand-alone treatment were distinguished from those that used a more comprehensive metacognitive therapy treatment approach. Raw data for the outcomes measured were extracted from the graphs by the second author (RM) with PlotDigitizer 2.6.3 for Windows (plotdigitizer.sourceforge.net/). Previous research has shown that data retrieval from graphs leads to reliable and valid results across several data extraction software tools (Drevon, Fursa, & Malcolm, 2017).

The methodological quality of each study was independently assessed by two authors (LR, RM), who used the 15 criteria of the Risk of Bias in N-of-1 Trials (RoBiNT) Scale (Tate et al., 2015), which assesses internal and external validity of each study by assigning 2 points for each criteria that is fully met, 1 point if partially met, and 0 points if not met. Thus, the maximum score is 30 points. Disagreements between the two authors who assessed the quality of the studies were discussed until an agreement was reached.

Three corresponding authors of four different studies were contacted because insufficient information was provided in the article regarding data extraction, or to make an accurate quality assessment. One author provided supplementary information.

Data analysis

Choice of meta-analytic strategy

The between-case standardized mean difference, more widely known as the *d*-statistic, was chosen because it allows for a classical meta-analysis using inverse variance weighting, representing the effects via a forest plot. The main limitations of *d*-statistic are detailed in Valentine, Tanner-Smith, Pustejovsky and Lau (2016). One of the restrictions regards its applicability to studies with at least three participants, which was the case for 7 of the 9 studies reporting anxiety (77.78%) and 9 of the 10 studies reporting depression outcomes (90.00%). These values indicate that the loss of information due to the use of *d*-statistic was relatively minor. Additionally, the percentage change index (Hershberger, Wallace, Green, & Marquis, 1999) is computed for all two-phase comparisons between an A (baseline) phase and a subsequent B (intervention) phase for all participants. The use of the percentage change index is well-aligned with the visual inspection of the data suggesting that there was a progressive improvement (i.e., a change in slope) in many of the data sets, given that it focuses on the last three measurements per phase.

Calculation of effect sizes

Meta-analyses were performed grouping the outcomes in four different ways: (a) outcomes referring to anxiety using the Beck Anxiety Inventory (Beck, Epstein, Brown, & Steer, 1988) and the Hospital Anxiety and Depression Scale (Zigmon & Snaith, 1983); (b) outcomes referring to depression using the Beck Depression Inventory (Beck,

Ward, Mendelson, Mock, & Erbaugh, 1961), Hospital Anxiety and Depression Scale and the Geriatric Depression Scale (Yesavage et al., 1982); (c) other psychopathological outcomes (labelled as "remaining" hereinafter). Note that in the study by McNicol, Salmon, Young, and Fisher (2013), only the total score of the Hospital Anxiety and Depression scale is used and thus the outcome was labeled as "remaining", as it did not refer to either depression or anxiety separately; (d) all outcomes (anxiety, depression, and the remaining ones) grouped together. In all cases, improvement is seen as a reduction in the measurements.

When meta-analyzing with the *d*-statistic, one effect size per study is obtained, as this index combines the results for several participants into a single quantification. The data from Wells (1990) and McNicol et al. (2013) were not taken into account as they include only one participant and the data from Wells et al. (1997) because they include only two participants. Additionally, Papageorgiou and Wells (2000) measured outcomes only related to depression and anxiety and, thus, this study was not included for the meta-analysis of the remaining outcomes. For the *d*-statistic, the analyses for each study were performed using the "scdhlm" (https://github.com/jepusto/scdhlm) package for R (R Core Team, 2015), also available at https://jepusto.shinyapps.io/scdhlm/, whereas meta-analysis was performed using random effects models via the R code presented in Manolov and Solanas (2016).

For the percentage change index, we computed one value for each AB comparison and then obtained an average percentage change index per study and per outcome (anxiety, depression, remaining, or all), using the number of measurements in the AB comparison as a weight. The meta-analysis of these one-per-study average percentage change index values was performed using the number of measurements available in the study as a weight, a reasonable approach according to Kratochwill et al. (2010), when

inverse variance weighting is impossible. The meta-analysis using the percentage change index was performed via the R code from Manolov and Rochat (2015).

Vote counting

Vote-counting was applied to the follow up data. That is, we tallied the number of follow-up results that the authors considered as positive (effective intervention), focusing on the most stringent of all the criteria reported by the primary authors and on the latest follow-up measure available. After the number of positive results was tallied, the proportion of positive results in relation to the total number of participants was compared to the chance proportion of 0.5 using the binomial test; we also constructed the confidence interval around the proportion observed via the expressions provided in Bushman and Wang (2009).

Assessment of the evidence for metacognitive therapy

We assessed whether metacognitive therapy can be considered an "evidence-based practice" via the 5-3-20 criterion (Kratochwill et al., 2013) requiring positive evidence for the intervention from at least 5 studies carried out by three different and independent research teams and including at least 20 participants.

Moderator analysis

We considered moderator analysis to be justified when significant results of the Q test for heterogeneity of effects were obtained and for I^2 values greater than 50% (medium relative heterogeneity) and especially for I^2>75% (large relative heterogeneity). We used the following moderator variables: Metacognitive therapy vs attention training technique as a standalone treatment, the mean level of the target behavior during baseline, the number of intervention sessions, the duration of the disease in months,

whether participants were taking or not psychotropic medication when the study was carried out, the age and gender of the participants, and the presence or absence of co-morbidity. A file with all the moderators by participants and studies is provided at https://osf.io/am77z/.

In order to perform moderator analyses for the *d*-statistic at the study level, as described by Shadish et al. (2014), a single quantification per study for each moderator has to be obtained. Therefore, we calculated the proportion of individuals displaying a comorbid psychopathology, taking psychotropic medication and being female and computed the average age, duration of symptoms, and the number of sessions during the intervention phase. In order to examine the importance of the moderator variables at the individual level, we used the moderators in simple linear regression analyses, weighted by the number of measurements in phases being compared, one per each moderator used as a predictor of the values of the percentage change index computed for outcomes related to anxiety and depression.

Assessment of publication bias

Publication bias was dealt with in three ways. First, a funnel plot was created by plotting the effect sizes on the abscissa and the standard error of the effect size index on the ordinate. Publication bias is usually inferred from a lack of symmetry in the distribution of effect sizes, with smaller studies providing only larger effects (Sterne, Egger, & Moher, 2008). Second, to counter the potential subjectivity of the funnel plot, we used Egger's regression test (Egger, Smith, Schneider, & Minder, 1997) for exploring whether the effect size of smaller studies differs systematically from the effect size found in larger studies using a nominal alpha of 0.10 (i.e., rejecting the null hypothesis if $p \leq 0.10$) instead of the common 0.05, as suggested by Egger et al. (1997) for meta-analyses including relatively few studies. Third, Duval and Tweedie's (2000)

trim-and-fill procedure was applied, which eliminates any existing asymmetric effect sizes to estimate the center of the funnel plot (i.e., the overall summary measure), replaces these asymmetric effect sizes and their symmetric counterparts and re-estimates the average and the variance of the summary measure. Therefore, an adjusted effect size is produced, accounting for the potentially missing studies (Duval & Tweedie, 2000).

Results

Selection of studies

A flow diagram of the study selection process is presented in Figure 1. The electronic database searches produced 70 records after removal of duplicates. After reviewing the titles and the abstract, we identified 22 potentially eligible records. Full-text versions of these articles were obtained and assessed for eligibility. This led to the inclusion of 14 single-case studies that compared metacognitive therapy or attention training technique as a stand-alone treatment to a baseline in a total of 52 participants (53 if counting a participant in the study by Fitt & Rees, 2012, who did not complete the intervention). Unpublished data were not identified.

--INSERT FIGURE 1 HERE--

Description of included studies

Characteristics of the included trials are presented in Table 1.

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Sample characteristics

The total sample comprised 53 outpatients, of whom 10 received attention training technique as a stand-alone treatment. Three participants were excluded because not enough data points (i.e., fewer than 3 per phase) were collected and one participant was excluded because of not completing the intervention (Fitt & Rees, 2012). The majority of the sample was female (80%). All participants were adults, with ages ranging from 20 to 71 years. The total sample size per study ranged from one to six patients. Eight of the 14 studies were conducted in patients with a primary diagnosis of anxiety disorder (OCD, panic, hypochondriasis), four in patients with depression (postpartum, recurrent

major depressive disorder, major depressive episode), one patient with paranoid schizophrenia, and one in a patient who was a cancer survivor with emotional distress (anxiety, depression, PTSD). The mean duration of symptoms was 95.19 months (range from 2 to 480 months). Twenty-two patients had a least one comorbidity and 19 had a concurrent psychotropic medication at the time of the intervention.

Intervention characteristics

Most studies examined the efficacy of metacognitive therapy by using the original manual developed by Wells (2009), whereas two studies (Bevan et al., 2013; Hutton, Morrison, Wardle, & Wells, 2014) adapted the intervention to a specific target population (postpartum depression and paranoid schizophrenia, respectively). In addition, four studies examined the efficacy of attention training technique as a stand-alone treatment. All interventions were conducted face to face (one patient, one practitioner) and one intervention was delivered by video conference (Fitt & Rees, 2012). Sessions were usually delivered on a weekly basis and the total number ranged from 4 to 14.

Design

Among the 14 studies, three used a single-case experimental design (e.g., multiple baseline across participants, reversal design) in that the structure of the design provided sufficient opportunity to adequately demonstrate the experimental effect (Andouz, Dolatshahi, Moshtagh, & Dadkhah, 2012; Wells, 1990; Wells et al., 1997). The remaining studies used a quasi-experimental design such as an AB design, which is not considered experimental in that it does not provide an added opportunity to demonstrate experimental effect, as there is only a unique comparison.

Outcomes

Outcome measures (an index of all outcomes is reported in Table 1) were administered as follows: depression in 30 comparisons, anxiety in 34 comparisons, remaining outcomes in 103 comparisons. All instruments had good psychometric properties. All studies provided at least one follow-up session ranging from 6 weeks to 41 months.

Methodological quality

The quality assessment scores ranged from 8 to 15 points on the RoBiNT scale (see Table 1). All studies fully met the criteria of raw data record and full description of the dependent variables, both criteria referring to external validity. Criteria referring to internal validity were the most poorly rated. Interrater agreement on the quality assessment was elevated (94%). Note that the RoBiNT Scale represents stringent standards (Tate et al., 2015) and was initially published in 2013, the same year as two of the studies included here and only prior to the publication of three of the 14 studies from the current meta-analysis.

Meta-analysis

d-statistic

The numerical results of the meta-analysis with the *d*-statistic can be consulted in Table 2. The effect sizes per study and the overall average, including confidence intervals, are represented in Figure 2 (for anxiety) and in Figure 3 (for depression). The forest plots for the remaining outcomes and for all outcomes considered together can be obtained from https://osf.io/am77z/. The values of the *d*-statistic indicate that in all studies (and consequently for all overall summary measures), the effect of the intervention has been in the desired direction. All four summary measures, most of the

effects per study, and the overall weighted average effect size could be labeled as "large" according to Cohen's (1992) criteria (i.e., greater than 0.8).[1]

--INSERT TABLE 2 ABOUT HERE--

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Percentage change index

Regarding the results of using the percentage change index, for all studies, the results for all participants, except for one participant in Hutton et al. (2014), indicate a reduction in the problematic behaviors/outcomes measured. The average reduction is approximately 60-68% and for very few studies, the averages are below 50%. Graphical representations of the results for the percentage change index are available from https://osf.io/am77z/.Therefore, metacognitive therapy is apparently effective, given the large reductions observed on three levels: individually, on average per study, and in terms of the overall weighted average for all studies.

Vote counting

The results for vote counting indicates that 41 of the 53 individuals (77.36%) maintained the improvement achieved during the intervention phase at the most stringent criteria for intervention effectiveness and at the longest available follow-up point (ranging from 6 weeks to 41 months). This percentage was statistically significantly different from 0.5 according to the binomial test ($p < .01$), and the

[1] The use of Cohen's benchmarks for single-case designs has been put in doubt (Parker et al., 2005) and Harrington and Velicer (2015) suggested referring to values between 1 and 2.5 as "medium effects." However, they used the within-case version and not the *d*-statistic created to be comparable to that obtainable from between-group designs (Hedges, Pustejovsky, & Shadish, 2013).

confidence interval for the percentage estimated according to the formulas from Bushman and Wang (2009) ranged between 0.707 and 0.804. Notably, for this analysis, the two participants for whom there were fewer than three baseline phase measurements were taken into account (Fitt & Rees, 2012; Wells et al., 1997), as was the participant who did not complete the intervention in Fitt and Rees' (2012) study.

Assessment of the evidence for metacognitive therapy

Considering the results represented in the forest plots and the number of participants with positive results at follow-up, the minimum of 20 individuals who benefitted from metacognitive therapy or attention training technique is achieved. Additionally, given that 14 studies are included in the current meta-analysis, the minimum of five studies is also met for all outcomes considered together. Regarding the need for these studies to be authored by at "least three research teams with no overlapping authorship at three different institutions" (Kratochwill et al., 2013, p. 33), this criterion is met.

Apart from these general conclusions, according to the percentage change index computed for depression, metacognitive therapy always leads to reduction of scores, specifically for 42 participants in 10 studies and three different research teams (Andouz et al., 2012; Fitt & Rees, 2012; and eight studies in which Adrian Wells contributed [Bevan et al., 2013; Callesen, Jensen, & Wells, 2014; Fisher & Wells, 2008; Hutton et al., 2014; Papageorgiou & Wells, 1998, 2000; Wells & Sembi, 2004; Wells et al., 2009]; and Andouz et al., 2012; Fitt & Rees, 2012, and eight studies in which Adrian Wells contributed [Bevan et al., 2013; Callesen et al., 2014; Fisher & Wells, 2008; Hutton et al., 2014; Papageorgiou & Wells, 1998, 2000; Wells & Sembi, 2004; Wells et al., 1997]). According to the percentage change index computed for anxiety, metacognitive therapy always leads to reduction of the scores, specifically for a total of

33 participants in 9 studies and two different research teams (Fitt & Rees, 2012, and eight studies in which Adrian Wells participated [Bailey & Wells, 2014; Bevan et al., 2013; Fisher & Wells, 2008; Hutton et al., 2014; Papageorgiou & Wells, 1998, 2000; Wells & Sembi, 2004; Wells et al., 1997]). Therefore, given that the "5-3-20 rule" is met considering all outcomes together, as well as for depression, there appears to be enough empirical support to refer to metacognitive therapy as an evidence-based practice.

Moderator analysis at the study level using the d-statistic

Heterogeneity descriptive statistics and the Q test results for heterogeneity are presented in Table 2. The Q test suggested that there is statistically significant heterogeneity in the effect sizes across the studies for all outcomes considered together, for depression, and for the remaining outcomes. Moreover, the proportion of heterogeneity that is due to the variation in true effects, rather than being sampling error, is large (i.e., $I^2 > 75\%$) for all outcomes and for the remaining outcomes and medium ($I^2 \approx 50\%$) for depression. Following the suggestion by Borenstein, Hedges, Higgins and Rothstein (2009), we also report τ, the standard deviation of the true effects, with values between 0.51 and 0.65 for all outcomes, depression, and remaining outcomes (apart from anxiety and depression), which are apparently large considering that the weighted average *d*-statistic values are between −1.72 and −1.20 for these outcomes. The range of *d*-statistics for the individual studies is also wide, although for depression, the range is affected by the extreme *d*-statistic value for the Papageorgiou and Wells (2000) study and for anxiety, it is affected by the extreme *d*-statistic value for the Papageorgiou and Wells (1998) study.

For all outcomes considered together, the number of sessions was statistically significantly related to the effect sizes (p = .044; R^2 = 45.64%) and this reduced I^2 from 81.02% to 68.11%, with more sessions being associated with larger differences between baseline and intervention conditions. The proportion of female participants in the sample was also statistically significantly related to the effect sizes (p = .031; R^2=54.79%) and this reduced I^2 from 81.02% to 62.50%, with studies with fewer female participants reporting greater differences between intervention and baseline. The two moderators are collinear (r = −.61, p = .059) and thus were not used jointly in the same model.

Finally, for depression as outcome, none of the moderators was statistically significantly related to the effect sizes and the moderator that reduced I^2 to a greater extent (from 55.94 to 49.93%) was the number of sessions, with more sessions being associated with larger differences between baseline and intervention conditions. For the remaining outcomes, none of the moderators was statistically significantly related to the effect sizes and the moderator that reduced I^2 to a greater extent (from 76.73% to 68.26%) was the proportion of female participants, with studies with fewer female participants reporting greater differences between intervention and baseline.

Moderator analysis at the individual level using the percentage change index

For anxiety, the average of the last three baseline measurements was statistically significantly related to the percentage change index values (b = 1.15, p = .001, R^2 = 32.91%). More specifically, lower baseline levels of anxiety were related to a greater decrease in symptoms. For depression, the duration of the symptoms was statistically significantly related to the percentage change index values (b = 0.045, p = .039, R^2= 11.95%), indicating that longer symptom duration was associated with worse results.

Similar to those for anxiety, lower baseline levels of depression were related to greater reductions (b = 0.62, R^2 = 9.70%), but this result was not statistically significant (p = .077). When the duration of the symptoms and the baseline level of depression were used as predictors in a multiple regression, these variables accounted for R^2 = 22.83% of the variability in the percentage change index values (p = .020 for the model).

Assessment of publication bias

The result of the funnel plots for the *d*-statistic are available from https://osf.io/am77z/. The distributions are practically symmetric, with one outlier for both anxiety and depression. Therefore, visually, there seems to be one extreme result included rather than solid evidence for publication bias for anxiety and depression. If the meta-analytic summary for anxiety is performed without the Papageorgiou and Wells (1998) study, the overall d is −1.31 (instead of −1.41) with a confidence interval ranging between −1.76 and −0.86. If the meta-analytic summary for depression is performed without the Papageorgiou and Wells (2000) study, the overall d is −1.08 (instead of −1.20) with a confidence interval ranging between −1.58 and −0.57. Thus, the conclusions do not change substantially after removing extreme values. More asymmetric results are apparently present when considering the remaining outcomes or all outcomes together.

In order not to rely on visual inspection only, we used Egger's regression test for funnel plot asymmetry. The following results were obtained: (a) for all outcomes, Z = −1.53, p = .13; (b) for anxiety, Z = −1.12, p = .26; (c) for depression, Z = −2.21, p = .03; and (d) for the remaining outcomes, Z = −0.80, p = .43. Therefore, statistically, there is an indication of asymmetry and potential publication bias for depression only. Nevertheless, we still applied the trim-and-fill method to obtain adjusted overall means

supposed to be free of publication bias. These means (for all outcomes −1.29 instead of −1.55, with a confidence interval ranging between −1.72 and −0.85; for the remaining outcomes, −1.45 instead of −1.72, with a confidence interval ranging between −1.98 and −0.92) continue indicating that a relatively large reduction of problematic outcomes has taken place.

Discussion

The main aim of this meta-analysis was to estimate the overall effects of metacognitive therapy on mental health outcomes in single-case studies. We used specific and appropriate analyses developed on the basis of statistical theory for single-case design outcomes so that they would be comparable to group-design outcomes, and the results indicated that metacognitive therapy has a large effect size on anxiety, depression, other psychopathological symptoms, and all outcomes considered together. Publication bias was statistically detected only for depression as outcome, probably due to one outlying effect size. However, we verified that the overall result (a large effect of meta-cognitive therapy) did not change substantially in either of two re-analyses: (a) when removing the outlying value from the meta-analysis; and (b) when obtaining an adjusted summary measure, imputing potentially missing effect sizes, according to the trim-and-fill method. Additional analyses revealed that effect sizes were moderated by the number of intervention sessions, the severity and duration of symptoms, and patient gender, but not by study quality or the type of intervention (i.e., attention training technique as a stand-alone treatment vs. the whole metacognitive therapy package).

Together, these results support the use of metacognitive therapy or attention training technique, as an evidence-based intervention to treat emotional disorders. This result corroborates previous meta-analyses conducted on group designs that showed a medium to large effect on mental health outcomes of metacognitive therapy or attention

training technique as a stand-alone treatment (Knowles et al., 2016; Normann et al., 2014). In addition, at the longest follow-up measure available, 77.36% of the individuals were considered recovered or improved, with the problem being eliminated or improvement being maintained during the intervention phase. This result is notable inasmuch as the effects of some psychological or psychopharmacological interventions tend to reduce or disappear after the withdrawal of treatment (e.g., Hollon, Thase, & Markowitz, 2002).

Regarding our second objective, which pertains to the examination of the efficacy of attention training technique as a stand-alone treatment, no significant effect of this moderator was found on the outcome measures. This result can be interpreted in two ways. First, corroborating the absence of an effect of an attention training technique component added to a cognitive behavior therapy program for reducing social phobia (McEvoy & Perini, 2009), some components of metacognitive therapy, such as detached mindfulness and worry or rumination postponement, may already promote a metacognitive mode of processing that interrupts the cognitive attentional syndrome by increasing attentional flexibility and executive control. From such a perspective, adding an attention training technique module to the package program may provide no additional benefit. Second, attention training technique as a stand-alone intervention may be in certain cases considered sufficient enough to challenge dysfunctional metacognitive beliefs and self-focused attention that perpetuate the cognitive attentional syndrome without requiring a more comprehensive metacognitive therapy package. This result is of particular interest inasmuch as attention training technique is considered a low-cost, easy-to-administer technique that provides an economic advantage compared with the full-package therapeutic program (Fergus & Bardeen, 2016). It remains to be determined, however, for which cases attention training technique as a stand-alone

treatment or metacognitive therapy as a whole treatment package (with or without an ATT component) fits best. Given that attention training technique has been shown to improve both self-reported mental flexibility and disengagement of attention from negative stimuli on a laboratory task (Callinan, Johnson, & Wells, 2015), such laboratory tasks (or questionnaires assessing related processes such as the Attention Control Scale; Derryberry & Reed, 2002) could tentatively be used to help identify individuals who would more likely benefit from this component treatment.

Regarding other moderators, the results indicated at the study level a significant dose-response relationship; that is, the greater the number of sessions, the better the outcomes. Although it is difficult to provide an optimal dosage of metacognitive therapy or attention training technique sessions, Knowles et al. (2016) and Wells (2009) generally considered that changes can be seen after a few weeks when weekly attention training technique sessions, guided by the therapist, are associated with daily personal practice at home. Nevertheless, Wells (2009) mentioned that more severe cases might require more intensive practice of the attention training technique. More generally, according to Hansen, Lambert, and Forman (2002), in the clinical trials literature, between 57.6% and 67.2% of patients with axis I disorders improve within an average of 12.7 sessions. The results also showed that gender moderated the efficacy of metacognitive therapy on the outcomes, as women showed worse outcomes than men did. Although this result should be interpreted with caution because of the overrepresentation of women (80%) in the studies included for the analyses, it is in line with the greater incidence and severity of internalizing symptoms (e.g., depression) in women than in men. Women may present a more inflexible and excessive cognitive attentional syndrome than men do, which in turn makes the treatment less efficacious for women. This assumption corroborates previous data showing that gender moderates

the link between self-focus attention and negative affect, in that being a woman increased the strength of this relationship (Mor & Winquist, 2002).

At the individual level, the moderator analysis showed that the longer the duration of the depression symptoms, the lower the efficacy of metacognitive therapy and/or attention training technique. These results indicate that metacognitive therapy and its various components might be particularly useful and relevant at an early stage of the development of psychopathological symptoms. Indeed, when the cognitive attentional syndrome and associated metacognitive beliefs and a nonadaptive self-focus strategy have been automatized and become excessively rigid and inflexible, it might be more challenging to have the person interrupt the cognitive attentional syndrome triggered by negative thoughts based only on metacognitive therapy and/or attention training technique interventions. Corroborating previous studies showing that chronicity has been associated with more severe psychopathological symptoms (e.g., Visser, van Oppen, van Megen, Eikelenboom, & van Balkom, 2014), the results also support that the lower the severity of anxiety and depression symptoms at baseline, the better the outcomes. Consequently, to maximize the effect of metacognitive therapy and/or attention training technique, these interventions might be advantageously proposed at an early stage of symptom development, or even in identified at-risk persons, children, or adolescents as a way to prevent further development of psychopathological symptoms. From this perspective, the metacognitive model has received preliminary empirical support in both clinical and nonclinical child or adolescent samples (e.g., Ellis & Hudson, 2010). Furthermore, a growing corpus of data supports that metacognitive therapy is suitable and efficacious for children or adolescents with GAD or OCD (e.g., Esbjørn, Normann, & Reinholdt-Dunne, 2015).

Finally, one important implication of the metacognitive model is that it generated theoretically based psychological interventions that are effective across emotional disorders and can be conceptualized within a transdiagnostic approach of psychopathology (Dudley, Kuyken, & Padesky, 2011). The transdiagnostic model emerged from the many limitations of the disorder-specific approach (e.g., high comorbidity between disorders, intradiagnostic heterogeneity, or poor construct validity of psychiatric diagnoses). It postulates that key psychological processes (cognitive, affective, motivational, interpersonal) are involved in the onset, perpetuation, and recurrence of psychopathological symptoms. In such a theoretical framework, the cognitive attentional syndrome, which was found to characterize a wide range of emotional disorders, can be viewed as a transdiagnostic pathogenic process that can be targeted by specific process-oriented interventions such as the metacognitive therapy and/or attention-training technique.

Some limitations to the study must be discussed. First, the methodological quality of the single-case studies included in the analyses is, in general, rather poor. In particular, all but three studies did not use an experimental design, but rather an A-B design, which reduced the opportunity to gather evidence of experimental control over the target behaviors as a result of the interventions. However, despite the poor general methodological quality regarding the internal validity of the studies included, methodological bias scores were not crucially related to the effect sizes. Further single-case studies should use experimental designs (e.g., a multiple baseline design across participants) to increase internal validity. Second, the analyses focused essentially on primary outcomes (e.g., various symptoms belonging to a diagnostic category), but not on underlying transdiagnosis factors such as measures of metacognitive beliefs or direct measures of the cognitive attentional syndrome (which could have been measured by

specific self-reports; see Wells, 2009). In most of the studies, measures of metacognitive beliefs were used but only administered in the pre- and posttests, thus preventing their inclusion in the analyses. Similarly, no included studies used laboratory tasks for assessing difficulties in executive control (e.g., mental flexibility) that is supposed to underlie the cognitive attentional syndrome. Use of both measures of transdiagnostic factors and objective measures of attentional control would have helped to further reveal the underlying mechanisms of change in metacognitive therapy and/or attention-training technique as a stand-alone treatment.

Conclusions

To the best of our knowledge, this is the first meta-analysis on single-case studies that evaluates the specific effects of a psychological intervention on mental health outcomes that used a specific and well-suited data analysis procedure. The current study first confirms the large effects of metacognitive therapy and/or attention-training technique across various mental health outcomes. Second, it effectively informs evidence-based practice in the clinical milieu. Indeed, according to Dattilio (2006), single-case designs provide clinicians with more immediacy than do group designs in which the context and details of clinical phenomena may be masked or ignored. Crucially, we observed that the potential of relying on a meta-analysis of single-case studies to advance knowledge in determining evidence-based interventions in clinical psychology research is largely under-exploited. This study thus emphasized the relevance of disseminating the use of meta-analyses on single-case studies in the field of psychopathological research. Indeed, this approach has unique strengths and well complements meta-analyses performed on group designs in examining the efficacy of a treatment, thereby improving clinical decision making.

Disclosure

The authors report no conflicts of interest. This research did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors.

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Table 1.

Characteristics of the included trials.

Study	RoBiNT Scale score	Number of patients	Diagnosis	Strategy intervention	Outcomes	Design
Andouz, 2012	13	6	Obsessive-compulsive disorder	MCT; 14 weekly sessions for 50 min	OCI-R; Y-BOCS; TFI; MCQ; BDI-II	AB
Bailey, 2014	12	4	Hypochondriasis	MCT; 6 to 9 sessions for maximum of 1 hr	WI; MCHQ; BAI	AB
Bevan, 2013	12	6	Postpartum depression	MCT; 8 to 12 sessions for 1 hr	EPDS; HADS-Depression; HADS-Anxiety.	AB
Callesen, 2014	10	4	Depression	MCT; 5 to 11 sessions for 1 hr	BDI-II; MDD-S (rumination time)	AB
Fisher, 2008	12	4	Obsessive-compulsive disorder	MCT; 12 to 14 sessions for 1 hr	BAI; BDI; PI	AB
Fitt, 2012	11	2	Obsessive-compulsive disorder	MCT; 8 sessions for 1 hr (by video conference)	Y-BOCS; DASS-21	AB
Hutton, 2014	11	3	Paranoid schizophrenia	MCT; 11 to 13 weekly sessions for 1 hr	BDI; BAI; PSYRATS delusions; PSYRATS voices; worry is harmful; worry is uncontrollable; analyzing problems brings answers; duration of worry	Multiple baseline
McNicol, 2013	9	1	Emotional distress (anxiety, depression, and posttraumatic stress) in a cancer survivor	MCT; 7 sessions of 45 to 60 min	HADS (total score); IES-R	AB
Papageorgiou, 1998	8	3	Hypochondriasis	ATT; 8 to 10 sessions for 30 min (including 15 min of ATT)	Frequency of health worry; illness beliefs; frequency of urge to seek reassurance; avoidance behaviors; bodily checking; BAI; GDS	Multiple baseline
Papageorgiou, 2000	10	4	Recurrent major depressive disorder	ATT; 5 to 8 weekly sessions for 30 min (including 15 min of ATT)	BDI; BAI	Multiple baseline

Wells, 1990	10	1	Panic disorder	Therapy 1 (B): ATT, 15 to 18 sessions for 15 min; Therapy 2 (C): standard autogenic exercises[a]; 12 sessions	Frequency of panic attacks; PQRST neck tension intensity rating	ABCB
Wells, 1997	8	2	Panic disorder	ATT; 3 to 4 sessions for 30 min (including 10 min of ATT)	Frequency of panic attacks; BAI; belief ratings	AB and ABA
Wells, 2004	13	6	Posttraumatic stress disorder	MCT; 8 to 11 sessions for 30 to 60 min	DTS; IES; BDI; BAI	AB
Wells et al., 2009	15	4	Major depressive episode	MCT; 8 to 11 sessions for 1 hr	BDI; time spent ruminating	Multiple baseline

Note.

MCT = metacognitive therapy; ATT = attentional training technique; RoBiNT Scale = Risk of Bias in N-of-1 Trials Scale; OCI-R = Obsessive Compulsive Inventory-Revised; Y-BOCS = Yale-Brown Obsessive-Compulsive Scale; TFI = Thought Fusion Inventory; MCQ = MetaCognitive Questionnaire; BDI/BDI-II = Beck Depression Inventory/Beck Depression Inventory-II; WI = Whiteley Index; MCHQ = Metacognitions about Health Questionnaire; BAI = Beck Anxiety Inventory; EPDS = Edinburgh Postnatal Depression Scale; HADS = Hospital Anxiety and Depression Scale; MDD-S = Major Depressive Disorder Scale; PI = Padua Inventory; PSYRATS = Psychotic Symptom Rating Scales; GDS = Geriatric Depression Scale; PQRST = Personal Questionnaire Rapid Scaling Technique; DTS = Davidson Trauma Scale; IES/IES-R = Impact of Events Scale/Impact of Events Scale-Revised; DASS-21 = Depression Anxiety Stress Scales.

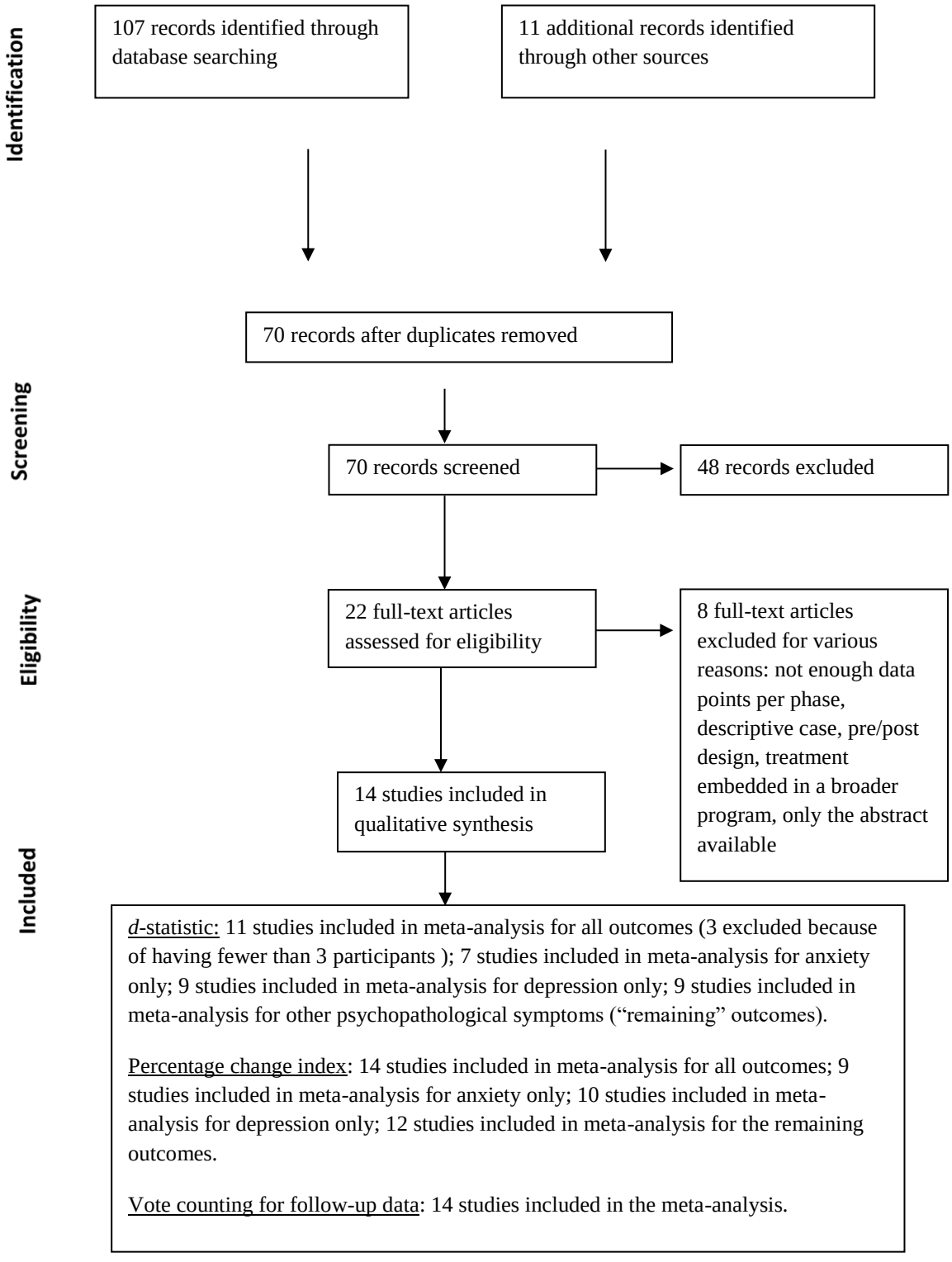
Table 2

Summary of the meta-analyses performed with the *d*-statistic.

Statistics	All outcomes	Anxiety	Depression	Remaining
d-statistic (standard error)	−1.55 (0.20)	−1.41 (0.21)	−1.20 (0.25)	−1.72 (0.24)
d-statistic confidence interval	(−1.99, −1.08)	(−1.93, −0.88)	(−1.76, −0.63)	(−2.27, −1.17)
Heterogeneity descriptive statistics	τ = 0.57,	τ = 0.20,	τ = 0.51,	τ = 0.65,
	I^2 = 81.02%,	I^2 = 14.45%,	I^2 = 55.97%,	I^2 = 76.73%,
	range: −2.58 to −0.60	range: −2.78 to −0.66	range: −2.91 to −0.28	range: −2.74 to −0.67
Heterogeneity test	Q(9) = 61.23, p < .01	Q(6) = 7.72, p = .26	Q(8) = 18.44, p =. 02	Q(8) = 49.23, p < .01

Note. Remaining outcomes include all outcomes except anxiety and depression.

Figure 1. Flowchart of the study selection process according to the PRISMA statement (Moher et al., 2009).



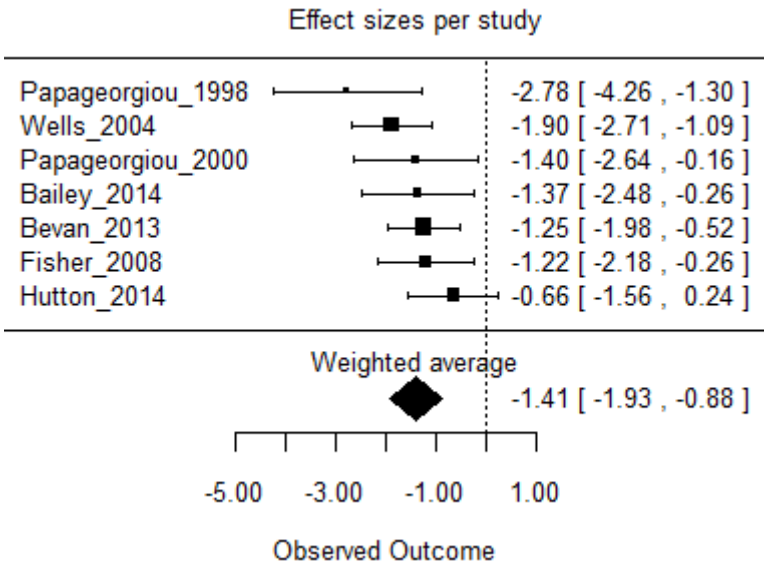


Figure 2. Forest plots for the *d*-statistic for outcomes measuring anxiety.

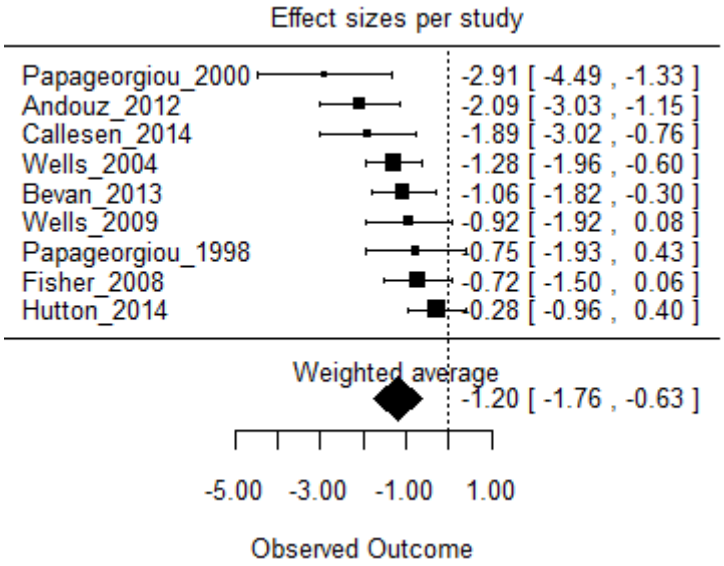


Figure 3. Forest plots for the *d*-statistic for outcomes measuring depression.