

# **LIVING WELL WITH BIPOLAR DISORDER**

INTEGRATING POSITIVE PSYCHOLOGY AND  
PERSONAL RECOVERY INTO THE TREATMENT OF  
BIPOLAR DISORDERS

JANNIS KRAISS

The background of the cover is an abstract composition of layered, organic shapes. At the top, there is a white space. Below it, a light blue horizontal band stretches across the width. To the right, a green shape with a speckled texture rises above the blue band. Below the green, a large brown shape with a mottled texture dominates the middle. At the bottom, a dark blue/black area with a marbled, textured appearance is visible, partially obscured by the brown shape.

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*Jannis Kraiss*

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# **LIVING WELL WITH BIPOLAR DISORDER**

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INTO THE TREATMENT OF BIPOLAR DISORDERS

## DISSERTATION

to obtain

the degree of doctor at the University of Twente,

on the authority of the rector magnificus,

Prof.dr. ir. A. Veldkamp,

on account of the decision of the Doctorate Board,

to be publicly defended

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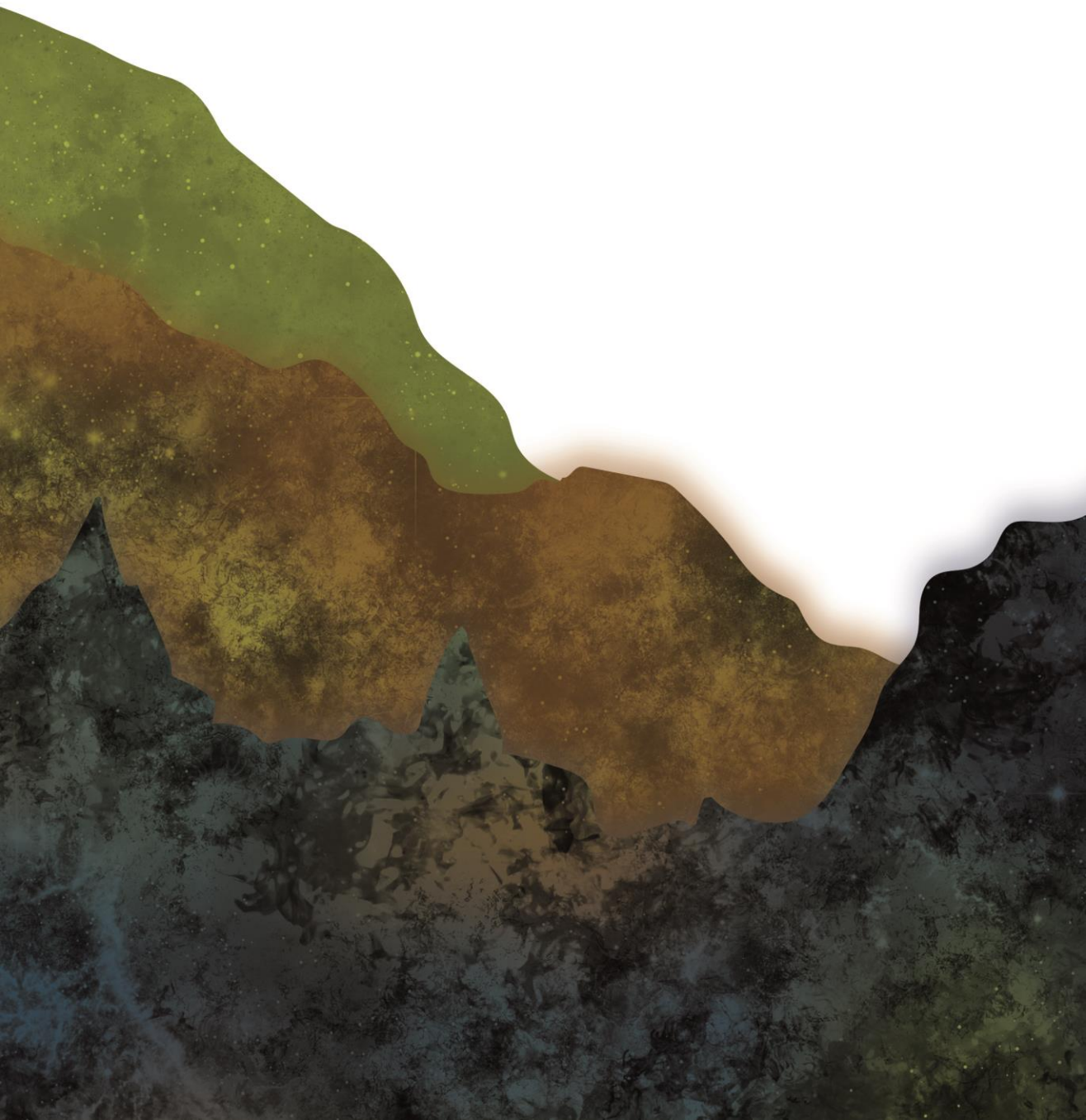
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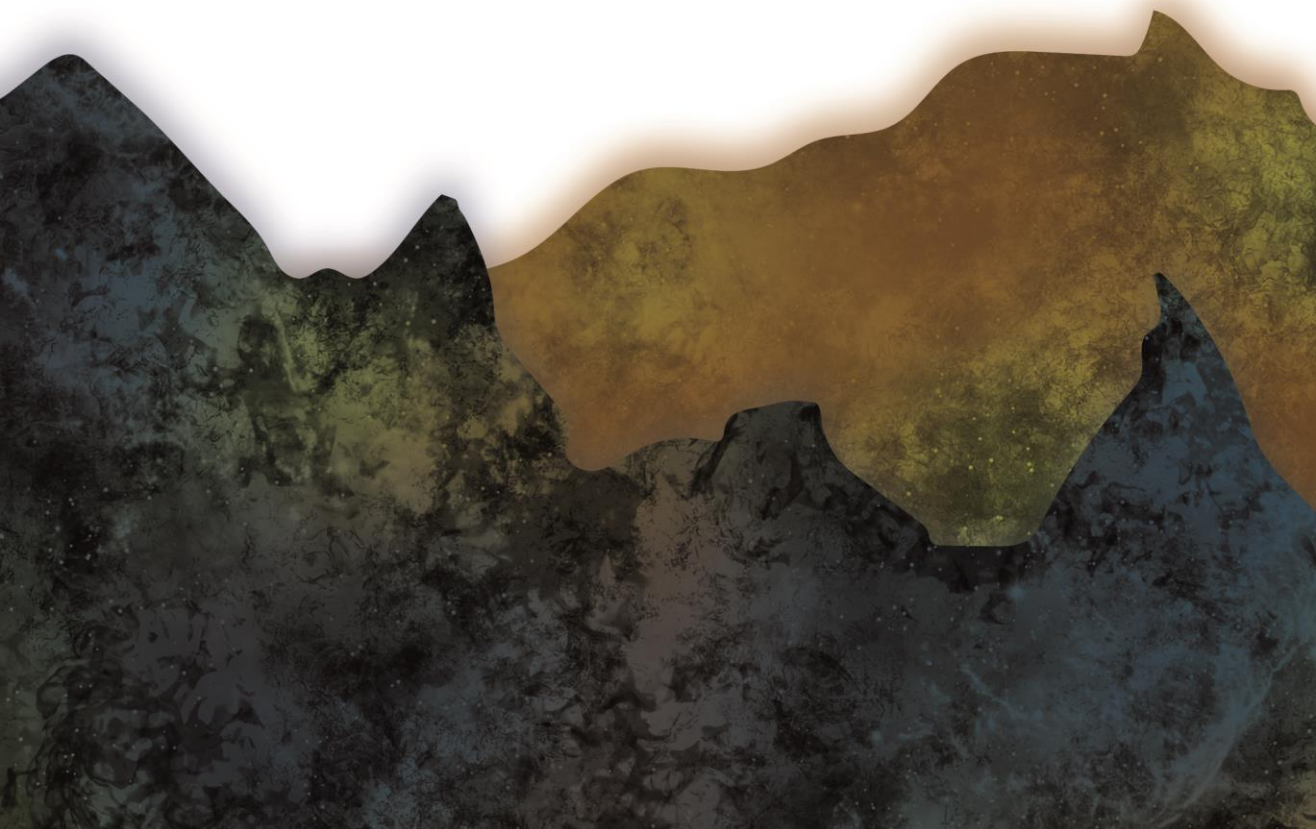
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# CHAPTER 1

General introduction





## BIPOLAR DISORDER

Bipolar disorder (BD) is a severe, lifelong and episodic illness that is characterized by depressive and (hypo)manic episodes. These acute mood episodes are alternated with euthymic states, in which patients are relatively free of symptoms (Goodwin & Jamison, 2007; Grande et al., 2015). The Diagnostic and Statistical Manual of Mental Disorders (DSM-5; American Psychiatric Association, 2013) makes a distinction between bipolar disorder type 1 (BDI), bipolar disorder type 2 (BDII), and several other subtypes. In BDII, an individual has experienced at least one depressive and one hypomanic episode in the course of the illness, but not a full-blown manic episode. In BDI, the person experiences at least one manic episode. Although depressive episodes are not necessary for the diagnosis of BDI, major depressive episodes are common in this group (American Psychiatric Association, 2013). A minority of BDI patients suffer from recurrent unipolar mania (Angst et al., 2019).

In the DSM-5, manic episodes are characterized by two core symptoms: abnormally elevated affect or irritable mood and abnormally and persistently increased goal-directed activity or energy for at least one week, most of the day and almost every day. Furthermore, at least three other manic symptoms need to be present, for example inflated self-esteem, decreased need for sleep, being easily distracted, racing thoughts, or engagement in risky behavior. If the mood is only irritable, at least four of the other manic symptoms have to present. Hypomanic episodes are less severe episodes of mania, in which the symptoms are identical, but are present for only four or fewer days, do not impair everyday functioning as in manic episodes, and are not severe enough to necessitate hospitalization. For depressive episodes, five symptoms of depression need to present for at least two weeks, such as sleeping problems, fatigue or suicidal ideations and at least one of these five symptoms needs to be a depressed mood most of the day or loss of interest or pleasure. Depressive symptoms have to cause clinically significant distress or impairment in social, occupational, or other important areas of functioning. In both depressive and (hypo)manic episodes the symptoms are not attributable to drugs or a medical condition (American Psychiatric Association, 2013; Grande et al., 2015).

### Prevalence and burden

Based on data from the World Mental Health Survey Initiative (Kessler & Ustun, 2008) pooled across 11 western and non-western countries, the worldwide lifetime prevalence was estimated at 0.6% for BDI and 0.4% for BDII (Merikangas et al., 2011). In the Netherlands, the lifetime and 12-month prevalence of BD was estimated at 1.3% and 0.8%, respectively (de Graaf et al., 2010). BD often manifests in early adulthood, indicating an early onset of the disorder. In terms of gender, and in contrast to unipolar major depressive disorder, BD affects

women only slightly more than men (de Graaf et al., 2010; Ferrari et al., 2016). Due to its early onset, chronic course and severe symptoms, BD is associated with substantial consequences for the individual and the society. Dilsaver (2011) estimated the societal costs of BD, including direct and indirect costs, at 151 billion dollars per year in the United States in 2009. A more recent study estimated the societal costs of BDI at around 202 billion dollars per year in 2015 in the United States (Cloutier et al., 2018). The total per annum costs of BD in 2002 were estimated at 1.83 billion US-dollars in the Netherlands. Most of these costs were associated with indirect costs, i.e. productivity loss (Hakkaart-van Roijen et al., 2004). In 2013, BD was the fifth leading cause of disability adjusted life years among all mental and substance abuse disorders (Ferrari et al., 2016). BD carries the highest risk of suicide among all affective disorders (Gonda et al., 2012), with suicide risk being up to 10-fold higher than in the general population (Crump et al., 2013). Furthermore, BD is associated with substantial impairments in cognitive functioning (Cotrena et al., 2016), quality of life (IsHak et al., 2012; Saarni et al., 2010), increased work-related issues (Fajutrao et al., 2009; Laxman et al., 2008), and high caregiver burden (Miller et al., 2014; Pompili et al., 2014).

## **Prognosis**

The natural course of BD includes periods of remission, but recurrent depressive or (hypo)manic episodes and subthreshold symptoms are common. Although BD has traditionally been thought of as an episodic illness, evidence suggesting that BD represents a chronic disorder and that residual subsyndromal symptoms often persist between episodes is accumulating. In a 15-year follow-up study of patients with BDI (Judd et al., 2002) and BDII (Judd et al., 2003), participants were symptomatically ill for almost half of the study period. Patients with BDI showed depressive symptoms for about one third of the study period and manic/hypomanic symptoms for about 9% of the time. In patients with BDII, depressive symptoms were more common compared to patients with BDI (50% of the time), while hypomanic symptoms were only present for about 1% of the study period. The amount of subsyndromal states was three times higher than the number of syndromal episodes (Judd et al., 2003; Judd et al., 2002). Residual symptoms after depressive episodes have been shown to increase the risk for relapse, suggesting that residual symptoms form an important risk factor for the recurrence of acute episodes (Judd et al., 2008).

## **Current treatment**

Several national and international guidelines have been published for the treatment of BD (Goodwin et al., 2016; Grunze et al., 2018; Kupka et al., 2015; National Institute for Health and Care Excellence [NICE], 2018; Yatham et al., 2018). In general, a distinction is made between treatment of acute episodes and long-term maintenance management of the disorder (Grande

et al., 2015). The primary goal of acute treatment is to ensure safety of the patient and achieve symptomatic remission. In this phase, pharmacotherapy is essential, with mood stabilizers and antipsychotics being the preferred choice of treatment (Grande & Vieta, 2015). In addition, electroconvulsive therapy has been shown to be effective for severe depression and treatment-resistant mania (Schoeyen et al., 2015). The main goal of maintenance treatment is to prevent recurrence of mood episodes, and to increase psychosocial functioning. The current approach to long-term management usually encompasses a combination of pharmacotherapy as well as supportive and psychological treatment. Lithium has been shown to be one of the most effective treatments for the prevention of recurrence during maintenance treatment (Miura et al., 2014). Effective psychological approaches include psychoeducation (Colom et al., 2003), cognitive behavioral therapy (Scott et al., 2006), interpersonal and social rhythm therapy (Frank et al., 1991) and family-focused therapy (Miklowitz et al., 2008). A recent meta-analysis suggests that individual psychological interventions led to fewer recurrences at posttest and follow-up compared to treatment as usual (Oud et al., 2016). Treatment in general is long-lasting and can shift rapidly depending on the course of symptoms and the phase of illness.

In the Netherlands, the guidelines for treatment for patients with BD (Kupka et al., 2015) are similar to international guidelines. During acute phases, treatment consists of pharmacotherapy and supportive treatment, while in maintenance phases pharmacotherapy in combination with enhancement of self-management skills (e.g. keeping a life chart), supportive sessions and psychoeducation is applied to improve symptoms and daily functioning and to prevent recurrences (Kupka et al., 2015). Currently, psychological treatments for BD have a primary focus on the reduction of symptoms and prevention of future mood episodes, and improvement of functioning related to disorder-specific impairments.

## PERSONAL RECOVERY

It is increasingly recognized that recovery from mental illness encompasses more than the alleviation of symptoms and restoration of functioning (Fava et al., 2007; Slade, 2009, 2010). An important component of recovery is personal recovery. Anthony (1993) defines personal recovery as *“a deeply personal, unique process of changing one’s attitudes, values, feelings, goals skills and/or roles [...] and a way of living a satisfying, hopeful and contributing life, even with the limitations caused by the illness”* (Anthony, 1993, p. 527). This definition implies that personal recovery is not complete merely because symptoms are reduced or under control (Macpherson et al., 2016). Personal recovery moves away from traditional clinical preoccupations, including managing current symptoms and avoiding future recurrence, and emphasizes personal goals and development (Slade, 2009). Furthermore, personal recovery is an individual process, which also means that the person affected by mental illness himself

determines what recovery means. The personal recovery movement has emerged from the lived experience of people with mental disorders, who highly value outcomes of personal recovery in treatment and for their recovery from mental illness (de Vos et al., 2017; Mead & Copeland, 2000; Pitt et al., 2007).

The initial definition of personal recovery remains relatively broad and lacks clarity on what personal recovery concretely means (Braslow, 2013; Le Boutillier et al., 2011). To further define and conceptualize personal recovery, Leamy et al. (2011) developed a conceptual framework of personal recovery from mental illness. They systematically searched the literature for studies that contained either a conceptualization of personal recovery or presented an original model or framework of recovery. Based on the synthesis of 97 studies, they identified several characteristics relevant for the recovery process. The most frequently named characteristics were that recovery is an active, individual, unique, and non-linear process and that recovery includes several stages, and is a struggle and a life-changing experience. Furthermore, they identified five core categories of personal recovery: connectedness, hope and optimism, identity, meaning in life, and empowerment. These five categories were comprised within the CHIME-framework, given by the first letters of these five recovery categories. Later, the validity and relevance has been tested using focus groups of people with mental disorders (Bird et al., 2014), who had to indicate whether they found the CHIME-framework meaningful and useful in the context of their own recovery experience. In general, they concluded that the main categories of the CHIME-framework adequately reflect the processes they find relevant for recovery (Bird et al., 2014).

Personal recovery is increasingly implemented as a guiding vision in mental health agendas and plans across the globe, for example in Australia (Department of Health and Ageing, 2009), Canada, and the United Kingdom (Department of Health and Social Care, 2011). Furthermore, several self-report questionnaires have been developed to assess the process of personal recovery. A systematic review by Shanks et al. (2013) identified 13 questionnaires measuring personal recovery, for example the Questionnaire about the Process of Recovery (QPR) (Neil et al., 2009), Maryland Assessment of Recovery (Drapalski et al., 2012), or the Recovery Assessment Scale (Corrigan et al., 1999). The QPR was the only questionnaire of which all items map to the five dimensions of the CHIME-framework (Shanks et al., 2013). However, the QPR has not yet been translated into Dutch nor has it been validated in persons with BD.

## **THE IMPORTANCE OF MENTAL WELL-BEING**

The concept of personal recovery aligns well with the growing interest in mental well-being as an important outcome of treatment of mental disorders. In this context, mental health is

increasingly considered as not merely the absence of psychopathology, but also the presence of mental well-being (Huppert, 2004, 2009; Keyes, 2005; Lamers et al., 2015). In this context, psychopathology and mental well-being are seen as two related, yet distinctive components of mental health (Keyes, 2005). Therefore, both should be considered when aiming to improve mental health (De Vos et al., 2018; Franken et al., 2018). According to Keyes (2005), mental well-being includes three different domains: emotional, social, and psychological well-being. Emotional well-being refers to the presence of positive affect, absence of negative emotions, and satisfaction with life (Diener & Ryan, 2009). Social well-being relates to an individual's functioning in community and social roles (Keyes, 1998). Psychological well-being relates to personal growth and self-actualization and includes processes such as self-acceptance, autonomy, positive relationships, and environmental mastery (Ryff, 2014; Ryff & Keyes, 1995). Similarly, mental well-being can be subdivided into hedonic and eudaimonic well-being. Hedonic well-being relates to the subjective experience of pleasure, positive emotions, and satisfaction with life, while eudaimonic well-being can be described as living a life of virtue and striving for human excellence and self-actualization (Deci & Ryan, 2008; Ryan & Deci, 2001). High levels of mental well-being are classified as flourishing, while low levels of mental well-being are described as languishing (Keyes, 2002).

Previous research emphasizes the importance of mental well-being for mental health. In a large longitudinal study, people with low levels of mental well-being at baseline were two times more likely to be depressed ten years later (Wood & Joseph, 2010), even after controlling for other factors, such as demographic variables, personality, or prior depression. Furthermore, the presence of mental well-being has been shown to serve as buffer against the onset and recurrence of mental disorders (Keyes et al., 2010; Schotanus-Dijkstra et al., 2017). Also, higher levels of mental well-being have been shown to increase the likelihood of recovery from mental illness (Iasiello et al., 2019; Schotanus-Dijkstra et al., 2019) and have been shown to be related with better physical health outcomes (Howell et al., 2007) and longevity in both healthy and physically ill people (Chida & Steptoe, 2008; Lamers et al., 2012). Results from cross-sectional studies suggest that higher mental well-being is related to reduced suicidality in college students (Keyes et al., 2012), improved work performance, and fewer physical complaints (Hone et al., 2015).

## **POSITIVE PSYCHOLOGY**

One field of psychology that specifically focuses on the improvement of mental well-being and personal recovery is positive psychology (Resnick & Rosenheck, 2006; Slade, 2010). In the last two decades, positive psychology emerged as a response to the predominant focus of clinical psychology on dysfunctioning and the negative aspects of mental functioning. Instead of



repairing the worst, positive psychology pursues to foster the things that make life worth living (Seligman, 2011; Seligman & Csikszentmihalyi, 2014; Seligman et al., 2006). Positive psychology interventions aim to improve positive behavior, cognitions, or emotions (Sin & Lyubomirsky, 2009). Various positive interventions have been developed, including gratitude, savoring, acts of kindness, strength-based, compassion or optimism exercises, or combined multicomponent interventions containing several exercises (Sin & Lyubomirsky, 2009). Both specific and multicomponent positive psychology interventions have been shown to be effective in improving mental well-being in the general public and clinical groups with psychiatric and somatic disorders (Bolier et al., 2013; Chakhssi et al., 2018; Geerling et al., 2020; Hendriks et al., 2020). The growing popularity of positive psychology has also led to the development of psychotherapeutic approaches that incorporate concepts of positive psychology (Rashid, 2015; Seligman et al., 2006). In the Netherlands, scholars also argue for an integration of mental well-being into psychological treatment when aiming for sustainable mental health (Bohlmeijer & Westerhof, 2020).

Interestingly, the concepts of mental well-being (Keyes, 2005) and personal recovery (Anthony, 1993; Leamy et al., 2011) share substantial overlap (Slade, 2010). Although both concepts evolved relatively independent, they similarly focus on aspects that make life meaningful and worth living. Furthermore, both concepts move beyond predispositions of clinical psychology that recovery and mental health should solely be defined as the absence of disease symptoms. Considering the overlap between mental well-being and personal recovery as well as the effectiveness of positive psychology interventions to improve mental well-being, one can reasonably assume that positive psychology might be an adequate approach to not only improve mental well-being in BD, but also enhance personal recovery (Resnick & Rosenheck, 2006; Slade, 2010).

## **POSITIVE EMOTION REGULATION**

Other processes that might also be relevant in people with BD in the context of mental well-being and recovery relate to emotion regulation. Emotion regulation is described as the way in which people modify their emotions unconsciously or consciously in order to achieve a certain goal (Gross, 1998). The field of emotion regulation has predominantly examined the role of responses to *negative* affect for mental health, for example rumination (Nolen-Hoeksema et al., 2008) or avoidance (Chawla & Ostafin, 2007; Hayes et al., 1996). However, accumulating evidence suggests that the way in which people alter and react to their *positive* affective states is similarly relevant for mental health (Carl et al., 2013; Quoidbach et al., 2010; Wood et al., 2003).

Two examples of responses to positive affect are dampening and positive rumination. Dampening has been described as the suppression of positive affect with the goal to decrease positive emotions (Quoidbach et al., 2010). Positive rumination is the tendency to react to positive emotions with recurrent thoughts about positive experiences. Regarding positive rumination, a distinction can be made between self-focused and emotion-focused positive rumination. While self-focused positive rumination describes ruminating about one's own positive qualities and strengths, emotion-focused is characterized by ruminating about positive feelings and somatic experiences (Feldman et al., 2008). To assess responses to positive affect, the Responses to Positive Affect questionnaire (RPA; Feldman et al., 2008) has been developed and also translated and validated in a Dutch-speaking Belgian community sample (Raes et al., 2010). Studies suggest that responses to positive affect might play an important role in persons with BD (Edge et al., 2013; Gilbert et al., 2013; Gruber et al., 2011; Johnson & Jones, 2009; Johnson et al., 2008; Johnson et al., 2016). However, the role of dampening and positive rumination and its relation to mental well-being and personal recovery has not yet been investigated. Also, the Response of Positive Affect Questionnaire has not yet been validated in individuals with BD.

## **PERSONAL RECOVERY AND POSITIVE PSYCHOLOGY INTERVENTIONS FOR BIPOLAR DISORDER: CURRENT EVIDENCE**

A few studies have recently been conducted that examined the effect of interventions aimed at personal recovery or mental well-being related outcomes in BD. Jones et al. (2015) conducted a pilot randomized controlled trial ( $N = 67$ ) to evaluate the feasibility and effectiveness of recovery-focused cognitive behavioral therapy compared with care as usual for people with BD. They found a significant between-group effect for personal recovery at posttest (Cohen's  $d = 0.62$ ), but no significant effects on outcomes of current mood symptoms. However, participants in the intervention group showed longer time to recurrence of a depressive or manic episode. Todd et al. (2014) conducted a feasibility randomized controlled trial ( $N = 122$ ) comparing a web-based self-management intervention for BD as an adjunct to care as usual with care as usual only. The intervention resulted in improved personal recovery and other outcomes, including physical and psychological quality of life, depression and social functioning. The significant effects were maintained until 6-month follow-up (Todd et al., 2014). Other studies did not specifically use personal recovery as outcome, but examined the effect of interventions on mental well-being related outcomes. Deckersbach et al. (2012) investigated the effect of Mindfulness-Based Cognitive Therapy in 12 euthymic BD patients in a noncontrolled pilot study and found significant improvements in positive affect and other

facets of mental well-being at posttest, including environmental mastery, purpose in life, self-acceptance, and positive relations (effect sizes ranging between 0.59 and 1.32). Another study investigated the effect of dialectical behavior therapy group skill training in a noncontrolled proof-of-concept study ( $N = 37$ ). They also found significant effects on psychological well-being at posttest ( $d = 1.02$ ) (Eisner et al., 2017).

Although some studies have examined the effect of psychological interventions in improving mental well-being or personal recovery, studies specifically investigating the effect of positive psychology interventions for BD are scarce. In a recent systematic review and meta-analysis of positive psychology interventions in severe mental illness, merely two pilot studies were identified that examined the effect of a positive psychology intervention for patients with BD (Geerling et al., 2020). One of the studies conducted by Painter et al. (2019) reported preliminary outcomes of a nine-week positive psychology group intervention in a noncontrolled pilot study ( $N = 16$ ). The described intervention specifically focused on the improvement of positive affect and positive emotion regulation strategies in patients with BD. Results showed significant improvements in positive and negative affect and emotion regulation skills from pre- to posttest. Participants rated the sessions as generally helpful, with scores ranging from 3 to 4.9 on a scale from 1 (not at all helpful) to 5 (extremely helpful). The other study (Celano et al., 2020) was a randomized pilot trial ( $N = 25$ ) with hospitalized BD patients. Participants in the intervention condition completed weekly positive psychology exercises for the intervention period of four weeks (e.g., gratitude letter, acts of kindness), while participants in the control condition were asked to recall three neutral events each day. In addition, both the intervention and control group received weekly phone calls with coaches. Significant between-group effects on optimism were found at posttest ( $d = 1.24$ ) and follow-up four weeks after the intervention ( $d = 1.10$ ). In addition, nonsignificant large effects were found for positive affect ( $d = 1.16$ ) and gratitude at posttest ( $d = 0.68$ ). The intervention was generally rated as helpful and feasible (Celano et al., 2020). Taken together, studies investigating the effect of interventions to improve personal recovery and mental well-being in BD remain scarce and have mainly used weak methodological designs or were not adequately powered. The potential of positive psychology as an additional psychological treatment modality to improve mental well-being and personal recovery in BD remains widely unknown and studies that investigated positive psychology interventions for this target group did not include personal recovery as an outcome and were only pilot studies.

## **The potential of positive psychology to improve mental well-being and personal recovery in bipolar disorder**

Prior positive psychology studies seem promising and their findings suggest that personal recovery and mental well-being could also be improved in this target group. For several reasons, positive psychology interventions may be particularly suitable to improve mental well-being and personal recovery in euthymic BD patients. Positive psychology exercises are relatively easy to complete and can be conducted individually or in groups. Therapists also only need minimal training to provide such interventions. Therefore, a positive psychology treatment could represent a simple, cost-effective addition to current treatment for euthymic BD patients. Considering that BD often has a chronic course, patients are also faced with substantial adaptive tasks. Consequently, learning to lead a good life despite the limitations caused by the illness remains an important, but also difficult to accomplish goal (Anthony, 1993; Leamy et al., 2011). Specifically enhancing personal resources such as self-compassion, positive emotions, optimism, strengths and positive relationships may support people with BD to reach this goal. These resources may provide crucial adaptive strategies that help to cope with challenges and limitations caused by the illness and to still lead a meaningful life (Bohlmeijer & Westerhof, 2020). Furthermore, current treatment for BD primarily focuses on functional and symptomatic recovery (Kupka et al., 2015). However, patients with severe mental disorders, such as BD, express dissatisfaction with current treatment outcomes (Mead & Copeland, 2000) and find outcomes related to mental well-being and personal recovery important (de Vos et al., 2017; Pitt et al., 2007). Clinical recovery alone misrepresents recovery for patients with BD (Mezes et al., 2020) and recovery for people with BD does not only mean being symptom-free (Todd et al., 2012). Therefore, a positive psychology treatment that focuses on personal recovery could be a valuable addition to current treatment for BD, both from a societal and care provider perspective, as well as from the perspective of patients.

### **AIM AND OUTLINE OF THIS THESIS**

The overall goal of this thesis is to contribute to the integration of positive psychology and personal recovery into the treatment of BD. For this, four aims are addressed in this thesis. First, an overview of the research field of positive psychology interventions for serious mental illness (SMI) and of economic studies for BD is given, aiming to summarize the current state of the art of positive psychology interventions for SMI and cost-effectiveness of non-pharmacological interventions for BD. This is done by two systematic reviews of the literature and a meta-analysis. Second, two measurement instruments of personal recovery and positive emotion regulation are psychometrically evaluated, namely the Questionnaire about the Process of Recovery (QPR) and the Responses to Positive Affect questionnaire (RPA). These

instruments assess the processes of personal recovery and positive emotion regulation, which both might constitute relevant outcomes in BD. Yet, these measurement instruments were not translated into Dutch before and/or have not been validated in people with BD. Third, this thesis aims to expand our knowledge of what contributes to personal recovery in BD, by exploring factors that are associated with personal recovery. Fourth, a novel positive psychology group treatment is developed aimed at improving personal recovery and mental well-being in euthymic BD patients and its effectiveness is evaluated in a pragmatic randomized controlled trial (RCT). Accordingly, the following four goals are addressed in this thesis:

- (1) To provide an overview of the research field regarding positive psychology interventions for patients with severe mental illness and economic studies in BD.
- (2) To examine the psychometric properties of a Dutch translation of the Questionnaire about the Process of Recovery (QPR) and the Responses to Positive Affect questionnaire (RPA) in people with BD.
- (3) To explore factors associated with personal recovery in BD.
- (4) To develop a specific positive psychology intervention for euthymic persons with BD aimed at mental well-being and personal recovery and evaluate its effectiveness in a pragmatic, multicenter randomized controlled trial.

These four goals will be addressed in four different parts containing seven chapters:

## **Part I: Overview of the research field**

The two chapters described in this part address the first goal of this thesis. These chapters provide an overview of the research field of positive psychology interventions for people with severe mental illness and economic studies in BD.

*Chapter 2* describes a systematic review and meta-analysis on positive psychological interventions for people with SMI. A systematic literature search was conducted for quantitative studies examining the effect of positive psychology interventions on mental well-being and psychopathology in SMI. Data on population, study, and intervention characteristics were extracted from the studies and summarized. Effects of the interventions on mental well-being and psychopathology were synthesized using meta-analytical techniques.

*Chapter 3* describes a systematic review on economic studies in BD. The literature was searched for both economic evaluations as well as cost-of-illness studies examining the cost-effectiveness of non-pharmacological interventions or costs associated with BD. Evidence on

the cost-effectiveness of psychological treatments for BD was summarized. In addition, recent cost-of-illness studies were summarized to derive information on the direct and indirect costs associated with BD.

## **Part II: Measurement of relevant constructs**

The two chapters described in this part address the second goal of this thesis. These chapters focus on psychometric evaluations of two widely used questionnaires for personal recovery and positive emotion regulation in people with BD.

*Chapter 4* describes a psychometric evaluation of a Dutch translation of the Questionnaire about the Process of Recovery (QPR) in a cross-sectional sample of people with BD. The QPR is a widely used measure of personal recovery. The factor structure, internal consistency and incremental validity of the QPR was examined. Furthermore, associations of personal recovery with social role participation and mental well-being were explored.

*Chapter 5* describes a psychometric evaluation of the Dutch Responses to Positive Affect questionnaire (RPA) in a cross-sectional sample of people with BD. Associations of dampening and positive rumination with mental well-being, personal recovery and symptomatology were examined. In addition, the assumed underlying three-factor structure of the RPA was validated and the internal consistency and incremental validity of the questionnaire was evaluated.

## **Part III: Determinants of personal recovery**

The chapter described in this part addresses the third goal of this thesis. *Chapter 6* describes an exploration of factors associated with personal recovery in BD. Using a split-sample regression approach, a parsimonious multiple regression model was built with personal recovery as dependent variable. The ability of the model to explain personal recovery was examined. Potentially relevant independent variables included sociodemographic variables, social role participation, psychopathology and positive emotion regulation.

## **Part IV: Intervention development and evaluation**

The two chapters described in this part address the fourth and final goal of this thesis. These chapters focus on the development and evaluation of a multicomponent positive psychology group intervention to enhance mental well-being and personal recovery in euthymic patients with BD.

*Chapter 7* describes the study protocol of the pragmatic randomized controlled trial. The protocol contains the rationale and design of the study, a description of the outcome and process measures and the intervention development and specific content of the intervention. Potential promises and limitations of the study and intervention were also discussed beforehand.

*Chapter 8* describes the effectiveness of the multicomponent positive psychology group intervention in patients with BD. Effects of the intervention on outcomes of mental well-being, personal recovery, psychopathology, and social role participation as well as positive emotions, self-compassion and positive relationships were examined. The effects were examined until 6-month follow-up.

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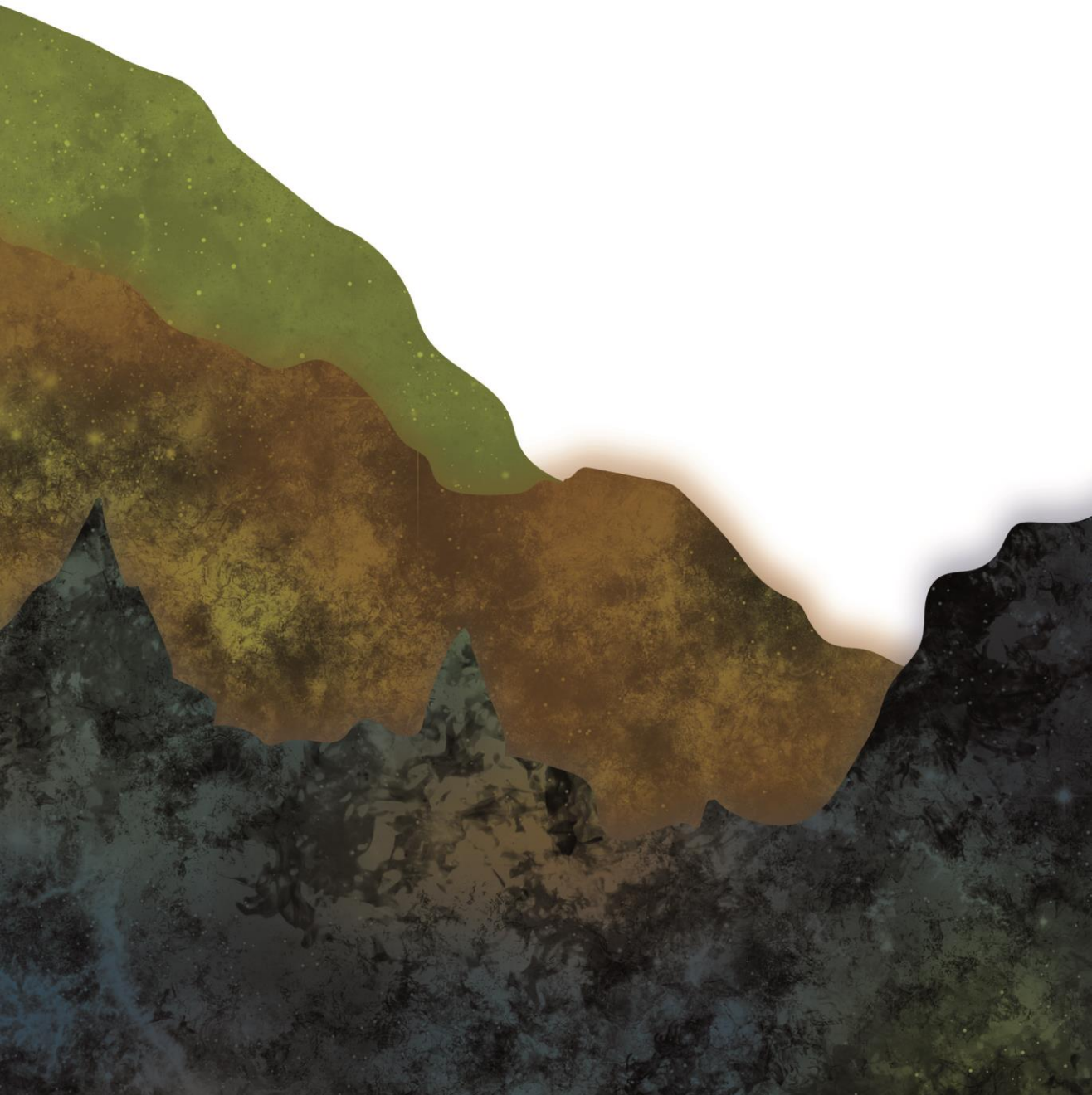
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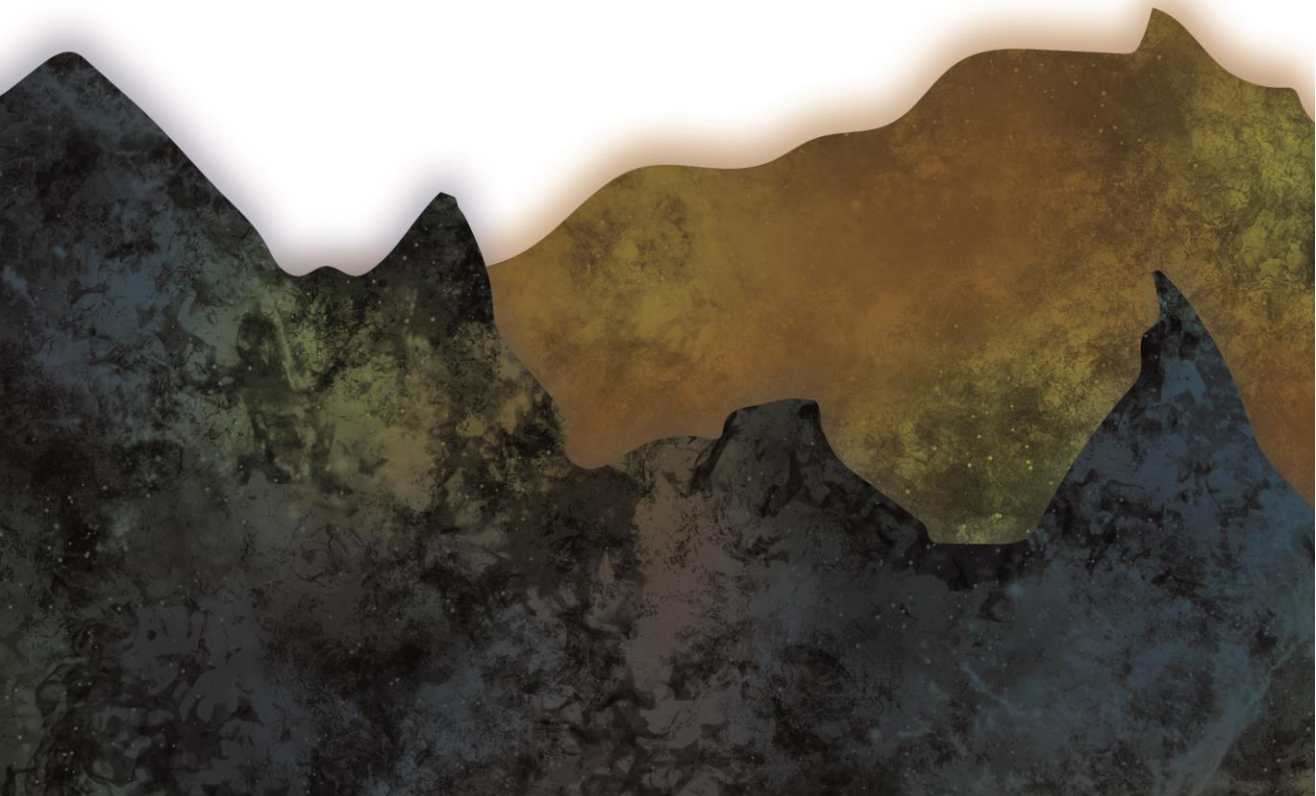
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# CHAPTER 2

The effect of positive psychology interventions on well-being and psychopathology in patients with severe mental illness: a systematic review and meta-analysis

Geerling, B., Kraiss, J. T., Kelders, S. M., Stevens, A. W. M. M., Kupka, R. W., & Bohlmeijer, E. T. (2020). The effect of positive psychology interventions on well-being and psychopathology in patients with severe mental illness: A systematic review and meta-analysis. *The Journal of Positive Psychology*, 15(5), 572-587. <https://doi.org/10.1080/17439760.2020.1789695>



## ABSTRACT

There is a growing number of studies evaluating the impact of positive psychology interventions (PPIs) in people with severe mental illnesses (SMIs). The results of these studies have not been quantitatively reviewed yet. In this study, we conducted a meta-analysis on the effects of PPIs on mental health in people with SMI across studies. Sixteen studies are included (nine RCTs), representing 729 patients. Meta-analyses were performed for well-being as the primary outcome at post-treatment. We found no significant effects on both well-being and psychopathology for PPIs in comparison with the control conditions. Within-group effects revealed a moderate effect (Hedge's  $g = 0.40$ ) on well-being and a large effect on psychopathology ( $g = 0.70$ ). Though there is at present no evidence that PPIs are more effective in comparison with other active interventions, our findings demonstrated that people with SMI do benefit from PPIs in terms of enhancement of mental health.

## INTRODUCTION

In an overview of the global burden of diseases, severe mental illnesses (SMIs) have a predominant place (Vos et al., 2015). The National Institute of Mental Health (NIMH) defines severe (or serious) mental illness (hereafter SMI) as “*a diagnosis of non-organic psychosis or personality disorder; duration characterized as involving prolonged illness and long-term treatment, operationalized as a two-year or longer history of mental illness or treatment*” (NIMH, 1987). This three-dimensional model (psychosis, chronic illness and long-term treatment) has been transformed into a two-dimensional model to also include nonpsychotic disorders (Ruggeri et al., 2000). Delespaul (2013) added the following criteria to define SMIs more strictly: a psychiatric disorder with severe functional problems, where the constraints are causal and consequential and which is not temporary, and there is a need for coordinated professional care (Delespaul, 2013). Examples of SMIs based on these criteria are schizophrenia, schizoaffective disorder, bipolar disorder (BD), personality disorder and major depressive disorders (MDD).

The burden of SMIs is extensive. For example, people with schizophrenia have a lower life expectancy compared to the average population (Hjorthøj et al., 2017), and the suicide rates are significantly higher for people with schizophrenia (20% – 50%) than for the general population (9% – 13%) (Pinikahana et al., 2003). As another example, the burden of BD is great, for both patients and caregivers (Erten et al., 2014; Vieta et al., 2013). BD contains a high risk of suicide compared to the general international population (Baldessarini & Tondo, 2003) and is associated with poor clinical and functional outcomes (Goodwin & Jamison, 2007). People with SMI also experience significantly less hope than the general population (Landein, 2000; Landein et al., 2000). Lower levels of hope are closely related to a lower quality of life (Hasson-Ohayon et al., 2009). In spite of this major negative impact on quality of life, evidence-based treatments have mainly focused on the reduction of symptoms and, to a lesser extent, on functional recovery and the improvement of well-being (Bensing, 2000).

Besides reduction and management of symptoms, there are also important personal recovery challenges and adaptive tasks for patients with SMI (Slade, 2010). Anthony (1993) describes personal recovery as “*a deeply personal, unique process of changing one’s attitudes, values, feelings, goals, skills, and/or roles*” and “*a way of living a satisfying, hopeful, and contributing life even within the limitations caused by illness*” (p. 527). With regard to well-being and personal recovery, many people with SMI have unmet needs (Goossens et al., 2007; Wiersma, 2006). Moreover, patients with SMI express dissatisfaction with current treatment approaches such as dire predictions about the course of the illness and medication as the only rational option for treatment and ‘learned helplessness’ (Malmström et al., 2016; Mead & Copeland,

2000). Patients also argue for the importance of well-being-related outcomes, such as meaningful activities, self-efficacy improvement, and reduced reliance on support networks. This underlines the need for a shift towards more well-being-oriented care (Bernstein, 2006; Fava et al., 2007; Malmström et al., 2016).

In recent years, there has been a growing interest in the potential benefits of positive psychology interventions (PPIs) for people with SMI (Parker et al., 2012; Russell & Moss, 2013). PPIs focus on the enhancement of positive feelings, behaviors, or cognitions (Sin & Lyubomirsky, 2009). Parks and Biswas-Diener (2013) underscored that evidence of effectiveness is an additional important criterion of a PPI (Parks & Biswas-Diener, 2013). For patients with SMI, PPIs aim to develop resources that are supportive in realizing personal recovery and well-being and in managing symptomatology (Slade, 2010; Slade et al., 2014).

Recent meta-analyses have found that PPIs have small to moderate significant effects on well-being and distress both in general populations (Bolier et al., 2013; Hendriks et al., 2020; Sin & Lyubomirsky, 2009) and in patients with mental or somatic illnesses (Chakhssi et al., 2018). Furthermore, a growing number of studies have examined the effects of PPIs on the mental health of people with SMI. However, evidence of their effectiveness has not been synthesized to date. In the context of this special issue of JOPP on the state-of-the-art of positive psychology, the aim of this study is, therefore, to make an initial assessment on the effects of PPIs in people with SMI across studies.

## **METHOD**

This systematic review and meta-analysis was conducted according to the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guidelines (Liberati et al., 2009; Moher et al., 2015).

### **Search strategy**

A systematic literature search was carried out in Scopus, PubMed, and PsycINFO. The initial search was conducted from 1998 (the start of the positive psychology movement) to 2017. We updated the search on January 8<sup>th</sup>, 2020. The search string consisted of terms referring to well-being and positive psychology constructs, which were combined with terms referring to SMI (Appendix A). In addition, we cross-checked included studies in prior meta-analyses and reviews conducted in this research field (Bolier et al., 2013; Chakhssi et al., 2018; Rashid, 2009). The search was restricted to peer-reviewed studies in the English language.

## Study selection

The search was aimed at identifying studies that focused on PPIs; it included a clinical sample with an SMI, and used a well-being outcome measure. Therefore, studies were included if they (1) examined the effects of an intervention aimed at raising positive feelings, positive cognitions or positive behavior (Sin & Lyubomirsky, 2009), (2) included adult participants (18 years or older), (3) included participants with one or more of the following DSM-IV or ICD-10 diagnoses: bipolar disorder, MDD, schizophrenia, psychosis, schizoaffective disorder or personality disorders, and (4) used outcome measures of well-being or related outcomes.

To explore the potential benefits of PPIs in people with SMI, we decided to include studies with various designs, including randomized controlled trials, quasi-experimental, and uncontrolled pre- post-test designs. We also included studies investigating the effect of interventions, which did not specifically call themselves positive psychology interventions but were also aimed at raising positive feelings, cognitions, and behavior (e.g., recovery-focused or compassion interventions). Articles were excluded if they (1) were not published in an English language peer-reviewed journal, or (2) studied an intervention consisting of physical exercises, or (3) concerned qualitative studies, or (4) concerned abstracts or study protocols.

Potentially eligible studies were screened based on the title in the first phase, on abstract in the second phase, and on full papers in the third phase. Two reviewers (BG & JK) selected the titles independently. The interrater-reliability was satisfactory (Cohen's kappa = 0.76,  $N = 2,189$ ). Abstracts and full texts were rated by the same reviewers, and disagreements were discussed until consensus was reached. The remaining issues were reviewed with the third and sixth authors (SK and EB).

## Data extraction

Population characteristics (age, sex, marital status and disorder), characteristics of the studies (design, sample size, measurements, controls and control intervention) and characteristics of the interventions (type of intervention, duration and additional treatment) were extracted. Five authors were contacted due to missing information, and four of them provided additional data on request.

## Primary and secondary outcomes

The main outcome of the meta-analysis was well-being at the end of treatment. If more than one measure of well-being was used, all measures related to well-being were combined.



Psychopathology was the secondary outcome, which included depression, mania, psychosis, negative symptoms and general symptom inventories.

## Meta-analytical procedures

For all included studies, the number of participants and means and standard deviations of outcomes of well-being and psychopathology was extracted. If possible, values were extracted based on the intention-to-treat principle, and if not, values were extracted based on a per-protocol basis. For all included studies, within-group effect sizes for the PPI groups were calculated by subtracting average scores at pre-treatment from average scores at post-treatment and dividing it through the standard deviation of the difference. For studies containing a control group, additional controlled effect sizes were computed by subtracting average scores at pre-treatment from average scores at post-treatment and dividing it through the pooled standard deviation. The difference in effect size was calculated by subtracting the effect size of the treatment group from the effect size of the control group.

Effect sizes were further analyzed in Comprehensive Meta-Analysis (CMA), version 2.2.064. If the correlation between pre-treatment and post-treatment scores was not provided in published papers, we followed recommendations by (Rosenthal, 1986) and used a conservative estimation of  $r = .7$  (Rosenthal, 1986). Since studies included in the meta-analysis were not functionally identical, effect sizes were pooled in CMA using a random-effects model instead of a fixed-effects model (Hedges & Vevea, 1998; Moses et al., 2002). As an indicator of effect sizes, Hedge's  $g$  and 95% confidence intervals were used. Hedge's  $g$  deals with bias caused by small sample size (Cumming, 2013).

To assess the effect of PPIs on well-being and psychopathology, separate meta-analyses were performed for outcomes of well-being and psychopathology for both within-group effect sizes and controlled effect sizes. In addition, exploratory subgroup analyses based on within-group effect sizes were conducted based on the following criteria: 1.) duration of treatment; short (< 8 weeks) vs. long ( $\geq 8$  weeks), 2.) treatment format; group vs. individual therapy, and 3.) diagnosis; MDD vs. schizophrenia vs. mixed populations (i.e., samples containing different types of diagnoses). Based on Lipsey and Wilson (1993), effect sizes were interpreted as small (0 – 0.32), moderate (0.33 – 0.55) and large (0.56 – 1.20). If one

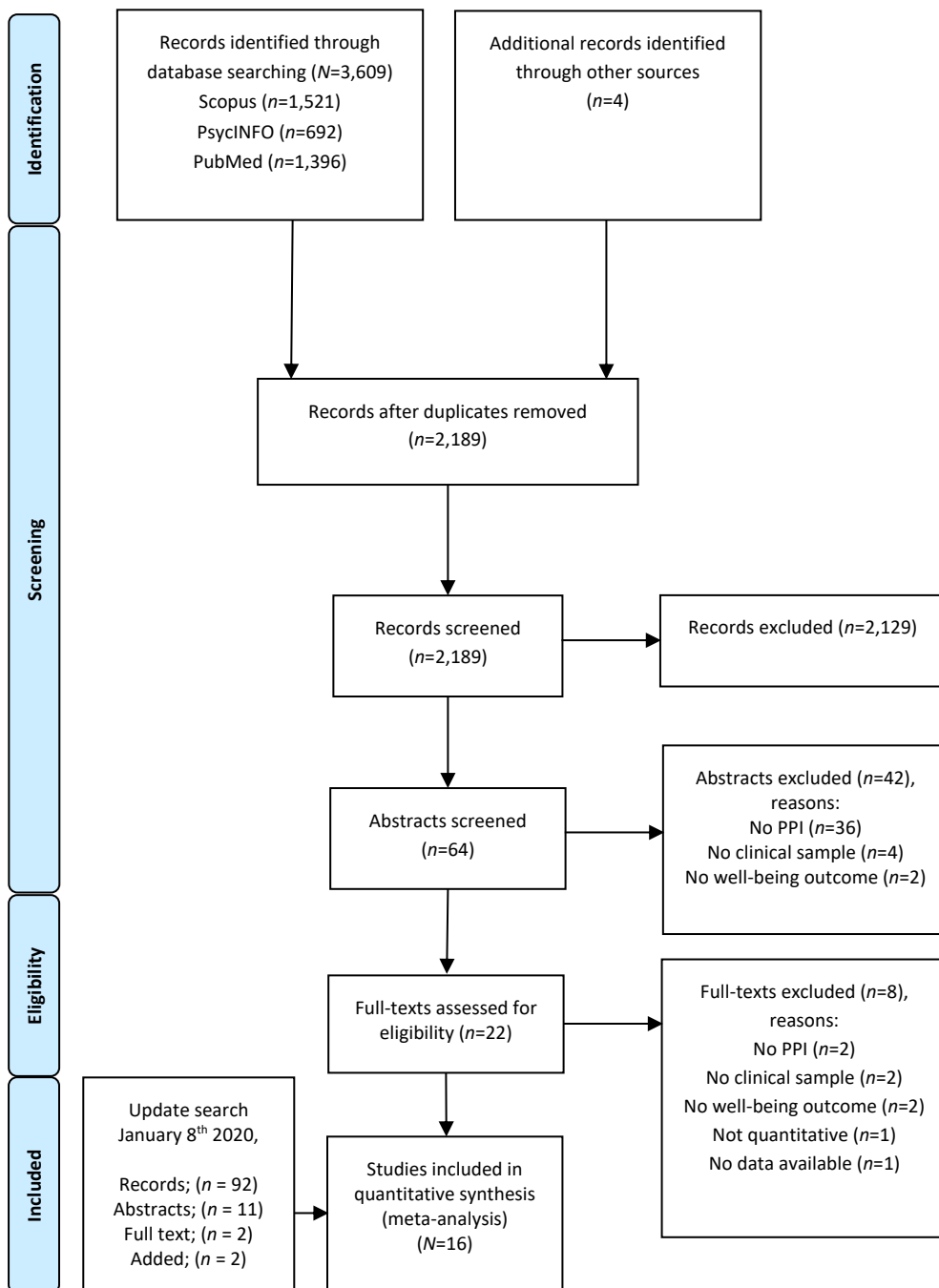


Figure 1. Flowchart of the study selection process

trial reported multiple outcomes of well-being or psychopathology, included effect sizes were averaged in CMA. The heterogeneity of effect sizes was assessed using  $Q$  and  $I^2$  statistics (Lipsey & Wilson, 1993). Significant  $Q$ -values are indicative of heterogeneity and test whether observed effect sizes are significantly more different from one another than would be expected on chance alone.  $I^2$  statistic shows the total variance across the included effect sizes, with higher values being indicative of more heterogeneity and zero of true homogeneity. Values of 25%, 50% and 75% show low, moderate and high heterogeneity, respectively (Higgins & Thompson, 2002). Due to the limited number of studies, we decided not to assess publication bias in this study.

## RESULTS

### Study selection

In total, we found 3,609 titles, and after the exclusion of duplicates, 2,189 titles remained. After screening of the titles, 64 abstracts remained and were reviewed, and 22 studies were found eligible for full-text screening. In the final analyses, 16 studies were included. The study selection process is summarized in Figure 1.

### Population characteristics

The 16 studies included 729 participants, of which 442 participants were in the PPI groups and 287 in the control groups. The mean age of the participants was 42.0 years in the intervention and 43.2 in the control groups. Furthermore, on average, 53.8% and 67.9% of the participants were female in the intervention and control groups, respectively. The dropout rates were 24.2% and 22.0%. The sixteen studies included 45.3% individuals with MDD, 35.4% with schizophrenia, 10.4% with bipolar disorder, 4.4% with borderline personality disorder, 2.3% with schizoaffective disorder, and 2.2% with other, not specified disorders. Demographical data for the included studies are displayed in Table 1.

### Study characteristics

Of the included studies, ten studies used a control group. Of these ten studies, nine studies utilized randomized controlled trials (Asgharipoor et al., 2012; Braehler et al., 2013; Carr et al., 2017; Celano et al., 2017; Celano et al., 2020; Farquharson & MacLeod, 2014; Feliu-Soler et al., 2017; Schrank et al., 2016; Seligman et al., 2006) and one study used a quasi-experimental design (Chaves et al., 2017). Six studies used a pre-post design without a control group (Favrod

et al., 2015; Ferguson et al., 2009; Johnson et al., 2011; Lai et al., 2015; Meyer et al., 2012; Painter et al., 2019). Study characteristics are summarized in Table 2.

**Table 1.** Sample characteristics of the included studies

	PPI group ( <i>n</i> = 442)	Control group ( <i>n</i> = 287)	Total group ( <i>N</i> = 729)
Age, years	42.0	43.2	42.5
Gender			
Female	238 (53.8%)	195 (67.9%)	433 (59.4%)
Marital status			
Single	277 (62.7%)	167 (58.2%)	444 (60.9%)
In relationship	165 (37.3%)	120 (41.8%)	285 (39.1%)
Drop-out rate	107 (24.2%)	63 (22.0%)	170 (23.3%)
Diagnosis			
Major depressive disorder	160 (36.2%)	170 (59.2%)	330 (45.3%)
Schizophrenia	186 (42.1%)	72 (25.1%)	258 (35.4%)
Bipolar disorder	55 (12.4%)	21 (7.3%)	76 (10.4%)
Borderline personality disorder	16 (3.6%)	16 (5.6%)	32 (4.4%)
Schizoaffective disorder	13 (2.9%)	4 (1.4%)	17 (2.3%)
Others not specified	12 (2.7%)	4 (1.4%)	16 (2.2%)

## Intervention characteristics

Intervention characteristics are summarized in Table 4. The interventions that were used in the included studies differed in both content and duration. The duration varied between 3 and 20 weeks, with an average duration of 8.8 weeks. Almost all interventions consisted of one session per week, with a length of one or two hours per session (two hours being the maximum). The Recovery Program (Lai et al., 2015) is an exception, incorporating five sessions weekly. In the 11 studies which described the duration of the interventions, the average total duration of the interventions was 17 hours (range 7 – 40 hours). Thirteen studies evaluated group programs, and three studies evaluated an individual approach. The majority of the interventions comprised PPIs in combination with other approaches such as elements of cognitive behavior therapy (Carr et al., 2017), defeatist thinking (Favrod et al., 2015) and positive goal setting (Meyer et al., 2012). Other interventions primarily focused on compassion (e.g., Braehler et al., 2013; Feliu-Soler et al., 2017) or goalsetting (Farquharson & MacLeod, 2014).

**Table 2.** Characteristics of included studies

First author (year)	Diagnosis	Female (%)	N	Mean age (SD)	PP intervention	Control group	Drop-out rate (%)	Outcome measures
Asgharipoor (2012)	MDD	72.2	18	26.4 (5.9)	Positive psychotherapy	CBT	NR	OHS, SWS, BDI-II, SUDS PANAS, BDI-II
Braehler (2012)	Schizophrenia, schizoaffective disorder, psychosis, bipolar disorder	45.0	40	41.8 (10.3)	Compassion-focused therapy	TAU	32.0	PANAS, BDI-II
Cairr (2017)	MDD	66.0	82	41.0 (NR)	Say yes to life	TAU + therapy	40.0	WEMBS, BDI-II, HRSD, MADRS PANAS, LOT-R, BHS, QIDS
Celano (2017)	MDD	69.0	65	44.0 (16.7)	Positive Psychology intervention	CF	10.0	PANAS, LOT-R, BHS, QIDS
Celano (2020)	Bipolar disorder	68.0	25	45.4 (13.0)	Positive Psychology intervention (+ telephone calls)	Recall neutral events (+ telephone calls)	20.0	PANAS, GQ-6, LOT-R, BHS, QIDS
Chaves (2017)	MDD	100.0	96	51.7 (10.4)	Positive psychology intervention	CBT	21.3	PANAS, PHI, PWBS, SWLS, LOT-R, BDI-II, BAI PANAS, SWLS
Farquharson (2014)	Schizophrenia, bipolar disorder, MDD	53.0	82	45.0 (10.4)	Goal-setting and planning	WL	51.0	PANAS, PHI, PWBS, SWLS, LOT-R, BDI-II, BAI PANAS, SWLS
Favrod (2015)	Schizophrenia, schizoaffective disorder	35.0	37	39.5 (20.0)	Positive emotions program for schizophrenia	NA	16.0	SBI, CDSS, SANS
Feliu-Soler (2017)	Borderline personality disorder	94.0	32	33.8 (7.2)	Loving-kindness and compassion meditation	MCT	NR	SCS, FSCRS, BSL-23
Ferguson (2009)	Schizophrenia, bipolar disorder	0.0	14	40.1 (10.8)	Well-being therapy	NA	2.0	PANAS, SWLS, FTT, HADS, BHS, NSS mDES, TEPS, PWBS, SWLS, THS, SBI, CAINS
Johnson (2011)	Schizophrenia, schizoaffective disorder	17.0	18	29.4 (10.2)	Loving-kindness meditation	NA	NR	

**Table 2.** Characteristics of included studies (continued)

First author (year)	Diagnosis	Femal e %	N	Mean age (SD)	PP intervention	Control group	Drop-out rate (%)	Outcome measures
Lai (2015)	Schizophrenia, bipolar disorder, MDD	40.0	63	NR	Recovery program	NA	5.2	CHS, CSWEMWBS
Meyer (2012)	Schizophrenia	56.0	16	39.6 (8.8)	Positive living	NA	25.0	SPWB, SBI, DHS, BSI
Painter (2019)	Bipolar disorder	50.0	12	45.3 (12.3)	Positive emotion regulation intervention	NA	25.0	AVI, SBI, SCS-SF, YMRS, HRSD
Schrank (2016)	Schizophrenia, schizoaffective disorder	45.0	94	42.5 (11.3)	Wellfocus positive psychotherapy	TAU	24.0	WEMWBS, PPI, SDHS, IHS, RSE-S, SBI, RES, SCS, BPRS
Seligman (2006), study 2	MDD	73.0	32	NR	Positive psychotherapy	TAU and TAU + medication	13.0	SWLS, PPTI, ZSRD

*Note:* AVI = Affect Valuation Index, BAI = Beck Anxiety Inventory, BDI-II = Beck Depression Inventory-II, BHS = Beck Hopelessness Scale, BSI = Brief Symptom Inventory, BSL-23 = Borderline Symptom List-23, BPRS = Brief Psychiatric Rating Scale, CAINS = Clinical Assessment Interview for Negative Symptoms, CBT = Cognitive Behavioral Therapy, CDSS = Calgary Depression Scale for Schizophrenia, CF = Cognition-focused, CHS = Chinese Hope Scale, CSWEMWBS = Chinese Short Warwick Edinburgh Mental Well-being Scale, DHS = Dispositional Hope Scale, FSCRS = Forms of Self-Criticism/Self-Attacking and Self-Reassuring Scale, FTT = Future Thinking Task, GQ-6 = Gratitude Questionnaire-6, HADS = Hospital Anxiety and Depression Scale, HIS = Integrative Hope Scale, HRSD = Hamilton Rating Scale for Depression, LOT-R = Life Orientation Test-Revised, MADS = Montgomery-Åsberg Depression Scale, MCT = Mindfulness Continuation Training, MDD = Major Depressive Disorder, mDES = Modified Differential Emotions Scale, NA = Not applicable, NR = Not reported, NSS = Negative Syndrome Scale, OHS = Oxford Happiness Scale, PPTI = Positive Psychotherapy Inventory, PWBS = Ryff's Psychological Well-being Scale, PHI = Pemberton Happiness Index, PPI = Positive Psychotherapy Inventory, QIDS = Quick Inventory of Depressive Symptomatology, RES = Rogers Empowerment Scale, RSE-S = Rosenberg Self-Esteem Scale, SANS = Scale for the Assessment of Negative Symptoms, SBI = Savoring Belief Inventory, SCS = Self-compassion Scale, SCS-SF = Self-compassion Scale-Short Form, SDHS = Short Depression Happiness Scale, SERS-SF = Self-esteem Rating Scale – Short Form, SUDS = Subjective Units of Distress Scale, SWLS = Satisfaction with Life Scale, SWS = Subjective Well-being Scale, TAU = Treatment as Usual, TEPS = Temporal Experience of Pleasure Scale, THS = Trait Hope Scale, WEMWBS = Warwick Edinburgh Mental Well-being Scale, WL = Waiting-list, YMRS = Young Mania Rating Scale, ZSRS = Zung Self-Rating Scale.

## Interventions in the control group

A variety of control conditions were used in ten studies: three studies used treatment as usual (Braehler et al., 2013; Schrank et al., 2016; Seligman et al., 2006) one study used a waitlist group (Farquharson & MacLeod, 2014). In the other six studies, PPIs were compared to active interventions. The control conditions comprised a program comparable to the intervention group (Carr et al., 2017; Celano et al., 2020), cognitive behavior therapy (Asgharipoor et al., 2012; Chaves et al., 2017), a cognition focused treatment (Celano et al., 2017) and a mindfulness continuation treatment (Feliu-Soler et al., 2017).

## Outcomes

In total, the included studies used 74 different measurement scales. In the meta-analysis, we used only scales with outcomes related to well-being (36) and symptomatology (20). Of the measurement scales focusing on well-being, the Satisfaction With Life Scale (SWLS), Positive And Negative Affect Schedule (PANAS), Savoring Belief Inventory (SBI), and Self-Compassion Scale (SCS) were used most often (5, 5, 4, and 4 times, respectively). Of the symptomatology scales, the Beck Depression Inventory (BDI), Beck Hopelessness Scale (BHS), and Hamilton Rating Scale for Depression were used in more than one study (4, 2, and 2 times, respectively). One included study (Lai et al., 2015) only assessed well-being related outcomes.

**Table 3.** Between- and within-group effects at post-intervention

Outcome measure	$N_{comp}$	Hedge's $g$	95% CI	Z
Between-group effects post-intervention				
Well-being	10	0.16	-0.08; 0.41	1.30
Psychopathology	10	-0.10	-0.49; 0.29	-0.50
Within-group effects post-intervention				
Well-being	16	0.40	0.25; 0.56	5.15***
Psychopathology	15	-0.70	-0.95; -0.44	-5.26***

Note.  $N_{comp}$  = Number of comparisons, CI = Confidence interval. \*\*\* $p < .001$ .

**Table 4. Characteristics of included interventions**

First author (year)	Intervention	Duration	Format	Goals	Modules/structure	Therapeutic techniques
Asgharipoor (2012)	Positive Psychotherapy	12 weeks, one session weekly	Group	Identifying potential capabilities and how to strengthen them	Awareness positive aspects of life, values in life, ranking, combine lifestyle with values. 1.) Orientation, 2.) Identify potential capabilities, 3. and 4.) Ways of appreciating positive affairs in life, 5.) Psycho education four life styles, 6. and 8.) Ranking activities of pleasure and meaningfulness, 8.-12.) Hierarchy of life-style and values.	Psycho-education, writing and planning
Braehler (2013)	Compassion-Focused Treatment	16 weeks, one 2 hour session weekly	Group	Building capacities in individuals suffering from psychosis to experience compassion	Based on compassion-focused therapy in high shame and self-critical individuals. 1.) Formation phase, sessions 1-5: insight in recovery, activation to build compassionate skills. 2.) Middle phase, sessions 6-13: developing compassion. 3.) End phase, sessions 14-16: reflect and integrate changes	Psycho-education, mindfulness and writing
Carr (2016)	Say Yes To Life Intervention	20 weeks, one 2 hour session weekly	Group	Learning a range of positive psychology and CBT-skills and providing a context for recovery	Positive psychology psychotherapy intervention for major depressive disorder. Positive psychology skills were included in the following sessions: 2.) Identifying personal strengths; 7.) Constructive use of humor, 13.) Strengthening adult attachments, 14.) Gratitude, 15. and 16.) Forgiveness, 17.) Strengthening social networks, 19.) Savoring positive experiences	Psycho-education, CBT exercises, homework and writing
Celano (2017)	Positive Psychology Intervention	6 weeks, weekly phone call	Individual	No specific goals were described	Telephone-based PP intervention for depressed patients hospitalized for a suicide attempt or suicidal ideation. Telephone session to review weekly sections of the treatment manual, highlighting a specific positive construct and its role in their recovery: 1.) Gratitude for positive events, 2.) Identifying and using personal strengths, 3.) Gratitude letter, 4.) Enjoyable and meaningful activities, 5.) Leveraging past success, 6.) Acts of kindness	Psycho-education, homework and writing
Celano (2020)	Positive Psychology intervention (+ telephone calls)	4 weeks, weekly phone call	Individual	Increasing PPE vocabularies, reinforcement of PPE, integrating PPI in daily life	Week 1 – Gratitude letter: Participants wrote a letter thanking someone for an act for which they were grateful. Week 2 – Using personal strengths: Participants used a strength in a new way. Week 3 Performing acts of kindness: Participants performed three acts of kindness in one day. Week 4 Imagining a best possible self: Participants wrote about their best possible life in the future.	Exercises, homework, reviewing exercises and writing



**Table 4. Characteristics of included interventions (continued)**

First author (year)	Intervention	Duration	Format	Goals	Modules/structure	Therapeutic techniques
Chaves (2017)	Positive Psychology Intervention	10 weeks, one 2 hour session weekly	Group	Designed to nurture components of hedonic well-being and components of eudaimonic well-being	Program based on positive psychology for treating major depression: 1.) Objectives, expectations and attitudes on treatment, 2.) Positive emotions, 3.) Savouring and Emotion regulation, 4.) Gratitude, optimism and best possible self, 5.) Positive relationships and kindness, 6.) Self-compassion, 7.) Personal strengths, 8.) Sense of living and goal setting, 9.) Resilience, 10.) Relapse prevention No further description of the intervention was given	Psycho-education, homework and writing
Farquharson (2014)	Goal-setting And Planning Intervention (GAP)	4 weeks, one 2 hour session weekly	Group	Targeting goal-setting and planning skills to improve well-being		No further description of the intervention was given
Favrod (2015)	Positive Emotions Program for Schizophrenia (PEPS)	8 weeks, one 1 hour session weekly	Group	Reducing anhedonia and apathy, helping to overcome defeatist cognitions and to increase the anticipation and maintenance of positive emotions	The intervention included meditation exercises, discussion of homework, exercise in challenging specific defeatist thoughts, develop an alternative and a more positive way of thinking, learn and practice a new skill to improve their anticipation or maintenance of pleasure. The sessions included: 1.) defeatist thinking, 2.) savouring pleasant moments, 3.) accentuating the behavioural expression of emotions, 4.) making the most of pleasant moments by sharing them with others, 5.) savouring past pleasant moments, 6.) anticipating pleasant moments, 7.) anticipating pleasant moments, 8.) review of all skills) Psycho-educational content from Gilbert's theoretical model of compassion, DBT techniques based on the use of kindness and affection. Other techniques included: affection in acceptance of negative emotions, loving-kindness meditation, and specific exercises from the Mindful Self-Compassion programme. LKM and compassion exercises were practiced during clinical sessions, and participants were strongly encouraged to practice daily	Visual and audio materials, psycho-education, homework and writing
Feliu-Soler (2016)	Self-compassion and Loving-kindness meditation	3 weeks, one session weekly	Group	Establishing motivation for LKM and compassion practice and provide an evolutionary understanding of compassion by explaining the three emotion regulation systems.		Psycho-education, audio material with LKM and compassion for practicing at home

**Table 4. Characteristics of included interventions (continued)**

First author (year)	Intervention	Duration	Format	Goals	Modules/structure	Therapeutic techniques
Ferguson (2009)	Goal-setting and Planning Intervention (GAP)	6 weeks, one session weekly	Group	Developing goal setting and planning skills with the aim to increase well-being	Focus on positive engagement with goals: 1.) Introduce the concept of well-being, role of goals and plans and their relation to well-being, identify valued personal goals, 2.) Select goal practice a visual imaging technique, discuss planning, 3.) Review plans of action and discuss common obstacles, 4.) Discuss action steps, obstacles and solutions, 5.) Discuss maintaining motivation, identify topics for final session, 6.) Revise some of the techniques used in the group and identify members of the team who can offer support	Psycho-education, writing, planning, homework
Johnson (2011)	Loving-kindness meditation	7 weeks, one 1 hour session weekly	Group	Cultivating kindness towards self and others, change the orientation to life experiences, aiming to broaden the range of emotional responses	Sessions incorporated 3 components: discussion, skill teaching, and practice. Practice began with mindfulness, participants then contemplated a person for whom they already feel compassion or a social situation where they felt kindness. Participants were led in guided meditations to sequentially extend these feelings to themselves as well as others. Participants were encouraged to practice LKM formally by listening daily to meditation audio material	Psycho-education, meditation, audio material for practice at home
Lai (2015)	Recovery Program	3 weeks, five 1 hour sessions weekly	Group	Promote successful recovery through goal setting, positive thinking, taking control and empowerment	Five elements as therapeutic modules: hope, support and managing symptoms, empowerment, relationship and coping, including 1.) Identification of hope and development of faith, 2.) Enhancement of personal responsibility and productivity, 3.) Promotion of self-management and autonomy, 4.) Importance of peer support and community life, 5.) Building frustration tolerance and learn forgiveness, 6.) Social acceptance and enhanced self-awareness; 7.) Adaptability and capacity to change, 8.) Situational applicability	Psycho-education
Meyer (2012)	Positive Living	10 weeks, one 90 minute session weekly	Group	No specific goals were described	Six behavioural exercises: Using your strengths, three good things, biography, gratitude visit, active/constructive responding, and savouring. Designed to increase positive emotions, as well as build character strengths and meaning. Also included positive goal and mindfulness minute	Homework, writing

**Table 4. Characteristics of included interventions (continued)**

First author (year)	Intervention	Duration	Format	Goals	Modules/structure	Therapeutic techniques
Painter (2019)	Positive emotion regulation intervention	9 weeks, 90 minute session weekly	Group	Improve emotional dysregulation in BD	The Learning Affective Understanding for a Rich Emotional Life (LAUREL): Week 1: introduction to bipolar disorder, Week 2: emotion education, Week 3: noticing and savouring positive experiences, Week 4: mindfulness, Week 5: reappraisal, Week 6: gratitude and small acts of kindness, Week 7: self-compassion, Week 8: setting and achieving attainable goals ,Week 9: feedback and implementation	Psycho-education, CBT, didactic skills, , daily log and homework
Schrank (2016)	Wellfocus Positive Psychotherapy	11 weeks, one 90 minute session weekly	Group	Increasing positive experiences, amplifying strengths, fostering positive relationships and creating a more meaningful self-narrative	Introduction, savouring, good things, identifying personal strengths, personal strength activity, strength activity with significant other, forgiveness, one door closes another door opens, gratitude, and positive responding. Sessions begin and close with a music savouring exercise. In contrast to standard PPT, this intervention has a reduced focus on literacy and didactics but instead includes more experiential and interactive components	Psycho-education, homework and writing, supporting and phone-calls
Seligman (2006), study 2	Positive psychotherapy	12 weeks and 14 sessions	Individual	No specific goals were described	1.) Orientation: lack of positive resources maintains depression, 2.) engagement: identifying signature strengths, 3.) Engagement/pleasure: cultivation of signature strength and positive emotions, 4.) Pleasure: good versus bad memories, 5.) Pleasure/engagement: forgiveness, 6.) Pleasure/engagement: gratitude, 7.) Pleasure/engagement: mid-therapy check, 8.) Meaning/engagement: satisficing instead of maximizing, 9.) Pleasure: optimism and hope, 10.) Engagement/meaning: love and attachment, 11.) Meaning: family tree of strengths, 12.) Pleasure: savouring, 13.) Meaning: gift of time, 14.) Integration: the full life	Psycho-education, homework

Note: CBT = Cognitive Behavioural Therapy, LKM = Loving-Kindness Meditation, PP = Positive Psychology Emotions.

## Post-test effects

For the ten studies including a control group, a small nonsignificant effect on well-being ( $g = 0.17, p = .19, n = 287$ ) and psychopathology ( $g = -0.10, p = .62, n = 287$ ) between the PPI and control groups was found. Analyses of within-group effects for all 16 included studies revealed a moderate significant effect on well-being (Hedge's  $g = 0.40, p < .001, N = 729$ ) and a large significant effect on psychopathology ( $g = -0.70, p < .001, n = 666$ ) within the positive psychology groups. An overview of the total effects on well-being and psychopathology can be found in Table 3. Individual effect sizes of the within-group effects are summarized in Figure B1 and Figure B2 (Appendix B).

## Subgroup analyses

Analyzing potential moderators of the within-group effects, no significant differences between subgroups for treatment duration and format were found for both well-being and psychopathology. For the diagnosis subgroups, PPIs had a significantly stronger effect on well-being in people with MDD compared to patients with schizophrenia ( $Q = 4.52, df = 1, p < .05$ ) and compared to studies with mixed samples ( $Q = 9.30, df = 1, p < .01$ ). Furthermore, PPIs revealed a significantly stronger effect on psychopathology in people with MDD compared to studies containing mixed samples ( $Q = 4.19, df = 1, p < .05$ ). Results of the subgroup analyses are summarized in Table 5.

**Table 5.** Subgroup analyses

Outcome	Criterion	Subgroup	$N_{comp}$	Hedge's $g$	95% CI	Z	
Well-being	Duration	<= 8 weeks	7	0.37	[0.13; 0.60]	3.05**	
		> 8 weeks	9	0.44	[0.22; 0.65]	3.95***	
	Format	Group	13	0.34	[0.19; 0.50]	4.25***	
		Individual	3	0.66	[0.32; 1.01]	3.74***	
		Diagnosis	MDD	5	0.73	[0.47; 0.99]	5.42***
Psychopathology	Duration	<= 8 weeks	6	-0.68	[-1.10; -0.25]	-3.10**	
		> 8 weeks	9	-0.72	[-1.06; -0.37]	-4.04***	
	Format	Group	12	-0.65	[-0.95; -0.35]	-4.26***	
		Individual	3	-0.90	[-1.52; -0.28]	-2.86**	
		Diagnosis	MDD	5	-1.08	[-1.56; -0.59]	-4.36***
			Schizophrenia	4	-0.84	[-1.33; -0.61]	-5.32***

Note.  $N_{comp}$  = Number of comparisons; CI = Confidence interval. \* $p < .05$ , \*\* $p < .01$ , \*\*\* $p < .001$ .

## DISCUSSION

The current explorative meta-analysis aimed to assess the effects of PPIs on well-being and psychopathology in people with SMI across studies. Ten controlled studies, nine of which used randomized allocation to conditions, and six non-controlled studies were included. For the ten controlled studies, a nonsignificant, small effect was found across studies between intervention and control groups for both well-being and psychopathology. Across all studies, a significant moderate pre-post intervention effect was found for well-being and a significant large effect for psychopathology.

These results demonstrate that people with SMI do benefit from PPIs in terms of well-being and psychopathology. However, at present, there is no evidence that PPIs are more effective in improving mental health when using controlled studies. One explanation for the absence of significant effects in the controlled studies is that the majority of these studies used an active intervention such as cognitive behavioral therapy, cognition therapy and mindfulness as control condition. It is a common finding in meta-analyses that therapeutic interventions are more effective in comparison to waiting-list groups or no-intervention conditions than active interventions (Davis et al., 2016; Sin & Lyubomirsky, 2009).

We also found that PPIs are more effective in improving well-being in patients with MDD compared to patients with schizophrenia and mixed diagnoses and that PPIs are more effective in reducing psychopathology in patients with MDD in comparison with mixed samples. These findings suggest that PPIs are the most impactful on recovery in patients with MDD. One possible explanation is that many PPIs are aimed at enhancing positive emotions and cognitions and that these processes have the most substantial impact in people with MDD since negative emotions and cognitions are the core symptoms of MDD. However, a depressed mood is not exclusive to individuals with an MDD diagnosis. Within the other SMI-diagnosis groups (e.g., BD and schizophrenia), depressive episodes are common as part of the diagnosis in BD or as a comorbid disorder in other diagnosis groups. A recent study found that affective symptoms had a greater influence on the personal recovery of patients with SMI than with psychotic symptoms (Van Eck et al., 2018). Another possible explanation is related to the characteristics of the disorders themselves. Schizophrenia is characterized by a complex set of symptoms and often has a chronic and life-long course (Miller et al., 2014). In addition, recovery rates are extremely low, and relapse rates are very high (Emsley et al., 2013; Ventura et al., 2011). Though PPIs may support the personal recovery of patients with schizophrenia, it may thus be harder to demonstrate the impact on well-being and symptomatology in this more persistent and complex disorder.

The findings of this meta-analysis can be placed in the context of recent pleas for a more balanced approach in clinical psychology (Bohlmeijer & Westerhof, 2020; Wood & Tarrrier, 2010). Until recently, the roles of positive emotions, cognitions and behavior in mental health have largely been ignored, but there is growing evidence demonstrating that including positive psychotherapeutic interventions in treatment models has the potential to increase the effects on mental health (Rashid, 2009; Rashid, 2015; Seligman et al., 2006; Wood & Tarrrier, 2010). Additionally, there is substantial evidence that well-being and psychopathology are two related but distinct dimensions of mental health (Franken et al., 2018; Lamers et al., 2011) and that positive functioning reduces the risk of future incidence of mental illness (Schotanus-Dijkstra et al., 2019; Schotanus-Dijkstra et al., 2017). Also, people with mental illness describe the development of positive functioning as an important outcome of their treatment (de Vos et al., 2017; Zimmerman et al., 2006). These studies underscore the need to further develop and evaluate PPIs in people with mental illness.

Several important limitations apply to this meta-analysis. First, only 16 studies were included in the review, and only nine studies used a randomized controlled design. Also, many studies included a limited number of patients, and the total number of included patients in this meta-analysis is relatively small. This warrants a cautious interpretation of the results. White et al. (2019) stated in a reanalysis of the two main meta-analyses in PPIs that, due to small sample size bias, the effects are possibly much smaller than previously reported (White et al., 2019). It is to expect that this bias also applies to our study. Second, there is a large variety in the studied types of interventions, and the limited number of studies precludes an assessment of the relative impact of specific types of interventions. Third, follow-up assessments were largely lacking, and therefore, the long-term impact of PPIs on mental health in people could not be assessed.

## Conclusion

At present, there is no evidence that PPIs are more effective in comparison with other active interventions and treatment as usual in improving mental health in people with SMIs. However, the results also show that people with SMIs do benefit from PPIs in terms of enhancement of well-being and reduction of psychopathology. To further broaden the evidence regarding the effect of PPIs in SMIs, we recommend to further evaluate the effectiveness of PPIs in these target groups by means of adequately powered, randomized controlled trials. Moreover, it is recommended to include measures of specific positive proximal outcomes, which are the primary focus of PPIs.

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## APPENDIX A

### Search string: Scopus

- #1 TITLE-ABS((well-being} OR wellbeing} OR well being} OR happiness OR happy OR life satisfaction} OR satisfaction with life} OR positive psych\*} OR positive emotion\*} OR positive feeling\*} OR positive cognition} OR positive behavio\*} OR compassion OR optimism OR gratitude OR kindness OR Savouring OR Strengths OR Flourishing OR resilience)
- #2 TITLE-ABS(severe mental illness} OR psychosis OR Schizophrenia OR major depressive disorder} OR bipolar disorder} OR Mood disorder} OR mania OR personality disorder))
- #3 TITLE-ABS(intervention\* OR therap\* OR treatment\* OR training\* OR program\* OR exercise)
- #4 TITLE-ABS(effect\* OR effic\* OR outcome\* OR evaluat\* OR feasibility OR acceptability)
- #5 #1 AND #2 AND #3 AND #4 (filters: English, article, limit to subject area psychology and social sciences)

### Search string: PubMed

- #1 ("well-being"[tiab] OR happiness OR happy OR "life satisfaction"[tiab] OR "satisfaction with life"[tiab] OR "positive psychology"[tiab] OR "positive emotion"[tiab] OR "positive feeling"[tiab] OR "positive cognition"[tiab] OR "positive behaviour"[tiab] OR compassion[tiab] OR optimism[tiab] OR gratitude[tiab] OR kindness [tiab]) OR Savouring [tiab] OR Strengths [tiab] OR Flourishing [tiab] OR resilience [tiab])
- #2 (Happiness[Mh] OR Positive Psychology[Mh] OR Well Being[Mh] OR Optimism[Mh] OR Life Satisfaction[Mh] OR Compassion[Mh] OR Optimism[Mh])
- #3 (intervention\*[tiab] OR therap\*[tiab] OR treatment\*[tiab] OR training\*[tiab] OR program\*[tiab] OR exercise[tiab])
- #4 (Therapy[Mh] OR Psychotherapy[Mh] OR Training[Mh] OR Exercise[Mh])
- #5 ("severe mental illness" [tiab] OR psychosis [tiab] OR Schizophrenia [tiab] OR "major depressive disorder" [tiab] OR "bipolar disorder" [tiab] OR "Mood disorder"[tiab] OR mania [tiab] OR "personality disorder"[tiab])
- #6 ("Severe mental illness" [Mh] OR Psychosis [Mh] OR "Disorder Schizophrenia" [Mh] OR "Major depressive disorder" [Mh] OR "Bipolar disorder" [Mh] OR "Mood disorder"[Mh] OR Mania [Mh] OR "Borderline personality disorder"[Mh])

- #7 (effect\*[tiab] OR effic\*[tiab] OR outcome\*[tiab] OR evaluat\*[tiab] OR feasibility [tiab] OR acceptability [tiab]) condition[tiab])
- #8 #1 OR #2
- #9 #3 OR #4
- #10 #5 OR #6
- #11 #8 AND #9 AND #10 AND #7 (filters: English, Adults)

## **Search string: PsycINFO**

- #1 ("well-being" OR happiness OR happy OR "life satisfaction" OR "satisfaction with life" OR "positive psych\*" OR "positive emotion\*" OR "positive feeling\*" OR "positive cognition" OR "positive behavio\*" OR compassion OR optimism OR gratitude OR kindness)
- #2 (DE "optimism" OR DE "well being" OR DE "life satisfaction" OR DE "happiness" OR DE "positive psychology" OR DE "gratitude") OR "Savouring" OR "Strengths" OR "Flourishing" OR "resilience")
- #3 (intervention\* OR therap\* OR treatment\* OR training\* OR program\* OR exercise)
- #4 (DE "Intervention" OR DE "Therapy" OR DE "Psychotherapy")
- #5 ("severe mental illness" OR psychosis OR Schizophrenia OR "major depressive disorder" OR "bipolar disorder" OR Mood disorder" OR mania OR "personality disorder")
- #6 (DE "severe mental illness" OR DE "psychosis" OR DE "Schizophrenia" OR DE "major depressive disorder" OR DE "bipolar disorder" OR DE "Mood disorder" OR DE "Mania" OR DE "Borderline personality disorder")
- #7 (effect\* OR effic\* OR outcome\* OR evaluat\*)
- #8 #1 OR #2
- #9 #3 OR #4
- #10 #5 OR #6
- #11 #8 AND #9 AND #10 AND #7 (filters: academic journals, adults English)

# APPENDIX B

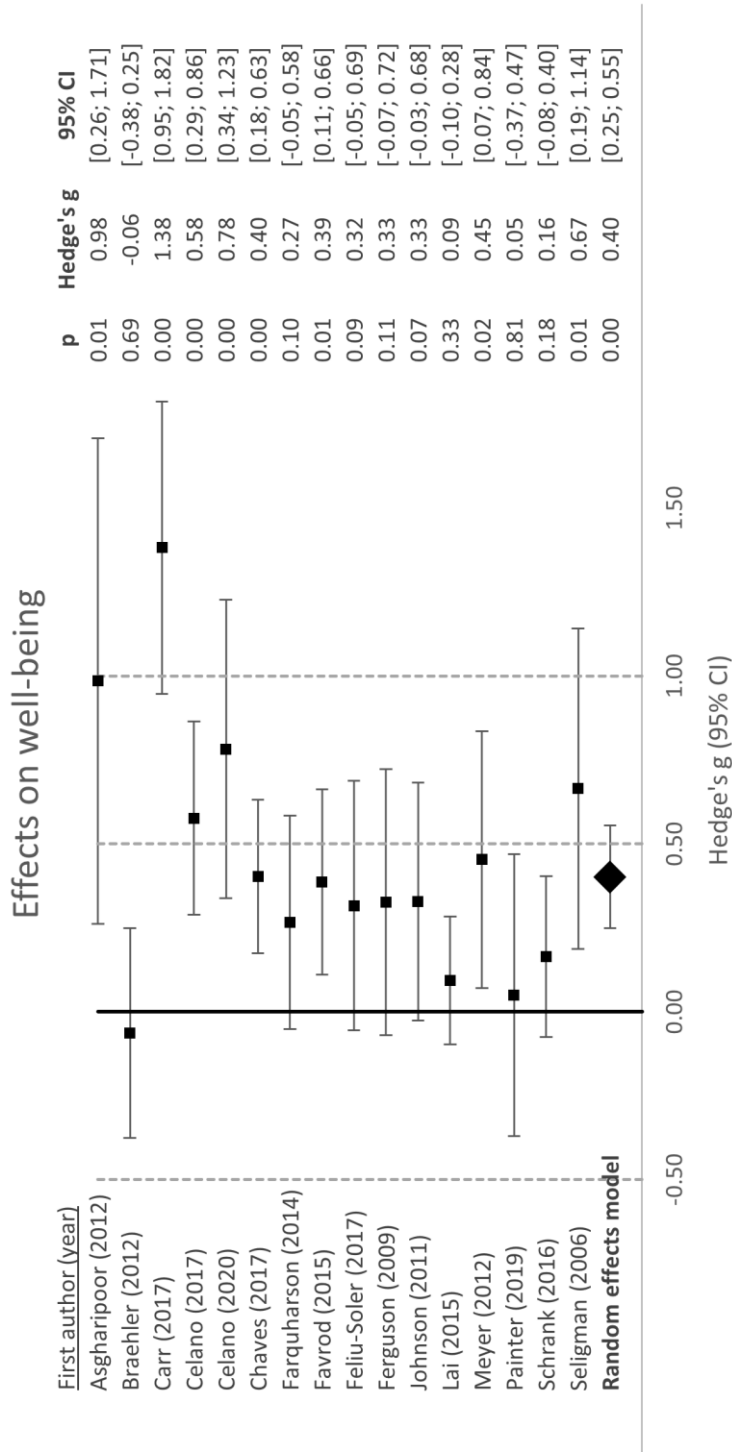
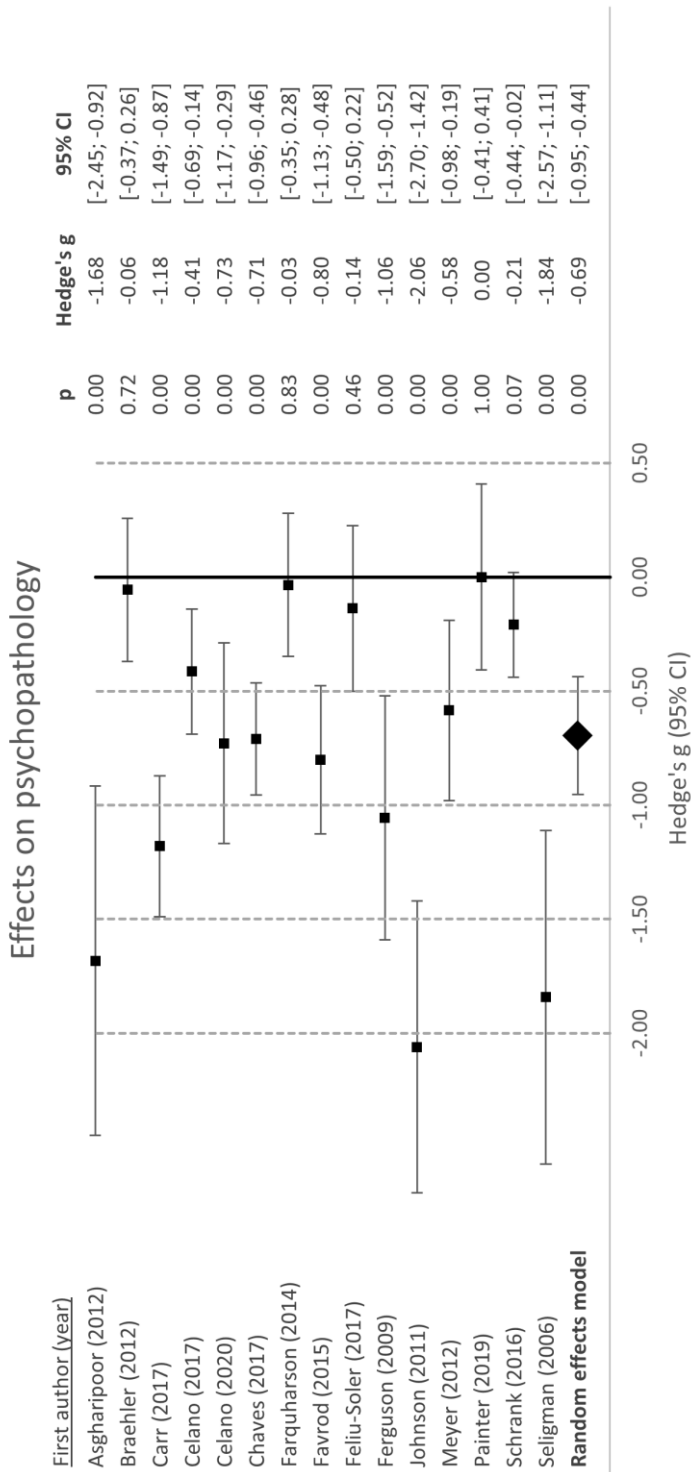


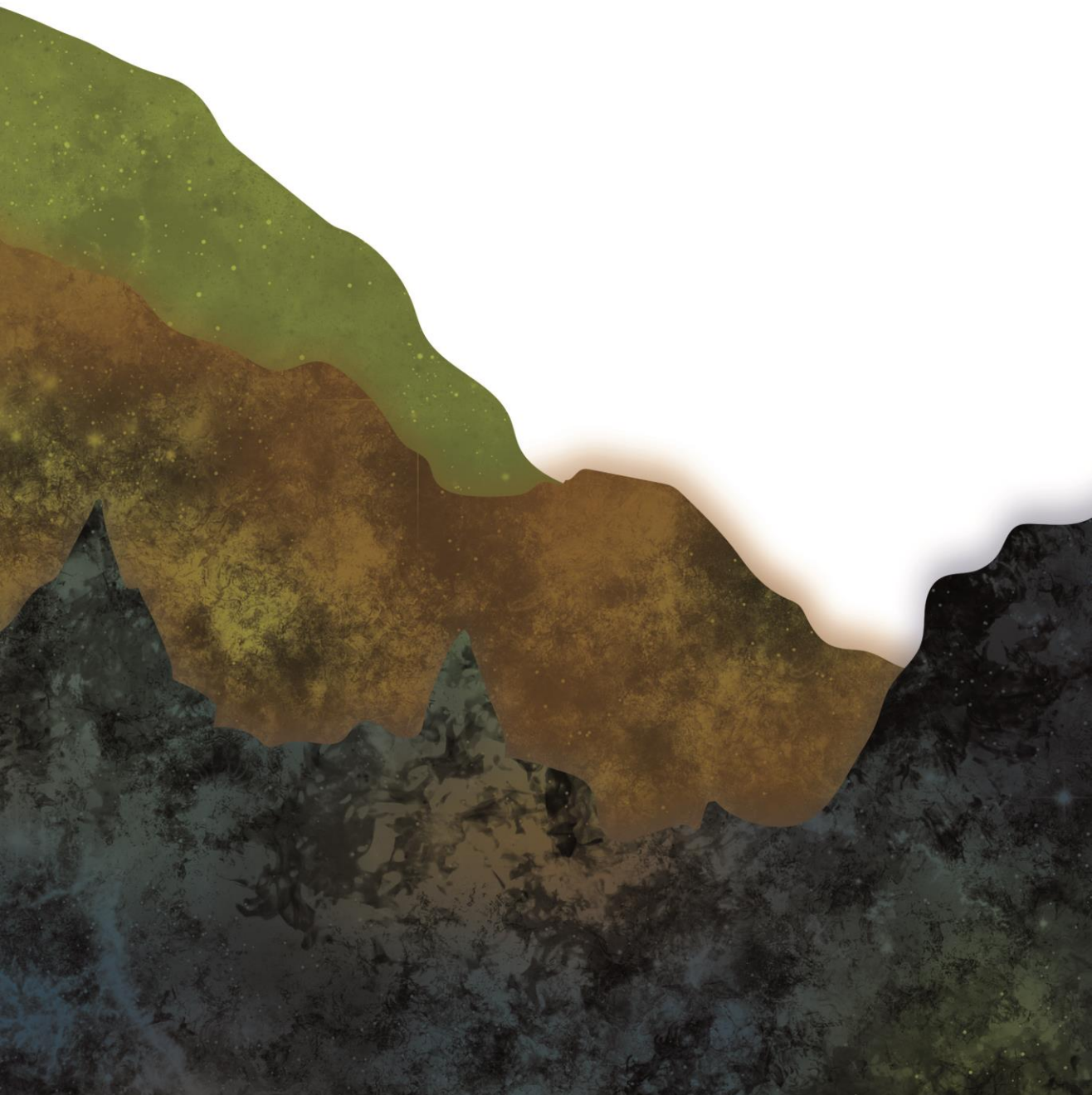
Figure B1. Forest plot of within-group effects on outcomes of well-being of the included positive psychology interventions





**Figure B2.** Forest plot of within-group effects on outcomes of psychopathology of the included positive psychology interventions

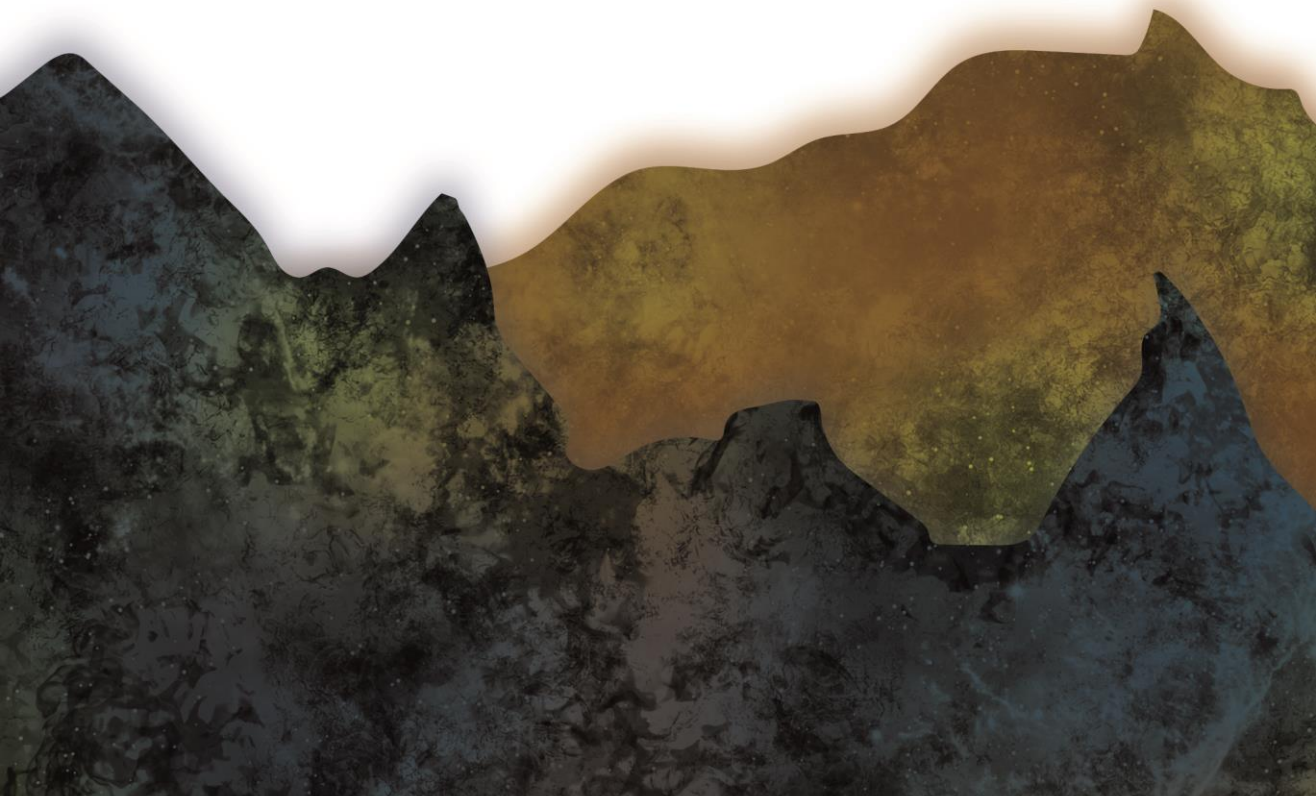




# CHAPTER 3

## Economic evaluations of non-pharmacological interventions and cost-of-illness studies in bipolar disorder: A systematic review

Kraiss, J. T., Wijnen, B., Kupka, R. W., Bohlmeijer, E. T., & Lokkerbol, J. (2020). Economic evaluations of non-pharmacological interventions and cost-of-illness studies in bipolar disorder: a systematic review. *Journal of Affective Disorders*, 276, 388-401. <https://doi.org/10.1016/j.jad.2020.06.064>



## ABSTRACT

Bipolar disorder (BD) is associated with substantial societal burden. Therefore, economic studies in BD became increasingly important. The goal of the current study is three-fold: (1) summarize the evidence regarding economic evaluations (EEs) of non-pharmacological interventions for BD, (2) summarize cost-of-illness studies (COIs) for BD published 2012 or later and (3) assess the quality of the identified studies. A systematic search was conducted in MedLine, EMBASE and PsycINFO. For both EEs and COIs, quality assessments were conducted and general and methodological characteristics of the studies were extracted. Outcomes included incremental-cost-effectiveness ratios for EEs and direct and indirect costs for COIs. Eight EEs and ten COIs were identified. The included studies revealed high heterogeneity in general and methodological characteristics and study quality. All interventions resulted in improved clinical outcomes. Five studies additionally concluded decreased total costs. For COIs, we found a wide range of direct (\$881-\$27,617) and indirect cost estimates per capita per year (\$1,568-\$116,062). High heterogeneity in terms of interventions, study design and outcomes made it difficult to compare results across studies. Interventions improved clinical outcomes in all studies and led to cost-savings in five studies. Findings suggest that non-pharmacological intervention for BD might be cost-effective. Studies on the costs of BD revealed that BD has a substantial economic burden. However, we also found that the number of EEs was relatively low and methodology was heterogenous and therefore encourage future research to widen the body of knowledge in this research field and use standardized methodology.

## INTRODUCTION

Bipolar disorder (BD) is a severe mood disorder characterized by recurrent manic and depressive episodes, often becoming manifest in early adulthood and extending over a lifetime (Goodwin & Jamison, 2007). In general, a distinction is made between bipolar I (BDI) and bipolar II (BDII). In the latter, a person experiences hypomanic and depressive episodes, but never a full-blown manic episode as in BDI (American Psychiatric Association, 2013). The lifetime prevalence of BD has been estimated at 0.6% for BDI and 0.4% for BDII, and the 12-month prevalence has been estimated at 0.4% and 0.3% respectively (Merikangas et al., 2011). Besides emotional distress and the impact on quality of life (Dean et al., 2004), negative social consequences (Calabrese et al., 2003) and high caregiver burden (Laxman et al., 2008; Miller et al., 2014), BD is also associated with a high economic burden. In 2010, the total costs including all direct and indirect costs (adjusted for purchasing power parity) of BD were estimated at €21.49 billion and the total costs per patient was estimated to be €7,183 in Europe (Olesen et al., 2012).

Economic evaluations (EEs) and cost-of-illness studies (COIs) contain important information to inform the healthcare sector about the cost-effectiveness of treatments and economic burden of a disease. The resources of our society are limited, and it is not possible to pay for every available intervention. Policy makers ought to be informed about the potential economic value of a new treatment or intervention. EEs aim to compare at least two treatment alternatives regarding costs and outcomes. Incremental analyses, which put the difference in effects in relation to the difference in costs between two competing interventions, are used to express the cost-effectiveness of an intervention towards a comparator. COIs are used to assess the economic impact of a disease and identify the associated costs to inform decision makers about the economic burden of the illness (Stuhldreher et al., 2012).

Summarizing studies regarding the cost-effectiveness of treatments and cost-of-illness of BD would guide policy makers in their decision making of which interventions should be prioritized. These decisions also have an impact on the work of clinicians, since decisions by the policy-makers set the agenda for treatment of mental disorders and determine which interventions are used and applied by clinicians. For academics, this is relevant as it provides an overview of the research field and identifies knowledge gaps. Therefore, it would help to obtain a comprehensive overview of this research field.

One review about EEs of pharmacological treatments has been published recently (Mavranouzouli & Lokkerbol, 2017). However, BD is typically treated with a combination of pharmacological and psychological interventions, both during acute episodes and during

preventive maintenance treatment of unlimited duration. Psychological interventions thus have an important place in the treatment of people with BD. To our knowledge, the only systematic review on EEs of psychological interventions specifically for BD was conducted by Abdul Pari et al. (2014). For COIs, two reviews on COIs in BD were conducted (Jin & McCrone, 2015; Kleine-Budde et al., 2014). Although the execution of the mentioned reviews was sound, the reviews only comprised studies until 2012 (Abdul Pari et al., 2014; Kleine-Budde et al., 2014) and 2013, respectively (Jin & McCrone, 2015) and an update is needed.

Thus, to our knowledge, no up-to-date comprehensive overview of EEs of non-pharmacological treatments for BD and of COIs exists. Summarizing this knowledge may help policy makers to prioritize interventions and academics to identify knowledge gaps. Therefore, the goal of the present study is three-fold: (1) to summarize the evidence regarding EEs of non-pharmacological interventions for BD in terms of their costs and outcomes, (2) to summarize COIs for BD published 2012 or later and summarize the estimated costs related to BD and (3) to assess the quality of the identified studies.

## **METHOD**

This systematic review was conducted in accordance with the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) statement (Moher et al., 2015). The review has been registered in PROSPERO, the International Prospective Register of Systematic Reviews (124044). Data sharing is not applicable to this article as no new data was created or analyzed in this study.

### **Search strategy**

A literature search was conducted in September 2018 for the period from January 1990 to September 2018 in the following databases: MedLine, EMBASE and PsycINFO. The search was repeated in January 2020 and results were updated. The search strategy was adapted from the search strategy of an earlier review about the cost-effectiveness of interventions for anxiety disorders (Ophuis et al., 2017). Search terms relating to economic studies were used from this earlier search and complemented with terms relating to bipolar disorder, that were derived from earlier reviews that were conducted to develop the Dutch multidisciplinary guidelines for treatment of BD (Kupka et al., 2015). The search strategy was supplemented with Medical Subject Headings (MeSH) for searches in MedLine and Thesaurus terms for searches in PsycINFO. The search string can be found in Appendix A. For EEs, the earlier review by Abdul Pari et al. (2014) was cross-checked. To check whether trials were available that were not identified through our original database search, we cross-checked three clinical trial registers

([www.clinicaltrialsregister.eu](http://www.clinicaltrialsregister.eu), [www.clinicaltrials.gov](http://www.clinicaltrials.gov), [www.isrctn.com](http://www.isrctn.com)) in January 2019 and again in January 2020. Checking trial registers is not likely to lead to additional studies, but we conducted this as an extra check for our literature search. Although there has been conducted a systematic review on EEs earlier, we decided to provide a complete overview of all EEs in this study, using a consistent quality appraisal tool. We considered it important to use a consistent quality assessment for all EEs, as outcomes of these studies might be especially relevant for policy-makers to prioritize interventions. If we only would have included more recent EEs, it would have been difficult to compare quality of studies with results from the earlier review (Abdul Pari et al., 2014). For COIs, the search was limited from 2012 to 2018, since we merely aimed to provide an update on the literature in this research field. Earlier reviews of COIs were cross-checked (Jin & McCrone, 2015; Kleine-Budde et al., 2014), to avoid including studies in the current review which were already included in an earlier review.

## Selection of studies

All identified articles were screened on title by the first and second author. Remaining abstracts and full-texts were independently screened by the first and second author and ambiguities were discussed until consensus was reached. Consensus was reached in every case, since any ambiguities were discussed until a collective decision could be made. Eligibility criteria were designed to cover the PICO characteristics (population, intervention, comparison and outcome). Both EEs and COIs had to contain a target population of people of any age diagnosed with BD. The target population could either be euthymic (i.e. stable) patients with BD or patients currently in an acute depressive or manic phase. EEs were included if they assessed the effectiveness of any non-pharmacological treatment for BD or any non-pharmacological treatment in combination with pharmacotherapy and conducted a full-economic evaluation. To be included in the review, included interventions should be aimed to elicit changes in individuals' emotions, behaviors or cognitions. Both model-based economic evaluations (MBEES) and trial-based economic evaluations (TBEEs) were included. Regarding the control group, it was not relevant which comparator was used in the included studies (e.g. treatment as usual, placebo, waiting-list or pharmacological treatment), as long as the intervention group contained a psychological intervention or a combination of psychological intervention with pharmacotherapy. Cost-utility analyses (CUAs), cost-effectiveness analyses (CEAs) or cost-benefit analyses (CBAs) were seen as full economic evaluations. COIs were included if the article reported the cost impact of BD, thus either direct (e.g. healthcare costs) or indirect (e.g. productivity loss) costs per individual and/or estimated direct or indirect societal costs. Studies needed to be in English, Dutch or German language. Possible COIs were excluded if the study reported the costs of an intervention only or if direct or indirect costs of BD were not reported or could not be derived. For both EEs and COIs, literature reviews, book



chapters and studies not published in peer-reviewed journals were excluded. Since studies published in peer-reviewed journals went through quality control by external reviews, we decided to exclude studies not published in peer-reviewed journals to ensure certain quality of studies. Furthermore, conference or dissertation abstracts, editorials, letters, commentaries and notes were excluded, as they did not provide enough details for their methodological quality to be judged.

## **Data extraction**

For EEs, data was extracted based on an adaptation of a pre-specified data extraction form (Wijnen et al., 2016) and included authors, publication year, country, type of economic evaluation (i.e. TBEE or MBEE), study type, analysis, valuation year, time horizon and funding. The type of perspective chosen by the authors of the papers (i.e. healthcare or societal perspective) was also extracted from the studies (Husereau et al., 2013). If the perspective was not explicitly mentioned, we decided whether a societal or healthcare perspective was chosen based on whether non-health related (i.e. productivity loss) outcomes were considered or not. Although societal and healthcare perspectives are most likely to be the most common chosen perspectives in economic studies for BD, other perspectives also may be relevant for this review, such as a health insurer perspective, health and social care or public sector perspective. For COIs, diagnostic criteria, approach (i.e. prevalence or incidence-based) and description of costs (i.e. per capita or national) were extracted. In addition, methodological characteristics were extracted for both EEs and COIs, including target population, sample size, comparators, mean age and gender. In case of EEs, the type of effect measurement used was also extracted. Any ambiguities in the data extraction were discussed between the first and second author until uncertainties could be resolved.

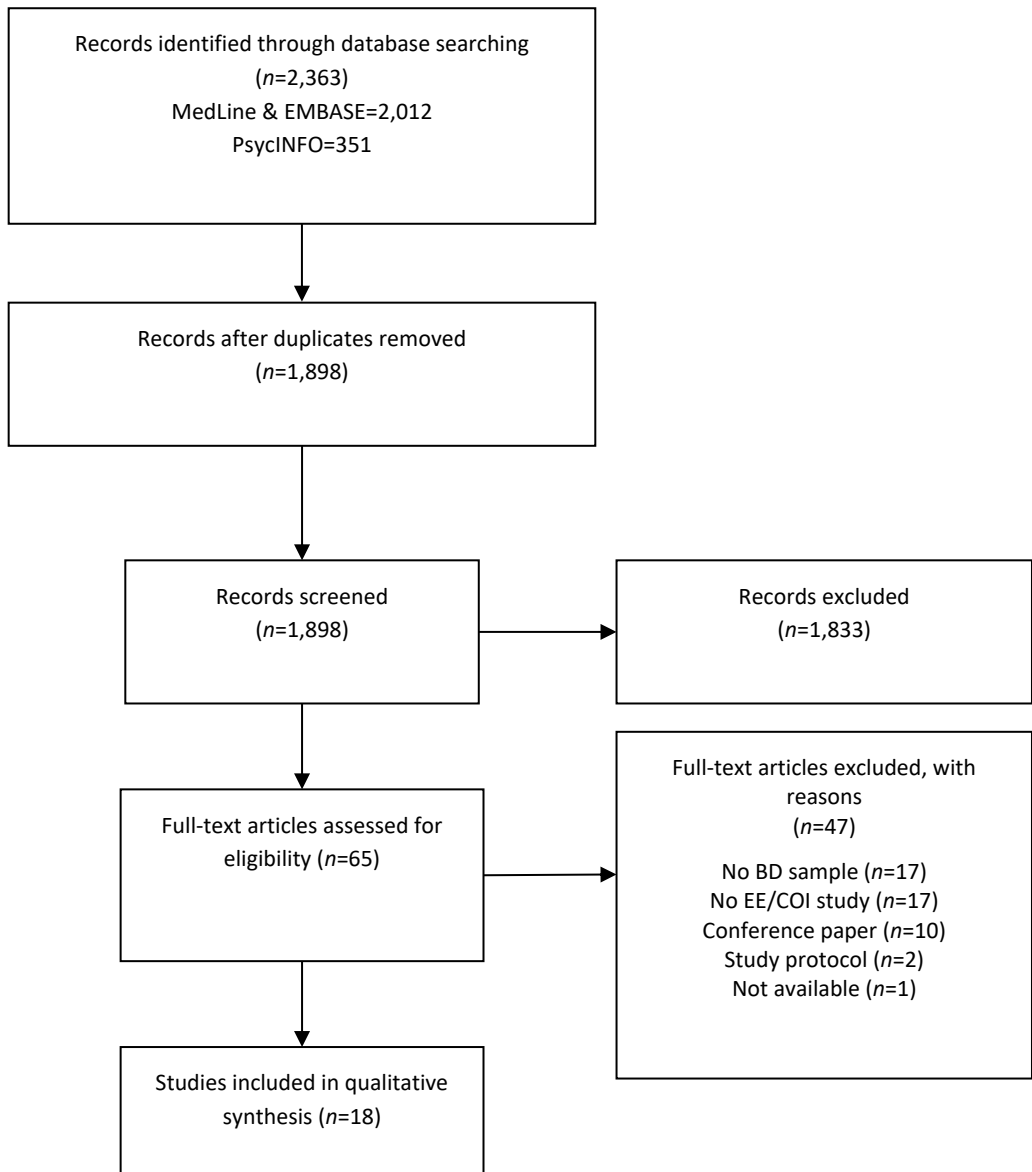
## **Quality assessment**

Quality of included EEs was assessed with the extended Consensus on Health Economic Criteria (CHEC) list (Evers et al., 2005; Odnoletkova et al., 2014), aiming to judge the internal and external validity of EEs. For this review, the CHEC-extended was used (Evers et al., 2005), which consists of the 20 CHEC-items and contains one extra item regarding model-quality (relevant in case of model-based economic evaluations). Item number seven of the CHEC (“Is the actual perspective chosen appropriate?”) was scored with 0 if the chosen perspective was not societal and no argumentation was provided for why a narrower perspective was chosen. We scored this item with 1 if the perspective was societal, or if the choice for a narrower perspective was properly motivated.

Since no standard instrument is available to assess the quality of COIs, included COIs were rated using a checklist provided in a previous systematic review on COIs in eating disorders (Stuhldreher et al., 2012). The checklist was developed based on recommendations by McGhan et al. (2009) and Drummond et al. (2015) and contains items on six different topics: (1) scope, (2) general economic criteria, (3) calculation of costs, (4) study design and analysis, (5) presentation of results and (6) discussion. Items of the CHEC and the quality assessment provided by Stuhldreher et al. (2012) were scored with *Yes* (score 1), *Suboptimal* (score 0.5), *No* (score 0), *NA* (not applicable) or *Uncertain* to obtain a more quantitative ranking. The first and second author independently assessed the quality of the included studies. The second author is a health economist and the first author a psychologist. Disagreements were discussed between the first and second author in consensus meetings until consensus was reached. Items were scored with *uncertain* only if the information was not clearly described in the article. Authors of the included studies were not contacted to clarify uncertainties.

## Outcomes

Incremental-cost effectiveness ratios (ICERs) were reported as the outcome for the included EEs. ICERs can be expressed as costs per quality-adjusted life year (QALY) gained or costs per any (clinical) outcome gained. If a study did not report an ICER, we presented the result of the EEs narratively. Since ICERs tend to be heterogeneous and are therefore not comparable, we did not pool the results. For COIs, direct, indirect and total costs reported in the studies were described for the BD group and (if applicable) also for a control group. If costs for a control group were also mentioned in the study, we also provided a ratio of costs between BD and the non-clinical group. Costs for the control group were only described if it concerned a non-clinical population. ICERs and costs were converted to 2018 international dollar (INT\$) by using Purchasing Power Parity (PPP) rates to account for varying price levels (Shemilt et al., 2010; World Bank, 2017) and then converted based on the Consumer Price Index provided by the US Bureau of Labor statistics (Bureau of Labor Statistics, 2018) to provide the most recent values adjusted for inflation rates.



**Figure 1.** Flowchart of the study selection process

## RESULTS

### Study selection

In total, the search yielded 2,363 studies and after removal of duplicates and screening of titles and abstracts, 65 full-texts were assessed for eligibility. Of these full-texts, 18 met inclusion criteria. The main reasons for exclusion of full-texts was that they did not present results specific for BD samples ( $n = 17$ ), were no CEA or COI studies ( $n = 17$ ), were conference papers or conference abstracts ( $n = 10$ ). Of the articles included in the final review, eight studies concerned EEs (Bauer et al., 2006; Camacho et al., 2017; Chisholm et al., 2005; Flood et al., 2006; Kessing et al., 2013; Lam et al., 2005; Scott et al., 2009; Simon et al., 2006) and ten studies were COIs (Broder et al., 2018; Cloutier et al., 2018; Correll et al., 2017; Degli Esposti et al., 2014; Ekman et al., 2013; Mennini et al., 2014; Pan et al., 2019; Parker et al., 2013; Somaiya et al., 2014; Wu et al., 2013). All studies investigating the cost-effectiveness of psychological interventions included in the earlier review on EEs in bipolar disorders (Abdul Pari et al., 2014) were also identified in the current review (Bauer et al., 2006; Chisholm et al., 2005; Lam et al., 2005; Simon et al., 2006). The study selection process is summarized in Figure 1.

### Overview of included EEs

#### *Main characteristics*

The most recent study was published in 2017 (Camacho et al., 2017) and the least recent studies in 2005 (Chisholm et al., 2005; Lam et al., 2005). All studies were published in western countries, namely the United Kingdom ( $n = 3$ ), United States ( $n = 2$ ), Denmark ( $n = 1$ ) and Spain ( $n = 1$ ). One study reported costs and outcomes globally for different regions (Chisholm et al., 2005). All EEs concerned RCTs, except one study which additionally conducted a MBEE to extrapolate results of a RCT (Camacho et al., 2017) and one study which applied a MBEE (Chisholm et al., 2005). Most studies used CEA, while two studies used CUA to investigate the cost-effectiveness of the intervention (Camacho et al., 2017; Chisholm et al., 2005). The perspective chosen by the studies concerned a healthcare perspective in most included studies, while one study explicitly stated that they chose a health and personal social services perspective (Camacho et al., 2017). Time horizon of the included EEs which applied TBEEs varied between 15 months (Flood et al., 2006) and five years (Scott et al., 2009). The only pure MBEE chose a life-time analytical horizon (Chisholm et al., 2005). The studies that reported their source of funding were not funded by industry and one study did not report their source of funding (Chisholm et al., 2005). Main characteristics of included EEs are described in Table 1.

**Table 1.** Main characteristics of included EEs

First author (year)	Country	Economic evaluation	Study type	Analysis	Perspective	Time horizon	Industry funding
Bauer (2006)	USA	TBEE	RCT	CEA	Healthcare	3 years	No
Camacho (2017)	UK	Combined <sup>a</sup>	Combined <sup>a</sup>	CUA	Health and Personal Social Services	96 weeks	No
Chisholm (2005)	Global	MBEE	Population model	CUA	Healthcare	Life-time analytical horizon	NR
Flood (2006)	UK	TBEE	RCT	CEA	Healthcare	15 months	No
Kessing (2013)	Denmark	TBEE	RCT	CEA	Healthcare	2 years	No
Lam (2005)	UK	TBEE	RCT	CEA	Healthcare	30 months	No
Scott (2009)	Spain	TBEE	RCT	CEA	Healthcare	5 years	No
Simon (2006)	USA	TBEE	RCT	CEA	Healthcare	2 years	No

Note. EEs = Economic evaluations, CUA = Cost-Utility Analyses, MBEE = Model-based-economic evaluation, NR = Not reported, RCT = Randomized controlled trial, TBEE = Trial-based-economic evaluation. <sup>a</sup>Both TBEE and MBEE components.

### *Methodological characteristics*

Included effect measurement concerned QALYs in one study (Camacho et al., 2017) and DALYs in one study (Chisholm et al., 2005). Other outcomes included admission to hospital ( $n = 2$ ), time spent in relapse or in any episode or hospitalized ( $n = 3$ ), relapse free years ( $n = 1$ ) and manic or depressive symptoms ( $n = 1$ ). Methodological characteristics of included EEs are summarized in Table 2.

Most studies involved people with a diagnosis of both BDI or BDII, while one study was specifically aimed at patients BDI experiencing frequent relapse currently not fulfilling criteria for a bipolar episode (Lam et al., 2005). One other study included patients with BD discharged from their hospital admission (Kessing et al., 2013). One study included veterans with BD (Bauer et al., 2006) and two studies included euthymic patients (Scott et al., 2009; Camacho et al., 2017), one of them patients at an increased risk for relapse (Camacho et al., 2017). Two studies contained any patients with BD (Chisholm et al., 2005; Flood et al., 2006) and one study patients with any diagnosis of bipolar spectrum disorder (Simon et al., 2006).

Economic evaluations included in this review used an array of different interventions for BD. The Bipolar Disorders Program (Bauer et al., 2006) was described as collaborative care program including for example enhancement of patient skills in self-managing the illness by psychoeducation. The joint crisis plan intervention (Flood et al., 2006) consisted of meetings with healthcare professionals aimed at generating a crisis plan. The systematic care program for BD (Simon et al., 2006) contained, for example, care planning and structured group psychoeducation. One study concerned an intervention program from an outpatient mood disorder clinical group program (Kessing et al., 2013). Two studies examined the effect of structured psychoeducation (Scott et al., 2009; Camacho et al., 2017) and one study used Cognitive Behavioral Therapy (CBT; Lam et al., 2005). None of the included articles mentioned that TAU was withheld from the intervention arm or constrained. Therefore it can be assumed that TAU was part of all the interventions included in this review.

Four of the included studies compared the effect of the psychological intervention versus treatment as usual only (TAU) only, while one study used unstructured psychoeducation and TAU as comparator (Scott et al., 2009) and another study used unstructured group peer-support and TAU as treatment alternative (Camacho et al., 2017). One study used standardized service information and TAU as treatment alternative (Flood et al., 2006) and

**Table 2.** Methodological characteristics of included EEs

First author (year)	Target population	Treatment alternatives (n)	% female	Mean age	Study type	Outcomes	Discount rate	Valuation year, original currency	Description of treatment alternatives
Bauer (2006)	Veterans with BD	I: Bipolar Disorders Program (166), II: TAU (164)	NR	NR	RCT	Weeks in any episode, mental and physical	3.0%	2004, USD	I: Collaborative care program with a specialty team, including the enhancement of self-management via group psychoeducation, evidence-based pharmacotherapy and access to care. II: Patients continued with TAU.
Camacho (2017)	Euthymic patients with BD at increased risk for relapse	I: Bipolar group structured PE + TAU (153), II: Unstructured bipolar group peer-support + TAU (151)	58.0	45.0	RCT	QoL (SF-36) EQ-5D-3L), relapse free years, relapse avoided	NR <sup>a</sup>	2012, pound	I: Weekly (21 sessions) structured bipolar group PE delivered by two healthcare professionals, each session included specific handouts and homework tasks. II: Weekly (21 sessions) unstructured peer-support groups delivered by healthcare professionals and a service-user facilitator, including a short manual.
Chisholm (2005)	Patients with BD	I: Lithium or valproic acid + psychosocial care; lithium or valproic acid alone, II: No treatment	NA	NA	Population model	DALYs	3.0%	2003, INT\$	I: Four treatments are included in the model representing either pharmacotherapy alone or in combination with psychosocial care: Lithium + psychosocial care, valproic acid + psychosocial care, lithium alone and valproic acid alone. II: The comparator is no treatment.
Flood (2006)	Patients with BD	I: Joint crisis plan + TAU (80), II: standardized service information + TAU (80)	53.0	39.1	RCT	Admission to hospital	NR <sup>b</sup>	2001, pound	I: During two sessions a joint crisis plan was developed, which contains a set of statements of what to do in a crisis. The patient was encouraged to bring a friend or advocate to the second meeting to work on the plan together. II: Patients received leaflets about local services and mental health.

**Table 2.** Methodological characteristics of included EEs (continued)

First author (year)	Target population	Treatment alternatives (n)	% female	Mean age	Study type	Outcomes	Discount rate	Valuation year, original currency	Description of treatment alternatives
Kessing (2013)	Patients with BD, discharged from hospital	I: Specialized mood disorder clinical group (72), II: TAU (86)	42.9	36.3	RCT	Readmission to hospital	NR	2006, EUR	I: Specialized out-patient clinic offering a combined treatment of evidence-based pharmacological treatment and group PE and psychotherapy for 2 years. Participants followed three sequential PE and psychotherapeutic group sessions. II: Patients continued with TAU.
Lam (2005)	Patients with BDI and frequent relapse	I: CBT (51), II: TAU (52)	56.5	44.0	RCT	Number of days in bipolar episode	NR	2001, pound	I: Cognitive therapy delivered by clinical psychologists for 12 to 18 individual sessions within 6 months and two additional booster sessions in the second 6 months. II: Patients continued with TAU.
Scott (2009)	Euthymic patients with a lifetime-diagnosis of bipolar disorder I or II	I: Structured PE + TAU (60), II: Unstructured PE + TAU (60)	63.3	34.1	RCT	Number of days spent in relapse or hospitalized	NR	2006, EUR	I: Structured group PE (21 sessions, 1.5 hours per session) run by two psychologists, aimed at improving illness awareness, treatment compliance, early detection of symptoms and lifestyle, II: Unstructured group meetings with the two psychologists, who tried to not give psychoeducational feedback except for what was necessary.
Simon (2006)	Patients with any diagnosis of bipolar spectrum disorder	I: Systematic care program for BD (212), II: TAU (229)	68.3	44.2	RCT	Manic and depressive symptoms (PSR; SCID interviews)	NR	2000, USD	I: Multicomponent intervention provided by three nurse care managers (24 months), aimed at care planning, structured monthly telephone calls, feedback to the mental health treatment team, structured group PE, as-needed support, education and care coordination. II: Patients continued with TAU.

*Note.* BD = Bipolar disorder, CBT = Cognitive Behavioral Therapy, DALYs = Disability-adjusted life-years, EQ-5D-3L = EuroQol 5D – 3 level version, EUR = Euro, ICFERs = Incremental cost-effectiveness ratios, INTd = International dollar, NA = Not applicable, NR = Not Reported, PE = Psychoeducation, PSR = Psychiatric Status Rating, RCT = Randomized controlled trial, SCID = Structured Clinical Interview for DSM, SF-36 = 36-item Short Form Survey, QALYs = Quality-adjusted life years, QoL = Quality of Life, TAU = Treatment as usual, USD = US dollar. <sup>a</sup>Due to the relatively short study period future costs and outcomes were not discounted. <sup>b</sup>Due to a lack of data on the timing of resource costs that occurred after the first 12 months of the trial costs could not be discounted.



one study investigated the effect of pharmacotherapy plus psychosocial care and pharmacotherapy only in comparison with no treatment (Chisholm et al., 2005). The studies that described TAU in their studies outlined it in a similar way, mainly consisting of pharmacotherapy, psychoeducation and supportive sessions.

### *Outcomes*

The main outcomes of included EEs are summarized in Table 3. Of the included EEs, four studies reported an ICER. Of these articles, one study estimated the costs for one additional QALY gained at \$75,106 and one relapse free year at \$13,187 for bipolar group structured psychoeducation and TAU compared to unstructured peer-supported psychoeducation and TAU (Camacho et al., 2017). One other study estimated the cost-effectiveness of joint crisis plans and concluded that the intervention led to less mean total costs per patient and was more effective in reducing admission to hospital compared to standardized service information in addition to TAU in the UK (Flood et al., 2006). Another study from Spain reported fewer total costs for structured psychoeducation and TAU compared with unstructured psychoeducation and TAU for the study period of 5 years. Also, the intervention resulted in less days in relapse and less mean hospital admissions. The intervention thus dominated the control group over the study period of five years (Scott et al., 2009). The Bipolar Disorders Program resulted in less weeks in any episode at higher outpatient costs compared to TAU only in the US. However, mean total costs were lower for the intervention arm over the study period of 3 years (Bauer et al., 2006). Treatment in the mood disorder clinic group resulted in fewer hospital admissions, decreased inpatient costs and lower two-year total treatment costs compared to TAU in Denmark (Kessing et al., 2013). The systematic care program for BD led to less manic symptoms, but not depressive symptoms at higher total costs compared to TAU in the US (Simon et al., 2006). One other intervention in the UK (cognitive behavioral therapy) led to fewer total costs at both 12 and 30 months after baseline. The higher costs of the intervention were offset by lower costs for other services. Cost-effectiveness analyses suggested that, if the value for one bipolar-free day was set at \$21, the probability that the intervention was cost-effective was about 85% for a period of 12 months and about 80% for a period of 30 months (Lam et al., 2005).

The only MBEE included in this review (Chisholm et al., 2005) compared the cost-effectiveness of four different interventions with no treatment; lithium plus psychosocial care, lithium alone, valproic acid plus psychosocial care and valproic acid alone.

**Table 3.** Outcomes of included EEs (in PPP-INT\$)

First author (year)	Incremental health benefits per patient	Incremental costs per patient	ICER	Description of outcomes
Bauer (2006)	6.2 fewer weeks in any episode; improvement in mental QoL	-\$3,879, 95%CI(-\$20,861-\$13,796)	Dominant	Intervention resulted in less weeks in any episode and improved mental functioning at lower 3-year total costs compared to the control group (\$79,901 versus \$83,780); ICER NR.
Camacho (2017)	0.023 QALYs; 0.131 relapse free years; 0.102 Relapse avoided during follow-up	\$1,727, 95%CI(\$396-\$3,056)	\$75,106/QALY; \$13,187/relapse free year; \$16,931/relapse avoided	Additional costs of \$1,727 in healthcare resources used for PE group; QALY gain of 0.023 in the PE group, 0.131 relapse free years and 0.102 relapse avoided; ICER group PE versus control; \$75,106/QALY gained; \$13,187/relapse free year and \$16,931/relapse avoided.
Chisholm (2005)	Fewer disability free days per year <sup>a</sup> : Li+psy (59.6-67.3); Va+Psy (58.4-63.3); Li (54.5-62.1); Va (53.1-60.2)	Costs per treated case, coverage rate of 50%: Hospital-based <sup>a</sup> : Li+psy (\$1,091-\$9,627); Va+Psy (\$1,208-\$9,409); Li (\$1,068-\$9,493); Va (\$1,181-\$9,235) Community-based <sup>a</sup> : Li+psy (\$719-\$5,599); Va+Psy (\$849-\$5,531); Li (\$697-\$5,465); Va (\$821-\$5,344)	Hospital-based <sup>a</sup> : Li+psy (\$4,096-\$47,611/DALY); Va+psy (\$5,650-\$48,572/DALY); Li (\$4,324-\$51,252/DALY); Va (\$5,959-\$52,540/DALY) Community-based <sup>a</sup> : Li+psy (\$2,894-\$28,245/DALY); Va+psy (\$4,212-\$29,779/DALY); Li (\$3,028-\$30,110); Va (\$4,403-\$31,036)	Hospital-based service model: Lithium + psychosocial care had a lower ICER than valproic acid + psychosocial care compared with no treatment; Community-based service model: Lithium + psychosocial care had a lower ICER than valproic acid + psychosocial care compared with no treatment. Lithium alone had a higher ICER than lithium + psychosocial care compared to no treatment in both models; Valproic acid alone had a higher ICER than valproic acid + psychosocial care compared to no treatment in both models.
Flood (2006)	0.69 fewer hospital admissions	-\$2,286, 95%CI(-\$10,445-\$5874)	Dominant: \$273/reduction of 1% of patients admitted to hospital	Decreased mean total costs per patient in the intervention group (\$15,161 versus \$17,448, nonsignificant); intervention was cheaper and more effective; ICER joint crisis plan versus control group; \$273 per 1% reduction in the proportion of patients admitted to hospital.
Kessing (2013)	0.61 fewer hospital readmissions after discharge	-\$4,036	Dominant	Intervention resulted in fewer hospital readmissions after discharge, decreased inpatient costs (\$18,302 versus \$27,175) and lower two-year total treatment costs (\$32,787 versus \$36,823); ICER NR.

**Table 3.** Outcomes of included EEs (in PPP-INT\$) (continued)

First author (year)	Incremental health benefits per patient	Incremental costs per patient	ICER	Description of outcomes
Lam (2005)	110 fewer days spent in bipolar episode	-\$2,881	Dominant	Intervention led to 110 fewer days with bipolar episodes (95.3 versus 201.0 days) in 30 months at lower total costs (\$21,739 versus \$24,620); if society is willing to pay \$21 per bipolar free day CBT is more cost effective with 85% probability over the study period of 30 months; ICER NR.
Scott (2009)	433 fewer days spent in relapse, 0.35 fewer admissions to the hospital	-\$5,626, 95%CI(-\$16,538-\$5,283)	Dominant: ICER based on outpatient costs: \$6,824/relapse free person and \$5,882/hospitalization-free person	Intervention resulted in less days in relapse (153.73 versus 586.45), less mean admission to the hospital (0.24 versus 0.59) at higher outpatient but lower total costs (\$29,734 versus \$35,361); Structured PE + TAU dominated the control group over the study period of 5 years.
Simon (2006)	Fewer manic symptoms; 5.5 fewer weeks with clinically significant mania symptoms	\$1,846, after adjustment for several baseline covariates: \$1,774, 95%CI (\$78-\$3,470)	NR	Intervention resulted in less manic symptoms; no difference was found for depressive symptoms; intervention group resulted in higher total costs (\$11,396 versus \$9,550); ICER NR.

Note. BD = Bipolar disorder, CI = Confidence interval, CBT = Cognitive Behavioral Therapy, EEs = Economic evaluations, DALYs = Disability-adjusted life-years, EQ-5D-3L = EuroQol 5D – 3 level version, ICER = Incremental cost-effectiveness ratio, Li = Lithium, Li+psy = Lithium plus psychosocial care, NA = Not applicable, NR = Not reported, PE = Psychoeducation, PSR = Psychiatric Status Rating, QALYs = Quality-adjusted life years, QoL = Quality of life, RCT = Randomized controlled trial, SF-36 = 36-item Short Form Survey, Va = Valproic acid, Va+psy = Valproic acid plus psychosocial care. <sup>a</sup>Numbers between brackets represent the range of estimates for different subregions and for the corresponding treatment alternative.

Compared to no treatment, the additional costs of lithium plus psychosocial care were estimated to be \$4,096 to \$47,611 per additional DALY averted in a hospital-based service model and at \$2,894 to \$28,245 in a community-based service model, depending on the subregion in which the costs are assumed to occur (e.g. developing or non-developing regions). Compared with no treatment, lithium plus psychosocial care had a lower ICER than lithium alone. Valproic acid plus psychosocial compared with no treatment was estimated to lead to \$5,650 to \$48,572 additional costs in a hospital-based service model and to \$4,212 to \$29,779 in a community-based service model. Valproic acid alone had a higher ICER than treatment with valproic acid and psychosocial care compared with no treatment. Among all treatment alternatives, lithium plus psychosocial care had the lowest ICER compared to no treatment. Furthermore, lithium alone had a lower ICER than valproic acid plus psychosocial care or valproic acid alone compared with no treatment.

## Overview of included COIs

### *Main characteristics*

Seven of the included COIs were conducted in western countries, including the United States (n = 3), Italy (n = 2), Sweden (n = 1) and Australia (n = 1). One study was conducted in India (Somaiya et al., 2014) and two in Taiwan (Pan et al., 2019; Wu et al., 2013). All articles, except one, reported diagnostic criteria for the target group included in the studies. Seven studies used criteria of the International Classification of Diseases (ICD) and two of the Diagnostic Statistical Manual of Mental Disorders (DSM). The approach to determine the costs of BD was prevalence-based in all of the included studies. Of all included COIs, five studies took a societal perspective and five a healthcare perspective. The perspective chosen was explicitly mentioned in two studies only (Cloutier et al., 2018; Ekman et al., 2013) and took a societal perspective in both cases. Six of the included COIs used national or insurance databases as data source. Two studies used databases from hospitals to estimate costs related with BD (Ekman et al., 2013; Somaiya et al., 2014), of which one study combined it with self-report questionnaire data from patients (Somaiya et al., 2014). Two studies used purely self-report questionnaire data to obtain costs (Mennini et al., 2014; Parker et al., 2013). The description of costs was based on per capita costs in all of the COI studies. One study additionally estimated national costs related with BD. Five of the studies were funded by industry, four studies were not (Pan et al., 2019; Parker et al., 2013; Somaiya et al., 2014; Wu et al., 2013) and one study did not report funding source (Mennini et al., 2014). Main characteristics of included COIs are summarized in Table 4.

**Table 4.** Main characteristics of included COIs

First author (year)	Country	Diagnostic criteria	Approach	Perspective	Data source	Valuation year, original currency	Description of costs	Industry funding
Broder (2018)	USA	ICD-9, ICD-10	P	Healthcare	Truven Health Analytics MarketScan databases	2016, USD	PC	Yes
Cloutier (2018)	USA	ICD-9, ICD-10	P	Societal	Truven Health Analytics MarketScan databases	2015, USD	PC/NC	Yes
Correll (2017)	USA	ICD-9	P	Healthcare	Premier Perspective Database	2014, USD	PC	Yes
Degli Esposti (2014)	Italy	ICD-10	P	Healthcare	Health-assisted Subjects' Database, Medications Prescription Database, Hospital Discharge Database, Mental Health Information System, Specialist Outpatient Services Database, No Charges Database	2010, EUR	PC	Yes
Ekman (2013)	Sweden	ICD-10	P	Societal	Management system Northern Stockholm Psychiatry, Swedish Prescribed Drug Registry of the National Board of Health and Welfare, Social Insurance Authority	2009, EUR	PC	Yes
Mennini (2014)	Italy	NR	P	Societal	Self-reported data from a longitudinal prospective study	2012, EUR	PC	NR
Pan (2019)	Taiwan	ICD-9	P	Healthcare	National Health Insurance Research Database	2008, NTW	PC	No
Parker (2013)	Australia	DSM-IV	P	Societal	Self-reported questionnaire data <sup>a</sup>	2011, AUD	PC	No
Somaiya (2014)	India	DSM-IV	P	Societal	Hospital statistics, bills, payment slips and self-report	2011, Indian rupee	PC	No
Wu (2013)	Taiwan	ICD-9	P	Healthcare	National Health Insurance Research Database	2007, USD	PC	No

Note. AUD = Australian Dollar, COIs = Cost-of-illness studies, DSM = Diagnostic Statistical Manual of Mental Disorders, EUR = Euro, ICD = International Classification of Diseases, NC = National costs, NTW = New Taiwan Dollar, P = Prevalence-based, PC = Per capita, USD = US Dollar. <sup>a</sup>Participants were only asked at one measurement point.

### *Methodological characteristics*

Methodological characteristics and outcomes of included COIs are summarized in Table 5. Four studies focused on BDI (Broder et al., 2018; Cloutier et al., 2018; Correll et al., 2017; Mennini et al., 2014). One of these studies (Correll et al., 2017) also included patients with BD and cardiometabolic comorbidities, but for the present review only the estimated costs for patients without cardiometabolic comorbidities were extracted. Five studies included samples with both BDI and BDII (Degli Esposti et al., 2014; Ekman et al., 2013; Pan et al., 2019; Parker et al., 2013; Somaiya et al., 2014). One study included patients with BD and intellectual disabilities and cost estimates for patients with BD only. We used only the latter estimates for this review (Wu et al., 2013). Another study estimated direct costs for two different age groups in Taiwan, namely for people with BDI and BDII aged 18-64 and aged 65 or older (Pan et al., 2019).

Direct costs related to BD were estimated in nine of the included COIs, one study merely estimated indirect costs but no direct costs (Mennini et al., 2014). Five studies estimated indirect costs. Total costs (direct and indirect costs combined) were estimated in four studies. In every study estimating direct costs, these costs included the costs of drugs and in six studies also outpatient and inpatient costs (Broder et al., 2018; Cloutier et al., 2018; Ekman et al., 2013; Pan et al., 2019; Parker et al., 2013; Wu et al., 2013). Three studies assessed costs of inpatient care only, instead of inpatient and outpatient costs (Correll et al., 2017; Degli Esposti et al., 2014; Somaiya et al., 2014).

All five studies which estimated indirect costs, included productivity loss in their cost estimation. Three studies also included other indirect costs, for example unemployment and caregiving (Cloutier et al., 2018) or disability pension (Parker et al., 2013). One study included a non-clinical control group to compare the costs associated with BD to a healthy population (Cloutier et al., 2018).

### *Outcomes*

Four studies reported costs for patients with BDI and three of these estimated direct costs, including a range of \$7,604 and \$27,617 per capita per year (Broder et al., 2018; Cloutier et al., 2018; Correll et al., 2017). Indirect costs for BDI ranged between \$9,065 in Italy (Mennini et al., 2014) and \$62,343 per year per capita in the US (Cloutier et al., 2018). One study also calculated the societal costs for BDI and estimated direct costs of \$59,050,087,989 and indirect costs of \$154,449,703,726 per year in the in the United States.

**Table 5. Methodological characteristics and outcomes of included COIs (in PPP-INT\$)**

First author (year)	Target population (n)	% female	Mean age	Bipolar disorder				Controls		Ratio BD / Control group
				Direct costs include	Direct costs per annum	Indirect costs include	Indirect costs per annum	Total costs Per annum	Total costs per annum	
Broder (2018)	People with BDI (51,480)	64.0	41.6	Outpatient, Inpatient, Drugs	\$27,617 per capita	-	-	-	-	-
Cloutier (2018)	I: People with BDI (2,477,737) <sup>a</sup> , II: Healthy US population	66.4	41.4	Outpatient, Inpatient, Drugs, Others	\$59,050,087,989 (societal <sup>b</sup> ); \$23,832 per capita <sup>c</sup>	Productivity loss Unemployment Carregiving Others	\$154,449,703,726 (societal <sup>b</sup> ); \$62,343 per capita <sup>c</sup>	\$213,499,791,715 (societal); \$86,175 per capita <sup>c</sup>	\$86,977,430,290 (societal <sup>b</sup> )	2.5
Correll (2017)	People with BDI (61,777) <sup>d</sup>	63.0 <sup>e</sup>	45.4 <sup>e</sup>	Inpatient, Drugs	\$7,604 per capita <sup>f</sup>	-	-	-	-	-
Degli Esposti (2014)	People with BD (5,486)	59.0	52.0	Inpatient, Drugs, Others	\$5,118 per capita	-	-	-	-	-
Ekman (2013)	People with BD (1,846)	59.0	50.1	Inpatient, Outpatient, Drugs	\$7,160 per capita	Productivity loss	\$24,477 per capita	\$31,637 per capita	-	-
Mennini (2014)	People with BDI (265)	54.9	50.0	-	-	Productivity loss	\$9,056 (low income class); \$17,585 (high income class)	-	-	-
Pan (2009)	I: People with BDI or BDII aged 18-64 (13,105), II: People with BDI or BDII aged ≥ 65 (2,149);	I: 60.3, II: 58.8	I: 40.0, II: 74.7	Inpatient, Outpatient, Drugs, Emergency attendances	I: 6,617 per capita, II: 10,425 per capita	-	-	-	-	-

**Table 5.** Methodological characteristics and outcomes of included COIs (in PPP-INT\$) (continued)

First author (year)	Bipolar disorder					Controls		Ratio		
	Target population (n)	% female	Mean age	Direct costs include	Direct costs per annum	Indirect costs include	Indirect costs per annum		Total costs Per annum	Total costs per annum
Parker (2013)	People with BDI (44) or BDII (102)	59.6	35.8	Outpatient, Inpatient, Drugs, Others	\$19,036 (BDI) per capita, \$12,699 (BDII) per capita	Productivity loss Disability pension Others	\$97,026 (BDI); \$55,492 (BDII)	\$116,062 (BDI) per capita; \$68,191 (BDII) per capita	-	-
Somaiya (2014)	People with BD (75)	30.7	34.3	Inpatient, Drugs, Others	\$881 per capita	Productivity loss Others	\$1,568 per capita	\$2,449 per capita	-	-

Note. BD = Bipolar disorder, COIs = Cost-of-illness studies. <sup>a</sup>The adult US BDI population was estimated based on a prevalence of 1.0%. <sup>b</sup>Societal costs = estimated annual costs for the US that is caused by BDI. <sup>c</sup>Costs per capita were not provided in the study and were calculated by dividing the societal costs by the assumed number of individuals with BDI. <sup>d</sup>Actual sample size was larger, but also included people with cardiometabolic comorbidities. For the present study, only costs for patients without cardiometabolic comorbidities were extracted. <sup>e</sup>Based on the whole sample (incl. cardiometabolic comorbidities). <sup>f</sup>Not clear for which time period the costs were calculated



Estimated costs were 2.5 times higher compared to a non-clinical US population (Cloutier et al., 2018). Another study reported the costs for BDI and BDII separately and estimated the direct costs of BDI at \$19,036 and for BDII at \$12,699 and indirect costs for BDI at \$97,026 and for BDII at \$55,492 per year per capita in Australia (Parker et al., 2013). Direct costs for samples including both BDI and BDII ranged between \$881 in India (Somaiya et al., 2014) and \$10,425 in Taiwan for people aged 65 or older (Pan et al., 2019) per capita per year. Indirect costs were estimated at \$1,568 in India (Somaiya et al., 2014) and at \$24,477 per capita per year in Sweden (Ekman et al., 2013). The study by Wu et al. (2013) estimated costs for patients with BD with and without intellectual disability and estimated the total direct costs at \$9,065 per year per capita in Taiwan for patients with BD without intellectual disability.

## **Quality assessment**

The quality assessment of included EEs revealed an average study quality of 74%, with a range of 68% to 83%. Across all included studies, the two items on 'appropriate discounting' (item 15) and 'generalizability of results' (item 18) had the lowest scores. Merely one study appropriately discounted future costs and outcomes and only two studies discussed generalizability of the findings properly. Items regarding the economic study design and the chosen time horizon had the highest scores. These items were considered appropriate for all included EEs.

For included COIs, the average study quality was 70.6%, with the quality of included studies ranging between 41% and 88%. The lowest scores were found for whether a non-diseased comparison group was used and whether a perspective was reported. Merely one study contained a non-clinical control group, while only two studies mentioned the perspective. The highest scores were found for the items on study objective, diagnostic criteria, analysis of costs that accrued from a particular disease, currency, source of healthcare utilization, sample size reported, sample characteristics described, and limitations discussed in detail. These items were rated as sufficient for all included COIs. Results of the quality assessment are summarized in Table A1 and A2 (Appendix A).

## DISCUSSION

In all of the included EEs, the interventions resulted in improved clinical outcomes and five studies additionally concluded decreased total costs (Bauer et al., 2006; Flood et al., 2006; Kessing et al., 2013; Lam et al., 2005; Scott et al., 2009). Thus, in five of the included studies, the intervention dominated its comparator over the study period. Overall, these findings indicate that psychological treatments may reflect a (cost)-effective option for the treatment of BD. However, the results should be interpreted with caution, as the studies used several different outcomes to assess the (cost)-effectiveness of their interventions, such as admissions to hospital, number of days with BD or symptomatology and also different types of comparators. Furthermore, healthcare costs were retrieved in different ways, for example by self-report questionnaires or computerized pharmacy registration data. This makes it difficult to compare the EEs included in this review and also makes generalizability and transferability to other settings difficult.

Of the included EEs, one study expressed the cost-effectiveness of an intervention as ICER in relation to QALYs (Camacho et al., 2017). In this study, structured group psychoeducation as addition to TAU led to an increase in QALYs compared to TAU in combination with unstructured peer-supported psychoeducation. One additional QALY gained was estimated to cost \$75,106. This is higher than the willingness-to-pay threshold (WTP-T) in the UK (approx. \$25,000-\$40,000), but still within the range of the WTP-T for the Netherlands (\$20,000-\$80,000), for example. The relatively small increase in QALYs could be explained by the included quality of life instrument (EQ-5D) or due to the lack of additional effect of the intervention. It is debatable whether the EQ-5D is sensitive to change in people with mental disorders and assesses all relevant aspects of quality of life for this target group (Brazier, 2010; Longworth et al., 2014). Another included study assessed quality of life with the SF-36, but they did not need to report an ICER (Bauer et al., 2006) as the intervention (Bipolar disorder program) resulted in fewer 3-year costs and a significant improvement in quality of life and thus was dominant compared to TAU.

The only MBEE included in this review estimated the cost-effectiveness of lithium and psychosocial care and valproic acid and psychosocial care compared to no treatment (Chisholm et al., 2005). One additional DALY averted was estimated to cost up to \$47,611 for lithium and psychosocial care compared to no treatment, while the other treatment alternatives (lithium alone, valproic acid plus psychosocial care and valproic acid alone) were estimated to have higher ICERs compared to no treatment. This is also in line with an earlier review on economic evaluations in BD, concluding that combined therapies (pharmacological and psychotherapy) might be important for this target group. Psychotherapy can be effective in enhancing the

patients' awareness, compliance and thereby prevent relapse and hence improve overall treatment outcomes (Abdul Pari et al., 2014).

While the current review was conducted, another systematic review about EEs of psychological interventions for people with BD and/or schizophrenia has been published (Shields et al., 2019). Interestingly, merely two studies that were included as EEs in our study were also included in their study (Camacho et al., 2017; Lam et al., 2005). One of the main reasons why this review has included more studies are the broader eligibility criteria regarding outcomes compared to the study by Shields et al. (2019), in which the authors decided to only include full EEs including an ICER or some measure of net benefit that incorporates health outcomes. Studies also had to include a 'cost-effectiveness acceptability curve' (CEAC) or explicitly report probabilities of being cost-effective to allow for an assessment of uncertainty (Shields et al., 2019). In general, however, the conclusions by Shields et al. (2019) were similar to the current study, namely that non-pharmacological interventions for BD might be cost-effective, but that studies remain scarce and that there was also great heterogeneity in study characteristics, which makes it difficult to reach strong conclusions.

One other review about EEs of pharmacological treatments for BD has been conducted recently (Mavranouzouli & Lokkerbol, 2017). In general, they concluded that pharmacological interventions are cost effective compared with no treatment. Specifically, antipsychotic drugs in combination with lithium or valproate appear to be most cost effective for acute manic, mixed or depressive phases compared with no treatment. For maintenance treatment, lithium appears to be cost effective compared with no treatment. However, they also note that results are difficult to compare across studies due to high heterogeneity regarding methodological and intervention characteristics, which is in line with our findings regarding non-pharmacological interventions. Combining their findings with results from the current review, it can be concluded that both pharmacological interventions and psychological interventions might be cost-effective compared to no treatment. However, specific conclusion about the cost-effectiveness between psychological and pharmacological treatment cannot be drawn. Although a direct comparison of psychological and pharmacological interventions would be difficult to research in acute phases, future research might address this question for example in maintenance phases of the disorder.

It is notable that all included EEs in the current review used a time horizon of one year or longer to estimate the cost-effectiveness of their intervention. This allows cost-effectiveness to be estimated over a substantial period of time. In prior reviews, for example on EEs in anxiety disorders (Ophuis et al., 2017), the percentage of studies using an appropriate time horizon of at least one year was substantially smaller. The average study quality was 74%, which is

comparable to a prior review on EEs in anxiety disorders (Ophuis et al., 2017) and suggests that studies adhered to most of the quality criteria as outlined by the CHEC. However, most studies lacked discounting and a clear discussion of the generalizability of the findings. Therefore, we encourage future research to properly discount the costs and discuss the generalizability of their results.

In total, ten COIs were included in the current review. We found a wide range of direct costs (\$881-\$27,617) and of indirect cost estimates per capita per year (\$1,568-\$116,062). These findings coincide with earlier reviews on this topic, which also identified a wide range of cost estimates and heterogeneity in characteristics of the studies (Jin & McCrone, 2015; Kleine-Budde et al., 2014). Compared to the findings of the review by Kleine-Budde et al. (2014), direct cost estimates per capita were relatively comparable for most studies. It is notable that only four out of 27 studies assessed indirect costs per capita in the prior review. Of these four studies, conducted in the US and the Netherlands, indirect cost estimates were considerably lower compared to findings in our review. When comparing our findings regarding COIs with the review by Jin and McCrone (2015), a similar picture emerges. Direct costs identified were comparable, but the authors also note that there has been inconsistency in the way the studies were conducted and limitations in their design. Similar to the review by Kleine-Budde et al. (2014), relatively few studies reported indirect costs. Since five out of nine studies in the current review reported indirect costs, this suggests that the number of studies reporting indirect costs has increased. In our study, the lowest costs were estimated by Somaiya et al. (2014), which might be attributed to the fact that this study was carried out in India, reflecting a different healthcare system compared to other included COIs. Another possible explanation for the wide range of costs might be that some studies assessed costs more comprehensively than others. For example, while some studies assessed outpatient, inpatient, drugs and other costs (e.g. Parker et al., 2013), some studies merely assessed hospitalizations and drugs as direct costs (e.g. Correll et al., 2017). Also, indirect costs were estimated in different detail. Similar to the findings regarding EEs, the heterogeneity in outcomes, specific target group (BDI or BDII), general characteristics, such as country and data source, makes it difficult to compare the results of the included COIs. One study included in this review estimated the national total costs of BDI in the United States (Cloutier et al., 2018). They estimated the costs at around \$213 billion per year and concluded that BDI leads to 2.5 times higher costs compared to a control population. These findings are higher than in a prior study by Dilsaver (2011), in which the total costs for BDI were estimated at approximately \$60 billion dollar in 2009 in the US. Cloutier et al. (2018) explain this discrepancy by the fact that the study by Dilsaver (2011) used data and assumptions from a study from 1995 (Wyatt & Henter, 1995), not taking into account any of the changes in the clinical management and classifications since then, which might be an explanation for the discrepancy in costs reported in those two studies.

In general, we could identify four additional studies investigating the cost-effectiveness of non-pharmacological interventions in BD since a prior review in this field has been undertaken (Abdul Pari et al., 2014). Also, we could identify ten additional COIs since the prior reviews have been conducted (Jin & McCrone, 2015; Kleine-Budde et al., 2014). This suggests that research has been conducted in this research field in the past years. However, the methodology of the studies remained heterogenous, which is in line with the earlier reviews. Especially for EEs, the evidence-base remains relatively small. Due to the lack of published studies including EEs of non-pharmacological interventions for BD interpretation of the findings should be done carefully. Our findings suggest that, for example, cognitive behavioral therapy, structured group psychoeducation or a joint crisis plan might be cost-(effective) treatment alternatives that might improve clinical outcomes and also lead to lower costs. However, considering that the eight included EEs only cover a small range of psychological treatment alternatives and revealed high heterogeneity in study characteristics, it is difficult to prioritize interventions and provide general advice for policy makers on which intervention they should choose. This heterogeneity in outcomes and study characteristics also makes it difficult to pool the results. In this context, it is also notable that the cost-effectiveness of interventions based on more novel developments in clinical psychology have not been conducted, such as third-wave cognitive behavioral therapies or recovery-based approaches (Hayes, 2004; Jones et al., 2012; Jones et al., 2015; Rashid, 2015). Although recently published study protocols are promising (e.g. Hanssen et al., 2019; Kraiss et al., 2018), there remains a lack of evidence in this research domain.

## **Strengths and limitations**

The current study has several strengths. One strength is that we followed the PRISMA-guidelines (Moher et al., 2015), currently reflecting the golden standard for conducting systematic reviews. Moreover, we followed rigorous guidelines for the preparation of our review (Wijnen et al., 2016) and conducted a comprehensive quality assessment (Evers et al., 2005; Stuhldreher et al., 2012).

Our study also has a number of limitations. First, for both COIs and EEs, a high heterogeneity was found for general and methodological characteristics. Therefore, the results are difficult to compare, and no attempt has been made to pool the results. In line with this, all EEs, apart from the study by Chisholm et al. (2005), and most COIs were conducted in western countries, which also limits the generalizability of the results outside western countries. However, the present review still provides a valuable aggregation of the existing evidence in this research field. Second, we merely included COIs explicitly describing the costs related with BD, but not studies that examined health care utilization only and also not government or institutional

reports or other grey literature, which might have helped to better understand the burden associated with BD. We also focused on EE literature only, that might have led to the exclusion of studies examining the effect of intervention on, for example, bed days or emergency department use. Third, due to the descriptive nature of this review, we could not check for the presence of publication bias. Since positive results are more likely to be published, the presence of publication bias (especially in EEs) cannot be excluded. Fourth, our search strategy did not contain an extensive list of terms related to COIs (e.g. burden, economic burden or expenditure) and we therefore might have missed studies reporting cost-of-illness in BD (Higgins et al., 2019). Also, other sources of health economic studies were not included in the search (e.g. the European Network of Health Economic Evaluation Databases), although also including them in the search might have led to the identification of additional studies.

## Implications

Several implications and recommendations arise from the current findings. A relatively low number of studies could be identified for EEs. This shows that the evidence in the field of economic evaluations in BD, specifically for non-pharmacological interventions, is sparse. We therefore encourage future research to investigate the cost-effectiveness of non-pharmacological interventions for BD. Although the quality of the included EEs was comparable to a prior review in anxiety disorders (Ophuis et al., 2017), we still found a high variability in study quality and also in which sections of the quality assessments were conducted properly. Conducting high quality studies improves the validity of research and we therefore encourage future research to take quality assessments, for example the CHEC (Evers et al., 2005), into account when conducting research on cost-effectiveness. This would increase standardization of methodology across studies and thereby making them less heterogenous. The study by Camacho et al. (2017) can be highlighted as positive example, since it had the highest quality according to our rating. This study can thus be seen as a valid example for future research. Moreover, we recommend that future research should clearly mention their perspective. In this context, a societal perspective is desirable since indirect costs (e.g. productivity loss) caused by BD are substantial and should therefore not be ignored. However, whether a healthcare or societal perspective should be chosen always depends on the context of the study and the decision-makers involved.

We also encourage future studies to use quality of life measures, for example the EQ-5D or SF-36 and report ICERs in relation to these measures. This would make the results more comparable across different studies and conditions, especially because WTP thresholds are available for QALYs, supporting decision making processes. When using the EQ-5D however, one should keep in mind that the sensitivity to change might be limited, specifically when

examining effects of intervention, and may want to use additional, more elaborative, quality of life instruments. Finally, one should be aware that, as with RCTs, EEs are prone to several biases which may threaten internal or external validity. A threat to the internal validity would be the exclusion of relevant cost parameters or an insensitive outcome questionnaire. The external validity may be threatened by differences across populations but also across different (health care) payment / reimbursement systems. In agreement with Abdul Pari et al. (2014) we emphasize the importance of transparent description of comparators, a broad societal perspective (including all relevant costs and effects), use of a suitable multi-attribute utility instrument, and a thorough description for all statistical issues (e.g. imputation of missing data or indexing).

## **Conclusion**

The current study provides a comprehensive overview of the literature on EEs of non-pharmacological interventions and COIs in BD. In all EEs the interventions resulted in improved clinical outcomes and in most cases also in less total costs. This suggests that non-pharmacological interventions might be cost-effective treatment alternatives for people with BD. The fact that all interventions led to improvements in clinical outcomes also indicates that they might be a valuable adjunct to TAU and might be considered by clinicians as addition to pharmacological treatments. However, for both EEs and COIs, studies were identified with a vast variety of general and methodological characteristics and outcomes. This makes it difficult to compare the results across studies. Therefore, we encourage future research to use standardized methodology, aim to widen the body of knowledge in this research domain and consider to investigate the cost-effectiveness of more novel psychological treatment alternatives for BD.

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# APPENDIX A

**Table A1.** CHEC quality assessment of included EEs

Item	First author							
	Bauer	Camacho	Chisholm	Flood	Kessing	Lam	Scott	Simon
1.) Is the study population clearly described?	1	1	0.5	0.5	1	1	1	1
2.) Are competing alternatives clearly described?	1	1	0.5	1	1	0.5	1	1
3.) Is a well-defined research question posed in answerable form?	1	1	0.5	1	1	1	1	0.5
4.) Is the economic study design appropriate to the stated objective?	1	1	1	1	1	1	1	1
5.) Are the structural assumptions and the validation methods of the model properly reported?	NA	0.5	0.5	NA	NA	NA	NA	NA
6.) Is the chosen time horizon appropriate in order to include relevant costs and consequences?	1	1	1	1	1	1	1	1
7.) Is the actual perspective chosen appropriate?	1	1	1	0	1	0	0	0
8.) Are all important and relevant costs for each alternative identified?	1	1	0.5	1	0	1	0.5	0.5
9.) Are all costs measured appropriately in physical units?	1	1	0	0.5	1	1	0.5	1
10.) Are costs valued appropriately?	1	1	1	1	1	1	1	0.5
11.) Are all important and relevant outcomes for each alternative identified?	1	1	1	0	1	1	1	1
12.) Are all outcomes measured appropriately?	1	1	1	1	1	1	1	1
13.) Are outcomes valued appropriately?	1	1	1	1	1	1	1	1
14.) Is an appropriate incremental analysis of costs and outcomes of alternatives performed?	0	1	0.5	1	0	0.5	0	1
15.) Are all future costs and outcomes discounted appropriately?	0.5	0	1	0	0	0	0	0
16.) Are all important variables, whose values are uncertain, subjected to sensitivity analysis?	0	1	0.5	1	0.5	0.5	0	0
17.) Do the conclusions follow from the data reported?	1	1	1	0.5	1	1	1	1
18.) Does the study discuss the generalizability of the results to other settings and groups?	0	0	0.5	0	1	0	0	1
19.) Does the article indicate that there is no potential conflict of interest?	0	1	1	1	1	1	1	1
20.) Are ethical and distributional issues discussed appropriately?	0	0	NA <sup>a</sup>	1	1	0	1	1
Score (%)	69	83	72	71	77	76	68	76

Note. NA = Not applicable. <sup>a</sup>Ethical considerations were not applicable, since this study concerns a MBEE.



**Table A2.** Quality assessment of included COIs

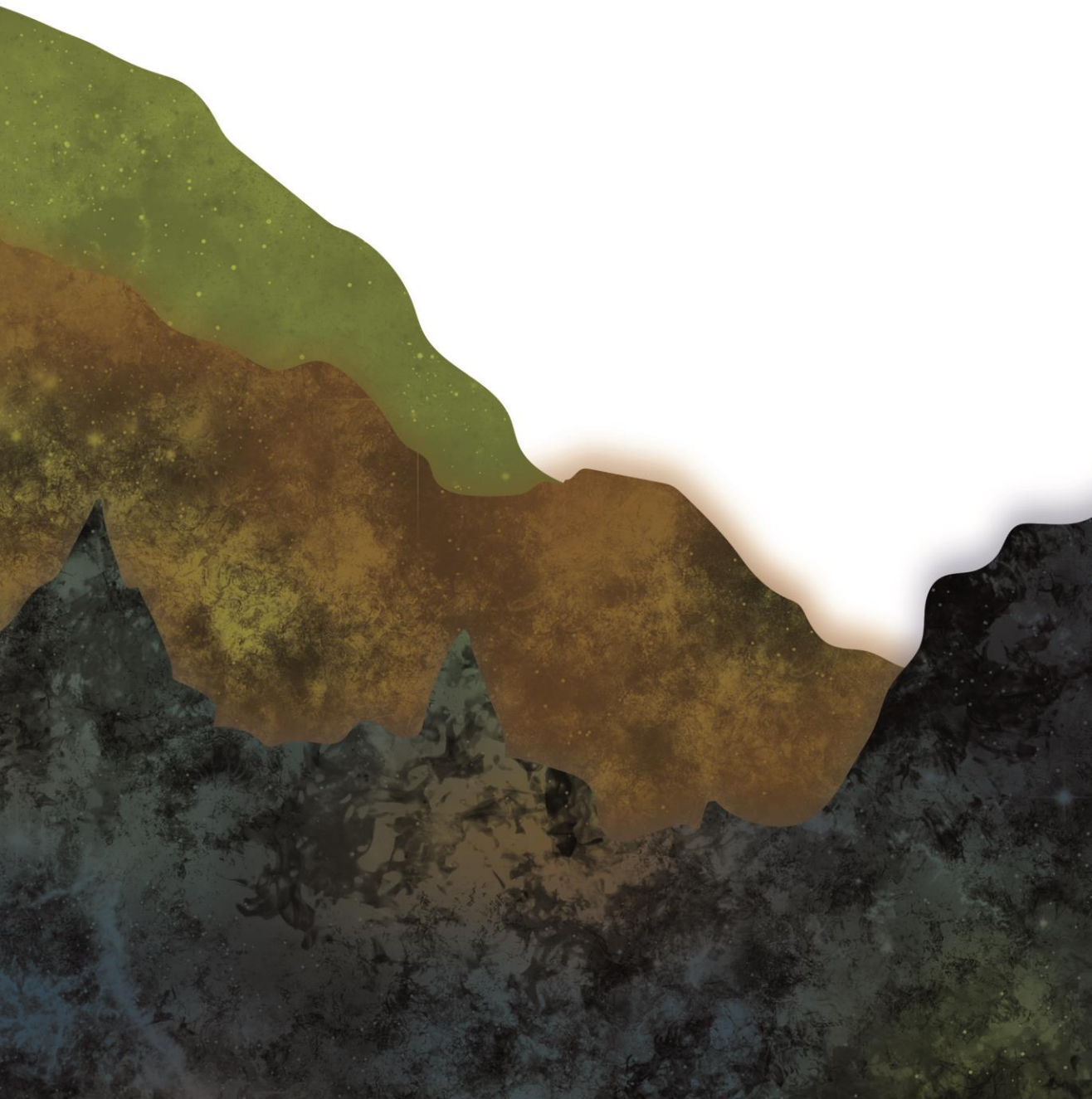
Item	First author				
	Broder	Cloutier	Corell	Degli Esposti	Ekman
1.) Is the study objective defined and were hypotheses given?	1	1	1	1	1
2.) Are clear and objective inclusion and exclusion criteria defined?	1	1	1	1	0.5
3.) Are the objective diagnostic criteria reported used to identify eligible participants?	1	1	1	1	1
4.) Does that study analyze costs that accrued from a particular disease?	1	1	1	1	1
5.) Does the study include a non-diseased control group to calculate excess cost or are the costs restricted to the disease of interest?	0	1	0	0	0
6.) Is the currency in which the costs are reported uncontroversial?	1	1	1	1	1
7.) Are all costs valued at the price level of a stated base year (and inflated if necessary)?	1	1	1	0	1
8.) Are the costs analyzed from the perspective of a patient, a payer or the society and was the perspective reported?	0	1	0	0	1
9.) Does the study estimate costs from the utilization of different kinds of health care services?	1	1	0.5	1	1
10.) Is the source of information on healthcare utilization reported?	1	1	1	1	1
11.) If data on healthcare utilization is reported, does the study report the source of unit costs?	0	1	1	1	1
12.) If costs are estimated for a period longer than one year, are the future costs and effects discounted and is the discount rate given?	0	1	0	0	0
13.) Is the proportion of missing data reported and is the imputation method described?	NA	NA	NA	NA	NA
14.) Are the statistical methods described and appropriate regarding the characteristics of cost data?	1	0	1	0	1
15.) Are all relevant parameters subjected to sensitivity analyses?	1	1	0	0	0
16.) Is the sample size of each group reported?	1	1	1	1	1
17.) Are the characteristics of the sample described?	1	1	1	1	1
18.) Are the cost estimated presented as arithmetic means?	1	1	1	1	1
19.) Are the standard deviations of cost estimates reported?	0	0	0	0	1
20.) Are the results discussed in relation to other studies on the same topic?	1	1	1	1	1
21.) Are the limitations (particular in calculation of costs) discussed in detail?	1	1	1	1	1
22.) Do the conclusion allow for the uncertainty inherent to the results?	0.5	0.5	0.5	0.5	1
<b>Score (%)</b>	<b>74</b>	<b>88</b>	<b>71</b>	<b>64</b>	<b>83</b>

**Table A2.** Quality assessment of included COIs (continued)

Item	First author				
	Mennini	Pan	Parker	Somaiya	Wu
1.) Is the study objective defined and were hypotheses given?	1	1	1	1	1
2.) Are clear and objective inclusion and exclusion criteria defined?	1	1	1	1	1
3.) Are the objective diagnostic criteria reported used to identify eligible participants?	1	1	1	1	1
4.) Does that study analyze costs that accrued from a particular disease?	1	1	1	1	1
5.) Does the study include a non-diseased control group to calculate excess cost or are the costs restricted to the disease of interest?	0	1	0	0	0
6.) Is the currency in which the costs are reported uncontroversial?	1	1	1	1	1
7.) Are all costs valued at the price level of a stated base year (and inflated if necessary)?	0	1	1	0	0
8.) Are the costs analyzed from the perspective of a patient, a payer or the society and was the perspective reported?	0	1	0	0	0
9.) Does the study estimate costs from the utilization of different kinds of health care services?	0	1	1	1	1
10.) Is the source of information on healthcare utilization reported?	1	1	1	1	1
11.) If data on healthcare utilization is reported, does the study report the source of unit costs?	0	0	1	0	0
12.) If costs are estimated for a period longer than one year, are the future costs and effects discounted and is the discount rate given?	0	0	0	NA	0
13.) Is the proportion of missing data reported and is the imputation method described?	0	0	0	0	NA
14.) Are the statistical methods described and appropriate regarding the characteristics of cost data?	0	0	1	1	1
15.) Are all relevant parameters subjected to sensitivity analyses?	0	0	0	0	0
16.) Is the sample size of each group reported?	1	1	1	1	1
17.) Are the characteristics of the sample described?	1	1	1	1	1
18.) Are the cost estimated presented as arithmetic means?	1	1	1	1	1
19.) Are the standard deviations of cost estimates reported?	0	1	0	1	1
20.) Are the results discussed in relation to other studies on the same topic?	0	1	1	1	1
21.) Are the limitations (particular in calculation of costs) discussed in detail?	0	1	1	1	1
22.) Do the conclusion allow for the uncertainty inherent to the results?	0	1	0	0.5	1
<b>Score (%)</b>	<b>41</b>	<b>77</b>	<b>68</b>	<b>69</b>	<b>71</b>

Note. NA = Not applicable.

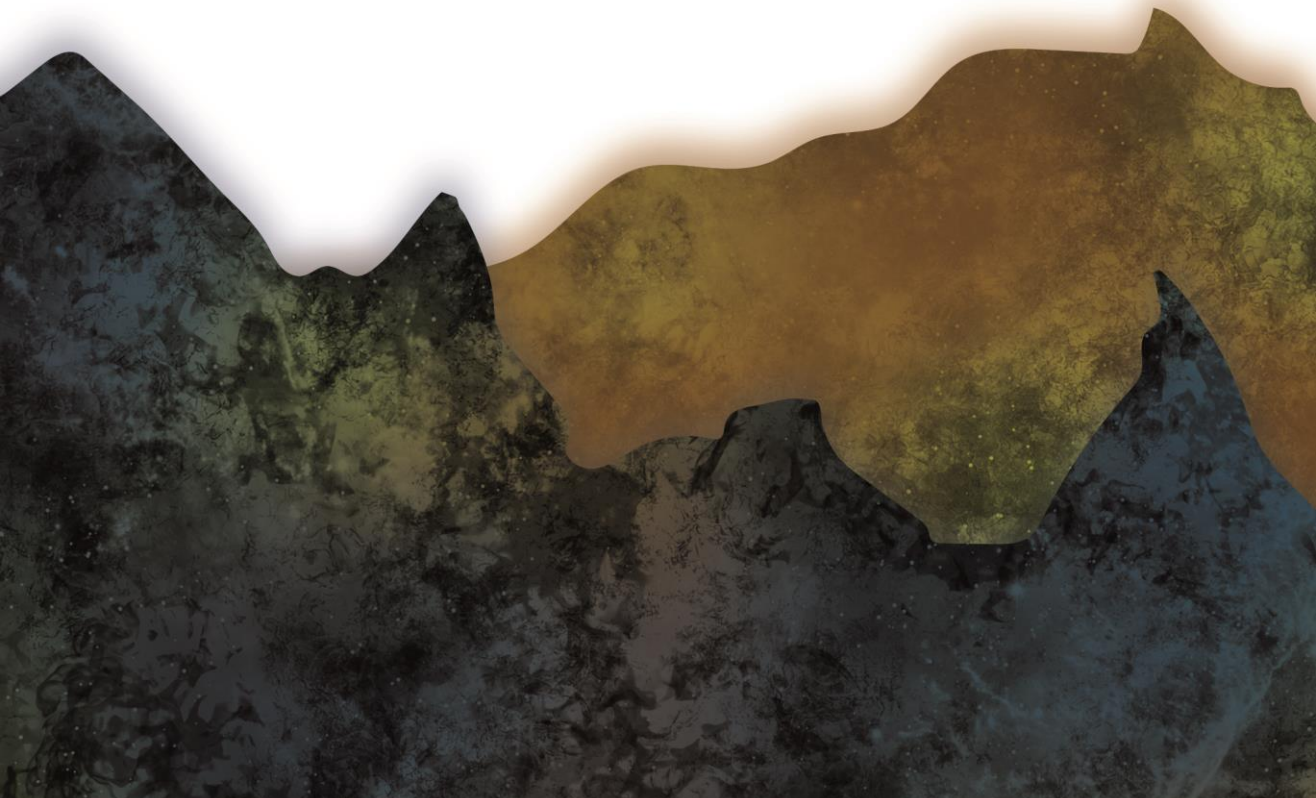




# CHAPTER 4

## Measuring personal recovery in people with bipolar disorder and exploring its relationship with well-being and social role participation

Kraiss, J. T., Ten Klooster, P. M., Chrispijn, M., Stevens, A. W., Kupka, R. W., & Bohlmeijer, E.T. (2019). Measuring personal recovery in people with bipolar disorder and exploring its relationship with well-being and social role participation. *Clinical Psychology & Psychotherapy*, 26(5), 540-549. <https://doi.org/10.1002/cpp.2371>



## **ABSTRACT**

The relevance of personal recovery receives increasing attention in mental health care and is also important for people with bipolar disorder (BD). There is a need for reliable and valid instruments measuring personal recovery. Therefore, the current study evaluated the psychometric properties of a Dutch translation of the Questionnaire about the Process of Recovery (QPR) in a sample of people with BD and explored the relationship with constructs of well-being, social role participation and psychopathology. A cross-sectional survey study was conducted, in which 102 people diagnosed with BD completed the QPR. Factor structure of the QPR was evaluated by conducting confirmatory factor analyses (CFA) and internal consistency was assessed by calculating reliability coefficients. Convergent validation measures assessed well-being, social role participation and symptomatology. Incremental validity was determined by evaluating the ability of the QPR to explain variance in symptomatology above and beyond measures of well-being and social role participation. Findings of the CFA supported a unidimensional factor structure and internal consistency estimates were excellent. Scores of the QPR showed strong correlations with convergent measures, but were only weakly associated with manic symptomatology. Moreover, personal recovery explained additional variance in symptoms of depression and anxiety above and beyond well-being, indicating incremental validity. The QPR appears to be a reliable and valid tool to assess personal recovery in people with BD. Our findings underline the importance of personal recovery in the context of treatment of BD. Personal recovery demonstrates a substantial overlap with well-being.

## INTRODUCTION

Bipolar disorder (BD) is a severe and chronic affective disorder, which characterized by shifting depressive and (hypo)manic mood episodes (Kupka et al., 2008). In general, a distinction is made between bipolar I (BDI) and bipolar II disorder (BDII). In BDII, an individual has never experienced a full manic episode but only milder hypomanic episode(s) (Grande et al., 2015). Prevalence estimates from a large community sample from 11 countries revealed a lifetime prevalence of 0.6% for BDI, 0.4% for BDII and 1.4% for subthreshold BD (Merikangas et al., 2007). The economic burden of BD was estimated at 151 billion dollars per year in the United States alone (Dilsaver, 2011). Suffering from BD is associated with negative social consequences (Calabrese et al., 2003), decreased quality of life (Dean et al., 2004), work-related issues (Fajutrao et al., 2009; Laxman et al., 2008) and a high burden for caregivers (Miller et al., 2014).

In mental health care, the concept of personal recovery is receiving increasing interest (Fava et al., 2007; Jones et al., 2013; Leamy et al., 2011) and especially Anglophone countries move towards supporting personal recovery in the treatment of people with mental disorders (Bird et al., 2014). In contrast to symptomatic and functional recovery, personal recovery has been defined as a *'deeply personal, unique process of changing one's attitudes, values, feelings, goals, skills and/or roles [and] a way of living a satisfying, hopeful and contributing life even with the limitations caused by illness'* (Leamy et al., 2011, p. 445). Leamy et al. (2011) identified five key processes important for personal recovery: connectedness, hope, identity, meaning and empowerment. These five processes are comprised in the CHIME framework of personal recovery (Leamy et al., 2011). People with severe mental disorders, such as BD, have highlighted the relevance of hope, meaning and connectedness in life as important outcomes of recovery in contrast to the traditional focus on symptomatic recovery (Jones et al., 2012; Mead & Copeland, 2000; Pitt et al., 2007; Slade, 2009).

Independent from, but related to the idea of personal recovery, the paradigm of focusing on mental health as opposed to mental illness has also received increased attention. Fava (2007) defines full recovery as the absence of psychopathology and the presence of psychological well-being (Ryff, 2014). In a similar way, Keyes (2005) defines mental health not merely as the absence of psychopathology, but also as the presence of well-being. In his conceptualization, well-being comprises three dimensions: (1) emotional (e.g. presence of positive emotions), (2) psychological (e.g. autonomy and environmental mastery) and (3) social well-being (e.g. social acceptance or social coherence). Research shows that the presence of well-being buffers against psychopathology (e.g. Keyes et al., 2010; Lamers et al., 2015; Schotanus-Dijkstra et al., 2016). Although personal recovery and well-being emerged as independent concepts, they

share substantial conceptual overlap (Slade, 2010). Besides personal recovery and well-being, social role participation is increasingly considered a key outcome in rehabilitation of people with a wide range of chronic impairments, including those with psychiatric conditions, and may be another important factor for recovery (Jaeger & Hoff, 2012; Whitley & Drake, 2010). It has been shown to be important to build and maintain self-esteem and autonomy (Gordeev et al., 2010) and plays a role in long-term mental health (Oude Voshaar et al., 2016).

Several measures exist to assess personal recovery. Shanks et al. (2013) systematically reviewed the recovery literature for measures of personal recovery and found 13 questionnaires assessing personal recovery. Of the identified measures, only from the Questionnaire about the Process of Recovery (QPR) all items matched with the processes of the CHIME framework and at the same time covered all five processes. Based on service users' accounts of recovery from psychosis, the original QPR comprised 22 items and contained the two dimensions (1) intrapersonal and (2) interpersonal recovery (Neil et al., 2009). Although originally developed and validated in people with experience of psychosis, all items of the QPR refer to generic, non-psychosis-specific processes of recovery that have been identified in the CHIME framework as relevant across mental illnesses.

The original English QPR has been translated and validated in samples with different or mixed mental disorders in several languages, including Chinese (Chien & Chan, 2013), Swedish (Argentzell et al., 2017) and Japanese (Kanehara et al., 2017). Further psychometric analyses by Law et al. (2014) showed that the most interpretable solution of the QPR was a unidimensional 15-item version. Williams et al. (2015) could confirm this conclusion by comparing the 22-item version with the 15-item version. They found that the interpersonal recovery subscale of the 22-item version underperformed in confirmatory factor analyses (CFA) and that the intrapersonal subscale showed substantial overlap with the 15-item version. The authors concluded that the 15-item was more robust and less burdensome compared to the 22-item version (Williams et al., 2015). Another specific measure of recovery experiences in BD is the Bipolar Recovery Questionnaire (BRQ; Jones et al., 2013). The BRQ has been developed in a sample of patients with BD and has been shown to be a reliable and valid instrument (Jones et al., 2013). However, the QPR represents a less burdensome and more feasible solution to assess recovery, since BRQ comprises 36 items. Moreover, the BRQ fits less well to the well-established and evidence-based CHIME framework compared to the QPR, of which every item maps to one of its dimensions (Shanks et al., 2013).

Although the QPR has been validated in several different languages and target groups, there are some important gaps in current knowledge. First, the QPR has not yet been validated in people with BD. The illness course of BD is often chronic (Fagiolini et al., 2013) and although

people with BD might recover symptomatically and functionally, this does not necessarily mean that they are personally recovered. The QPR can be a potentially suitable instrument to assess personal recovery in BD, since it was the only questionnaire identified of which every item maps a dimension of the CHIME framework (Shanks et al., 2013). Moreover, it has been developed in cooperation with mental health service users experiences of recovery from psychosis (Neil et al., 2009). Even though it has not been specifically developed with people with BD, many important personal recovery challenges such as meaning or identity can be seen as transdiagnostic across various serious mental illnesses (Jones et al., 2010; Mead & Copeland, 2000; Pitt et al., 2007). Hence, the QPR may also be appropriate to assess recovery in BD. Second, the relationship between the concepts of personal recovery, well-being, social role participation and symptomatology has not yet been explored in people with BD. Third, the QPR has not yet been translated into Dutch. Translating the QPR into Dutch would be an important step for assessment of personal recovery in the Netherlands.

Therefore, the goal of the current study is three-fold: (1) to confirm the unidimensional factor structure and internal consistency of a Dutch translation of the QPR, (2) to investigate convergent validity of the QPR with measures of well-being, social role participation and psychopathology and (3) to determine the incremental validity of the QPR in explaining variance in symptoms of anxiety and depression above and beyond scores of well-being and social role participation in a sample of people with BD.

For convergent validity, we hypothesized strong positive correlations between personal recovery and well-being, since these two constructs show substantial conceptual convergence (Slade, 2010) and earlier studies have shown strong positive relationships between personal recovery and well-being related outcomes, such as optimism (Neil et al., 2009) and self-esteem (Law et al., 2014; Neil et al., 2009) and a moderate positive correlation with well-being (Williams et al., 2015). In particular, strong positive relationships between personal recovery and the emotional and psychological dimensions of well-being were expected. For example, similar to emotional and psychological well-being, personal recovery focuses on an individual's experience of positive emotion and sense of autonomy, self-acceptance and meaning. Furthermore, we hypothesized moderate to strong positive correlations between personal recovery and social role participation, since social role participation plays an important role in long-term mental health (Gordeev et al., 2010; Oude Voshaar et al., 2016) and has been widely recognized as important part of recovery (Jaeger & Hoff, 2012; Whitley & Drake, 2010). Finally, we hypothesized moderate to strong negative correlations between personal recovery and symptomatology. This would be in line with an earlier study, showing moderate correlations with hopelessness and strong correlations with depressive symptomatology (Law et al., 2014).

# METHOD

## Procedure

A cross-sectional validation survey study was conducted. Participants were gathered through convenience sampling via the Dutch patient association for BD. The study was promoted in the newsletter and in the journal of the patient association. Data was gathered via the online survey tool Limesurvey (<https://www.limesurvey.org/>). At the beginning of the survey, respondents were informed about the aim of the study, that they could stop the survey at any moment and that their data were processed anonymously and confidentially. Ten shopping vouchers of 50 euro were raffled among all participants. The study was approved by the Ethics Committee of the University of Twente.

## Measures

Participants were asked to provide basic demographical data, including gender, age, marital and employment status, ethnicity and educational background. People were also asked to specify their type of diagnosis (BDI or BDII) and if they were in psychological or psychiatric treatment at the moment of participation. Moreover, they were asked if they were taking medication in the context of their BD and whether there were any recent adaptations in their medication. The following questionnaires were used to assess the relevant constructs:

### *Personal recovery*

For the current study, the 15-item version of the QPR (Law et al., 2014; Neil et al., 2009) was used to measure personal recovery. The 15 items of the QPR (e.g. “*I feel better about myself*” or “*I can actively engage with life*”) can be answered on a 5-point Likert scale, ranging from 0 (*disagree strongly*) to 4 (*agree strongly*). More personal recovery is indicated by higher scores on the questionnaire. For this study, the QPR was translated from English into Dutch through forward and backward translation by the first and second author. The English 15-item version of the QPR showed high internal consistency in a sample of psychotic individuals ( $\alpha = .89$ ; Williams et al., 2015) and in a sample of people with schizophrenia spectrum disorder ( $\alpha = .93$ ; Law et al., 2014).

### *Well-being*

The 14-item Mental Health Continuum-Short Form (MHC-SF; Lamers et al., 2011) aims to assess well-being on three dimensions: (1) emotional well-being (three items), (2) psychological well-being (six items) and (3) social well-being (five items). Respondents rate the frequency of feelings on a 6-point Likert scale in the past month, ranging from 0 (*never*) to 5

(*every day*). We used the Dutch version of the MHC-SF, which showed high internal consistency for the total scale ( $\alpha = .89$ ) and for the scales emotional ( $\alpha = .83$ ) and psychological well-being ( $\alpha = .83$ ) and adequate reliability for the social dimension ( $\alpha = .74$ ) (Lamers et al., 2011). The MHC-SF has also shown convergent validity by correlating well with aspects of well-being and positive functioning. Cronbach's alpha in the current study was .91 for the total scale and .89 and .87 and .67 for the emotional, psychological and social well-being subscale, respectively.

### *Social role participation*

The Short-Social Role Participation Questionnaire (S-SRPQ; Oude Voshaar et al., 2016) was used to assess social role participation. The 12-item S-SRPQ contains two subscales: (1) satisfaction with role performance and (2) experienced difficulties. Respondents are asked to specify their satisfaction with their social role and experienced difficulties in relation to six social roles (e.g. intimate relationship or employment). The items are scored on a 5-point Likert scale, reaching from 0 (*not satisfied at all or no difficulties at all*) to 4 (*very much satisfied or not possible*). More satisfaction and more experienced difficulties are indicated by higher scores on the corresponding subscales. Both subscales of the questionnaire have shown good internal consistency ( $\alpha = .86$ ) (Oude Voshaar et al., 2016). Cronbach's alpha in the current study was adequate for the satisfaction subscale ( $\alpha = .74$ ) and good for the experienced difficulties subscale ( $\alpha = .82$ ).

### *Symptoms of depression and anxiety*

To assess depressive and anxious symptomatology, the Hospital Anxiety and Depression Scale (HADS; Zigmond & Snaith, 1983) was used. This 14-item questionnaire assesses the presence of symptoms of depression (7 items) and anxiety (7 items). Respondents are asked to rate the frequency of symptoms over the last week, with scores ranging from 0 (*not at all*) to 3 (*very often*). Prior psychometric validations have shown good and adequate reliability of the anxiety ( $\alpha = .84$ ) and depression subscale ( $\alpha = .79$ ) of the Dutch HADS in the general population (Spinhoven et al., 1997). In the current study, Cronbach's alpha was .85 and .73 for the anxiety and depression subscale, respectively.

### *Manic symptomatology*

Symptoms of mania were assessed using the Altman Self-Rating Mania Scale (ASRM; Altman et al., 1997). This measure comprises 5 items assessing manic symptoms (e.g., inflated self-confidence or increased cheerfulness) over the past week. Each item contains five answering options representing the severity of the symptoms. Higher scores are indicative of more manic symptoms. Prior psychometric evaluations haven shown good test-retest reliability (Altman et



al., 1997) and sensitivity to change in a clinical group (Altman et al., 2001). Cronbach's alpha in the current study was .73.

## Statistical analyses

Statistical analyses were performed using Mplus version 7.11 (Muthén & Muthén, 2010), RStudio (R Core Team, 2018) and the statistical package for social sciences version 25 (SPSS). CFA was conducted in Mplus to confirm the previously established unidimensional factor structure of the QPR (Argentzell et al., 2017; Law et al., 2014; Williams et al., 2015). For this, we fitted a strict one-factor solution, in which all 15 items loaded on one single latent factor. We used the robust diagonally weighted least square mean and variance adjusted estimation method (WLSMV), given the relatively small sample size and ordinal nature of the data (Flora & Curran, 2004; Moshagen & Musch, 2014). Factor loadings  $> .40$  were considered satisfactory (Floyd & Widaman, 1995; Hair et al., 2009). Chi-square statistics ( $\chi^2$ ) were used to assess the model fit, where a smaller and significant  $\chi^2$  value is indicative for a better model fit and a ratio between the  $\chi^2$  value and the degrees of freedom should be  $< 5$  for an acceptable and around  $< 2$  for a good model fit (Kline, 2015; Watkins, 1989). Additionally, we used the comparative fit index (CFI), Tucker-Lewis Index (TLI), weighted root-mean-square residual (WRMR) and root-mean-square error approximation (RMSEA) to evaluate the model fit (Hu & Bentler, 1998). Values  $\geq 0.90$  are seen as acceptable and  $\geq 0.95$  as good model fit for the CFI and TLI (Hu & Bentler, 1999). In addition, WRMR values  $< 1$  were seen as good model fit (DiStefano et al., 2018; Yu, 2002) and RMSEA values  $\leq 0.08$  were considered as acceptable and  $\leq .05$  as good model fit (Browne & Cudeck, 1992). The evaluation of the model was first based on a restrictive model assuming uncorrelated error terms. Although model fit can often be increased by allowing errors terms to correlate, we decided to only allow error correlation(s) to improve model fit if an initial evaluation of the model indicated unacceptable fit and if it made substantial sense (Jöreskog, 1993). The difference in fit between the 1-factor model without error correlations and the model where error correlations were allowed, was statically tested with the DIFFTEST function of Mplus, which adequately deals with  $\chi^2$  difference testing of nested models.

Internal consistency was determined by calculating Cronbach's alpha ( $\alpha$ ) and categorical McDonald's omega ( $\omega$ ) (Dunn et al., 2014). Since Cronbach's  $\alpha$  assumes tau-equivalence and may thus be deficient for evaluating the internal consistency of congeneric models, McDonald's  $\omega$  was calculated as an alternative estimate of internal consistency. We used the MBESS package (Kelley, 2018) in RStudio to calculate  $\alpha$  and categorical  $\omega$  coefficients with 95% bias corrected and accelerated confidence intervals (CI) based on 1000 bootstrap samples.

Estimated values  $> .70$  were seen as acceptable and  $> .80$  as good reliability (Cicchetti, 1994). In addition, we calculated item-total correlations for each item of the QPR.

Construct validity of the QPR was determined by calculating Pearson's correlation coefficients between scores of the QPR and convergent measures of well-being (MHC-SF), social role participation (S-SRPQ) and depressive (HADS-D), anxiety (HADS-A) and manic symptomatology (ASRM). Values between  $.1$  and  $.3$  were considered as weak, between  $.3$  and  $.5$  as moderate correlation and larger than  $.5$  as strong correlation. To determine incremental validity, we conducted two separate multiple hierarchical regression analyses to test the ability of the QPR to explain variance in psychopathology above and beyond well-being and social role participation. In the first step, scores of the MHC-SF and S-SRPQ were entered, respectively and in the second step total scores of the QPR were entered to the model. Significant F-changes ( $p < .05$ ) in the second step of the model were indicative for incremental validity of the QPR.

**Table 1.** Sample characteristics ( $N = 102$ )

Variable	Category	N	%
Gender	Female	80	78.4
	Male	22	21.6
Marital status	Married	53	52.0
	Never married	26	25.5
	Divorced	22	21.6
	Widowed	1	1.0
Employment status	Not capable to work	38	39.6
	Paid work	24	25.0
	Voluntary work	14	14.6
	Retired	9	9.4
	Housewife/houseman	4	4.2
	Self-employed	4	4.2
	Student	3	3.1
Education	Other	6	5.9
	Low	13	12.9
	Moderate	32	31.7
Diagnosis	High	56	55.4
	BDI	41	40.2
	BDII	47	46.1
Currently in psychological treatment	Unknown	14	13.7
	Yes	84	82.4
	No	18	17.6
Currently taking medication	Yes	97	95.1
	No	5	4.9

# RESULTS

## Description of the sample

Mean age of the 102 respondents was 52 years (SD = 11.17, range 23-77). More than three quarters of the sample was female, and half of the sample was married, whereas the rest has never been married or was divorced. A large number of participants were not currently working and approximately half of the sample had a high educational background. The mean score of the QPR in the current sample was 37.66 (SD = 11.14). This is relatively low compared with a prior study by Law et al. (2014), in which they included a mixed sample of mental health service users and found a mean QPR score of 50.13 (SD = 11.56). A detailed overview of the sample characteristics can be found in Table 1.

**Table 2.** Standardized factor loadings and corrected item-total correlations for the 15 items of the QPR

Item	Factor loading	Corrected item-total correlation
QPR1 I feel better about myself.	.79	.70
QPR2 I feel able to take chances in life.	.86	.75
QPR3 I am able to develop positive relationships with other people.	.83	.71
QPR4 I feel part of society rather than isolated.	.80	.69
QPR5 I am able to assert myself.	.64	.55
QPR6 I feel that my life has a purpose.	.80	.75
QPR7 My experiences have changed me for the better.	.70	.65
QPR8 I have been able to come to terms with things that have happened to me in the past and move on with my life.	.71	.66
QPR9 I am basically strongly motivated to get better.	.49	.42
QPR10 I can recognise the positive things I have done.	.65	.54
QPR11 I am able to understand myself better.	.65	.55
QPR12 I can take charge of my life.	.84	.78
QPR13 I can actively engage with life.	.90	.81
QPR14 I can take control of aspects of my life.	.85	.73
QPR15 I can find the time to do the things I enjoy.	.62	.56
<b>McDonald's omega (95% CI)</b>	.95 (.91 - .97)	
<b>Cronbach's alpha (95% CI)</b>	.92 (.90 - .93)	

Note. N = 102, QPR = Questionnaire about the Process of recovery, CI = Confidence interval.

## Factor structure and internal consistency

The one-factor solution revealed acceptable CFI (0.964), TLI (0.958) and WRMR values (0.895) and all items showed satisfactory factor loadings. However, RMSEA (0.105) indicated a poor fit to the data. The chi-square test of model fit was significant ( $\chi^2 = 191.11$ ,  $df = 90$ ,  $p < .001$ ), but the ratio between the  $\chi^2$  value and degrees of freedom was smaller than 3, indicating an acceptable fit. Based on the modification indices, we allowed an error correlation between items 3 and 4, which led to a slight improvement of fit indices (CFI = 0.969; TLI = 0.963; RMSEA = 0.098;  $\chi^2 = 176.03$ ,  $p < .001$ ). The model in which the error correlation between item 3 and 4 ( $r = .661$ ) was allowed, showed a significantly better fit based on a chi-square difference test ( $\Delta\chi^2 = 15.08$ ,  $\Delta df = 1$ ,  $p < 0.01$ ). However, since the improvement of fit on the other fit indices was negligible and CFI and TLI fit indices already indicated a good fit without error correlations allowed, we decided to adhere to the restrictive 1-factor model without error correlations allowed. Internal consistency of the QPR was excellent in the present sample ( $\alpha = .92$  and  $\omega = .95$ ) and corrected item-total correlations were high for all items. Table 2 gives an overview of the standardized factor loadings, reliability parameters and corrected item-total correlations.

**Table 3.** Descriptive statistics and bivariate Pearson's correlations between the QPR and criterion measures

Measure	M (SD)	QPR
QPR ( $n = 102$ )	37.66 (11.14)	-
MHC-SF ( $n = 102$ )		
Emotional well-being	7.40 (3.96)	.77**
Social well-being	8.36 (4.94)	.58**
Psychological well-being	13.58 (7.19)	.80**
Total score	29.34 (14.48)	.80**
S-SRPQ ( $n = 98$ )		
Satisfaction with role	14.62 (5.41)	.63**
Experienced difficulty	16.77 (5.41)	-.53**
HADS ( $n = 98$ )		
Anxiety symptoms	8.71 (4.64)	-.50**
Depression symptoms	9.61 (3.99)	-.71**
ASRM ( $n = 98$ )		
Total score	2.99 (3.23)	-.21*

*Note.* Variations in  $n$  due to missing data, QPR = Questionnaire about the Process of Recovery; MHC-SF = Mental Health Continuum-Short Form; S-SRPQ = Short-Social Role Participation Questionnaire; HADS = Hospital Anxiety and Depression Scale; ASRM = Altman Self-Rating Mania Scale. \* $p < .05$ , \*\* $p < .01$ .

## Convergent validity

A detailed overview of the descriptive statistics of the validation measures and correlations with the QPR can be found in Table 3. We found a particularly strong relationship between personal recovery and emotional ( $r = .77$ ) and psychological well-being ( $r = .80$ ) and also with total scores of the MHC-SF. In addition, we found a lower, but still strong positive relationship with social well-being ( $r = .58$ ). Moreover, a strong positive relationship was found with satisfaction with role performance of the S-SRPQ ( $r = .63$ ) and a strong negative relationship with experienced difficulty with a social role ( $r = -.53$ ). QPR scores were strongly negatively correlated with depressive symptoms ( $r = -.71$ ) and with anxious symptomatology ( $r = -.50$ ). Finally, a weak negative, but significant relationship between the QPR and manic symptoms was found ( $r = -.21$ ).

## Incremental validity

To investigate the incremental validity of the QPR, we tested whether scores of the QPR explained a significant amount variability in psychopathology above and beyond scores of well-being. Therefore, two separate multiple hierarchical regression analyses with scores of psychopathology as criterion variables were conducted. In the first step, subscale scores of the MHC-SF were entered. In the second step, scores of the QPR were entered.

In the first model, scores of the QPR explained 3% additional variance in depressive symptoms above and beyond well-being ( $p < .01$ ) and personal recovery significantly explained depressive symptoms above and beyond well-being. In the second model, scores of the QPR explained 4% additional variance in anxiety symptoms ( $p < .05$ ) above and beyond well-being. After controlling for well-being, personal recovery was uniquely related to anxiety symptoms ( $p < .05$ ). An overview of the regression analyses can be found in Table 4 and Table 5.

**Table 4.** Summary of hierarchical regression analysis for MHC-SF subscales and QPR and depressive symptoms (HADS-D)

Variable	B	SE	$\beta$	t	$\Delta R^2$
Step 1					
Constant	15.53	.61		25.66***	.59***
Emotional well-being (MHC-SF)	-.66	.10	-.65	-6.31***	
Psychological well-being (MHC-SF)	-.09	.07	-.16	-1.23	
Social well-being (MHC-SF)	.02	.08	.02	.24	
Step 2					
Constant	17.65	.99		17.91***	.03**
Emotional well-being (MHC-SF)	-.54	.11	-.54	-4.89***	
Psychological well-being (MHC-SF)	.00	.08	.00	0.01	
Social well-being (MHC-SF)	.02	.08	.02	.21	
Personal recovery (QPR)	-.11	.04	-.31	-2.68**	

Note. MHC-SF = Mental Health Continuum-Short Form; QPR = Questionnaire about the Process of Recovery; HADS-D = Hospital Anxiety and Depression Scale – depression subscale.

\*\* $p < .01$ , \*\*\* $p < .001$ .

**Table 5.** Summary of hierarchical regression analysis for MHC-SF subscales and QPR and anxiety symptoms (HADS-A)

Variable	B	SE	$\beta$	t	$\Delta R^2$
Step 1					
Constant	12.52	.97		12.86***	.21***
Emotional well-being (MHC-SF)	-.39	.17	-.33	-2.31*	
Psychological well-being (MHC-SF)	-.17	.11	-.26	-1.50	
Social well-being (MHC-SF)	.16	.12	.18	1.32	
Step 2					
Constant	15.52	1.60		9.71***	.04*
Emotional well-being (MHC-SF)	-.22	.18	-.19	-1.23	
Psychological well-being (MHC-SF)	-.05	.12	-.07	-.37	
Social well-being (MHC-SF)	.16	.12	.17	1.32	
Personal recovery (QPR)	-.16	.07	-.38	-2.34*	

Note. MHC-SF = Mental Health Continuum-Short Form; QPR = Questionnaire about the Process of Recovery; HADS-A = Hospital Anxiety and Depression Scale – anxiety subscale.

\* $p < .05$ , \*\*\* $p < .001$ .

## DISCUSSION

The current study is the first to evaluate the psychometric properties of the QPR in a sample of people with BD and to assess the relationship of personal recovery with well-being, social role participation and psychopathology. Several measures exist to assess personal recovery. In a recent review, Shanks et al. (2013) identified only one questionnaire assessing all processes of the CHIME-framework of personal recovery, namely the QPR. Originally designed as a 22-item questionnaire (Neil et al., 2009), the QPR was recently shortened to a 15-item version (Law et al., 2014; Williams et al., 2015). The QPR has been translated into several languages, including Swedish (Argentzell et al., 2017), Chinese (Chien & Chan, 2013) and Japanese (Kanehara et al., 2017). However, to date the QPR had not been translated into Dutch and has not been validated in people with BD.

Overall, our findings provide preliminary support for the reliability and validity of the Dutch QPR for measuring personal recovery in people with BD. Findings of the CFA revealed a good fit for a unidimensional model, based on several fit indices. This is in line with studies investigating the 1-factor model of the QPR (Law et al., 2014; Williams et al., 2015). Furthermore, all items showed high factor loadings. This supports the conclusion of prior studies suggesting an overall recovery score rather than distinct subscales (Law et al., 2014; Williams et al., 2015). The RMSEA fit statistic did not meet the cut-off for acceptable model fit. It should be noted though that fit indices may not perform uniformly across different conditions. Different factors, such as sample size or parameter estimation methods can affect fit indices in different ways (Cook et al., 2009). Since all the other fit indices indicated a good model fit, we concluded that there was sufficient support for a unidimensional factor structure overall. Reliability values, including Cronbach's alpha and McDonald's omega were excellent, coinciding with earlier studies (Law et al., 2014; Williams et al., 2015).

QPR scores were significantly related with each validation measure in this study. Most of these relationships were in line with our hypotheses, such as the strong positive correlation between personal recovery and well-being. This strong relationship can be explained by the high conceptual overlap between well-being and personal recovery. Slade (2010) outlined the similarity between these two concepts and how they can complement each other. Furthermore, a strong relationship between personal recovery and the two subscales of social role participation was found. This supports the idea that social role participation (e.g. being able to work) can be seen as a relevant factor for recovery (Jaeger & Hoff, 2012; Whitley & Drake, 2010).

Findings regarding the relationship between personal recovery and symptomatology were mixed. We found a strong negative relationship between personal recovery and both symptoms of anxiety and depressive symptoms, which is in line with earlier studies (Law et al., 2014; Neil et al., 2009). This may imply that either personal recovery will yield further symptom reduction or that symptom reduction yields personal recovery. Surprisingly, the relationship between personal recovery and symptoms of mania was only weak. One possible explanation might be that the presence of manic symptoms might not necessarily be an obstacle for personal recovery. Possibly, manic symptoms might actually increase the experience of personal recovery. Another explanation might be the positively skewed and relatively low average ASRM scores in the present sample which might have suppressed the correlation with personal recovery.

Results of the multiple hierarchical regression analyses suggest incremental validity of the QPR, which explained variance in symptoms of depression and anxiety above and beyond well-being. This is a surprising finding, since research indicates that well-being already is a strong predictor of symptomatology (Wood & Joseph, 2010; Wood et al., 2008) and we also found very strong relationships between well-being and personal recovery in the present study. This finding is in line with the conceptual overlap between personal recovery and well-being (Slade, 2010). While the total variance in depressive and anxiety symptoms only marginally increased in the second step of the models (3% and 4%, respectively), the variance explained by well-being substantially decreased when adding personal recovery to the model and the QPR remained independently associated with symptoms. In other words, although there is a strong overlap between these concepts, personal recovery appears to be sufficiently distinct to warrant assessment.

## Limitations and future research

Our study also has several limitations which should be considered. First, we used a cross-sectional design and thus cannot make any inferences about the longitudinal relationship of the included constructs and we could not evaluate psychometric properties such as sensitivity to change and test-retest reliability of the QPR. Future research could focus on the longitudinal relationships between personal recovery, well-being and symptomatology and could evaluate psychometric properties such as sensitivity to change and test-retest reliability of the QPR. Especially the relationship between personal recovery and well-being might be interesting to further clarify the differences and commonalities between these two concepts. Second, we used a relatively small sample especially for factor analysis purposes and multivariate regression analyses. For example, following recommendations by Hu and Bentler (1999) to include at least 10 participants per free parameter in the model, 150 people would be needed



for the 1-factor model evaluated in the present study. Thus, these results should be interpreted with some caution. It must be noted though, that our sample contains a clinical group and provides the first evaluation of the Dutch QPR. However, future research should try to evaluate the psychometric properties of the Dutch QPR in larger clinical samples. Third, diagnosis of BD was based on self-report only and we did not confirm the diagnosis based on a structured clinical interview. However, participants were recruited via the patient association for BD and 95% of the sample stated that they were taking medication in the context of their BD. It can thus be assumed that the vast majority of the sample actually had BD.

## **Practical and scientific implications**

Several implications for both clinical practice and research arise from these findings. Our results give a first indication that the Dutch QPR is a reliable and valid tool and appears to be a promising instrument to assess personal recovery in BD. Bipolar disorder is a prevalent and highly disabling condition, in which the concept of personal recovery is particularly important, since the course of the disorder is often chronic and recurrent (Fagiolini et al., 2013). Moreover, patients with serious mental illness, such as BD, express the need for personal recovery outcomes (de Vos et al., 2017; Jones et al., 2012; Mead & Copeland, 2000; Pitt et al., 2007; Slade, 2009). Therefore, one of the main advantages of using the QPR in clinical practice is to foster collaboration and improve engagement by demonstrating to the patient that personal recovery is part of the recovery process (Neil et al., 2009). Although scores of the QPR should be interpreted as one overall recovery score rather than distinct subscores for assessment or monitoring purposes (Law et al., 2014; Williams et al., 2015), the QPR might also be used to individually tailor the recovery process and find out which aspects of recovery are important for the patient (Neil et al., 2009). This step is important, since the recovery process should be seen as highly individual and unique process (Leamy et al., 2011), in which therapists should pay attention to the individual needs of the patient. In this context, the QPR can also be used to set individual treatment goals (Neil et al., 2009). The current study now also provides the opportunity to use the QPR for above named purposes in the Netherlands. In a research context, the QPR provides standardized scores, which gives the opportunity to compare the effectiveness of interventions across different studies. Law et al. (2014) suggest that a medium effect size of 0.4 would be equivalent to a change of 4.63 points on the overall 15-item QPR score, which is comparable to the findings in the present sample. Trials focusing on personal recovery in the future could thus use the QPR as outcome measure and contribute to the body of recovery research.

## Conclusion

The present study suggests that the QPR is a reliable and valid tool to assess personal recovery in people with BD. Although personal recovery seems to have much overlap with well-being, it appears to be uniquely related with measures of symptomatology. The QPR can be used in both a clinical and research context to assess personal recovery and can be used by clinicians as a tool to improve the process of personal recovery.

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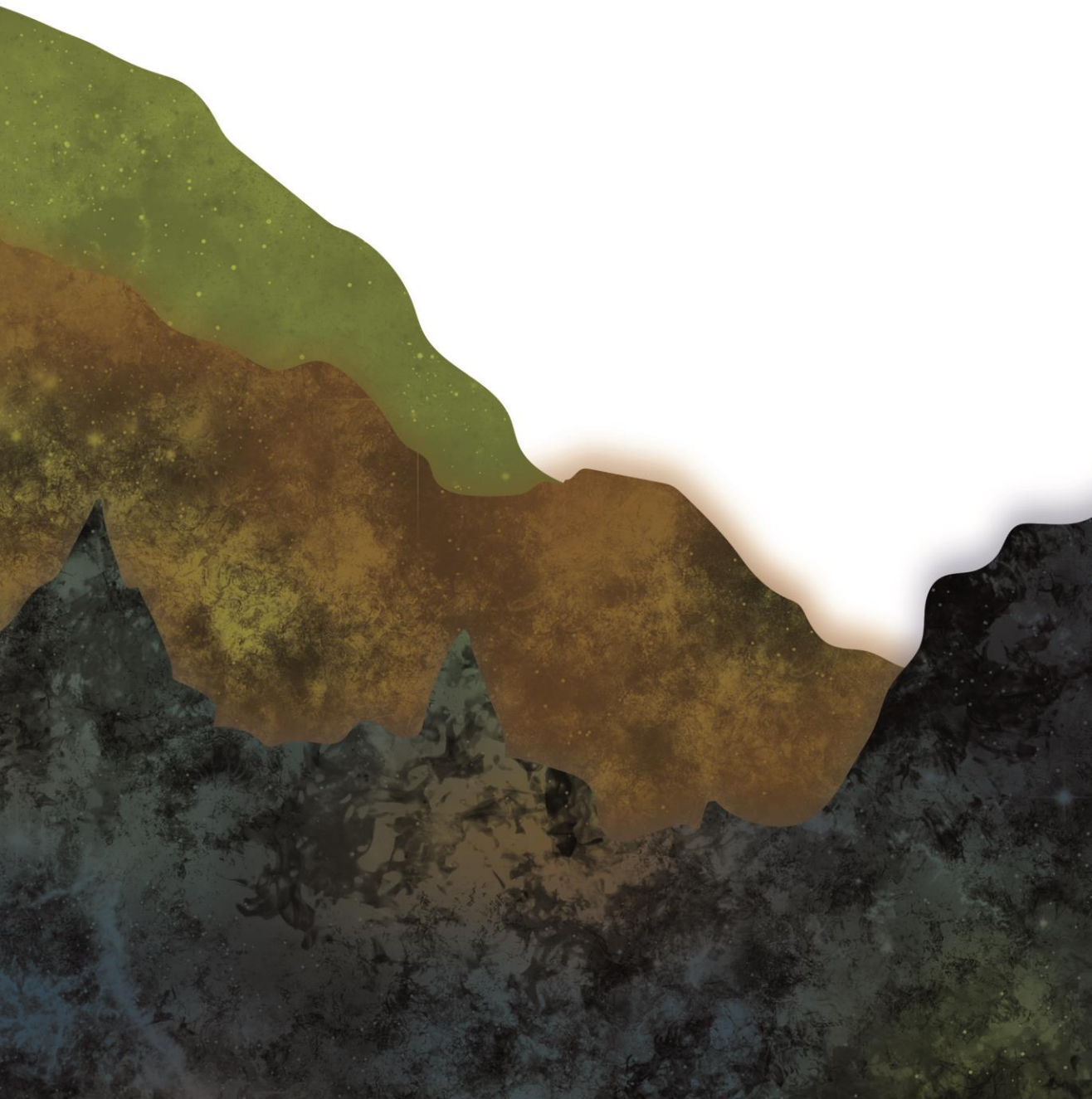
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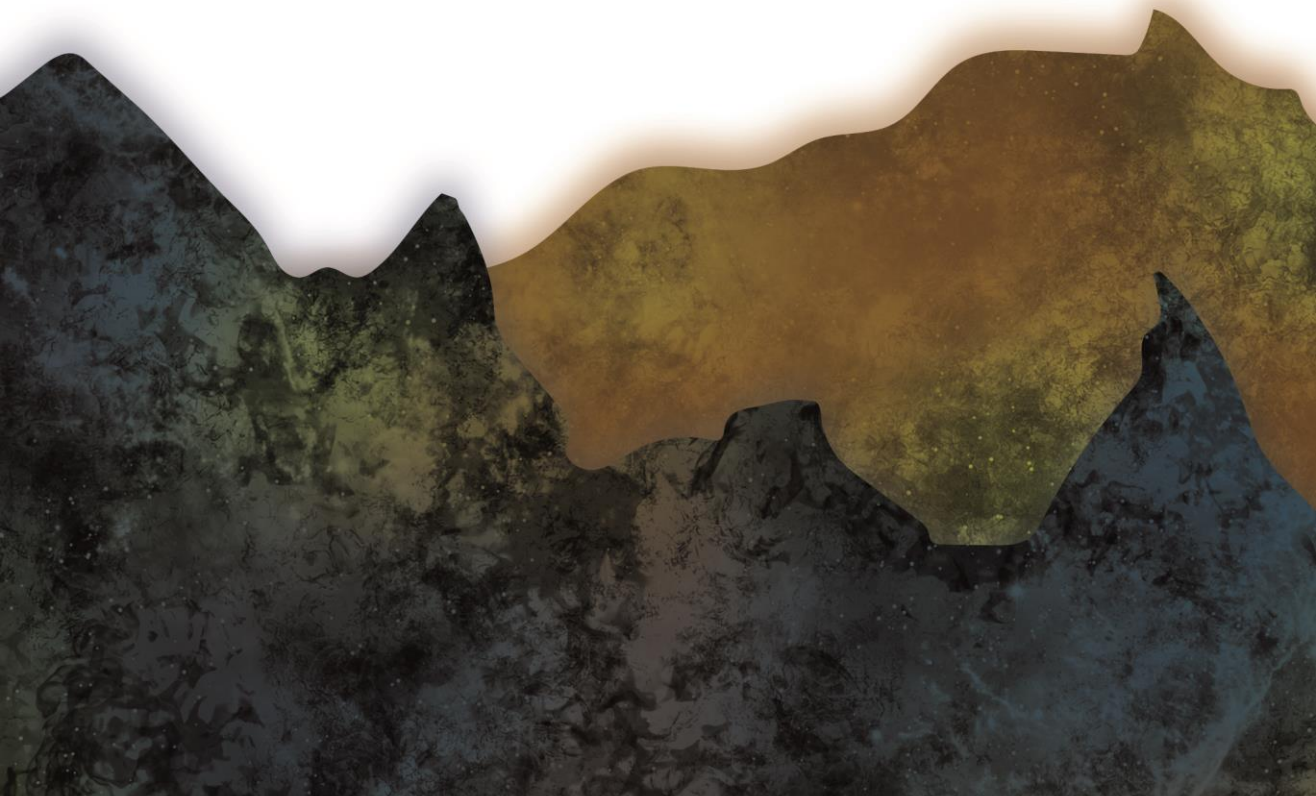




# CHAPTER 5

## Measuring personal recovery in people with bipolar disorder and exploring its relationship with well-being and social role participation

Kraiss, J. T., Ten Klooster, P. M., Chrispijn, M., Stevens, A. W., Kupka, R. W., & Bohlmeijer, E.T. (2019). Measuring personal recovery in people with bipolar disorder and exploring its relationship with well-being and social role participation. *Clinical Psychology & Psychotherapy*, 26(5), 540-549. <https://doi.org/10.1002/cpp.2371>



## **ABSTRACT**

The objective of this study was to evaluate the psychometric properties of the Responses to Positive Affect questionnaire (RPA) in a sample of persons with bipolar disorder (BD). A cross-sectional survey study with 107 persons with BD was conducted. The original 3-factor-model of the RPA was compared with a 2-factor-model. Construct validity was determined with measures of well-being, personal recovery, social role participation and measures of psychopathology and incremental validity was evaluated. Fit of the 3-factor-model was acceptable for most fit indices. Subscores of the RPA revealed a significant relationship with aspects of well-being, personal recovery and psychopathology. Dampening and self-focused positive rumination explained additional variance in personal recovery above and beyond well-being. The RPA is an internally consistent and valid tool to assess positive emotion regulation processes in persons with BD. Specifically the processes of dampening and emotion-focused positive rumination seem to play an important role in BD.

## INTRODUCTION

Bipolar disorder (BD) is a chronic mood disorder characterized by recurring depressive and (hypo)manic episodes (Grande et al., 2015; Kupka et al., 2008). In the fifth version of the Diagnostic and Statistical Manual of Mental Disorders (DSM-5; American Psychiatric Association, 2013), a distinction is made between bipolar I (BDI) and bipolar II (BDII) disorder. In BDII, a person experiences hypomanic and depressive episodes but never a full manic episode. Prevalence estimates reveals a lifetime prevalence of 0.6% for BDI and 0.4% for BDII (Merikangas et al., 2011). The economic burden of BD was estimated at 151 billion dollars per year in the United States (Dilsaver, 2011) and the illness is associated with decreased quality of life (Dean et al., 2004), negative social consequences (Calabrese et al., 2003), work-related problems (Fajutrao et al., 2009; Laxman et al., 2008), and high caregiver burden (Miller et al., 2014; Reinares et al., 2006).

In mental health care, the importance of personal recovery is becoming increasingly emphasized in the treatment of patients with mood disorders (Fava et al., 2007; Jones et al., 2013; Slade, 2010). Personal recovery can be defined as the ability to live a meaningful, hopeful, and contributing life, even in the presence of mental illness (Leamy et al., 2011). Closely related to this conception is the notion of mental health (Keyes, 2002; Keyes, 2005), defined as the absence of psychological symptoms, but also as the presence of well-being. Well-being comprises an emotional component (feeling well, e.g. the presence of positive emotions) and a social (e.g. contribution to society) and psychological dimension (living well, e.g. personal growth). Research indicates that well-being protects against the recurrence of psychopathology (Keyes et al., 2010; Lamers et al., 2015; Schotanus-Dijkstra et al., 2016; Trompetter et al., 2017). Furthermore, social role participation is increasingly seen as an important factor of recovery and refers to the ability to fulfill social roles (Jaeger & Hoff, 2012; Whitley & Drake, 2010). Social role participation has been shown to be important for building and maintaining self-esteem and autonomy (Gordeev et al., 2010) and contributes to long-term mental health (Oude Voshaar et al., 2016).

Research emphasizes the relevance of cognitive emotion regulation processes for the onset and recurrence of mood disorders (Nolen-Hoeksema, 1991). The impact of cognitive response to negative affect has been extensively studied (Aldao et al., 2010; Nolen-Hoeksema et al., 2008). However, less research has focused on the role of cognitive responses to positive affect, even though these processes might be equally important to understand emotion regulation processes in mood disorders (Carl et al., 2013; Wood et al., 2003). Two types of cognitive responses to positive affect appear especially relevant in the context of BD. *Dampening* refers to the suppression of positive affect to reduce the intensity of a positive mood (Quoidbach et

al., 2010). *Positive rumination* can be described as the tendency to respond to positive affective states with recurrent thoughts about positive experiences (Feldman et al., 2008). Paradoxically, dampening is associated with risk for mania (Johnson et al., 2008), and is heightened among people with BD (Edge et al., 2013; Johnson et al., 2016). Furthermore, dampening has been shown to predict depressive, and interestingly also manic symptoms over the course of six months in remitted persons with BD (Gilbert et al., 2013). Dampening positive emotions may thus be a particular maladaptive strategy (Gilbert et al., 2013). In addition, dampening has been shown to have a negative impact on life satisfaction (Quoidbach et al., 2010) and correlates positively with depressive symptoms and self-esteem (Feldman et al., 2008; Raes et al., 2010). Positive rumination strategies have been shown to be related with increases in different aspects of well-being, such as positive affect and life satisfaction (Quoidbach et al., 2010), lower levels of depressive symptoms (Feldman et al., 2008; Raes et al., 2010), but also with higher mania lifetime frequency (Gruber et al., 2011) and risk for manic or hypomanic episodes (Johnson & Jones, 2009). Emotion-focused positive rumination has been shown to be positively related to lifetime diagnoses of mania or hypomania and self-focused positive rumination has been related to current manic symptom severity (Johnson et al., 2008).

To assess cognitive responses to positive affect, the Responses to Positive Affect questionnaire (RPA) has been developed (Feldman et al., 2008). An initial psychometric evaluation of the questionnaire in a student sample yielded three underlying factors: (1) dampening, (2) emotion-focused positive rumination and (3) self-focused positive rumination (Feldman et al., 2008). Emotion-focused positive rumination refers to rumination on positive moods and somatic experiences with the aim to intensify current positive mood states, while self-focused rumination is signified by ruminating about positive qualities or personally relevant goals (Dempsey et al., 2011; Feldman et al., 2008; Olofsson et al., 2014). The factor structure of the RPA has been replicated in a Dutch community sample (Raes et al., 2010) as well as Swedish (Olofsson et al., 2014) and Chinese (Yang & Guo, 2014) student samples. The RPA revealed convergent validity in relation to measures of emotion regulation and self-esteem and incremental validity to depressive and manic symptoms.

Although responses to positive affect may be relevant for people with BD, there are some important gaps in current knowledge. The RPA has not been validated in a clinical sample of persons with BD. Also, the relationship between responses to positive affect and constructs relating to personal recovery and well-being have not been explored yet. The aim of the present study was to: 1) investigate the factor structure and internal consistency of the RPA in a clinical group of persons with BD, 2) study the construct validity of the RPA by investigating the relation with personal recovery, well-being, social role participation and symptomatology

and (3) determine the incremental validity of the RPA in explaining variance in personal recovery above and beyond measures of well-being and social role participation.

Concerning the factor structure of the RPA, we hypothesized that the 3-factor structure of the RPA found in earlier studies (Feldman et al., 2008; Nelis et al., 2016; Raes et al., 2010) will be replicated in the current clinical sample. With respect to construct validity, prior studies have shown negative associations between dampening and well-being related outcomes, such as life satisfaction and self-esteem (Quoidbach et al., 2010; Raes et al., 2010) and a positive relationship between positive rumination strategies and well-being outcomes (Quoidbach et al., 2010). Hence, it was hypothesized that dampening would correlate weakly to moderately with well-being. Although the relation between positive emotion regulation and personal recovery has not been explored yet, well-being and personal recovery share substantial conceptual overlap (Slade, 2010). Thus, we expected similar correlations with personal recovery. Since dampening might hamper people with BD to have a meaningful and contributing life, we expected a weak to moderate negative correlation between dampening and social role participation. Furthermore, we anticipated dampening and depressive symptoms to be moderately positively correlated (Feldman et al., 2008; Raes et al., 2010; Raes et al., 2011) and expected a low to moderate correlation between dampening and anxiety symptoms, since dampening was moderately related with anxiety (Olofsson et al., 2014) and ruminative thinking and brooding (Feldman et al., 2008; Raes et al., 2010). Furthermore, we expected that both self-focused and emotion-focused positive rumination show small to moderate correlations with constructs of manic symptoms (Gruber et al., 2011; Johnson & Jones, 2009; Johnson et al., 2008). Finally, we assumed that positive rumination would reveal a positive small to moderate correlation with constructs of personal recovery, well-being and social role participation (Quoidbach et al., 2010).

## METHOD

### Procedure

The study was approved by the Ethics Committee of the Faculty of Behavioral, Management and Social Sciences (BMS) of the University of Twente. Data were collected between April and July 2018. The survey was conducted via the online survey tool LimeSurvey (<https://www.limesurvey.org/>). Participants were gathered through convenience sampling via the Dutch patient association for BD, where the study was advertised through a notice in the newsletter of the patient association. Enrollment of participants was based on self-selection. Diagnoses of the participants were self-reported only and not confirmed by a clinical interview. At the start of the survey, participants were informed about the scope of the study and that

participation was voluntary, could be stopped at any moment, and that data were processed anonymously and confidentially. Ten shopping vouchers of 50 euro were raffled among all participants.

## Measures

Participants were asked to specify their gender, age, marital and employment status, ethnicity and educational background and also their type of diagnosis (BDI or BDII). Moreover, they were asked to state whether they were in psychological or psychiatric treatment and if they were taking medications in the context of their BD. Finally, participants were asked whether there were any recent adaptations in their medication and if they experienced relapse into a mood episode in the past 6 months. The following questionnaires were used to assess relevant constructs:

*Responses to positive affect.* The Responses to Positive Affect questionnaire (RPA; Feldman et al., 2008; Raes et al., 2010) consists of 17 items and measures cognitive responses to positive affective states. Respondents rate the items on a 4-point Likert scale, ranging from 1 (*almost never*) to 4 (*almost always*). The RPA consists of three subscales: (1) emotion-focused positive rumination (five items), (2) dampening (eight items) and (3) self-focused positive rumination (four items). Cronbach's  $\alpha$  in a prior psychometric evaluation was .80 for the subscales self-focused positive rumination and dampening, and .72 for the emotion-focused positive rumination (Raes et al., 2010).

*Personal Recovery.* The 15-item version of the Questionnaire about the Process of Recovery (QPR; Law et al., 2014; Neil et al., 2009) was used to assess personal recovery. Items of the QPR are scored on a 5-point Likert scale, ranging from 0 (*disagree strongly*) to 4 (*agree strongly*) and higher total scores indicate better personal recovery. For the purpose of this study, the QPR was translated by the first and second author of this article into Dutch through forward and backward translation. The English 15-item version of the QPR showed high internal consistency in a sample of psychotic individuals ( $\alpha = .89$ ; Williams et al., 2015) and in a sample of people with schizophrenia spectrum disorder ( $\alpha = .93$ ; Law et al., 2014). In the present study, Cronbach's  $\alpha$  was .93.

*Well-being.* The Mental Health Continuum-Short Form (MHC-SF; Lamers et al., 2011) is a 14-item self-report questionnaire assessing well-being on three dimensions: (1) emotional well-being (three items), (2) psychological well-being (six items) and (3) social well-being (five items). On a 6-point Likert-scale, respondents rate the frequency of feelings in the past month. For this study, the Dutch version of the MHC-SF was used, which revealed high internal

consistency for the subscales emotional ( $\alpha = .83$ ) and psychological well-being ( $\alpha = .83$ ) and adequate reliability for social well-being ( $\alpha = .74$ ; Lamers et al., 2011). Cronbach's  $\alpha$  in the current study was .89, .87 and .67 for emotional, psychological and social well-being, respectively.

*Social role participation.* Social role participation was assessed using the short version of the Social Role Participation Questionnaire (S-SRPQ; Oude Voshaar et al., 2016). This 12-item questionnaire measures the influence of mental health on six social roles along two dimensions: (1) satisfaction with role and (2) experienced psychological difficulty. Items are scored on a 5-point Likert Scale, reaching from 0 (*not satisfied at all* respectively *no difficulties at all*) to 4 (*very much satisfied* respectively *not possible*). Higher scores indicate more satisfaction and more experienced difficulties with a social role. A recent psychometric evaluation by Oude Voshaar et al. (2016) revealed high internal consistency for both subscales ( $\alpha = .86$ ). Cronbach's  $\alpha$  in the current study was .75 and .82 for the subscales satisfaction with role and experienced psychological difficulty, respectively.

*Depression and anxiety symptoms.* The 14-item Hospital Anxiety and Depression Scale (HADS; Zigmond & Snaith, 1983) assesses the presence of psychopathology in two domains: anxiety (7 items) and depressive symptoms (7 items). Respondents rate the frequency of symptoms over the last week from 0 (*not at all*) to 3 (*very often*), with higher scores indicating more psychopathology. Scores of 8 or higher on one of the subscales are seen as cut-off score for caseness (Bjelland et al., 2002). A psychometric evaluation by Spinhoven et al. (1997) found acceptable internal consistency for the depression subscale ( $\alpha = .79$ ) and good reliability for the anxiety subscale ( $\alpha = .88$ ). Cronbach's  $\alpha$  in the current study was .73 for the depression and .85 for the anxiety subscale.

*Manic symptoms.* The Altman Self-Rating Mania Scale (ASRM; Altman et al., 1997) is a 5-item self-rating scale measuring symptoms of mania in the past week. Items include symptoms of mania (e.g. inflated self-confidence). Each item provides five response options with increasingly severe descriptions. Total scores are calculated by summing up the scores on each item and higher scores indicate more manic symptoms. Scores of 6 or higher are an indication for the presence of meaningful manic symptoms (Altman et al., 1997). The ASRM has been shown to have high test-retest reliability (Altman et al., 1997), to be sensitive to changes in a clinical group (Altman et al., 2001) and to predict related outcomes in student samples (Meyer et al., 2004). Cronbach's  $\alpha$  was .73 in the current study.



## Statistical analyses

Statistical analyses were performed using Mplus version 7.11 (Muthén & Muthén, 2010), RStudio (R Core Team, 2020) and the statistical package for social sciences version 25 (SPSS). First, confirmatory factor analyses (CFA) were conducted with Mplus to investigate whether the dimensionality of the RPA found in earlier studies in non-clinical samples could be confirmed in the current BD sample. Therefore, we fitted the original 3-factor-model, in which 5 items load on the emotion-focused positive rumination factor, 4 items on the self-focused positive rumination factor and 8 items on the dampening factor. Afterwards, we tested a 2-factor-model, in which the 9 items of the two positive rumination subscales loaded on a single latent positive rumination factor and 8 items on the dampening factor. Fit of the models was based on the restrictive assumption that error terms of items were uncorrelated. Since correlated error terms are indicative of model misspecification, but are not unusual in psychological assessment instruments (Byrne et al., 1995), we decided to only allow error correlations if this made substantive sense and if none of the restrictive models achieved acceptable fit (Jöreskog, 1993). Considering the ordinal nature of the data and the small sample size, we followed the recommendations by Flora and Curran (2004) and Moshagen and Musch (2014) and used a robust diagonally weighted least square mean and variance adjusted (WLSMV) estimation method. In the literature, factor loadings higher than .30 or .40 are usually seen as satisfactory (Floyd & Widaman, 1995; Hair et al., 2009) and we used  $> .35$  as compromise. Model fit for both models was assessed using chi-square statistics ( $\chi^2$ ), in which a smaller value indicates better model fit and the ratio between  $\chi^2$  and degrees of freedom should be  $< 3$  for an acceptable fit (Kline, 2015). Furthermore, the comparative fit index (CFI), Tucker-Lewis Index (TLI), weighted root-mean-square residual (WRMR) and root-mean-square error approximation (RMSEA) were calculated to determine the model fit (Hu & Bentler, 1998). Values  $\geq 0.90$  were seen as acceptable and values  $\geq 0.95$  as good model fit for the CFI and TLI, whereas RMSEA values  $\leq 0.80$  and  $\leq 0.50$  were considered as acceptable and good model fit, respectively (Browne & Cudeck, 1992; Hu & Bentler, 1999). For the WRMR, estimates around 1 were seen as good model fit (DiStefano et al., 2018; Yu, 2002). The difference in fit between the 3-factor and 2-factor-model was statistically tested using the Mplus DIFFTEST procedure, which computes differences in  $\chi^2$  values of nested models.

Internal consistency was evaluated by calculating Cronbach's alpha ( $\alpha$ ) and categorical McDonald's omega ( $\omega$ ) for the subscales of the 2-factor and 3-factor-model (Dunn et al., 2014). McDonald's  $\omega$  deals with the limitation of Cronbach's  $\alpha$ , which assumes tau-equivalence and thus shows deficiencies for congeneric models (Kelley & Pornprasertmanit, 2016). Using the MBESS package (Kelley, 2018),  $\alpha$  and  $\omega$  coefficients with 95% bias corrected and accelerated bootstrap confidence intervals (CI) based on 1000 bootstrap samples were calculated. Alpha

and omega estimates  $> .70$  and  $> .80$  were seen as acceptable and good internal consistency (Cicchetti, 1994).

For examining construct validity, bivariate Pearson's correlation coefficients were calculated between the three subscales of the RPA and criterion measures. Correlation coefficients between  $.1$  and  $.3$  were interpreted as weak, coefficients larger than  $.3$  and smaller than  $.5$  as moderate and larger than  $.5$  as strong correlations (Cohen, 1988). To determine incremental validity of the RPA in explaining variance in personal recovery, multiple hierarchical regression analyses were performed with total scores of the QPR as dependent variable. In the first step, scores of the MHC-SF, S-SRPQ, HADS and ASRM were entered, respectively. Scores of the subscales of the RPA were entered in step 2. Significant changes in explained variance after the second step ( $p < .05$ ) were seen as indicative for incremental validity.

**Table 2.** Sample characteristics ( $N = 107$ )

Variable	Category	N	%
Age	Mean = 52 (range 23 – 77)		
Gender	Female	82	76.6
	Male	25	23.4
Marital status	Married	56	52.3
	Never married	28	26.6
	Divorced	22	20.6
	Widowed	1	0.9
Employment status	Unable to work	39	36.4
	Paid work	26	24.3
	Unpaid/voluntary work	14	13.1
	Retired	10	9.3
	Housewife/houseman	5	3.7
	Self-employed	4	4.7
	Student	3	2.8
	Other	6	5.6
Education	Low	14	13.2
	Moderate	35	33.1
	High	57	53.7
Diagnosis	BDI	42	39.3
	BDII	51	47.7
	Unknown	14	13.0
Relapse into mood episode (past 6 months)	Yes	59	55.1
	No	48	44.9
Currently in psychological or psychiatric treatment	Yes	89	83.2
	No	18	16.8
Currently taking medication	Yes	92	95.3
	No	5	4.7

# RESULTS

## Description of the sample

Mean age of the 107 participants was 52 years (SD = 11.26, range 23 – 77). Of the sample, 55.1% ( $n = 59$ ) experienced a relapse into a depressive or manic episode in the past 6 months and 50.5% had adaptations to their medications in the past 6 months ( $n = 54$ ). Eight participants were administered to a psychiatric hospital due to symptoms related to BD. Of the participants who completed the HADS, 61 (62.2%) scored above the clinical threshold for anxious symptomatology and 68 (69.4%) for depression. For manic symptomatology, 20 (20.4%) participants scored above the cut-off for manic symptomatology. Sample characteristics are summarized in Table 1.

## Factor structure and internal consistency

An initial CFA with the original 3-factor-model and 2-factor-model revealed a very low factor loading of 0.13 in both models for item number 6 (“*Think this is too good to be true*”). We decided to drop this item for further analyses, a strategy which is in line with earlier studies (Kim & Kwon, 2014; Nelis et al., 2016). Afterwards, we again fitted a 2-factor-model and 3-factor-model, in which only 7 items load on the latent dampening factor. Fit indices for the two- and three factor models without item 6 are presented in Table 2.

**Table 2.** Goodness of fit indices for two models tested in CFA

Model	$\chi^2$	$df$	CFI	TLI	WRMR	RMSEA (90% CI)
2-factor-model	194.70	103	.908	.893	1.098	.091 (.071 – .111)
3-factor-model	181.55	101	.919	.904	1.032	.086 (.066 – .106)

*Note.*  $N = 107$ , CFI = comparative fit index, CI = Confidence interval,  $df$  = degrees of freedom, RMSEA = root-mean-square error of approximation, TLI = Tucker-Lewis Index, WRMR = weighted root-mean-square residual,  $\chi^2$  = chi-square statistics.

Overall model fit indices were slightly better for the 3-factor-model. This was confirmed by the chi-square test for difference testing, which revealed that the two models significantly differed in their fit ( $\Delta\chi^2 = 11.32$ ,  $\Delta df = 2$ ,  $p < 0.01$ ). CFI estimates were acceptable for both tested models, but the TLI was acceptable only for the 3-factor-model. The ratio between  $\chi^2$  and degrees of freedom was  $< 2$  for both models, indicating good fit. For the 2- and 3-factor-model, WRMR estimates (1.098 and 1.032, respectively) and RSMEA values (.091 and .086, respectively) fell short of the criterion for adequate fit. However, it should be noted that the WRMR is an experimental test statistic and cut-offs for fit are still debated. Modification

indices revealed an improvement of the 3-factor-model, if the error correlation between item 10 (“*Remind yourself that these feelings won’t last*”) and 15 (“*I am lucky for now, but this will end soon*”) was allowed ( $r = .647$ ). In this model, CFI and TLI values increased to .924 and .938, respectively, RMSEA estimates improved to .076 and the chi-square value decreased ( $\chi^2 = 161.80$ ,  $df = 100$ ,  $p < .001$ ). Besides the WRMR, all fit indices revealed an adequate fit in this model and the chi-square test for difference indicated a significant better fit of the 3-factor-model with error correlation compared to the original 3-factor-model without error correlation ( $\Delta\chi^2 = 19.75$ ,  $\Delta df = 1$ ,  $p < .001$ ). However, since the 3-factor-model without error correlation was already showing acceptable fit, we decided to adhere to this model and not allow error correlations for further analyses.

Standardized factor loadings for the 3-factor-model (without error correlation) and 2-factor-model and corresponding Cronbach’s  $\alpha$  and categorical McDonald’s  $\omega$  coefficients are shown in Table 3. All standardized factor loadings were above .35 and McDonalds’s  $\omega$  coefficients revealed good and adequate internal consistency for dampening ( $\omega = .86$ ), emotion-focused ( $\omega = .78$ ) and self-focused positive rumination ( $\omega = .77$ ).

## Construct validity

Table 4 presents the mean values and bivariate intercorrelations between the subscales of the RPA and criterion measures. Dampening was weakly negatively associated with emotional, psychological well-being and overall well-being. Higher levels of dampening were found to be moderately associated with less personal recovery and with more anxiety symptoms. Scores of emotion-focused positive rumination were moderately positively correlated with all facets of well-being and also with personal recovery. Weak negative correlations were found between emotion-focused positive rumination and experienced difficulty with social roles and anxiety symptoms and a moderate negative association with depressive symptoms. No significant relation was found between self-focused positive rumination and the subscales emotional ( $p = .06$ ), social ( $p = .11$ ) and psychological well-being ( $p = .06$ ). Surprisingly, self-focused positive rumination was moderately associated with both higher levels of personal recovery and symptoms of mania.

## Incremental validity

To determine incremental validity of the RPA, we conducted several multiple hierarchical regressions analyses with scores of the QPR as criterion variable. The RPA explained 4% additional variance in personal recovery above and beyond measures of well-being ( $p < .01$ ).

**Table 3.** Standardized factor loadings for the two tested models and corresponding Cronbach's alpha and categorical McDonald's omega for subscales of the RPA

RPA Item	3-factor-model			2-factor-model		
	D	EF	SF	D	PR	PR
<b>Dampening</b>						
Think about things that could go wrong (rpa9)	.57	-	-	.58	-	-
Remind yourself that these feelings won't last (rpa10)	.77	-	-	.78	-	-
Think "People will think I am bragging." (rpa11)	.54	-	-	.53	-	-
Think about how hard it is to concentrate (rpa12)	.63	-	-	.62	-	-
Think "I don't deserve this." (rpa14)	.65	-	-	.64	-	-
Think "I am lucky for now, but this will end soon." (rpa15)	.84	-	-	.84	-	-
Think about the things that have not gone well for you (rpa17)	.66	-	-	.66	-	-
<b>Emotion-focus</b>						
Notice how you feel full of energy (rpa1)	-	.72	-	-	-	.69
Focus on enjoying this moment (rpa2)	-	.73	-	-	-	.68
Think about how happy you feel (rpa7)	-	.48	-	-	-	.45
Think about how strong you feel (rpa8)	-	.78	-	-	-	.75
Think about how proud you are of yourself (rpa16)	-	.71	-	-	-	.68
<b>Self-focus</b>						
Think "I am getting everything done." (rpa3)	-	-	.84	-	-	.82
Think how you feel ready to do anything (rpa4)	-	-	.82	-	-	.77
Think "I am the best I could be." (rpa5)	-	-	.64	-	-	.61
Think "I am achieving everything I could want." (rpa13)	-	-	.80	-	-	.77
<b>Cronbach's alpha (95% CI)</b>						
	.80 (.73-.86)	.77 (.69-.83)	.75 (.65-.82)	.80 (.73-.86)	.85 (.80-.89)	
<b>McDonald's omega (95% CI)</b>						
	.86 (.74-.91)	.78 (.66-.85)	.77 (.65-.83)	.86 (.74-.91)	.89 (.80-.93)	

Note. D = Dampening, EF = Emotion-focused positive rumination, SF = Self-focused positive rumination, PR = Positive rumination, CI = Confidence interval.

**Table 4.** Descriptive statistics and bivariate correlations between the RPA and criterion measures

Measure	M (SD)	Dampening	Emotion-focus	Self-focus
RPA ( <i>n</i> = 107)				
Dampening	13.35 (3.98)	-	-	-
Emotion-focus	12.30 (2.90)	-.26**	-	-
Self-focus	8.21 (2.63)	-.15	.62**	-
MHC-SF ( <i>n</i> = 102)				
Emotional well-being	7.40 (3.96)	-.22*	.42**	.19
Social well-being	8.36 (4.94)	-.18	.34**	.16
Psychological well-being	13.58 (7.19)	-.21*	.37**	.19
Total score	29.34 (14.48)	-.20*	.41**	.20*
QPR ( <i>n</i> = 102)				
Total score	37.66 (11.14)	-.33**	.42**	.31**
S-SRPQ ( <i>n</i> = 98)				
Satisfaction with role	14.62 (5.41)	-.17	.21	.11
Experienced difficulty	16.77 (5.41)	.20*	-.22*	-.08
HADS ( <i>n</i> = 98)				
Anxiety symptoms	8.71 (4.64)	.47**	-.24*	-.04
Depression symptoms	9.61 (3.99)	.20*	-.39**	-.19
ASRM ( <i>n</i> = 98)				
Total score	2.99 (3.23)	.14	.19	.34**

*Note.* Variations in *n* due to missing data, ASRM = Altman Self-Rating Mania Scale, HADS = Hospital Anxiety and Depression Scale, MHC-SF= Mental Health Continuum-Short Form, QPR = Questionnaire about the Process of Recovery, RPA = Responses to Positive Affect questionnaire (without item 6), S-SRPQ = Short-Social Role Participation Questionnaire.

\* $p < .05$ , \*\* $p < .01$ .

Dampening and self-focused positive rumination significantly explained variance in personal recovery independently of the MHC-SF subscales. Furthermore, the RPA explained 14% additional variance in personal recovery above and beyond social role participation ( $p < .001$ ). Dampening significantly explained variance above and beyond personal recovery independent of the scores of social role participation ( $p < .05$ ). Findings regarding incremental validity are summarized in Table 5 and Table 6.

**Table 5.** Summary of hierarchical regression analysis for MHC-SF and RPA subscales and personal recovery (QPR)

Variable	B	SE	$\beta$	t	$\Delta R^2$
Step 1					
Constant	19.17	1.43		13.44***	.69***
Emotional well-being (MHC-SF)	1.08	.25	.39	4.31***	
Psychological well-being (MHC -SF)	.75	.17	.49	4.52***	
Social well-being (MHC-SF)	.03	.19	.01	.15	
Step 2					
Constant	21.80	3.77		5.47***	.04**
Emotional well-being (MHC-SF)	1.02	.25	.36	4.16***	
Psychological well-being (MHC -SF)	.72	.16	.46	4.53***	
Social well-being (MHC-SF)	.02	.18	.01	.10	
Dampening (RPA)	-.38	.15	-.14	-2.48*	
Emotion-focus (RPA)	-.18	.29	-.05	-.62	
Self-focus (RPA)	.69	.29	.17	2.42*	

Note. MHC-SF = Mental Health Continuum-Short Form, QPR = Questionnaire about the Process of Recovery, RPA = Responses to Positive Affect questionnaire. \* $p < .05$ , \*\* $p < .01$ , \*\*\* $p < .001$ .

Additionally, we investigated whether the RPA explained variance in personal recovery above and beyond measures of symptomatology. The RPA explained 9.6% additional variance above and beyond depressive symptoms ( $F_{\text{change}}(1, 96) = 7.21, p < .001$ , adjusted  $R^2$  step 2 = .577). More specifically, self-focused positive rumination ( $p < .05$ ) and dampening ( $p < .001$ ) independently explained personal recovery above and beyond depressive symptomatology. The RPA explained 17.5% additional variance in personal recovery outcomes above and beyond anxiety symptoms ( $F_{\text{change}}(1, 96) = 8.85, p < .001$ , adjusted  $R^2$  step 2 = .374). Only self-focused positive rumination independently explained variance in personal recovery above anxious symptomatology ( $p < .01$ ). Finally, the RPA explained 27.4% additional variance above and beyond ASRM scores ( $F_{\text{change}}(1, 96) = 12.46, p < .001$ , adjusted  $R^2$  step 2 = .289). In this model, dampening ( $p < .001$ ) and emotion-focused positive rumination ( $p < .05$ ) independently explained variance in personal recovery above and beyond manic symptomatology.

**Table 6.** Summary of hierarchical regression analysis for S-SRPQ and RPA subscales and personal recovery (QPR)

Variable	<i>B</i>	<i>SE</i>	$\beta$	<i>t</i>	$\Delta R^2$
Step 1					
Constant	26.28	6.84		3.84***	.41***
Satisfaction with role (S-SRPQ)	1.12	.25	.52	4.74***	
Experienced difficulty (S-SRPQ)	-.34	.22	-.16	-1.50	
Step 2					
Constant	19.38	7.62		2.73**	.14***
Satisfaction with role (S-SRPQ)	1.07	.22	.47	4.87***	
Experienced difficulty (S-SRPQ)	-.22	.20	-.11	-1.08	
Dampening (RPA)	-.52	.20	-.19	-2.62*	
Emotion-focus (RPA)	.65	.35	.17	1.90	
Self-focus (RPA)	.65	.37	.16	1.78	

Note. QPR = Questionnaire about the Process of Recovery, RPA = Responses to Positive Affect, Questionnaire, S-SRPQ = Short-Social Role Participation Questionnaire. \* $p < .05$ , \*\* $p < .01$ , \*\*\* $p < .001$ .

## DISCUSSION

The present study set out to evaluate the psychometric properties of the RPA in a clinical sample of persons with BD. The RPA was designed as a measure of positive emotion regulation (Feldman et al., 2008). Growing evidence of responses to positive affect in mental disorders emphasizes the importance of these processes in mood disorders (Carl et al., 2013; Edge et al., 2013; Gruber et al., 2011; Johnson et al., 2008). Several prior studies evaluated the psychometric properties of the RPA (Feldman et al., 2008; Kim & Kwon, 2014; Olofsson et al., 2014; Raes et al., 2010; Yang & Guo, 2014), but used community or student samples.

Results of the CFA showed that the 3-factor-model had a significant better fit to the data than the 2-factor-model, coinciding with prior studies (Feldman et al., 2008; Raes et al., 2010; Yang & Guo, 2014). Most fit indices showed an adequate fit, but the RMSEA fell short of the criterion for acceptable fit. However, it should be noted that fit indices are differentially sensitive for the type of model and several misspecifications. For this reason, Hu and Bentler (1998) recommend to report several different fit indices to assess the overall quality of a model. The relevance of distinguishing between emotion-focused and self-focused positive rumination was also apparent from their differential associations with criterion measures. In the course of the CFA, we decided to remove item number 6 of the RPA (*"This is too good to be true"*) from the further analyses. One possible explanation for the low factor loadings of this item might be the position in the questionnaire, since the five items before and the two items after the



item belong to positive rumination. Alternatively, the formulation of the item could be considered in a non-dampening manner (Nelis et al., 2016).

Results of the correlational analyses showed that more dampening was moderately associated with reduced personal recovery and weakly with lower well-being, coinciding with our hypotheses (Quoidbach et al., 2010). This finding can be interpreted as dampening being an obstacle for people with BD to reach personal recovery. Mansell (2016) argues that the fear of feeling too good and becoming manic, might be one of the greatest problems in BD. This fear might lead to dampening and inhibit people from leading a meaningful life. This hypothesis is supported by a moderate correlation between dampening and anxiety symptoms in the present study. Moreover, earlier studies suggest that persons with BD tend to avoid rewarding activities to prevent mania (Edge et al., 2013) and that dampening is associated with poorer well-being (Quoidbach et al., 2010) and quality of life (Edge et al., 2013). Interestingly, our results revealed only a weak relationship between dampening and depressive symptoms. It would be interesting for future research to specifically explore the role of dampening in relation to well-being and symptomatology in this group.

Emotion-focused positive rumination was moderately correlated with measures of well-being and personal recovery, which is in line with earlier research (Gilbert et al., 2013; Quoidbach et al., 2010). We found a nonsignificant relationship between emotion-focused positive rumination and manic symptoms, but a significant relationship with more anxiety and depressive symptoms. Interestingly, for self-focused positive rumination, this relationship was inverted, as it showed a significant relationship with manic symptoms, but not with depression or anxiety symptoms. One possible explanation for this finding might be that self-focused positive rumination is associated with manic symptoms because it focusses on personal qualities and specific actions and, while emotion-focused positive rumination specifically focusses on affective processes. Also, the low proportion of participants meeting the threshold for manic symptoms, might be a possible explanation for some of the null results around manic symptoms.

Scores of the RPA showed incremental validity by explaining variance in personal recovery above and beyond well-being. This is a remarkable finding with respect to the utility of the RPA, since the concept of well-being shares several commonalities with personal recovery (Slade, 2010). Specifically, dampening and self-focused positive rumination explained additional variance in personal recovery. This supports the assumption that tackling dampening might be an effective way to improve personal recovery. We also found that dampening explained variance in personal recovery above and beyond social role

participation, again underscoring the importance of dampening in relation to personal recovery.

## Implications

Some relevant implications for both research and clinical practice arise from these findings. First, when using the RPA for assessment purposes, caution should be paid to item number 6. We advise that this item is not used in calculating the dampening subscale score. Future research could examine if adjusting the position of the item in the questionnaire results in a better fit with its assumed factor. Since the dampening subscale also revealed good internal consistency without this item, it could also be considered to remove the item altogether. Second, our findings suggest that positive emotion regulation is an important factor in BD. Health care professionals working with patients with BD should try to tackle the tendency to dampen positive emotions and at the same time try to foster positive rumination, especially when aiming to achieve personal recovery and improved well-being.

## Limitations and future research

Our study has several limitations, giving opportunities for future research. First, the sample size was relatively small. For example, Kline (2015) recommends a sample of at least 200 participants for sound structural equation modeling procedures. Although we tried to compensate for the relatively small sample by using WLSMV estimation, the modest sample might have distorted the fit indices of the factor models. Future research could investigate if the superiority of the 3-factor-model can be confirmed and whether the factor structure is invariant across subgroups of patients. Second, no conclusions about causality can be drawn from the study. For example, it cannot be concluded that dampening actually leads to less personal recovery. A reverse causal direction might also be plausible. Future research should explore the relationship between these concepts in a longitudinal design. Third, diagnosis of BD was based on self-report, which leaves the possibility that not all participants had a clinically confirmed diagnosis of BD. However, since participants were recruited via the patient association for BD, most people indicated a specific type of BD and 95% of the sample stated that they were taking medication in the context of their BD, it can be assumed that the vast majority of the sample actually had BD.

## Conclusion

Our findings suggest that the RPA is an internally consistent and valid tool to assess positive emotion regulation processes in persons with BD. Furthermore, dampening and positive rumination seem to play an important role in BD patients and specifically in relation to

personal recovery and well-being. This relationship deserves closer examination in the context of longitudinal studies.

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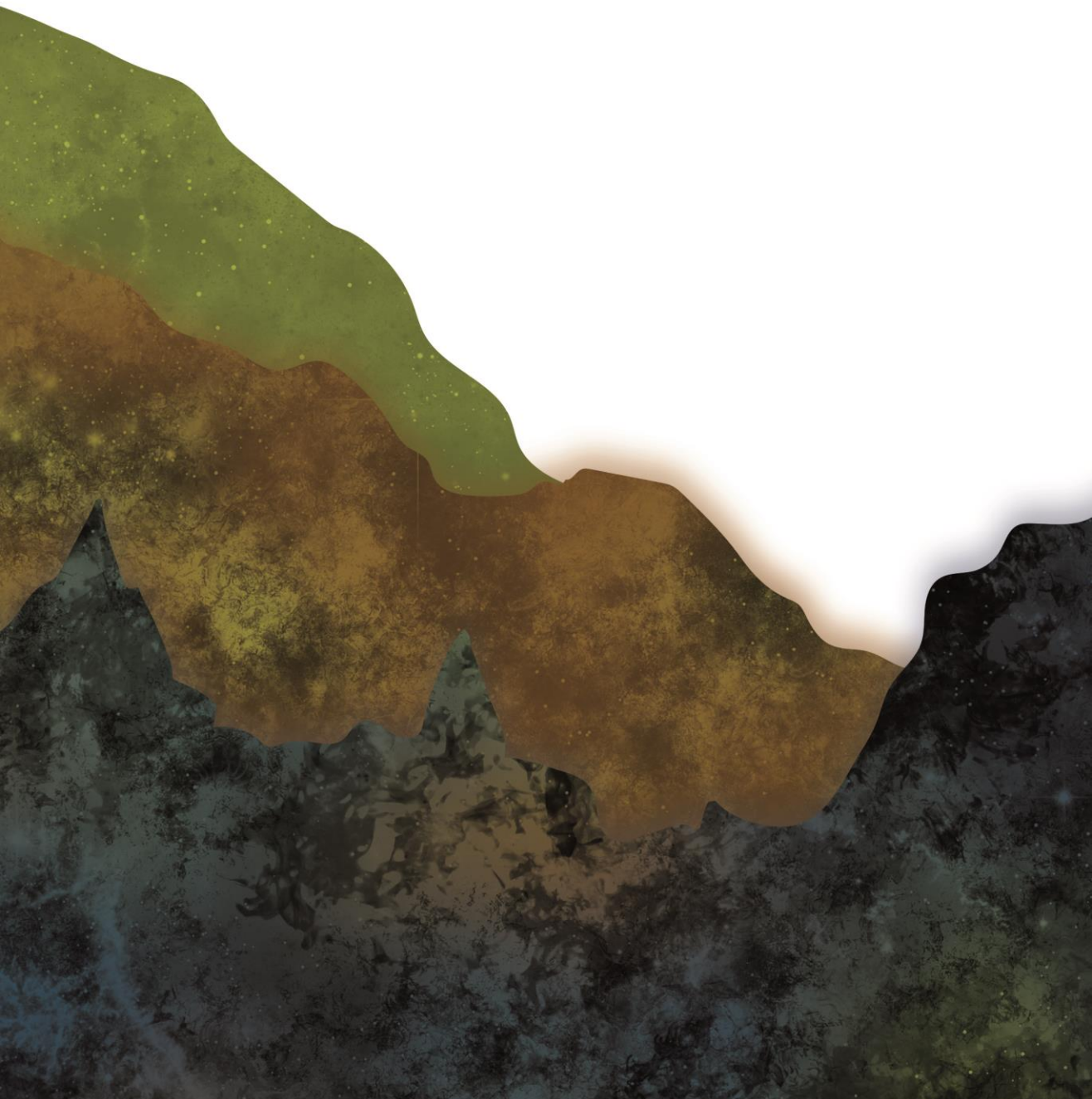
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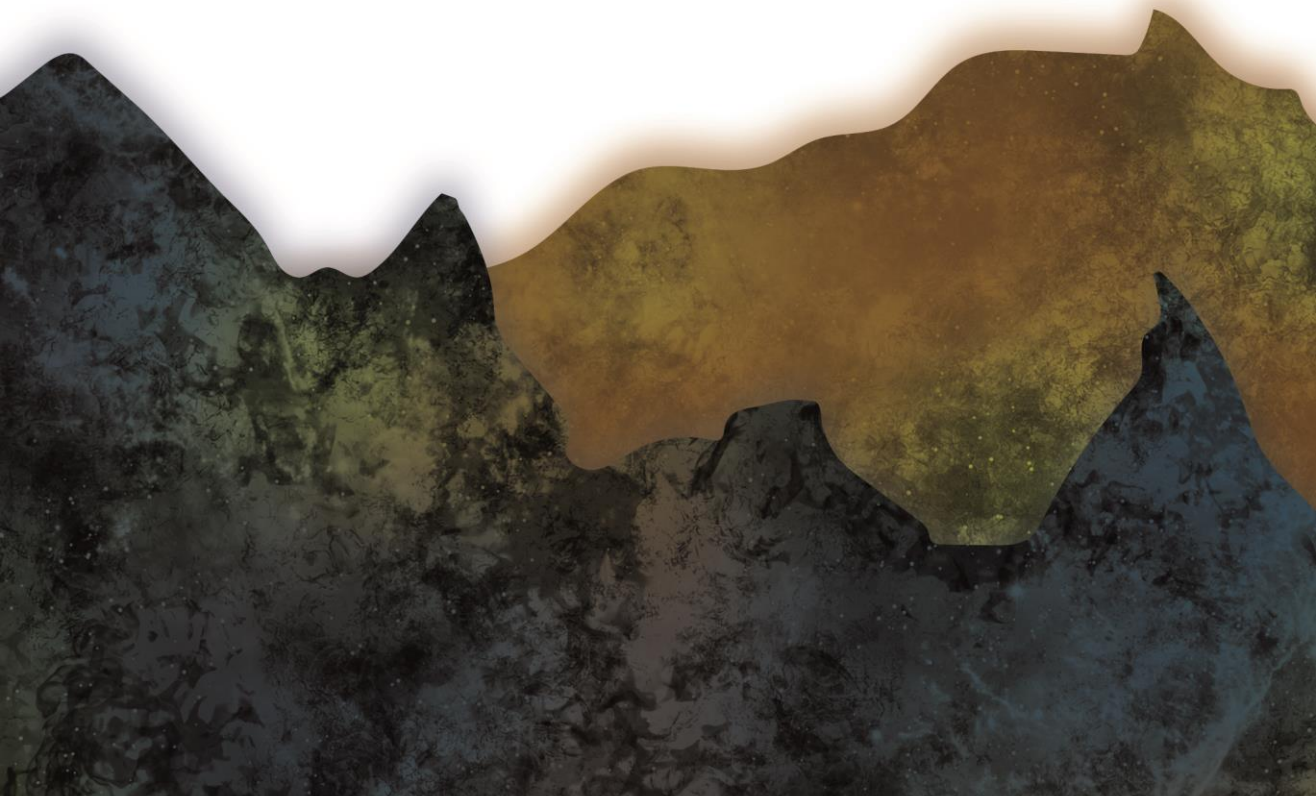
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# CHAPTER 6

## Exploring factors associated with personal recovery in bipolar disorder

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## ABSTRACT

Personal recovery is increasingly recognized as important outcome for people with bipolar disorder (BD), but research addressing associated factors of personal recovery in this group remains scarce. This study aimed to explore the association of sociodemographic variables, social participation, psychopathology, and positive emotion regulation with personal recovery in BD. Baseline data from a randomized controlled trial and survey data were combined ( $N = 209$ ) and split into a training ( $n = 149$ ) and test sample ( $n = 60$ ). Block-wise regression analyses and model training were used to determine the most relevant predictors. The final parsimonious model was cross-validated in the test sample. In the final parsimonious model, satisfaction with social roles ( $\beta = .44, p < .001$ ), anxiety symptoms ( $\beta = -.33, p < .001$ ), manic symptoms ( $\beta = .28, p < .001$ ), and emotion-focused positive rumination ( $\beta = .26, p < .001$ ) were independently associated with personal recovery. The model explained 57.3% variance in personal recovery (adjusted  $R^2 = .56$ ) and performed well in predicting personal recovery in the independent test sample (adjusted  $R^2 = .49$ ). Our findings suggest that especially social participation, anxiety and positive rumination might be relevant treatment targets when aiming to improve personal recovery.

## INTRODUCTION

Bipolar disorder (BD) is a chronic mental disorder characterized by recurrent depressive and (hypo)manic mood episodes, intertwined with euthymic phases, in which patients are relatively symptom-free. A distinction is made between bipolar I (BDI) and bipolar II (BDII) disorder. In BDII, an individual experiences depressive and hypomanic episodes, but never a full-blown manic episode (Goodwin & Jamison, 2007). BD carries the highest suicide risk compared to all other psychiatric illnesses (Miller & Black, 2020; Plans et al., 2019) and is associated with impairments in cognitive functioning (Cotrena et al., 2016), quality of life (Sylvia et al., 2017), high caregiver burden (Blanthorn-Hazell et al., 2018), and substantial societal costs (Cloutier et al., 2018; Kraiss et al., 2020).

Besides clinical and functional recovery, the importance of improving personal recovery in people with mental disorders is increasingly recognized. Personal recovery has been defined as *“a deeply personal, unique process of changing one’s attitudes, values, feelings, goals skills and/or roles [...] and a way of living a satisfying, hopeful, and contributing life, even with limitations caused by the illness”* (Anthony, 1993, p. 527). Especially anglophone countries emphasize the importance of recovery-focused approaches in their policies and health services (Bird et al., 2014). A literature review identified five key components of personal recovery in mental disorders, namely connectedness, hope, identity, meaning, and empowerment. The authors of this review also concluded that more research is needed about contributing factors for personal recovery (Leamy et al., 2011). Personal recovery related outcomes are highly valued by people with mental disorders and are considered important objectives of treatment (de Vos et al., 2017; Leamy et al., 2011; Mead & Copeland, 2000), also increasingly for people with BD (Jones et al., 2015). People with BD describe similar factors as relevant for personal recovery. Respect, hope and empowerment (Tse et al., 2014), becoming the director of your own life (Warwick et al., 2019) and social support and companionship (Mansell et al., 2010) were described as important parts of recovery. Also, research suggests that recovery in BD is not only about being symptom-free (Todd et al., 2012) and that clinical recovery alone may misrepresent treatment success from the perspective of people with BD (Mezes et al., 2020). However, quantitative research about factors that are associated with personal recovery in BD remains scarce.

One factor that might be an important determinant of personal recovery from chronic psychiatric conditions is social role participation (Jaeger & Hoff, 2012), referring to the degree to which an individual is able to fulfil social roles, such as having intimate relationships or being a working employee (Oude Voshaar et al., 2016). Social functioning is described as integral part of recovery (Lieberman, 2009). Having less satisfaction with social roles has been associated

with increased depression and stress, and greater role limitations were associated with increased health care utilization (Gignac et al., 2013). Also, the degree to which an individual is able to fulfil social roles has been shown to be important for building and maintaining self-esteem (Gordeev et al., 2010). Although the direct link between personal recovery and social participation has not yet been investigated in BD, increased health care utilization and depression symptoms as well as lower self-esteem might be indicative of low recovery. Furthermore, being able to work has been described as relevant factor for recovery by people with BD (Borg et al., 2013). Therefore, social role participation might also be a relevant determinant for personal recovery in BD.

Another important process for the onset, persistence, and recurrence of mental disorders is positive emotion regulation (Carl et al., 2013), which is defined as the way in which people react to their positive affective states. Two important positive emotion regulation strategies are *dampening* and *positive rumination*. Dampening is described as the suppression of positive moods to reduce the intensity of positive affect (Quoidbach et al., 2010), while positive rumination has been described as the tendency to respond to positive emotions with recurrent thoughts about positive experiences (Feldman et al., 2008).

Research suggests that dampening might be a particular maladaptive strategy in BD (Gilbert et al., 2013). Dampening has been shown to be heightened among people with BD (Edge et al., 2013; Johnson et al., 2016) and to predict depressive and also manic symptoms over the course of six months in remitted persons with BD (Gilbert et al., 2013). Mansell (2016) describes the fear of becoming manic and experiencing relapse as one of the greatest problems in BD. One consequence of this fear might be dampening of positive emotions and avoiding positive experiences (Edge et al., 2013), which may prevent people from leading a meaningful life and hinder recovery. Dampening could thus eventually be an obstacle for personal recovery in BD. Dampening is negatively associated with quality of life (Edge et al., 2013) and positively associated with depressive symptoms in BD (Gilbert et al., 2013). Both decreased quality of life and increased depressive symptoms might be an indication for decreased recovery and dampening might thus also be a relevant determinant of personal recovery in BD. Positive rumination has been shown to be positively related with higher lifetime frequency of mania (Gruber et al., 2011) and lifetime diagnoses of mania or hypomania (Johnson et al., 2008). Interestingly, research shows that extreme appraisal of positive emotions is related with worsened illness course in BD (Ford et al., 2015), suggesting that positive rumination might be a maladaptive strategy in BD. Conversely, research also shows that increased positive emotions are associated with better functioning in BD (Johnson et al., 2016). Considering that positive rumination has been linked to increased positive emotions (Quoidbach et al., 2010), positive rumination, depending on the level of rumination, might thus also be an adaptive

strategy that has the potential to improve functioning. This makes positive rumination an interesting candidate to examine as potential determinant of personal recovery in BD.

Although personal recovery is an important outcome for people with severe mental disorders, little is known about factors contributing to personal recovery in people with BD. One recent study explored the role of negative beliefs about mood swings and self-referent appraisals of mood-related experiences as correlates of personal recovery in BD, but social role participation or positive emotion regulation were not included in their study (Dodd et al., 2017). However, previous research suggests that positive emotion regulation and social role participation might be important correlates of personal recovery. To our knowledge, the actual association of these processes with personal recovery in BD remains unknown. Also, it has not yet been investigated whether these processes are associated with personal recovery above and beyond symptomatology. Furthermore, little is known about the role of sociodemographic factors for personal recovery in BD, but there is evidence suggesting that for example active work status is beneficial for people with BD (Dodd et al., 2017).

Increasing our knowledge about factors associated with personal recovery in BD will inform clinical practice and help to enhance recovery-focused therapies, which may eventually lead to more effective treatments and improve prognoses for individuals with BD. The current study aims to widen the body of knowledge in this field by exploring an array of sociodemographic variables, as well as social participation, positive emotion regulation and psychopathology as potential correlates of personal recovery in BD. In the present study, personal recovery is operationalized as a generic and comprehensive construct, including all five dimensions from the evidence-based CHIME-framework (Leamy et al., 2011). Based on prior research, we hypothesized that social role participation will be positively associated with personal recovery. Further, we expected that positive rumination will be positively associated with personal recovery and that overall symptomatology and dampening show a negative relationship with personal recovery.

## METHOD

### Procedure

Data from two studies were combined for the current study. The first dataset ( $n = 119$ ) comes from an online cross-sectional survey study conducted between April and July 2018 (Kraiss et al., 2019b). The second dataset ( $n = 90$ ) was collected between September 2018 and March 2020 for the baseline assessment of a randomized controlled trial (RCT) on the effectiveness of a psychological intervention for patients with BD (Kraiss et al., 2018). The Ethics Committee



of the corresponding University approved the first study. The second study has been approved by a Medical Ethical Research Committee. Merging both datasets for the current study was not originally planned, but decided later in the process of the RCT study, since both studies include similar outcome measures and this way the statistical power could be increased.

For the survey study, adult participants were gathered through convenience sampling via the Dutch patient association for people with bipolar disorder. Diagnoses were based on self-report and not confirmed by a clinical interview. For the RCT, participants had to sign an informed consent before participation. The most relevant inclusion criteria for the RCT were: (1) diagnosis of BDI or BDII, (2) ages 18-65, and (3) currently not in an acute depressive or (hypo)manic episode. A detailed overview of the procedure and in- and exclusion criteria of the RCT can be found in the study protocol (Kraiss et al., 2018).

## Measures

Participants were asked to provide demographical features, including age, gender, relationship status and education. Participants were asked whether they took medication in the context of BD and whether they had been admitted to a psychiatric hospital because in the past months. The following constructs were assessed:

*Personal recovery* was assessed with the 15-item Questionnaire about the Process of Recovery (Neil et al., 2009). Respondents were asked how they felt in the past 7 days (e.g. "I feel better about myself") on 5-point Likert scale ranging from 0 (*strongly disagree*) to 4 (*strongly agree*), with higher scores being indicative of more personal recovery. The range of possible total QPR scores ranges from 0 to 60. Although a specific measure of personal recovery for BD has been developed, the Bipolar Recovery Questionnaire (BRQ; Jones et al., 2013), we chose the QPR in our studies because it represents a less burdensome and more feasible alternative to the 36-item BRQ and fits well with the evidence-based generic CHIME-framework (Leamy et al., 2011), since every item of the QPR maps to one of its dimensions (Shanks et al., 2013). The Dutch version of the QPR has been translated and validated in patients with BD (Kraiss et al., 2019a). Cronbach's alpha in the current (combined) sample was 0.93.

*Social role participation* was measured with the Short-Social Role Participation Questionnaire (S-SRPQ; Oude Voshaar et al., 2016) containing the two subscales satisfaction with role performance (six items) and experienced difficulties with role performance (six items). The S-SRPQ asks participants to specify their satisfaction and experienced difficulties in relation to six different social situations (e.g., work, relationships) on a 5-point Likert scale ranging from 1 (not satisfied at all / no difficulties at all) to 5 (very much satisfied / not possible). Higher scores

on the corresponding subscales are indicative of more satisfaction with role performance or more experienced difficulties with role performance, respectively. In the current study, we used mean scores for both subscales, as this is the usual way of scoring the S-SRPQ. Therefore, the scores can range from 1 to 5 in the current study. Cronbach's alpha in the current sample was .73 for the subscale satisfaction with social role performance and .77 for the subscale difficulties with social roles.

*Anxiety symptoms* were assessed with the anxiety subscale of the Hospital Anxiety and Depression Scale (HADS-A; Spinhoven et al., 1997; Zigmond & Snaith, 1983). The HADS-A measures the presence of mild forms of anxiety with 7 items. The presence of symptoms in the past week is rated on a scale from 0 (not at all) to 3 (very often). The scores of the HADS-A can range from 0 to 21. Higher scores are indicative of increased anxiety. Cronbach's alpha in the current study was .86 for the anxiety subscale.

*Manic symptoms* were assessed using 5-item Altman Self-Rating Mania Scale (ASRM; Altman et al., 1997), which consists of statements representing manic symptoms, such as inflated self-confidence or increased chattiness. Items are rated on a 5-point Likert scale containing different answering categories. The scores of the ASRM can range from 0 to 20. The total score is obtained by summing up all items and higher scores are indicative of more manic symptomatology. The ASRM revealed good rest-retest reliability (Altman et al., 1997). Cronbach's alpha in the current study was .78.

*Positive emotion regulation* was assessed with the 17-item Responses to Positive Affect questionnaire (RPA; Feldman et al., 2008). This questionnaire assesses the positive emotion regulation strategies dampening and positive rumination on three different subscales: (1) dampening (e.g. "I don't deserve this"), (2) self-focused positive rumination (e.g. "I am achieving everything") and (3) emotion-focused positive rumination (e.g. "Savor this moment"). Respondents were asked to rate how frequently they have these cognitions when experiencing positive emotions on a Likert-scale ranging from 1 (almost never) to 4 (almost always). Higher scores indicate more dampening and positive rumination, respectively. For the current study, one item of the dampening subscale ("This is too good to be true") was removed from the analyses. This is in line with two previous studies (Kraiss et al., 2019b; Nelis et al., 2016) that found that this item shows low factor loadings. In the current study, the scores for the dampening subscale can range from 7 to 28, for self-focused positive rumination from 4 to 16, and for emotion-focused positive rumination from 5 to 20. The Dutch RPA has shown satisfying psychometric properties in patients with BD (Kraiss et al., 2019b). Alpha in the current study was .79 for the emotion-focused and self-focused subscales and .83 for the dampening subscale.

## Statistical analyses

All statistical analyses were conducted in R (R Core Team, 2020; RStudio Team, 2020). Missing values (3.2%) were handled using random forest imputation using the MissForest package (Stekhoven & Bühlmann, 2012). This imputation method adequately deals with mixed-type data and is superior to other common imputation techniques in terms of imputation error and maintenance of predictive ability (Waljee et al., 2013). The number of missing values ranged from 1 to 18 for different variables. Categorical variables were dummy coded. The original dataset ( $N = 209$ ) was then split into a training ( $n = 149$ ) and a test set ( $n = 60$ ) using the caret package (Kuhn, 2020).

Three blocks of variables were considered for the multiple regression model. In the first block, the sociodemographic variables age, gender, education, work status and marital status were included. In the second model, the two variables satisfaction and difficulties with social roles were considered (S-SRPQ). In the third block, medication and admission to a psychiatric hospital, anxiety (HADS-A) and manic symptoms (ASRM), self-focused and emotion-focused positive rumination and dampening of positive emotions (RPA) were additionally considered for the multiple regression model. These potential predictors had to pass three preselection criteria to be included in the multiple regression model. First, categorical variables without sufficient variability ( $> 80\%$  in one response category) were excluded (Kuhn & Johnson, 2013). Second, simple univariate regression analyses were conducted for each predictor and the criterion personal recovery. Predictors with  $p > .10$  were excluded. Third, Pearson intercorrelations between potential predictors were examined to avoid multicollinearity. In case of  $r > .70$  or  $r < -.70$  between two predictors, the variable with the weaker correlation with the QPR was excluded. To examine whether combining the two datasets introduced bias, we created a dummy variable indicating to which dataset a participant belongs. Regression models were run to check whether this dummy interacts with any of the variables in explaining personal recovery.

All predictors that passed these criteria were included in the block-wise multiple regression analyses with personal recovery measured with the QPR as outcome variable. The first model contained sociodemographic variables only, while the second model contained social participation variables. In the third model, psychopathology and emotion regulation variables were entered. To check for significant outliers, Cook-distances were examined. Cook-distances greater than 1 were considered as noteworthy and would require further investigation (Cook & Weisberg, 1982). The variance inflation factor (VIF) was calculated to check for multicollinearity. Values that exceeded 5 were seen as problematic (James et al., 2013). To visually check linearity and homoscedasticity of residuals, normal Q-Q plots, residuals versus

fitted and residuals versus leverage plots were examined and histograms of residuals were plotted and inspected. To statistically test for normal distribution of residuals, Shapiro-Wilk test was conducted. A nonsignificant Shapiro-Wilk test is indicative for a normal distribution of the residuals. To compare the fit of the different models, the root-mean-square-error (RMSE) and mean absolute error (MAE) were calculated. Lower RMSE and MAE values are indicative of better model fit.

To create a parsimonious model that comprises fewest variables as possible without compromising predictive ability, we used the train function from the caret package with 10-fold-cross-validation. The caret package contains functions for regression and classification training. The train function tests different combinations of variables for each possible size of a model and finds the combination of predictors that minimizes the error (Kuhn, 2020). This parsimonious fourth model was then compared with the more complex third model using F-statistics to determine whether it performs comparably well in explaining variance in personal recovery. To further check whether combining the two datasets leads to confounded findings, we conducted a sensitivity analysis by using the final parsimonious model on both datasets (survey and RCT) separately and by examining whether the explained considerably differed in both datasets. Afterwards, the parsimonious model was cross-validated to test the accuracy of the model. For this, the unstandardized regression coefficients from the final parsimonious model were used to calculate predicted values for the QPR in the test sample. The predicted values from the test sample were then regressed on the observed values from the test sample to examine the accuracy of the predicted values. A  $\beta$ -coefficient  $> .70$  was considered a strong association and indicative for high model-accuracy. To further evaluate the final model, the  $R^2$  of the test sample was inspected and examined whether it falls within the 95% confidence interval of the  $R^2$  from the training sample. If the  $R^2$  fell within the 95% confidence interval, this was considered indicative of a comparable performance of the model in the test sample.

## RESULTS

### Sample characteristics

Table 1 summarizes the sample characteristics. Mean age of the 149 respondents in the training sample was 50.2 ( $SD = 11.4$ ) and about 70% were female. About half of the sample was highly educated (51%) and a relatively high proportion of people was unable to work, had no work or was retired (47.0%). 66 respondents (44.3%) had a diagnosis of BD I, while 70 had a diagnosis of BD II. Almost every respondent (94.6%) took medication because of their BD and almost no one was admitted to a psychiatric hospital in the past 3 months (94.6%).

**Table 1.** Sample characteristics (*N* = 209)

Variable	Category	Training sample	Test sample
		( <i>n</i> = 149)	( <i>n</i> = 60)
		<i>n</i> (%)	<i>n</i> (%)
Age		<i>M</i> = 50.2 ( <i>SD</i> = 11.36)	<i>M</i> = 49.5 ( <i>SD</i> = 10.47)
Gender	Male	45 (30.2)	11 (18.3)
	Female <sup>c</sup>	104 (69.8)	49 (81.7)
Education	Low	23 (15.4)	6 (10)
	Moderate	50 (33.6)	24 (40)
	High <sup>c</sup>	76 (51)	30 (50)
Work status	Working	79 (53)	33 (55)
	Not working	70 (47)	27 (45)
Marital status	Married or registered relationship <sup>c</sup>	67 (45)	26 (43.3)
	Never been married	42 (28.2)	13 (21.7)
	Divorced	31 (20.8)	14 (23.3)
	Other	9 (6)	7 (11.7)
Diagnosis	BDI <sup>c</sup>	66 (44.3)	30 (50)
	BDII	70 (47.0)	24 (40)
	Other/unknown	13 (8.7)	6 (10)
Currently taking medication <sup>b</sup>	Yes	141 (94.6)	56 (93.3)
	No	8 (5.4)	4 (6.7)
Admitted to psychiatric hospital in the past 3 months <sup>b</sup>	Yes	8 (5.4)	1 (1.7)
	No	141 (94.6)	59 (98.3)
		<b><u>M (SD)</u></b>	<b><u>M (SD)</u></b>
QPR		35.99 (10.55)	35.31 (10.60)
S-SRPQ difficulties		2.67 (0.83)	2.54 (0.78)
S-SRPQ satisfaction		2.37 (0.79)	2.57 (0.81)
HADS-A		8.42 (4.43)	8.54 (4.79)
ASRM		2.17 (2.83)	2.39 (2.79)
RPA emotion-focused PR		12.11 (3.05)	12.46 (3.04)
RPA self-focused PR		7.87 (2.84)	7.60 (2.65)
RPA dampening		15.1 (4.71)	15.29 (4.46)

*Note.* ASRM = Altman Self-Rating Mania Scale, HADS-A = Hospital Anxiety and Depression Scale-Anxiety Subscale, *M* = Mean, PR = Positive rumination, QPR = Questionnaire About the Process of Recovery, RPA = Responses to Positive Affect questionnaire, *SD* = Standard deviation, S-SRPQ = Short-Social Role Participation Questionnaire. <sup>a</sup>Includes participants that were self-employed, students, housewives or housemen or participants that were doing unpaid voluntary work. <sup>b</sup>Variables removed for further analyses because of lack of variance. <sup>c</sup>Reference category in regression analyses.

## Simple linear regression analyses

Table 2 summarizes the outcomes of the simple linear regression analysis. From the sociodemographic variables, age was negatively related with personal recovery ( $\beta = -.20, p < .05$ ). Currently working versus currently not working was positively related with personal

recovery ( $\beta = .31, p < .001$ ). Being divorced versus being in a relationship or being married was negatively associated with personal recovery ( $\beta = -.18, p < .05$ ). Social participation and psychological variables were all significantly related with personal recovery. Intercorrelations between predictors did not exceed .70 for any of the variables.

## Interaction effects

No significant interaction was found for any of the predictors and type of diagnosis or gender, suggesting that the relationship between predictors and personal recovery was independent of type of diagnosis and gender. No significant interaction effects were found for the dataset dummy variable, suggesting that the effect of the independent variables did not depend on the dataset. The following interaction terms were found for the interaction with the dataset variable: satisfaction with social roles ( $b = -1.28, p = .45$ ), difficulties with social roles ( $b = -1.96, p = .27$ ), anxiety symptoms ( $b = -0.13, p = .70$ ), manic symptoms ( $b = -0.80, p = .39$ ), emotion-focused positive rumination ( $b = -0.53, p = .31$ ), self-focused positive rumination ( $b = -0.18, p = .77$ ) and dampening ( $b = 0.55, p = .13$ ).

**Table 2.** Outcomes of simple univariate regression analyses between predictors and the criterion variable personal recovery

Block	Predictor	<i>b</i>	<i>SE</i>	$\beta$	F-value	<i>p</i>
1	Age	<b>-0.187</b>	<b>0.075</b>	<b>-.201</b>	<b>6.19</b>	<b>.014</b>
	Gender <sub>Female</sub>	1.478	1.885	.065	0.62	.434
	Education <sub>Low</sub>	-0.081	2.401	-.003	0.00	.973
	Education <sub>Moderate</sub>	-2.739	1.823	-.123	2.26	.135
	Work status <sub>Working</sub>	<b>6.439</b>	<b>1.655</b>	<b>.306</b>	<b>15.14</b>	<b>.000</b>
	Marital status <sub>Never married</sub>	<b>4.671</b>	<b>1.889</b>	<b>.200</b>	<b>6.12</b>	<b>.015</b>
	Marital status <sub>Divorced</sub>	<b>-4.568</b>	<b>2.103</b>	<b>-.176</b>	<b>4.72</b>	<b>.031</b>
	Marital status <sub>Other</sub>	0.602	3.640	.014	0.00	.869
2	Difficulties with social roles	<b>-6.762</b>	<b>0.892</b>	<b>-.530</b>	<b>57.51</b>	<b>.000</b>
	Satisfaction with social roles	<b>8.541</b>	<b>0.852</b>	<b>.637</b>	<b>100.6</b>	<b>.000</b>
3	Anxiety symptoms	<b>-1.202</b>	<b>0.169</b>	<b>-.505</b>	<b>50.4</b>	<b>.000</b>
	Manic symptoms	<b>0.907</b>	<b>0.298</b>	<b>.243</b>	<b>9.24</b>	<b>.003</b>
	Self-focused positive rumination	<b>1.165</b>	<b>0.292</b>	<b>.313</b>	<b>15.98</b>	<b>.000</b>
	Emotion-focused positive rumination	<b>1.369</b>	<b>0.262</b>	<b>.395</b>	<b>27.21</b>	<b>.000</b>
	Dampening of positive emotions	<b>-0.701</b>	<b>0.176</b>	<b>-.313</b>	<b>15.95</b>	<b>.000</b>

Note. Outcomes printed in bold were considered for multiple regression analyses.

**Table 3.** Model summary

Model	R <sup>2</sup>	95% CI	R <sup>2</sup> adjusted	RMSE	MAE	ΔR <sup>2</sup>	F change	Sig. F change
1	.131	.034 to .228	.107	0.654	0.537	.131	5.42	.000
2	.452	.340 to .564	.429	0.519	0.403	.321	41.61	.000
3	.605	.515 to .695	.573	0.441	0.352	.153	10.62	.000
4	.573	.474 to .672	.561	0.458	0.365	.032	1.59	.144

Note. Model 1 = Sociodemographic variables, Model 2 = Sociodemographic + Social variables, Model 3 = Sociodemographic + Social + Psychological variables, Model 4 = Parsimonious model after model training was applied.

## Multiple regression analyses

The model summary for the block-wise multiple regression analyses can be found in Table 3. Variables in the first model explained 13.1% variance in personal recovery, while the second model explained 32.1% additional variance. The third model explained 15.3% additional variance compared to the second model.

Findings show that in the first model, a significant independent predictor of personal recovery was if a participant was currently working. In the third model, satisfaction with social roles and manic symptoms were positively related with personal recovery. VIF scores ranged from 1.050 to 2.592. Cook-distances for the third model did not indicate that there were noteworthy outliers that would require further investigation. Shapiro-Wilk test indicated a normal distribution of the residuals ( $W = 0.99, p = 0.76$ ).

## Building a parsimonious model

In order to create a parsimonious model based on the third model, we used the train function from the caret package to determine the combination of variables that represent the best fit to the data with the fewest number of variables. This model contained the four variables satisfaction with social roles ( $\beta = .44, p < .001$ ), anxiety symptoms ( $\beta = -.30, p < .001$ ), manic symptoms ( $\beta = .22, p < .001$ ) and emotion-focused positive rumination ( $\beta = .22, p < .001$ ). The parsimonious fourth model explained 57.3% variance in personal recovery. The RMSE was 0.46 and the MAE was 0.37. Comparing the more complex third model with the parsimonious fourth model showed that the parsimonious model did not perform significantly worse in predicting personal recovery ( $\Delta R^2 = .03, Fchange(7, 137) = 1.59, p = .144$ ). Using the final parsimonious model on both datasets separately, showed a similar variance explained in both datasets (adj.  $R^2 = 0.54$  in the survey data and adj.  $R^2 = 0.57$  in the RCT data). This indicates that the final model performed comparably well in both datasets.

**Table 4.** Multiple regression analyses with personal recovery as criterion

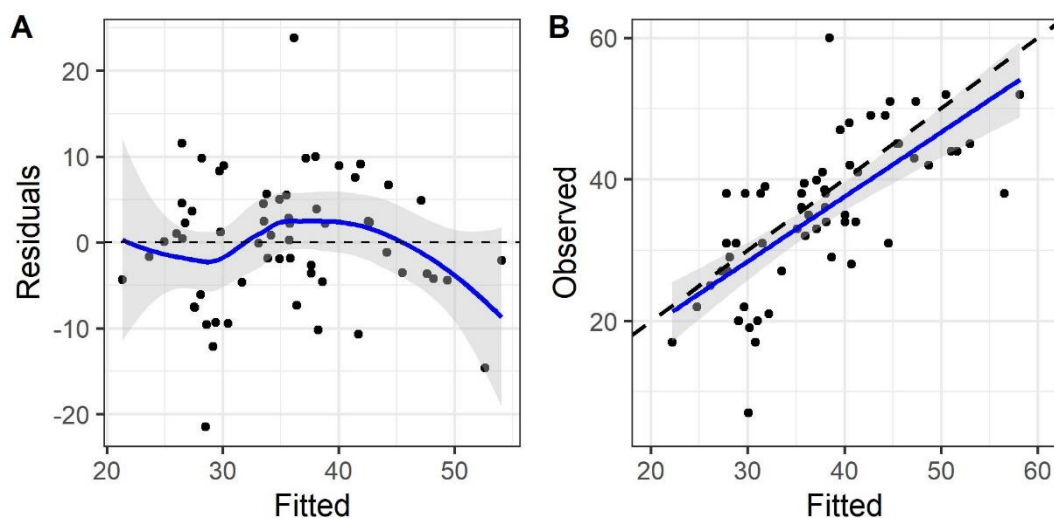
Model	Predictor	<i>b</i>	<i>SE</i>	$\beta$	<i>t</i>	<i>p</i>
1	Age	-0.088	0.078	-.095	-1.13	.261
	Work status <sub>Working</sub>	5.178	1.742	.246	2.97	.003
	Marital status <sub>NeverMarried</sub>	2.604	1.999	0.111	1.30	.195
	Marital status <sub>Divorced</sub>	-2.128	2.167	.111	-0.98	.328
2	Age	-0.030	0.063	-.032	-0.47	.640
	Work status <sub>Working</sub>	0.424	1.494	.020	0.28	.777
	Marital status <sub>NeverMarried</sub>	3.692	1.611	.158	2.29	.023
	Marital status <sub>Divorced</sub>	0.391	1.777	.015	0.22	.826
	Difficulties with social roles	-1.821	1.223	-.143	-1.62	.107
	Satisfaction with social roles	7.039	1.185	.525	5.94	.000
3	Age	0.013	0.056	.014	0.24	.811
	Work status <sub>Working</sub>	-0.085	1.338	-.004	-0.06	.949
	Marital status <sub>NeverMarried</sub>	2.401	1.449	.103	1.66	.100
	Marital status <sub>Divorced</sub>	0.797	1.550	.031	0.52	.608
	Difficulties with social roles	-1.039	1.057	-.081	-0.98	.327
	Satisfaction with social roles	5.278	1.058	.394	4.99	.000
	Anxiety symptoms	-0.597	0.178	-.251	-3.35	.001
	Manic symptoms	0.853	0.225	.229	3.79	.004
	Self-focused positive rumination	0.337	0.322	.091	1.05	.296
	Emotion-focused positive rumination	0.503	0.273	.145	1.85	.067
4	Dampening of positive emotions	-0.226	0.137	-.101	-1.65	.101
	Satisfaction with social roles	5.917	0.847	.442	6.99	.000
	Anxiety Symptoms	-0.704	0.148	-.296	-4.75	.000
	Manic symptoms	0.829	0.208	.222	3.98	.000
	Emotion-focused positive rumination	0.773	0.197	.223	3.92	.000

## Cross-validation

To test the model accuracy of the parsimonious model, we used the regression equation of this model to predict personal recovery in the test sample. We then regressed the predicted values on the observed values from the test sample. Visual inspection of Figure 1 suggests a symmetric distribution of error terms around zero (Plot A) and a relatively strong linear relationship between the predicted and observed values (Plot B). The correlation between predicted and observed values was high ( $b = 0.91$ ,  $\beta = .71$ ,  $t = 7.62$ ,  $p < .001$ ), suggesting high model accuracy. The fitted values explained 50% variance in the observed values ( $R^2 = .50$ , 95% CI = .34 to .66, adj.  $R^2 = .49$ ). The explained variance was somewhat lower than in the training sample, but the estimate fell within the 95% confidence interval of the  $R^2$  estimate in the training sample, indicating that the final model performed comparably well in the independent



cross-validation sample. The RMSE of the model in the test sample was 0.520 and the MAE was 0.397, which was only marginally higher than in the training sample.



**Figure 1.** Distribution of error terms (plot A) and relationship between predicted and observed personal recovery scores in the test sample (plot B)

## DISCUSSION

The aim of the current study was to explore whether several sociodemographic, social and psychological factors relate to personal recovery in BD. Four variables were identified as most relevantly associated with personal recovery, including satisfaction with social roles, anxiety symptoms, manic symptoms and emotion-focused positive rumination. We found that our final parsimonious model with these four variables explained a considerable amount of variance in personal recovery and did not perform worse than the more complex model. This was confirmed in an independent test sample using cross-validation.

### Main findings

The degree to which an individual was satisfied with the performance of social roles was the strongest correlate of personal recovery and was even stronger related with personal recovery than anxiety symptoms. Prior studies found that satisfaction with social roles is associated with an array of psychological and health-related outcomes in medical groups (Gignac et al., 2013) as well as with personal recovery and symptomatology in people with schizophrenia (Giusti et al., 2015). The current findings are relevant, as they further solidify the importance of social

role participation and suggest that it is an important focus when striving for personal recovery in BD. Social participation might therefore also be a relevant treatment target in this group, for example by specifically aiming at the ability to work or being able to have intimate relationships. This might increase feelings of relatedness and meaning in life, which are seen as two of the main components of personal recovery (Leamy et al., 2011). Interventions that have been shown to improve social role functioning in BD are collaborative care (van der Voort, van Meijel, Hoogendoorn, et al., 2015) and cognitive therapy (Lam et al., 2005). Although not yet evaluated in BD, other possible treatment alternatives to improve these outcomes might be vocational rehabilitation (Twamley et al., 2003) or supported employment (Modini et al., 2016).

One other relevant factor that was included in our final model were anxiety symptoms. This finding coincides with previous studies that similarly found significant negative associations between symptomatology and personal recovery in BD (Dodd et al., 2017; Jones et al., 2013). Although clinical recovery is generally considered as being distinct from personal recovery (Best et al., 2020; Macpherson et al., 2016), our findings suggest that anxiety symptoms were still independently associated with personal recovery. Therefore, residual anxiety symptoms might also be a relevant focus of treatment when aiming to improve personal recovery. This might help patients to lead a good and meaningful life by reducing anxiety as possible obstacles for personal recovery. In this context, it is interesting that dampening did not remain a significant correlate in the final model, although we assumed that it is a relevant mechanism for recovery in BD. This finding might be explained by the fact that the bivariate correlation between dampening and anxiety symptoms was significant. Therefore, it is likely that dampening could not independently explain variance in personal recovery beyond anxiety symptoms. This is also relevant for clinical practice, as it indicates that reducing anxiety symptoms might decrease dampening as they seem to overlap. However, the nature of the current data does not allow conclusions about causality, so this remains purely speculative.

Surprisingly, self-reported manic symptoms were positively related with personal recovery. Previous studies did not report a significant association between manic symptoms and recovery (Dodd et al., 2017; Jones et al., 2013). One possible explanation might be that up until a certain level the feelings and cognitions that are associated with (hypo)manic phases are actually positive. Thus, an individual might feel good, energized and may experience inflated self-confidence, which might also result in a relatively high score in personal recovery. For therapists and patients, our findings suggest that they may not need to be too concerned of positive moods per se and that improving these positive aspects is also relevant when striving for recovery. Actually, feelings similar to hypomanic experiences are reported by the majority of people in the general population (Jones et al., 2006; Udachina & Mansell, 2007), suggesting

that positive mood itself may not always be main problem in BD. This corresponds with Mansell (2016), who argues that positive moods should be accepted and cherished in BD, but that it is also important to distinguish between the destructive and constructive power of positive mood. A qualitative study by Russell and Moss (2013) shows that it is possible for people with BD to develop this insight, which may be an important step towards recovery. Nonetheless, it should also be mentioned that (hypo)manic episodes can have devastating effects. This might make it difficult for patients, therapists and their environment to impartially accept and cherish positive feelings and find the right balance between fostering and controlling positive feelings. It is also important to mention that the present study predominantly captured patients with subsyndromal manic symptomatology. This is indicated by the mean ASRM score in the current study, which was relatively low and comparable to previous studies that included euthymic patients with BD (van der Voort, van Meijel, Goossens, et al., 2015; Zyto et al., 2016). Furthermore, full-blown manic episodes are relatively rare and the course of BD is usually dominated by depressive episodes and euthymic states (Judd et al., 2003; Judd et al., 2002; Kupka et al., 2007). This makes it impossible to make inferences regarding the relationship between full-blown manic symptoms and personal recovery in the current study.

Emotion-focused positive rumination was also included as relevant predictor in the final model. Research regarding the role of positive rumination for personal recovery in BD remains absent and to our knowledge, this was the first study to explore the association between positive emotion regulation and personal recovery in BD. Our findings are in line with prior studies in community and college samples that similarly found that positive rumination is positively related with recovery-related outcomes, such as self-esteem (Feldman et al., 2008) and life satisfaction (Quoidbach et al., 2010) and negatively related with depressive symptoms (Nelis et al., 2016). Our findings further solidify the relevance of positive rumination and generalize the relevance of this process to the concept of personal recovery in BD. This finding is also relevant for clinical practice, since positive rumination might be trained by practicing with savoring strategies (Quoidbach et al., 2010). Some examples of savoring strategies that have been shown to be related with increased experience of positive emotions include capitalizing (i.e. communicating and celebrating positive events) (Langston, 1994) and mental time travel (i.e. vividly remembering positive events) (Suddendorf & Corballis, 2007).

Sociodemographic variables explained a relatively small amount of variance in personal recovery, suggesting that personal recovery is reachable independent from gender, age and education. Interestingly, whether someone was working had an independent positive impact on personal recovery in the first model. This indicates that employment status contributes to personal recovery, which corresponds to previous research that found that employment status was significantly associated with personal recovery in BD, even after controlling for other

clinical outcomes (Dodd et al., 2017). Working status was not a significant predictor anymore after including social role participation in the model. This was most likely because the social role participation questionnaire also includes items about whether the respondent is able to work and therefore actual employment status could not explain much additional variance beyond perceived social role participation. It is also important to mention that people were also classified as working in the current study if they had unpaid voluntary work. This implies that also having unpaid work might already be beneficial for recovery.

## Implications

Our findings show that social role participation as well as anxiety symptoms and positive rumination appear to be relevant predictors for personal recovery in BD. Focusing on these outcomes during treatment might help to improve recovery. Therapies such as Cognitive Behavioral Therapy or Acceptance and Commitment Therapy may be beneficial to cope with anxiety. Collaborative care (van der Voort, van Meijel, Hoogendoorn, et al., 2015), vocational therapy (Twamley et al., 2003) or supported employment (Modini et al., 2016) has been found to be effective for social functioning and functional recovery. Exercising savoring strategies and positive psychology interventions might represent promising ways to cultivate positive emotions and increase positive rumination. Also, a positive psychology intervention has recently been shown to be effective in improving recovery-related outcomes such as optimism in a randomized controlled pilot trial in people with BD (Celano et al., 2020). Combining these interventions might help to comprehensively increase recovery in BD. Nonetheless, it is also desirable that future research further investigates the effectiveness of novel forms of psychotherapy to improve recovery (Murray et al., 2017), including third-wave therapies or positive psychotherapy.

## Limitations

The current study has several limitations. First, the data was cross-sectional. Therefore, no conclusions about causality and direction of effect can be drawn. Second, about one half of the sample did not have a confirmed diagnosis of BD, as these participants were gathered via convenience sampling for an online survey. However, they were gathered through the Dutch patient association for people with BD and were asked to self-report their diagnosis. Therefore, it can be assumed that a great part of the sample actually had BD. Third, our study only included a specific selected set of predictors. Therefore, no complete overview of all possible factors that might contribute to personal recovery can be given. For example, we merely included anxiety symptoms as predictor, but not depressive symptoms. The reason for this is that depression was measured with different scales in the two datasets, making it impossible to compare depression scores of participants between the two datasets. Considering that

depression might also be present in remission (Vieta et al., 2008) and that there is a link between depression and recurrence (Pinto et al., 2020), depression may also be related to personal recovery we may thus miss an important factor contributing to personal recovery in the current study.

## **Conclusion**

The current findings suggest that psychopathology as well as social role participation and positive rumination are important independent predictors for personal recovery in BD. These insights widen the knowledge of what contributes to personal recovery in this specific group. The results also have implications for clinical practice, as including these factors into treatment might improve personal recovery in people with BD. We encourage future research to further explore predictors of personal recovery in BD and examine which interventions effectively enhance personal recovery. This might lead to more effective recovery-based treatments and help to enhance recovery beyond clinical and functional recovery.

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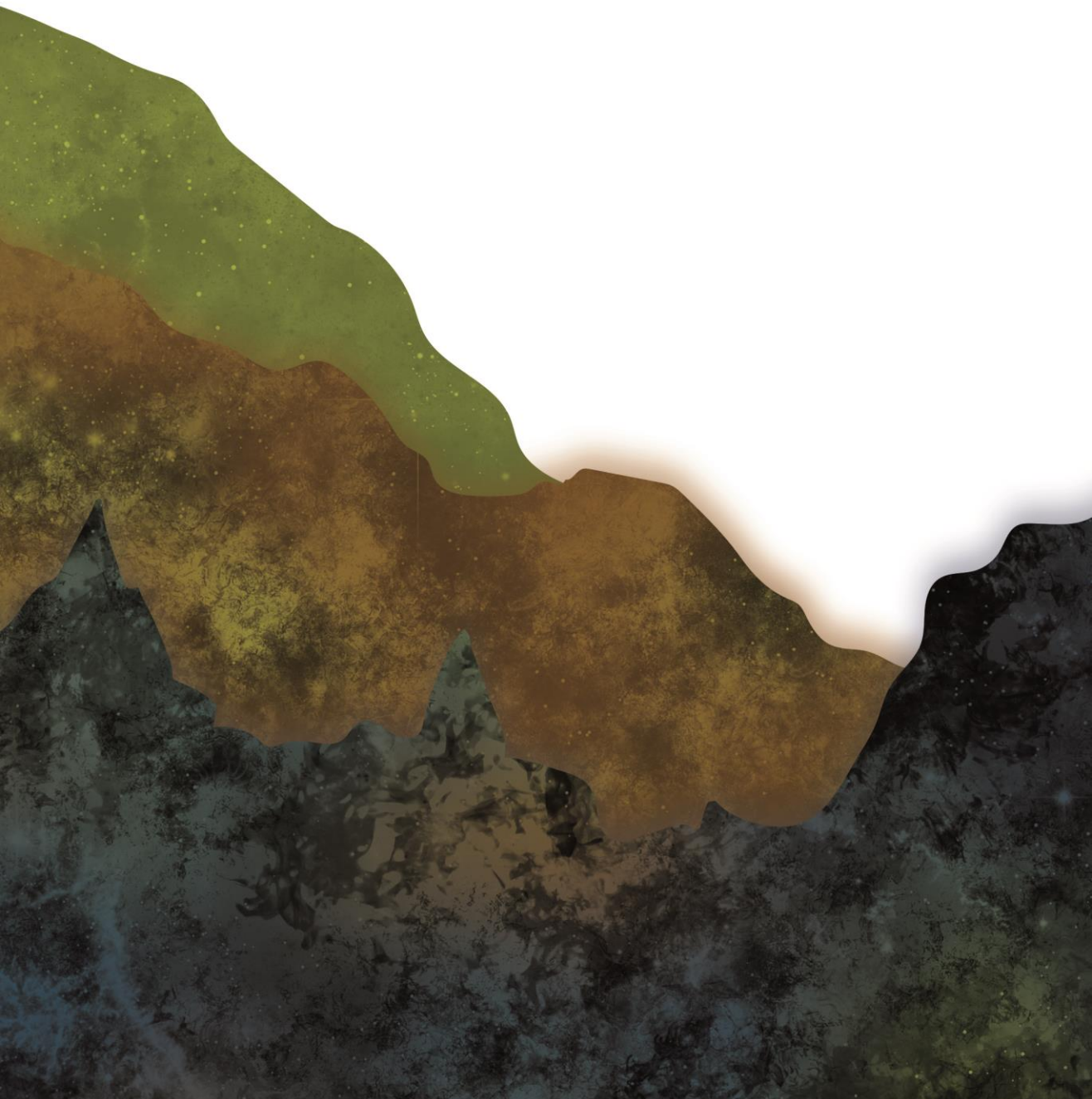
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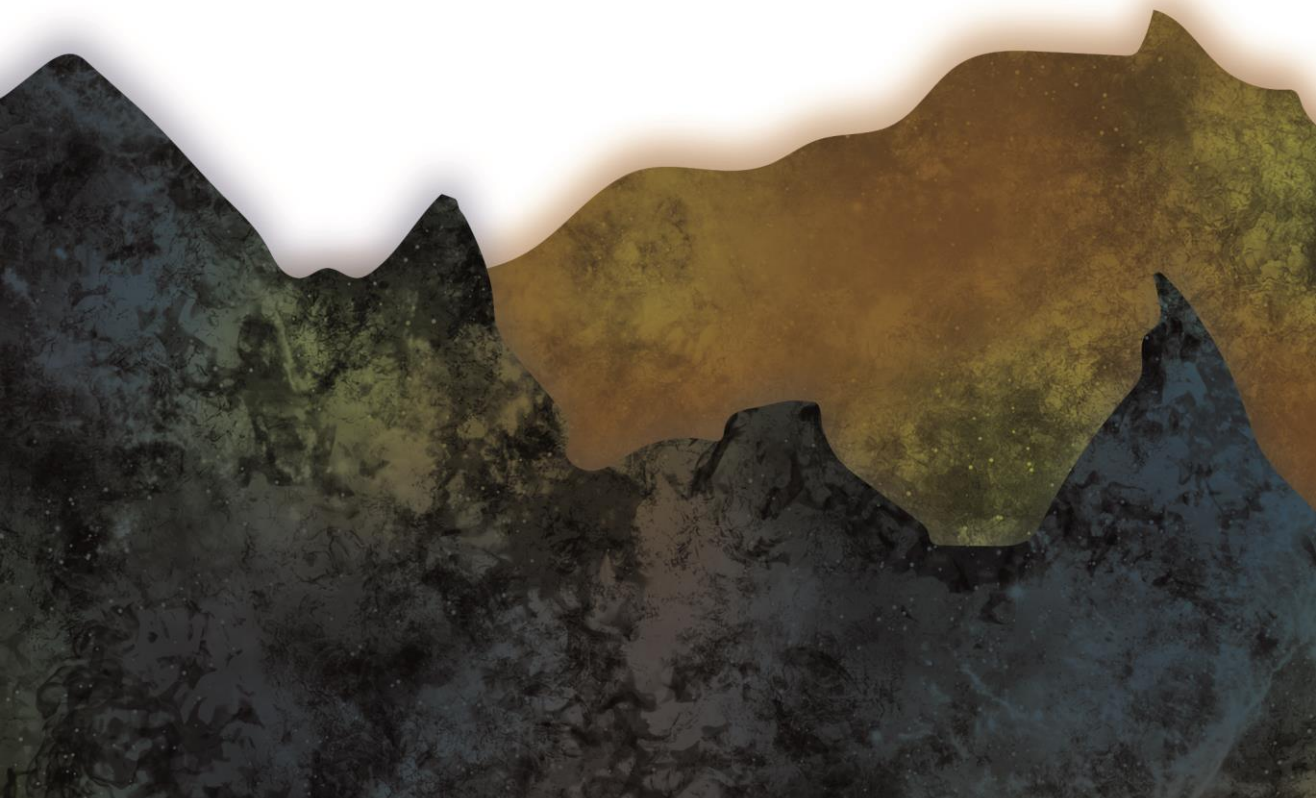
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# INTERMEZZO

Signposts in everyday life: Interview about the meaning of positive psychology for people with bipolar disorder







## SIGNPOSTS IN EVERYDAY LIFE

### *Interview about the meaning of positive psychology for people with bipolar disorder*

We met at the entrance of Mediamarkt at the edge of the center of a city. "That is next to a parking garage and nice and easy", she wrote in the email. She participated in the *Living Well With Bipolar Disorder* course. I understood from the course leaders that she was enthusiastic about the course and it seemed worthwhile to interview her about her experiences. I hear a cordial greeting and when I turn around, I see a woman at ten meters away walking towards me with a firm step. We walk into the city center in search for a cafe. While we are walking, she already starts to tell. "We started the group with eight people, but four dropped out after the first meeting. One noticed that he was still too depressed, the other found it difficult to talk in a group. The four that remained formed a very nice group. Quite luxurious, in a small group like this." The *Living Well With Bipolar Disorder* course focuses on promoting personal recovery and mental well-being. It is mainly based on positive psychology with extra attention for self-compassion. It is intended for people who are relatively stable and feel able to manage their symptoms.

Once we are equipped with a cappuccino and café latte, I turn on the recording device. In the beginning of the conversation, I speak about her illness, but I notice that it doesn't feel quite right. "What do you call it?" I ask. She doesn't know exactly. But illness or disorder, she also thinks, is not entirely correct with positive psychology. I mention that in the course we also speak of vulnerability. I tell her that for me positive psychology is about finding joy, meaning and involvement in your life, even in the presence of a psychological or physical vulnerability. This appeals to her and we talk about vulnerability from now on.

I first ask her to tell me something about herself and her life. She has worked as a nurse for 20 years. After her studies she mainly traveled and worked all over the world. "Afterwards I realized that this is where all my unrest was, I kept pushing my limits." She says that the turmoil and traveling were also related to her childhood. She experienced her parents as unloving. "My parents had an unhappy marriage, the village mentality and the Catholic faith were stifling to me. No joy." That's how she sums it up. She says there was no closeness and that she was never touched. It also took a long time for her to allow that. There was a strong bond with her brother though, who has also worked all over the world and struggles with the same vulnerability. At 40, she can no longer keep it up. She is burned out and experiences a deep loneliness. She gets into crises several times, has psychotic experiences and is admitted to a closed ward. "A very bad experience, and traumatic, but it was also necessary." She also got

EMDR and "afterwards bipolar disorder came out of it." The diagnosis was only made two years ago.

Candidly and soberly she talks about her experiences and complaints. She is doing well now ("relatively stable"), but she has to keep paying close attention. She compares her vulnerability to diabetes. You have to monitor continuously and sometimes from minute to minute you have to be able to respond quickly. She works with the color system *green, orange and red*. As soon as she notices that certain experiences are increasing and she turns orange, she actively takes measures. What she thinks is really "false" though is that she sometimes thinks that she can still assess it properly. And then this turns out not to be the case. Through falling and getting up she learned to trust her boyfriend. She has also signed that he and her brother may intervene if they see it as necessary. They may contact each other and also the psychiatrist if they think things are not going well. What she experiences as helpful is the structure in her life. In addition to working on the care farm, she regularly babysits a friend's children. She also has a cleaning job, does a lot in her own garden and is member of a singing association. "It is structure with meaning," she says. "What is crucial is that I am free to organize my own time. Also on the care farm. I lug around wood and earth. That physical work is good for me, but I have to be able to determine it myself."

At the beginning of 2019, her practitioner proposes to her to participate in the course *Living well with bipolar disorder*. First she hesitated. "Will that *positive psychology* take root? Isn't it all just tricks?" were her reactions. But she was also curious. "And then at the first meeting I had to introduce myself and tell what my goal was for the course. Then it suddenly popped out of nowhere that I wanted to be kinder. I didn't know if that was the intention, but that was exactly my wish. And I think I also achieved this goal," she says with a broad smile. The course turned out to be a great success. The course book offered her a clear and pleasant framework. "It contains a lot of information, I thought that was very nice and affirming. It is actually full of signposts that can help you in everyday life." As an example, she gives the exercise to observe your environment with extra attention and to appreciate what is good. Then she notices that she easily falls into negativity. "Then I cycled to work in the winter and I saw a woman walking a dog and I thought: how annoying in this weather. Or I saw a beautiful house in the nature. And then I said to myself: you should only live here in the winter." She can now break the pattern of negative thoughts quicker and quicker. And she continues: "The book explains that positive emotions contribute to flexibility and increase creativity. That's very important to me." When she now experiences something nice, she takes the time to appreciate it more. "Recently I saw a woman taking the time to help a vagrant and she offered him a cup of coffee. I loved that." She gives another example. "I read in the book that there are ten positive emotions. I started looking for that in my diary. Then I discovered that I was always missing a few: hope

and gratitude. I then started paying more attention to that. Very healing. It is also important that it is always mentioned that negative emotions may also exist and that it is not the goal to completely remove them. I then noticed that it is not a hocus-pocus story."

During the conversation I realized how serious she took the course. She has the workbook with her and it is full of answers and discoveries. Often she got up an hour earlier to work on it. "To really let it sink in and sharpen it," as she calls it. She is keen to systematically discuss the eight weeks of the course and share what each topic means to her. After one hour, as I try to get to a conclusion by asking what the course has meant to her, she corrects me. "First the last chapters, if you don't mind?" We order another round of coffee. Downstairs the buzzing of the coffee machine and music. On the first floor where we are sitting, a young woman is doing homework, it seems. "That self-compassion was also such a signpost," she continues. "We had to write down our favorite pessimistic thought. A lot of fun to do. I often have thoughts like "I don't matter anymore" or "I feel undervalued" and then we had to practice with thoughts like "I am valuable." That was quite a job, because sometimes I had the feeling that it was bottomless. But it kept getting better." She also found the questions to discover strengths very stimulating. "What is the best compliment you have received?" is such a question. And she talks gratefully about the boy she regularly babysits. "On Wednesday evening he tells his mother that he would like to go to bed early, because then he can go to me sooner to play. I thought that was so special." The babysitting is no effort for her. Playfulness and creativity are her strengths, she carefully says. She also laughs about the exercises that sometimes mislead you. "Then I first had to fill in who inspires me. And then I had to write down what I recognize about them in myself. I came to be authentic, enlightening, adventurous, courageous and creative. It was the first time that I said this out loud to myself. And that was a huge gift." The forgiveness exercise to write a letter to her mother was also a special moment. The course leader suggested that. She decided to structure the letter on the basis of four themes: 'I see you; I forgive you; I thank you; I love you'. The spiritual practice of Ho'oponopono was the source of inspiration for this. She has not sent the letter, but it has been "clean" since she wrote it. The anger has gone and she no longer feels the need to keep talking about her parents.

We conclude the conversation with the theme of victimization and self-determination. "I think that self-determination is important and you miss it if you continue to feel like a victim." The image of the broken vase that you can try to repair or that you can also reconstruct into a new mosaic really appeals to her. She experiences that she sometimes feels like a victim, but that she also has many options to be self-determined. "There is a beautiful poem that says you are both; I'll send you that." And the next day I receive an email with the poem.

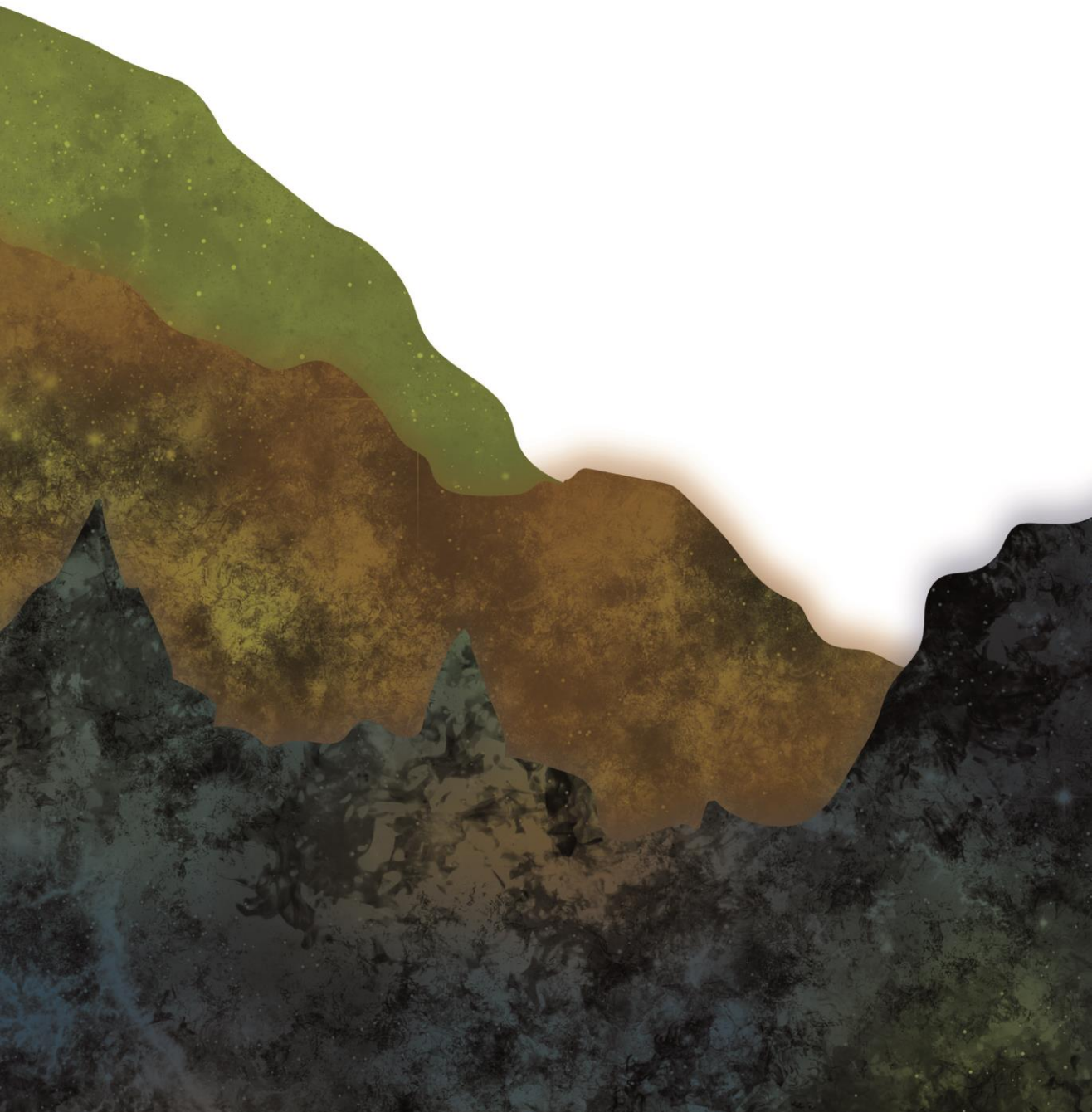
*You will know Life and be acknowledged by it according to your degree of transparency, your capacity, that is, to vanish as an end, and remain purely as a means.*

(From: *Markings*, Dag Hammarskjöld, 1963)

The poem is followed by a final 'signpost' that she forgot to mention during the conversation. It's about needs and motivation. As a need she first chose *recognition*. Until she realized that there is an extrinsic motivation behind it. "Others sometimes sell me short, I don't feel fully seen." But then she wonders what she really cares about. Her main need from intrinsic motivation is "wholeness". And that requires a completely different attitude. For recognition she wrote down "dead end", and for wholeness she wrote down "keep it up". We arrived at the final question of what the course as a whole meant to her. "In one word: positivity," she says. The broken vase had sharp edges, which I kept cutting myself on. I sanded it a lot and now the sharp edges of the disease are gone. It has become more of a vulnerability from a disability. That is a huge gain." It is quiet for a moment. She continues: "And that I can address important qualities more: sensitivity, compassion, more attunement. I never thought that of myself." In everything she has achieved I see her goal of becoming kinder and that she has made great strides. "And oh yes, I am also more balanced because of the new signposts, I have gone from two to one and a half pills a day. Hopefully some of my memory will come back ... and more creativity ... what a gain that is!"

And then again, I realize the enormous impact bipolar disorder has. Due to the condition and medication, functions that are so normal to others disappear: your memory, your spontaneity, your concentration. I didn't even notice that much during the conversation. And I realize that it shows that positive psychology offers a lot to find connection, meaning and joy again. I am impressed with her story. She has had a lot on her plate and living with bipolar disorder is very tough. But she is able to live life to the fullest within the limits of her illness. I learn something from that. I'm driving home. Grateful for her openness and inspiration. And for her generosity to share this story.

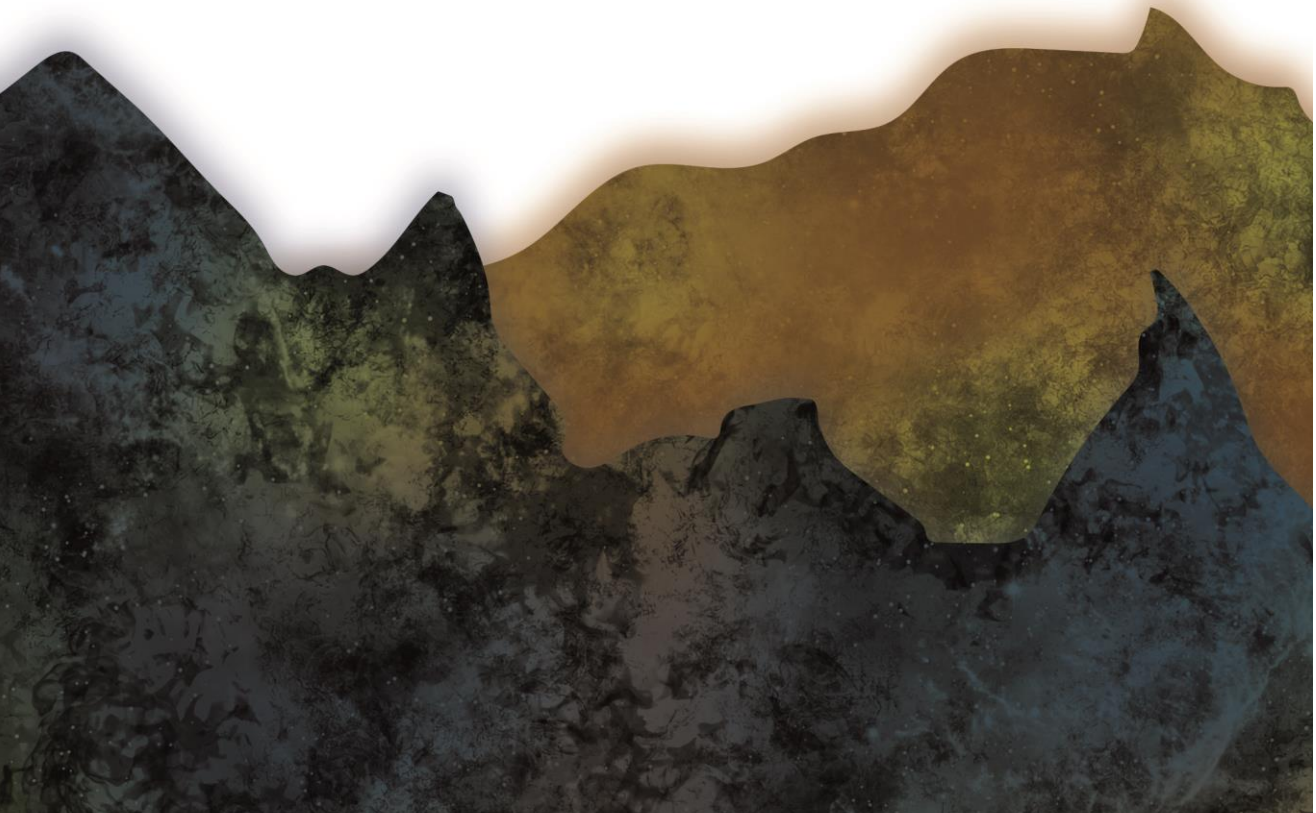




# CHAPTER 7

B-positive: a randomized controlled trial of a multicomponent positive psychology intervention for euthymic patients with bipolar disorder – study protocol and intervention development

Kraiss, J. T., Ten Klooster, P. M., Chrispijn, M., Trompetter, H. R., Stevens, A. W., Neutel, E., Kupka, R. W., & Bohlmeijer, E. T. (2018). B-positive: a randomized controlled trial of a multicomponent positive psychology intervention for euthymic patients with bipolar disorder: study protocol and intervention development. *BMC Psychiatry*, 18, 335. <https://doi.org/10.1186/s12888-018-1916-3>





## ABSTRACT

Bipolar disorder (BD) is characterized by recurrent (hypo)manic and depressive episodes, alternating with euthymic states in which patients are relatively symptom free. Besides clinical recovery, it is important to also strive for improvement of mental well-being and personal recovery. One prominent field focusing on the improvement of well-being is positive psychology. However, studies assessing the effects of positive psychology or personal recovery interventions for people with BD are scarce and have used weak methodological designs. The study described in this protocol article aims to assess the effectiveness of a multicomponent positive psychology intervention (“Living well with bipolar disorder”) adjusted for people with BD in the euthymic phase to improve well-being and personal recovery. The study concerns a pragmatic randomized multicenter trial. The principle objective of the study is to assess whether the positive psychology intervention offered to BD patients in remission in addition to usual care (CAU) is more effective than CAU. The study will include 112 participants randomized to either the experimental condition receiving the intervention in addition to CAU or the control condition receiving CAU. The study population are patients with BD I or II in the euthymic phase. The inclusion criteria are 1) diagnosis of BD I or BD II, 2) between the ages of 18-65, 3) four or more supportive sessions in the last year, and 4) only residual depressive or manic symptoms. Patients are excluded if they are in a depressive or manic episode, have current addiction problems or have optimal levels of well-being. Measurements take place at baseline, post-intervention and follow-up 6 and 12 months from baseline. Outcomes of measures include positive well-being, personal recovery, psychopathology, self-compassion, positive relationships, dampening of positive affect and relapse. The outlined study will be the first RCT examining the effects of a multicomponent positive psychology intervention for patients with bipolar disorder. Several limitations, including generalizability of the results and possible attrition issues, are discussed in advance. This study has been registered in the Netherlands Trial Register (NTR6729) on 12 October 2017.

## INTRODUCTION

Bipolar disorder (BD) is a severe mood disorder and is characterized by recurrent (hypo)manic and depressive episodes, alternating with euthymic phases in which patients are relatively symptom free (Grande et al., 2015; Kupka et al., 2008). BD is subclassified as bipolar I and bipolar II disorder. In the latter, patients solely experience hypomanic episodes but never a full manic episode (American Psychiatric Association, 2013; Berk & Dodd, 2005). Prevalence estimates from the Netherlands reveal a lifetime prevalence of 1.3% and 12-month prevalence of 0.8% for BD I and II (de Graaf et al., 2010). The economic burden in 2009 was estimated at 151 billion dollars per year in the United States (Dilsaver, 2011). BD is associated with decreased quality of life (Dean et al., 2004), negative social consequences, such as a disturbed social life and disrupted family interactions (Calabrese et al., 2003), issues related to work-performance and productivity (Fajutrao et al., 2009; Laxman et al., 2008) and high caregiver burden (Miller et al., 2014; Reinares et al., 2006). Current treatment for euthymic BD patients in the Netherlands includes pharmacotherapy, supportive treatment, psycho education and enhancement of self-management skills, and psychotherapy if indicated (Kupka et al., 2015; Van der Voort et al., 2011).

In addition to current symptom-focused treatment, it becomes increasingly important to also focus on personal recovery (Fava et al., 2007; Jones et al., 2013; Slade, 2010). Leamy et al. (2011) created a conceptual framework for personal recovery in mental health, containing five processes of personal recovery: connectedness, hope and optimism about the future, identity, meaning in life and empowerment (giving the acronym CHIME) as important factors for personal recovery (Leamy et al., 2011). Anthony (1993) operationalizes personal recovery in the context of psychopathology and describes it as the ability to live a meaningful, hopeful and contributing life, even in the presence of mental illness (Anthony, 1993). Similarly, Keyes (2002) defines mental health recovery as the presence of well-being and not merely the absence of mental illness. Well-being, in turn, includes subjective well-being (i.e. positive affect and life-satisfaction), psychological well-being (i.e. meaning, goals in life, mastery, positive relationships) and social well-being (i.e. contributing to society) (Keyes, 2002).

Personal recovery and well-being are particularly important in BD for several reasons. Residual subthreshold symptoms often persist in the interval between mood episodes (Fagiolini et al., 2005; Judd et al., 2008; Kaya et al., 2007). Moreover, up to 35% of BD patients do not completely recover from a depressive or manic episode (Fagiolini et al., 2013), which is an important risk factor for relapse (Fava et al., 2007). Research also indicates that improvement of well-being protects against the recurrence of mental illness (Keyes et al., 2010; Lamers et al., 2015; Trompetter et al., 2017). Patients with serious mental illness, such as BD also express

dissatisfaction with current primary targets of treatment and instead argue for the importance of personal recovery outcomes (Jones et al., 2010; Mead & Copeland, 2000).

One prominent field of psychology focusing on the improvement of well-being and positive capacities is positive psychology (Seligman & Csikszentmihalyi, 2014). Positive psychology interventions focus on the enhancement of positive feelings, behaviors, or cognitions and aim to improve well-being (Sin & Lyubomirsky, 2009). The key processes and goals of positive psychology are similar and central to personal recovery (Resnick & Rosenheck, 2006; Slade, 2010) making positive psychology interventions potentially useful for improving both well-being and personal recovery in people with mental illness. The effect of positive psychology interventions has been shown in meta-analyses for both general and clinical populations (Bolier et al., 2013; Sin & Lyubomirsky, 2009).

To date, however, no study has been conducted assessing the effect of positive psychology interventions for the treatment of people with BD and only a few studies investigated the effect of interventions focusing on the improvement of personal recovery. Deckersbach et al. (2012) report on a small uncontrolled clinical trial with 12 euthymic participants diagnosed with BD using Mindfulness-Based Cognitive Therapy (CBT). Analysis from pre- to follow-up indicated significant moderate to large improvements in outcomes of depressive symptoms (Cohen's  $d = .75$ ), positive affect and aspects of well-being. Eisner et al. (2017) conducted a proof-of-concept pilot study with 37 participants with BD who did not have a current major depressive, manic or mixed episode. Significant large improvements were obtained from baseline to post-treatment in psychological well-being, emotion regulation, and emotion reactivity. Finally, Jones et al. (2015) investigated the effectiveness of recovery-focused CBT in a randomized controlled pilot trial ( $n = 67$ ) with care as usual as control. Personal recovery significantly improved from baseline to 6 and 12 months follow-up ( $d = .62$ ) in comparison with a control group receiving treatment as usual. Although no significant effects were obtained in average mood symptoms, patients in the recovery-focused CBT group showed significantly longer time to relapse into depression or mania over a 15-month period compared to patients only receiving care as usual. For example, 32 patients relapsed to either depression or mania (20 CAU v. 12 recovery-focused CBT) and median survival times were longer for recovery-focused CBT (56 weeks) compared to CAU (18 weeks).

In summary, no studies exist investigating the effectiveness of positive psychology interventions for BD patients. Furthermore, studies examining the effect of interventions aimed to enhance personal recovery or well-being for patients with BD are scarce and mostly used underpowered and weak methodological designs. For this reason, we aim to develop, implement and thoroughly investigate the effects of a multicomponent positive psychology

intervention for the improvement of well-being and personal recovery for people with BD in an adequately powered randomized controlled trial. This will be the first study specifically evaluating a positive psychology intervention for patients with bipolar disorder and will use a more sophisticated methodological design than studies before.

The primary objective is to evaluate the effectiveness of an eight-week multicomponent intervention “Living well with bipolar disorder” added to usual care (CAU) in BD patients in the euthymic phase. Primary outcome is the short and long-term improvement of well-being and personal recovery. Second, the study aims to investigate whether the intervention in addition to CAU is more effective in improving social participation, and in improving depressive, manic, and anxiety symptoms. Third, we will explore possible working mechanisms for intervention effects, including positive emotions, self-compassion, positive relationships and dampening of positive affect. Fourth, the study aims to assess whether the intervention combined with CAU is more effective than CAU in reducing recurrence of depressive and (hypo)manic episodes in patients with BD in the long term. Finally, we aim to evaluate the cost-effectiveness of the intervention in addition to CAU for the treatment of euthymic patients with BD compared to CAU.

## METHOD

### Study design

A pragmatic, parallel-group randomized non-blinded multicenter trial is used to investigate the effectiveness of a multicomponent positive psychology intervention to improve well-being and personal recovery in patients with BD. Since the outcome in BD I and BD II disorder may be different, the sample will be stratified accordingly. Patients in the experimental condition receive “Living well with bipolar disorder” in addition to CAU. Participants in the control condition receive CAU only. Both participants in the control and experimental condition receive CAU according to the Dutch multidisciplinary guideline for BD (Kupka et al., 2015), consisting mainly of self-monitoring of mood and supportive group sessions focusing on functional problems, and psychopharmacotherapy. The study duration is 12 months for each individual and includes five measurement points. Immediately prior to the start of the intervention a baseline measurement is completed (T0), four weeks after the start of the intervention a mid-treatment measurement takes place (T1) and immediately following the intervention a post measurement will be conducted (T2). In addition, two follow-up measures will be conducted, six months (T3) and twelve months after baseline (T4). Figure 1 shows the intended flow of participants.

## **Participants, eligibility and screening**

In order to be eligible to participate in this study, participants must meet the following inclusion criteria: (1) diagnosis of BD I or BD II (assessed using the MINI-international neuropsychiatric interview (Hergueta et al., 1998)); (2) between the ages of 18-65; (3) four or more supportive sessions in the previous year with a psychiatrist or psychologist; and (4) presence of residual subsyndromal symptoms. Participants are included if they score between 2 (*minimal symptoms*) and 4 (*moderate symptoms*) for depressive symptoms and between 2 (*minimal symptoms*) and 3 (*mild symptoms*) for manic symptoms on the Clinical Global Impression Scale – Bipolar (CGI-BP). Exclusion criteria are: (1) currently in a depressive or (hypo)manic episode; (2) currently in treatment for addiction problems, or (3) having already optimal levels of well-being. Participants are assumed to have an optimal level of well-being if they score 4 or 5 on at least one item of the emotional well-being subscale together with a score of 4 or 5 on at least 6 of the 11 remaining items of the Mental Health Continuum-Short Form (MHC-SF; Keyes, 2002).

## **Randomization and treatment allocation**

Randomization will be centrally conducted by the principal investigator using stratified (by center) block randomization. For this purpose, randomization lists will be generated beforehand (one list for each participating treatment center) with an online tool (<https://sealedenvelope.com/>). The lists contain a random sequence of treatment allocations (i.e., participants are either allocated to the intervention or control condition according to the corresponding record) and are divided in blocks of allocations (20 allocations per block). By using blocks of allocations, 20 participants can be allocated to either the intervention or control condition after which the following block is used. This ensures that the group sessions in each center can start as soon as sufficient participants are randomized. The first participant included in the study is allocated according to the first record on the list, the second participant according to the second record on the list and so forth.

## **Recruitment**

Participants will be recruited from at least four mental health centers with six locations in the east and west of the Netherlands and will start in September 2018. Possible participants are informed by means of flyers and posters distributed in these centers. In order to include sufficient participants, mental health professionals will play an active role, by informing potentially eligible patients about the study and handing out information folders.

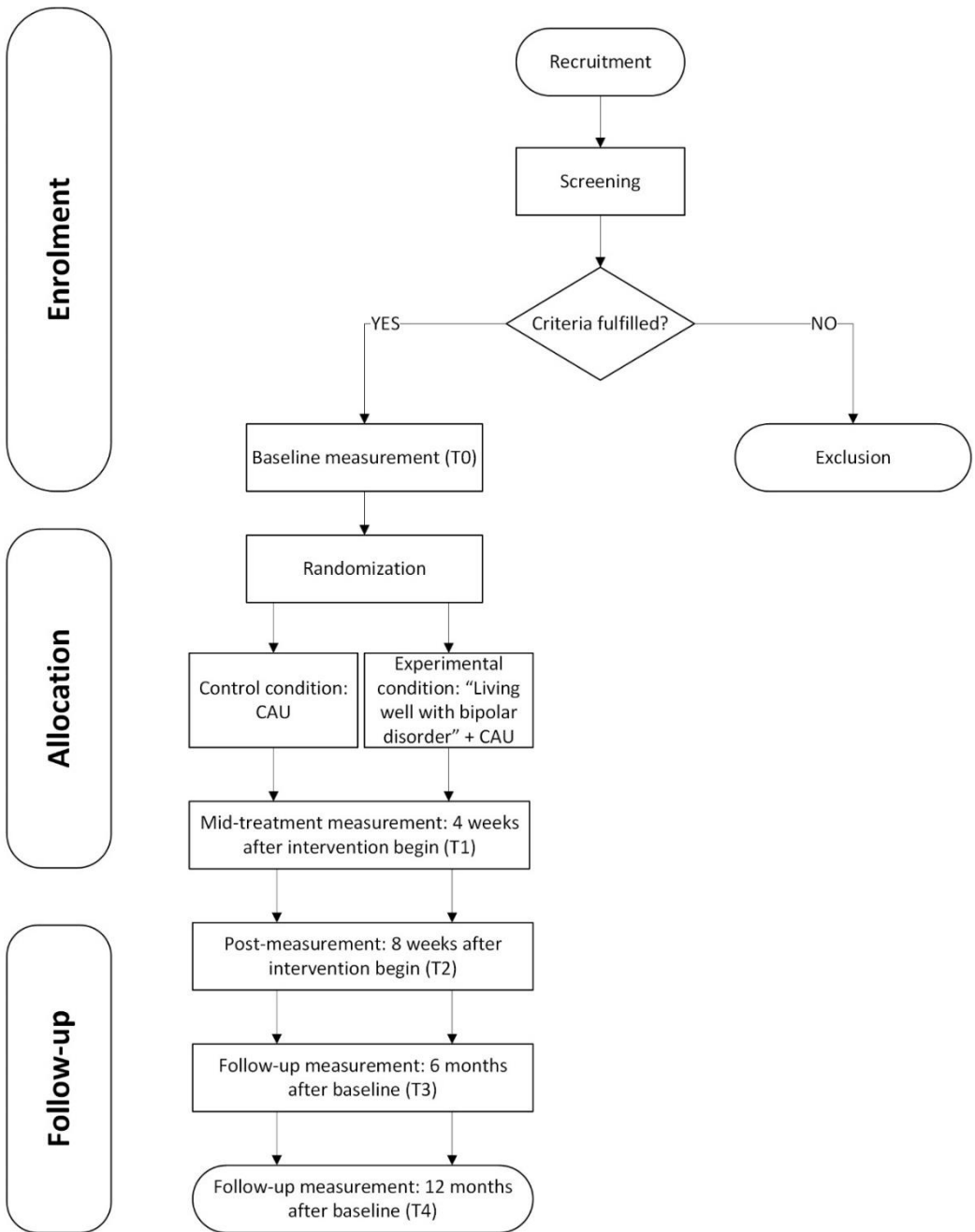


Figure 1. Participant timeline

## Intervention

We will adapt the positive psychology intervention “This is your life” (Schotanus-Dijkstra et al., 2015; Schotanus-Dijkstra et al., 2017) for patients with BD in the euthymic phase aiming to enhance personal recovery and well-being.

Originally developed as a self-help book, the intervention is primarily based on the well-being theory of Seligman (Seligman, 2011) and Ryff’s theory of psychological well-being (Ryff, 1989) and comprises different modules focusing on six key components: positive emotions; discovering and using strengths; optimism and hope; self-compassion; resilience and post-traumatic growth and positive relations. Each module contains psychoeducation and a range of different positive psychology exercises, such as the “three good things exercise” (Bolier et al., 2013) or the “best possible self” (Meevissen et al., 2011). All modules include proven strategies for improving well-being. In a recent randomized controlled trial, the intervention as guided self-help with email support revealed moderate to large effects on well-being ( $d = 0.66$ ) and effectively reduced subclinical symptoms of anxiety ( $d = 0.63$ ) and depression ( $d = 0.43$ ) in a non-clinical sample (Schotanus-Dijkstra et al., 2017).

## Intervention adjustments for euthymic BD patients

Since “This is your life” was not specifically developed for individuals with BD, we decided to customize the intervention content and delivery to the needs of this target group. The adjusted intervention is called “Living well with bipolar disorder”. We changed the mode of treatment delivery from individual self-help to group meetings (8-10 people per group), including eight sessions of two hours conducted by a specially trained therapist. This setup is believed to give participants the possibility to share experiences with fellow patients and benefit from the presence of other group members.

Each week, a key positive psychology component will be covered and corresponding exercises will be introduced. Participants are also encouraged to keep training with those exercises at home (for 15-30 minutes per day). All intervention participants receive the self-help book “This is your life” (Bohlmeijer & Hulsbergen, 2013), since several parts of the modules refer to chapters or exercises in the book. The adapted intervention consists of eight different modules covering different topics, such as positive emotions, positive relationships or personal goals. Homework differs depending on the module, but every week participants are encouraged to keep training with the exercises at home. Experiences with the homework is discussed at the beginning of each next session and possible obstacles or other benefits can be shared with the therapist and other group members. In case the module contains a collective exercise, a short

debriefing session will take place immediately after the exercise, to discuss experienced benefits or difficulties. Every module contains a short break of 15 minutes. Every session is finished by a short conclusion in which the therapist summarizes the session and where uncertainties can be broached and questions can be asked.

In spring 2017, a formative user evaluation of “This is your life” was carried out to evaluate the original intervention contents. Five individuals with BD read and practiced a selection of the original exercises and rated them on a scale from 1 (*not at all*) to 5 (*extremely*), representing the relevance and usability of the exercises. Results of this group were used to further adapt and tailor the intervention to the needs of individuals with BD by taking into account the ratings of exercises, preferences of the patients and also their critical responses.

For several reasons, we decided to put additional emphasis on fostering self-compassion skills by including two sessions of self-compassion. Research on emotion regulation (Gruber, 2011; Gruber et al., 2012; Phillips & Vieta, 2007) emphasizes the importance of disturbed positive emotion regulation (i.e., how an individual reacts to positive emotions) for the onset, maintenance and illness course of BD (Carl et al., 2013). Suppression of positive emotions is heightened among BD patients and predicts depressive and also manic symptoms (Edge et al., 2013; Gilbert et al., 2013). Additionally, negative thinking about high moods (e.g. “I will lose control if I get excited”) is also elevated in individuals with BD, compared with nonclinical controls (Alatiq et al., 2010; Mansell et al., 2011; Mansell et al., 2010), individuals with remitted unipolar depression (Alatiq et al., 2010; Mansell et al., 2011) and individuals who have had hypomanic experiences, but no psychological disorder (Mansell et al., 2011) and predicts mood symptoms in BD over the period of one month (Dodd et al., 2011) and six months (Gilbert et al., 2013). Studies show that self-compassion is positively related with adaptive emotion regulation processes, including acceptance and positive reappraisal and negatively correlated with maladaptive strategies, such as thought suppression and rumination (Allen & Leary, 2010; Crosskey & Curry, 2011; Leary et al., 2007). An overview of the intervention contents can be found in Table 1.



**Table 1.** Content of “Living well with bipolar disorder” and corresponding example exercises adapted for euthymic BD patients

Module	Contents	Example (home) exercises
1. Introduction & compassion	<ul style="list-style-type: none"> <li>▪ Participants are welcomed and become familiar with each other</li> <li>▪ Psychoeducation about personal recovery and compassion</li> <li>▪ Collective compassion exercise</li> <li>▪ Homework: Fill in handout about personal goals and optimism</li> </ul>	<ul style="list-style-type: none"> <li>▪ Wish yourself something good: be mindful and identify needs and use your inner voice to repeat your compassionate wish (Gilbert, 2010).</li> <li>▪ Common humanity: Realize that negative feelings and experiences are universal (Neff, 2003a; Neff &amp; Germer, 2013).</li> </ul>
2. Personal goals & optimism	<ul style="list-style-type: none"> <li>▪ Based on the handouts, participants talk about personal goals and wishes and specify personal goals in the group</li> <li>▪ Individually adjusting personal goals</li> <li>▪ Collective optimism exercise</li> <li>▪ Reading and discussing letter from an experienced person with BD</li> <li>▪ Homework: Working on personal goals, writing a letter from the future</li> </ul>	<ul style="list-style-type: none"> <li>▪ Imagine your best possible self: Visualize yourself in a future where everything has turned out in the most optimal way (Meevissen et al., 2011; Peters et al., 2010).</li> <li>▪ Letter from the future: write yourself a letter from a future perspective</li> </ul>
3. Positive emotions	<ul style="list-style-type: none"> <li>▪ Read out letter from the future</li> <li>▪ Psychoeducation about positive emotions</li> <li>▪ Collective positive emotions exercises</li> <li>▪ Homework: Taking a photo of a positive moment or experience, working on personal goals</li> </ul>	<ul style="list-style-type: none"> <li>▪ Three good things: Think about three good things that went well today and savor these moments (Fredrickson, 2013).</li> <li>▪ Expressing gratitude: identify what you are grateful for in the context of your illness and share those experiences (Lyubomirsky &amp; Layous, 2013).</li> </ul>
4. Coping with fear of relapse	<ul style="list-style-type: none"> <li>▪ Sharing photos of positive experiences and talking about the photos</li> <li>▪ Talking about participant’s fears, how fear is experienced and internal barriers</li> <li>▪ Psychoeducation about fear and (un)healthy emotion regulation strategies</li> <li>▪ Collective exercises on how to efficiently cope with fear</li> <li>▪ Homework: Do something you find exciting, perform personal strengths exercise, completing mid-treatment measurement</li> </ul>	<ul style="list-style-type: none"> <li>▪ Learn to tolerate and accept fear as important part of life and learn to regulate positive mood and gain a more open view towards them (Bohimeijer &amp; Hulsbergen, 2013).</li> <li>▪ Compassionate coping with inner fear: learn to be compassionate towards yourself, your emotions and your negative experiences (Gilbert, 2010).</li> </ul>

**Table 1.** Content of “Living well with bipolar disorder” and corresponding example exercises adapted for euthymic BD patients (continued)

Module	Contents	Example (home) exercises
5. Personal strengths	<ul style="list-style-type: none"> <li>▪ Identifying strengths</li> <li>▪ Psychoeducation about personal strengths</li> <li>▪ Personal goals and strengths: which strengths can be used to achieve personal goals?</li> <li>▪ Homework: Keep training with exercises, record possible benefits and barriers while performing the exercises</li> </ul>	<ul style="list-style-type: none"> <li>▪ Identifying strengths: Describe an activity you enjoy to someone else and he/she names strengths deriving from this activity.</li> <li>▪ Top 5 strengths: Choose your top 5 strengths that give you energy and pleasure (Linley et al., 2010; Wood et al., 2011).</li> </ul>
6. Positive relationships	<ul style="list-style-type: none"> <li>▪ Participants name skills they gathered in the course of the intervention so far and described one example from the last week</li> <li>▪ Psychoeducation about positive relationships</li> <li>▪ Participants describe a relationship they want to reinforce positively</li> <li>▪ Collective positive relationship exercises</li> <li>▪ Homework: Keep training with positive relationship exercises</li> </ul>	<ul style="list-style-type: none"> <li>▪ Acts of kindness: Performing unexpected acts of kindness for someone else (Otake et al., 2006).</li> <li>▪ Active-constructive responding: Respond positively to good news shared by someone else. Use active communication skills (Reis &amp; Gable, 2003; Reis et al., 2010).</li> <li>▪ Expressing gratitude (Lyubomirsky &amp; Layous, 2013).</li> </ul>
7. Compassion	<ul style="list-style-type: none"> <li>▪ Psychoeducation about emotional systems and evolutionary background</li> <li>▪ Collective exercises mindfulness and compassion</li> <li>▪ Coping with thoughts of inferiority and self-critique</li> <li>▪ Homework: Writing a response to the letter from the second session, fill in questionnaires for post-measurement</li> </ul>	<ul style="list-style-type: none"> <li>▪ Develop a compassionate inner voice: Write about situation in the past week where you showed self-compassion (Gilbert, 2010).</li> <li>▪ Grandma exercise: Imagine a person you feel comfortable with. Concentrate on how it feels to be together with this person and savor this moment (Bohlmeijer &amp; Hulsbergen, 2013).</li> </ul>
8. Conclusion	<ul style="list-style-type: none"> <li>▪ Talking about results of the questionnaires and figuring out which aspects are going well and which should receive some extra attention in the next weeks</li> <li>▪ Read out response to the letter from the second sessions</li> <li>▪ Participants thank each other</li> </ul>	<ul style="list-style-type: none"> <li>▪ Not applicable.</li> </ul>

## **Control group**

Participants in the comparison group will receive CAU for BD as described in the Dutch multi-disciplinary guideline for bipolar disorder (Kupka et al., 2015) which comprises supportive sessions with a psychologist or psychiatric nurse and maintenance pharmacological treatment by a psychiatrist. Most patients receive 2-12 supportive sessions per year. CAU includes many psychoeducational elements that have the following aims: to give patients information about the illness in the context of the patients' life-history, to learn to identify early warning signals and prodromal symptoms, to develop and implement strategies to cope with prodromal symptoms, and to develop plans for acute crisis and stabilizing one's mood. For some patients, CAU may additionally include psychotherapeutic treatments such as cognitive behavioral therapy and interpersonal therapy. For all patients, current CAU does not primarily focus on personal recovery in terms of emotional and psychological well-being (i.e. meaning, purpose in life, positive relationships)

## **Therapists training and treatment manual**

Therapists carrying out the intervention will receive a one-day workshop covering the central ideas of "Living well with bipolar disorder", including background, goals and possible exercises of the intervention. A treatment manual will be prepared to guarantee a standardized execution of the intervention. The manual includes explanations on each module and also on corresponding exercises. Short session handouts (1-2 pages) will be developed for patients participating in the intervention, explaining and summarizing the topics of each module and will include affiliated exercises and can be used to further exercise at home.

## **Study procedure**

Initial screening for participation will be performed by therapists working at the treatment centers. To assess symptom severity of possible participants, the Clinical Global Impression Scale – Bipolar (CGI-BP) is used. After possible participants sign the informed consent, the principal investigator contacts the patient and agrees on a time and date to conduct the additional screening.

To verify the diagnosis of BD, the MINI international neuropsychiatric interview (Hergueta et al., 1998; Hergueta & Weiller, 2013) is used and to assess well-being, participants are asked to complete the Mental Health Continuum-Short Form (MHC-SF) (Lamers et al., 2011). Eligible participants are then asked to complete the first test battery at baseline. Since the MHC-SF has already been completed for screening reasons before, participants are not asked to complete it again at baseline. Baseline measurements should take approximately 40 minutes to

complete. Afterwards, allocation of participants to the intervention or control group takes place.

The first intervention group will start approximately in fall 2018 and the last group will finish in fall 2019. Four weeks after the intervention begins (T1) and after the intervention has finished (T2), participants in the intervention and control condition are asked to fill out a test battery again. Six (T3) and 12 (T4) months after baseline, participants in the experimental and control group will be asked to complete the follow-up measures. We assume that completing the test battery takes approximately 35 minutes on average at each measurement point. At T4 participants will be approached for a semi-structured telephone interview with the goal to retrospectively assess relapse into mood episodes in the past nine months. To perform the interviews, a guideline will be prepared and the interviews will be conducted by student assistants according to a fixed scheme. The trained student assistants are blind to the treatment condition of the participants. One interview will take approximately 30 minutes.

## Study measures

Almost all data being gathered during the trial are self-reported data that will be collected via an online survey program (<https://www.qualtrics.com>). In addition, one semi-structured telephone interview will be conducted twelve months after baseline to assess relapse. Participants will be asked to report demographical data including gender, age, marital and employment status, ethnicity and education, as well as the information about the past course of BD, at baseline. The primary outcome is well-being and secondary outcomes include personal recovery, social role participation and symptoms of depression, mania and anxiety. Additionally, processes of positive emotions, self-compassion and positive relationships are assessed and economic outcomes are used to calculate the cost-effectiveness. An overview of the study measures at the different time points can be found in Table 2.

*Global illness severity.* To screen for the presence of depressive and (hypo)manic symptoms and determine eligibility of possible participants, the Clinical Global Impression – Bipolar (CGI-BP; Spearing et al., 1997) scale will be used. This scale comprises three different measures, including severity of depressive and (hypo)manic symptoms, change from preceding phases and change from worst phase of illness. For this study, only the measure assessing the severity of symptoms will be used ranging from 1 (normal, not ill) to 7 (very severely ill). The CGI-BP showed excellent interrater reliability in prior studies (Spearing et al., 1997).

*Well-being.* The Mental Health Continuum-Short Form (MHC-SF) is a comprehensive well-validated measure of well-being (Lamers et al., 2011). The MHC-SF measures three dimensions

of well-being: 1) emotional well-being (three items), defined in terms of the presence of positive feelings, the absence of negative feelings and satisfaction with life; 2) psychological well-being (six items), defined in terms of positive functioning in individual life in terms of e.g. self-acceptance, personal goals, positive relationships, and environmental mastery; 3) social well-being (five items), defined in terms of positive functioning in social life in terms of e.g. social integration and social contribution. Participants rate the frequency of feelings in the last week. A total score can be created by summing all 14 items, where higher scores indicate better positive well-being. The Dutch version of the MHC-SF showed high internal consistency for total scores ( $\alpha = 0.89$ ) and for the subscales *emotional* ( $\alpha = 0.83$ ) and *psychological well-being* ( $\alpha = 0.83$ ) and adequate reliability for the subscale *social well-being* ( $\alpha = 0.74$ ) and correlates well with corresponding aspects of well-being and functioning, showing convergent validity (Lamers et al., 2011).

*Personal recovery.* To comprehensively assess personal recovery, the 15-item version of the Questionnaire about the Process of Recovery is used (QPR; Law et al., 2014; Neil et al., 2009). The scale aims to assess personal recovery in the last 7 days (e.g. “I feel better about myself” or “I can actively engage with life”), with items being scored on a 5-point Likert scale, ranging from 0 to (*disagree strongly*) to 4 (*agree strongly*) and higher scores being indicative of recovery. The internal consistency of the 15-item version has been found to be high ( $\alpha = 0.89$ ) in a sample of psychotic patients (Williams et al., 2015) and in a group of individuals with a schizophrenia spectrum diagnosis ( $\alpha = 0.93$ ) (Law et al., 2014). For this study, the QPR has been translated into Dutch via forward and backward translation.

*Social role participation.* The Social Role Participation Questionnaire (SRPQ; Davis et al., 2011) assesses social role participation. For this study, the short version of the questionnaire (S-SRPQ; Oude Voshaar et al., 2016) will be used, which consists of 12 items, measuring the influence of (psychological) health in the past on six social roles (e.g. intimate relationship or employment) along two dimensions: (1) satisfaction with role performance and (2) experienced physical / psychological difficulty. Items are scored on a 5-point Likert Scale, reaching from 0 (*not satisfied at all / no difficulties at all*) to 4 (*very much satisfied / not possible*), with higher scores indicating more satisfaction respectively more experienced difficulties with a social role. The psychometric qualities of the Dutch s-SRPQ were found to be good for both subscales ( $\alpha = 0.86$ ) (Oude Voshaar et al., 2016).

*Depressive symptoms.* The self-report version of the Quick Inventory of Depressive Symptomatology (QIDS-SR) (Rush et al., 2003; Wardenaar et al., 2010) assesses depressive symptoms in the past on 16 items. The scale requires individuals to rate different depression symptoms, such as sad mood, concentration, suicidal ideation, general interest,

energy/fatigue, sleep, appetite and weight. Items are scored on a 4-point Likert Scale with different answering categories. A total score can be obtained by summing all items, with higher scores indicating more depressive symptomatology. The QIDS-SR has shown to be internally consistent ( $\alpha = 0.86$ ) (Rush et al., 2003).

*Manic symptoms.* Current manic symptoms are measured using the Altman Self-Rating Mania Scale (ASRM) (Altman et al., 1997). The scale consists of five statements that represent different manic symptoms, including feeling happier, self-confident and talkative than normal. All five items are rated on a 5-point Likert scale with different answering categories. A total score can be obtained by summing all items, with higher scores indicating more manic symptomatology. The ASRM has high test-retest reliability (Altman et al., 1997), has been shown to be sensitive to changes of clinical states (Altman et al., 2001) and to predict related measures in non-clinical student samples (Meyer et al., 2004).

**Table 2.** Overview of study parameters and measurement points

Questionnaire	Outcome	Screening	T0	T1 <sup>a</sup>	T2	T3 <sup>b</sup>	T4 <sup>c</sup>
CGI-BP <sup>d</sup>	Global illness severity	X					
MINI	Diagnosis BD	X					
MHC-SF	Positive well-being	X	X	X	X	X	X
QPR	Personal recovery		X		X	X	X
S-SRPQ	Social participation		X		X	X	X
QIDS-SR	Depressive symptoms		X		X	X	X
ASRM	Manic symptoms		X		X	X	X
HADS-A	Anxiety symptoms		X		X	X	X
PANAS	Positive emotions		X	X	X		
SCS-SF	Self-compassion		X	X	X		
SPWB	Positive relationships		X	X	X		
RPA	Dampening of positive affect		X	X	X		
Telephone interviews	Relapse						X
EQ-5D-5L	Quality of Life		X				X
TiC-P	Costs associated with psychiatric illness		X				X
Sociodemographic data	Gender, age, education, marital status, living situation, ethnicity		X				

*Note.* <sup>a</sup>4 weeks after intervention begin. <sup>b</sup>6 months after baseline. <sup>c</sup>12 months after baseline. <sup>d</sup>Clinician reported.

*Anxiety symptoms.* The anxiety subscale of the Hospital Anxiety and Depression Scale (HADS-A; Zigmond & Snaith, 1983) is used to assess anxious symptomatology. The HADS-A aims to measure anxiety symptoms in 7 items. Participants rate the frequency of symptoms (e.g. “Worrying thoughts go through my mind”) on scale ranging from 0 (“not at all”) to 3 (“very often”) and higher scores indicate higher anxiety symptoms. The Dutch version of

the HADS-A (Spinhoven et al., 1997) has been shown good internal consistency in a sample from the general population ( $\alpha = 0.84$ ) and in a sample of psychiatric outpatients ( $\alpha = 0.81$ ).

*Positive emotions.* The Positive and Negative Affect Schedule (PANAS) (Watson et al., 1988) measures current emotions on two different dimensions: (1) positive and (2) negative affect and includes 20 items describing emotional states (e.g. “*active*” or “*anxious*”). Participants can score those states on a 5-point Likert scale, representing the extent to which they experience an affect at this moment or have experienced in the past week, reaching from 1 (*very slightly or not at all*) to 5 (*extremely*). The scores can be summed up to gain scores for positive and negative affect respectively, with higher scores indicating higher affectivity. For this study, the Dutch version of the PANAS and only the positive affect subscale will be used, which showed acceptable reliability ( $\alpha = .79$ ) (Engelen et al., 2006).

*Self-compassion.* The Self-Compassion Scale – Short Form (SCS-SF) (Neff, 2003b; Raes et al., 2011) measures the process of self-compassion on six dimensions: (1) self-kindness, (2) self-judgment, (3) common humanity, (4) isolation, (5) mindfulness and (6) over-identification and contains twelve items (e.g., “*When I fail at something important to me I become consumed by feelings of inadequacy*”). Each dimension is assessed by two items, which are scored on a 7-point response scale ranging from 1 (*almost never*) to 7 (*almost always*), representing the extent to which an individual experiences certain aspects of self-compassion. Higher scores indicate an increased degree of self-compassion. Reliability of the total Dutch SCS-SF was shown to be good ( $\alpha = .87$ ). Solely total scores of the SCS-SF will be used for further analyses, since the psychometric properties of the subscales were questionable (Raes et al., 2011).

*Positive relationships.* The concept of positive relations is assessed using the Scales of Psychological Well-Being (SPWB; Ryff & Keyes, 1995), which assesses psychological well-being on six different dimensions (e.g. environmental mastery, self-acceptance). For this study, the subscale *positive relations* will be used measuring the extent to which an individual experiences meaningful intrapersonal relationships with other people (e.g. “*People would describe me as a giving person, willing to share my time with others*”). Items are scored on a scale ranging from 1 (strongly disagree) to 6 (strongly agree) with higher scores indicating more positive relations with others. Different versions of the SPWB exist within literature, differing in number of items per subscale (reaching from 3 items to 20 items per subscale). For feasibility reasons and since the short version of this subscale (3-items) showed unacceptable internal consistency ( $\alpha = .44 - .52$ ), we decided to use the 9-item version of the *positive relationship* subscale, which showed acceptable internal consistency in two previous studies ( $\alpha = .77$ ) in samples of psychology students and professionals from divers occupation background

*Dampening of positive affect.* To assess the process of dampening the Responses to Positive Affect questionnaire (RPA; (Feldman et al., 2008; Raes et al., 2010)) is used, which consists of 17 items and measures cognitive responses to positive affective states. Respondents rate the items on a 4-point Likert scale, ranging from 1 (*almost never*) to 4 (*almost always*). For this study, only the subscale *dampening* is used (e.g., “I don’t deserve this”), which assesses the tendency to cognitively avoid or suppress positive emotions (eight items). Scores of the scales are calculated by summing up the scores on the items. The Dutch version, which is used in this study, showed satisfactory internal consistency ( $\alpha = 0.80$ ) for the dampening subscale (Raes et al., 2010).

*Relapse.* Semi-structured telephone interviews will be performed with people of both the intervention and control group. Goal of the interviews is to retrospectively illustrate the mood development in the time after the intervention and to capture depressive or manic mood swings with the Life Chart Method (Denicoff et al., 2000). The interviews allow to graphically score severity of mood swings, the time they appeared (i.e., in which month) and which type of mood swings appeared (e.g., rapid cycling). The interview has been applied successfully in a previous study to measure relapse (Van der Voort et al., 2011).

## **Economic measures**

*Quality of Life.* The EQ-5D-5L (Herdman et al., 2011) is a quality of life measure consisting of five items representing five dimensions (mobility, self-care, usual activities, pain/discomfort and anxiety/ depression). For each dimension/item, individuals rate the extent of problems ranging from ‘no problems’ to ‘extreme problems’.

*Costs associated with psychiatric illness.* The Trimbos and iMTA questionnaire on costs associated with psychiatric illness (TiC-P) (Bouwman et al., 2013) is a measure of health care utilization and production loss in patients with psychiatric disorders. Items are generic and not related to a specific psychiatric disease. A first part of the TiC-P includes 9 structured no/yes items on medical consumption (e.g. contact with specific mental health care providers). A second part (13 items) consists of the Short Form-Health and Labor Questionnaire, a generic instrument to collect data on productivity losses due to health problems (e.g. absence from work).



## Data collection, management and storage

Data will be handled confidentially in accordance with the Dutch Personal Data Protection Act. For the purpose of this study, a data management plan has been created with DMPonline (<https://dmponline.dcc.ac.uk>). Details of data management procedures can be requested from the first author of this manuscript. Quality checks, including double data entry and range check for data values will be performed by the first author (JK) and an additional researcher. Personal data will be coded with an individual ID-code, which is not relatable to the participant. All collected data will be stored in a file containing only the identification code. The coded research data will be stored at the BMS Datalab of the University of Twente for a period of 15 years. In this time period, data is accessible to other researchers. After the period of 15 years, data will be stored in long time storage at Data Archiving and Networked Service by the Royal Dutch Academy of Sciences. Participants who want to be informed about their personal data or who want their data deleted can send a request to the principal investigator.

## Study integrity

The study protocol has been designed in accordance with the SPIRIT STATEMENT (Chan et al., 2013) and the study has been approved by the Medical Ethical Committee Twente (Proposal No: NL62997.044.17). The study will be carried out according to the principles of the Declaration of Helsinki (64<sup>th</sup> WMA General Assembly, Fortaleza, Brazil, October 2013) and the Medical Research Involving Human Subjects Act (WMO).

## Statistical methods

### *Power calculation*

The sample size calculation for this trial is conservatively based on the ability to detect at least a moderate effect of Cohen's  $d = 0.60$  in the post-hoc tests on the primary outcome (well-being) at post-intervention (T2). For a two-sided independent t-test with 80% power and  $\alpha = 0.05$ , this requires 45 patients for both treatment groups. Taking a maximum drop-out rate of 20% into account, a total of 112 patients will need to be included for the per-protocol analysis.

### *Statistical analyses*

Analyses will be done on both intention to treat (ITT) and per-protocol basis. The primary ITT analyses will be performed using linear mixed modelling (LMM) that adequately deals with missing at random data and the nested structure of repeated-measures data. LMMs with time, treatment and time-by-treatment interactions will be performed to test the effectiveness of

the intervention in improving continuous primary and secondary outcomes of well-being, personal recovery, psychopathology, self-compassion, positive relationships and dampening. Post-hoc independent t-tests with Holm-Bonferroni correction will be performed to test for significant between-group differences at all time-points. Based on estimated marginal means and corresponding standard errors from the LMM models, between-group standardized effect sizes will additionally be expressed as Cohen's d with 95% confidence intervals (CI). Binary relapse data from the interviews will be analyzed with Kaplan Meyer survival estimates to compare the time to relapse and relapse rates between the intervention and control group. Differences in the proportion of relapsed patients and predictors of relapse will be additionally examined using generalized (binary) LMMs with post-hoc chi-square tests and relative risks (RR) with 95% CI to examine the significance and magnitude of differences at each follow-up point. Moderation and mediation analyses will be conducted to explore possible working mechanisms of the interventions. To calculate the cost-effectiveness of the intervention, quality adjusted life years (QALYs) will be taken into account as primary utility measure. QALYs will be calculated from the EQ-5D-5L. The incremental cost-utility ratio (ICUR) will be calculated by dividing the difference in costs calculated from the TiC-P by the difference in the QALYs produced by the two groups. The ICUR is expressed as costs per QALY gained.

## DISCUSSION

The presented study aims to examine the short- and long term effectiveness of the multicomponent positive psychology intervention "Living well with bipolar disorder" for euthymic BD patients to enhance personal recovery and well-being. In the present article, we described the intervention development process and several adaptations made to the original program to tailor the intervention for the target group. The study will be conducted in at least four different outpatient treatment centers in the Netherlands, all specialized in with the treatment of BD. The intervention group will receive the intervention program in addition to CAU and the control group will receive CAU only.

Several potential limitations of the study have to be considered in advance. First, since participants in this intervention are euthymic (i.e. not in a syndromal depressive, hypomanic or manic episode), results of the trial cannot be generalized to the entire patient group in all phases of BD. Second, the study will not be able to determine which specific elements of the interventions lead to possible effects of the intervention. Non-specific factors (e.g. social contact during group lessons or placebo effect) can thus not be ruled out as possible confounding variables. However, since we mainly aim to investigate the effect of the intervention in real-life clinical practice and our study concerns a pragmatic randomized trial, this limitation is not considered to impair the value of our study (Chalkidou et al., 2012).

Further research could investigate the effectiveness of the intervention to determine whether the intervention is equally effective when taking non-specific factors into account. Third, non-adherence to the intervention or dropout from the assessment might occur. Both types of attrition are likely to bias the results (Eysenbach, 2005).

Our study will be the first to explore the effectiveness of a positive psychology intervention for the treatment of people with BD. In addition, it will be the first study to be implemented that focusses on the improvement of primarily personal recovery and well-being and uses a more sound design than existing pilot studies. The results of the study are expected to broaden the evidence base of clinical positive psychology and personal recovery and potentially generate novel treatment methods for people suffering from BD.

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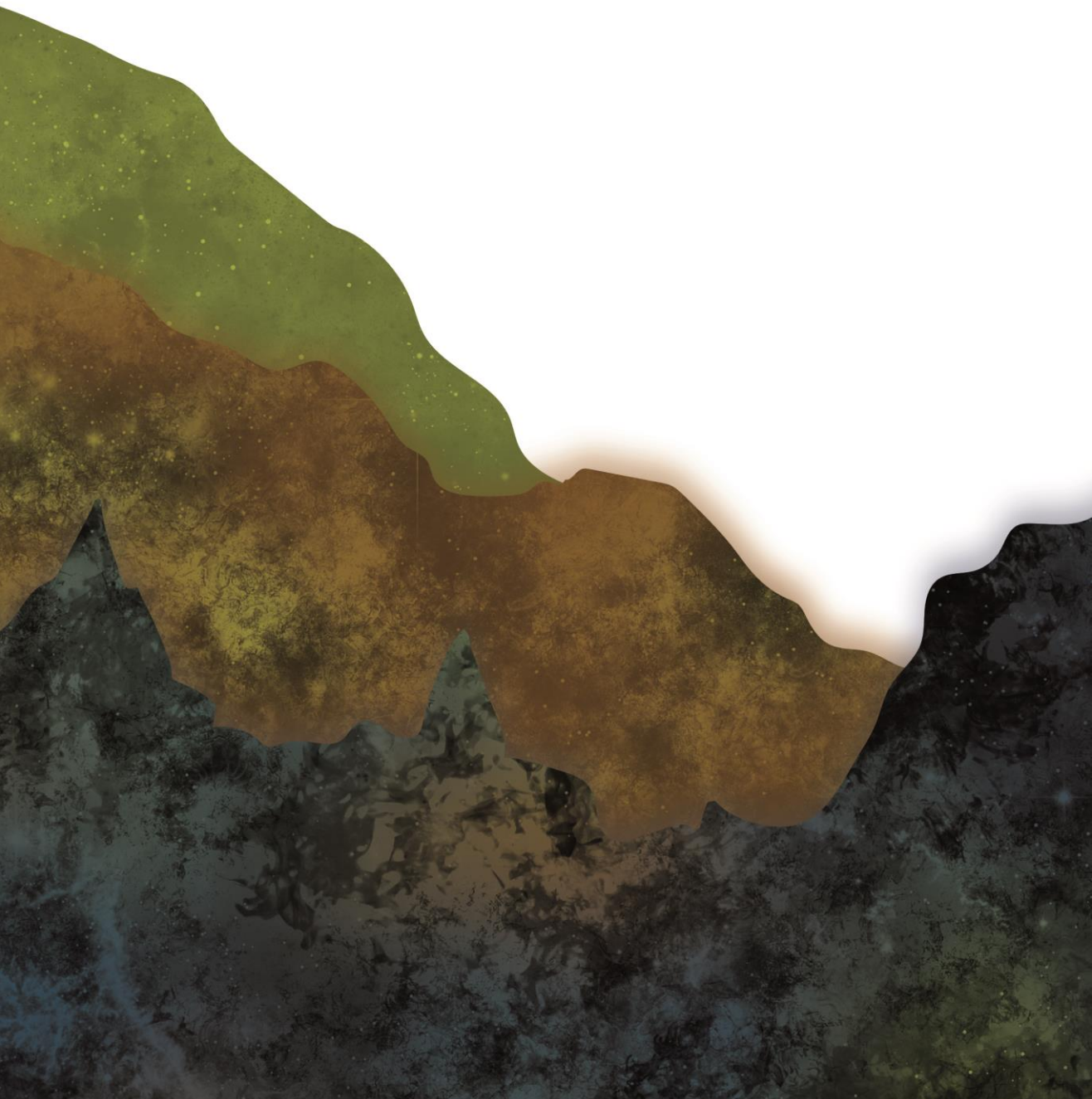


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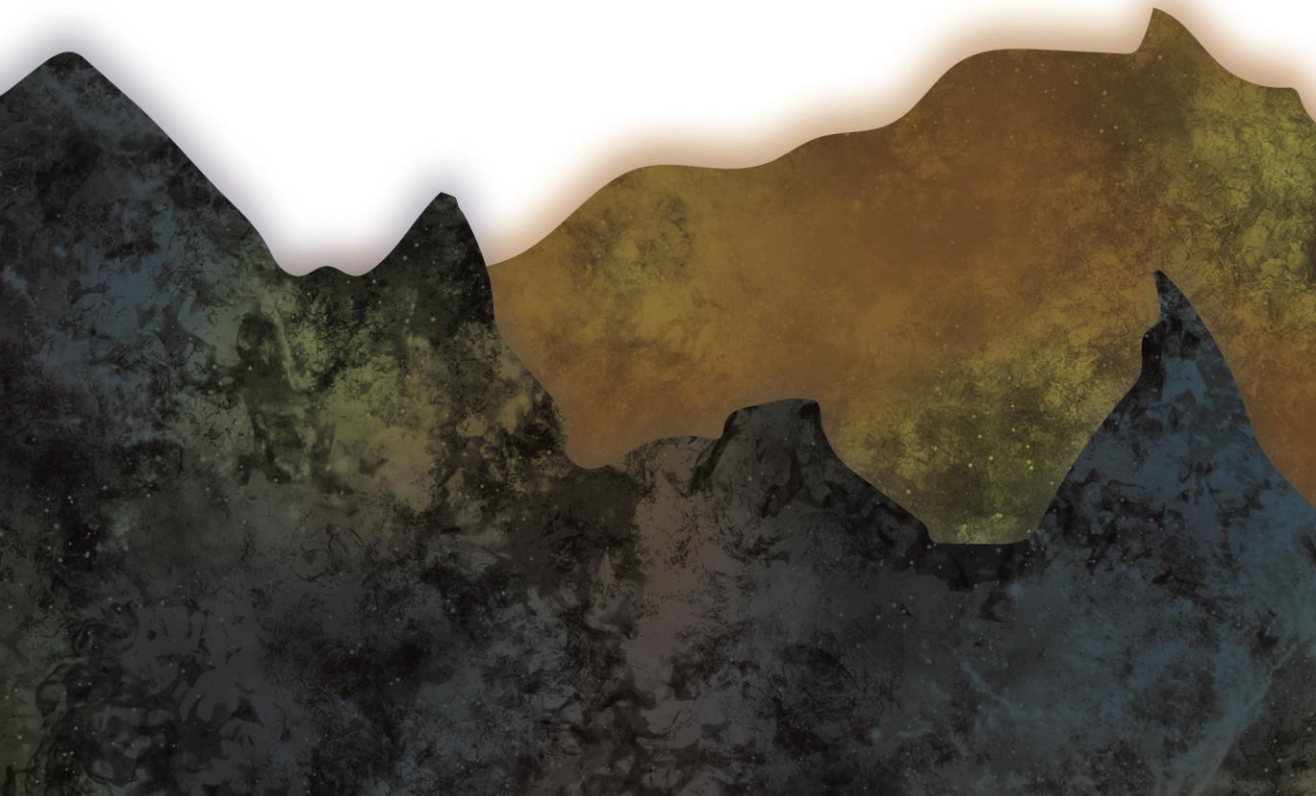
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# CHAPTER 8

The effectiveness of a multicomponent positive psychology group intervention for euthymic patients with bipolar disorder: a pragmatic randomized controlled trial

Kraiss, J.T., ten Klooster, P.M., Crispijn, M. Stevens, A.W.M.M., Doornbos, B., Kupka, R.W., & Bohlmeijer, E.T. (2021). The effectiveness of a multicomponent positive psychology group intervention for euthymic patients with bipolar disorder: a pragmatic randomized controlled trial. *Manuscript in preparation.*



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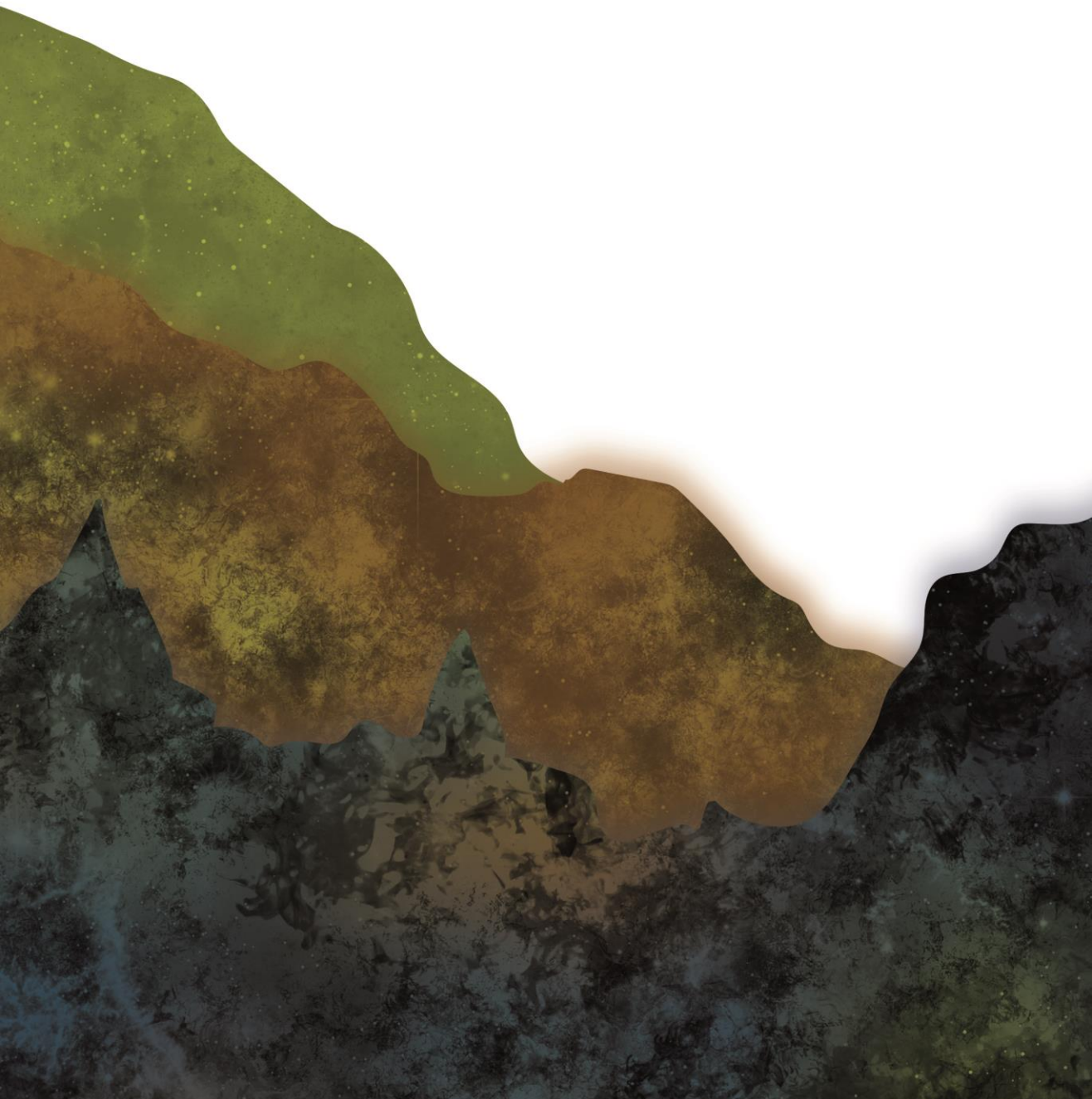


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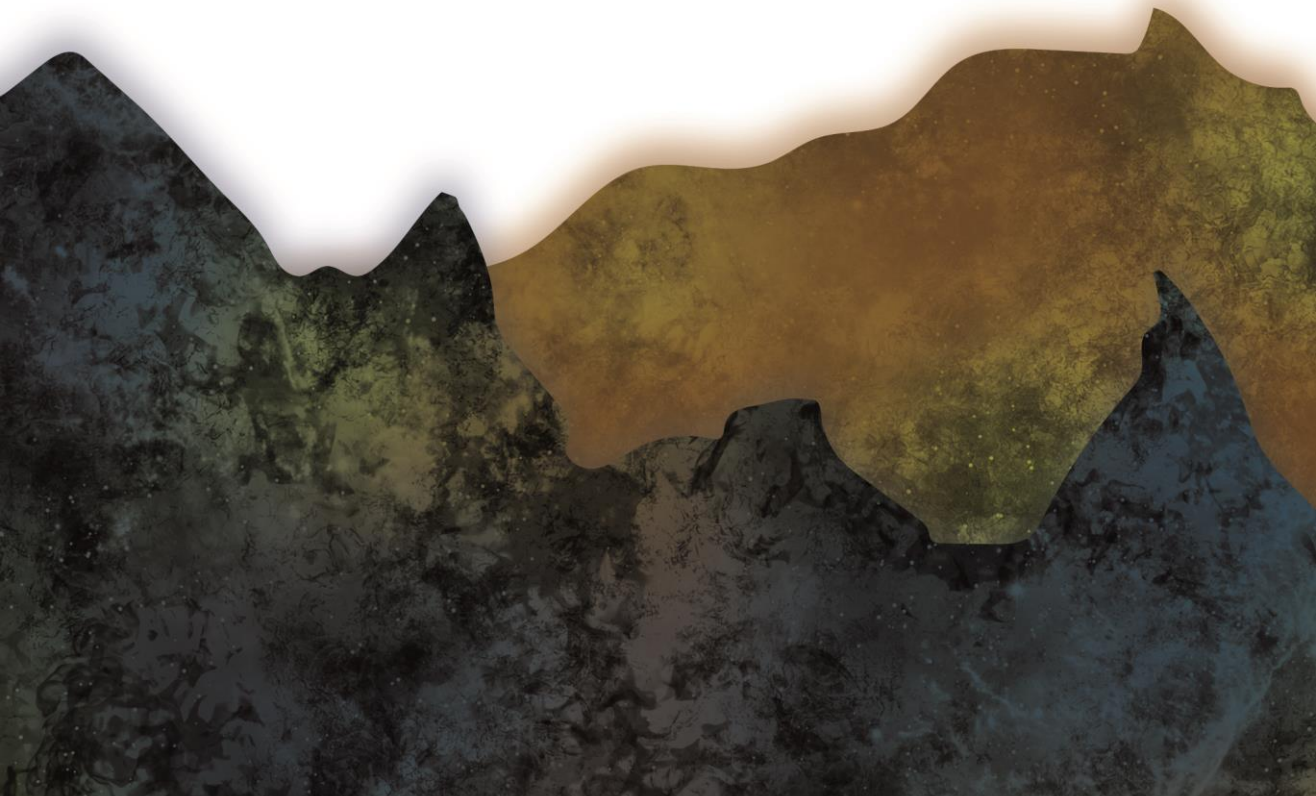
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# CHAPTER 9

Summary & General Discussion





## INTRODUCTION

Bipolar disorder (BD) is a recurrent mood disorder (Goodwin & Jamison, 2007; Grande et al., 2015) associated with substantial negative consequences for the individual and high societal burden (Cloutier et al., 2018; Crump et al., 2013; Ferrari et al., 2016; Gonda et al., 2012). Current treatment guidelines for BD primarily focus on symptomatic and functional recovery (Grande et al., 2015; Kupka et al., 2015). However, treatment outcomes related to mental well-being and personal recovery are increasingly seen as an important part of recovery from mental disorders (Bohlmeijer & Westerhof, 2020; Fava et al., 2007; Leamy et al., 2011; Slade, 2009, 2010), also in patients with BD (Bonnín et al., 2019; Dodd et al., 2017). Mental well-being and mental illness can be seen as related, yet distinctive parts of mental health (Keyes, 2005), and comprises the presence of emotional, social and psychological well-being (Diener & Ryan, 2009; Keyes, 2005; Keyes, 1998; Ryff & Keyes, 1995). Leamy et al. (2011) conceptualized personal recovery as containing five key components, namely connectedness, hope, identity, meaning and empowerment. One field of psychology that specifically focuses on the improvement of mental well-being and personal recovery is positive psychology (Resnick & Rosenheck, 2006; Slade, 2010). Patients with BD themselves consider outcomes related to mental well-being and personal recovery as important (Mezes et al., 2020; Todd et al., 2012) and research suggests that both mental well-being and psychoatholgo should be improved in people with mental disorders when aiming to enhance mental health (De Vos et al., 2018; Franken et al., 2018; Trompetter et al., 2017). Despite the importance of mental well-being and personal recovery for BD, studies examining the effect of interventions focusing on personal recovery and mental well-being in BD remain scarce, as well as studies examining the effect of positive psychology treatments in BD.

The overall goal of this dissertation is to contribute to the integration of positive psychology and personal recovery into the treatment of BD. Four aims are addressed in this thesis. First, an overview of the research field of positive psychology interventions for serious mental illness (SMI) and of economic studies for BD is provided. This aims to summarize the current state of the art of positive psychology interventions for SMI and cost-effectiveness of non-pharmacological interventions for BD. Two systematic reviews of the literature, of which one study also used meta-analysis techniques, are carried out. Second, two measurement instruments, the Questionnaire about the Process of Recovery (QPR) and the Responses to Positive Affect questionnaire (RPA), are psychometrically validated. Although these constructs constitute relevant outcomes for BD, the two measurement instruments were not translated into Dutch before and/or have not been validated in people with BD. Third, this thesis aims to widen our knowledge of what contributes to personal recovery in BD, by exploring factors that are associated with personal recovery in people with BD. Fourth, a novel positive psychology

group treatment is developed aimed at improving personal recovery and mental well-being in euthymic BD patients. The effectiveness of this treatment is evaluated in a pragmatic randomized controlled trial (RCT). Accordingly, the following four goals are addressed in this thesis:

- (1) To provide an overview of the research field regarding positive psychology interventions for patients with severe mental illness and economic studies in BD.
- (2) To examine the psychometric properties of a Dutch translation of the Questionnaire about the Process of Recovery (QPR) and the Responses to Positive Affect questionnaire (RPA) in people with BD.
- (3) To explore factors associated with personal recovery in BD.
- (4) To develop a specific positive psychology intervention for euthymic persons with BD aimed at mental well-being and personal recovery and evaluate its effectiveness in a pragmatic, multicenter randomized controlled trial.

These four goals are addressed in the following four parts in this dissertation: (I) overview of the research field, (II) measurement of relevant constructs, (III) determinants of personal recovery, and (IV) intervention development and evaluation. This last chapter, the general discussion, summarizes the seven studies presented in this thesis and then discusses the main findings obtained in each of these studies. Afterwards, implications, general strengths and limitations of the thesis and future directions are outlined. Finally, a conclusion about the overall findings of this dissertation will be drawn.

## **SUMMARY OF STUDIES**

### **Part I: Overview of the research field**

In *chapter 2* we provided an overview of the research field regarding positive psychological interventions for SMI. The effects of these interventions on outcomes of mental well-being and psychopathology were synthesized using meta-analysis techniques. SMIs can be defined as a psychiatric disorder with severe functional problems, where the constraints are causal and consequential and which is not temporary, and there is a need for coordinated professional care (Delespaul, 2013). Examples of SMIs are schizophrenia, schizoaffective disorder, personality disorder, BD and major depressive disorder. In our review, we identified 16 studies ( $N = 729$ ), of which nine studies were RCTs. The meta-analyses revealed nonsignificant between-group effects for outcomes related to well-being ( $g = 0.16$ ) and psychopathology ( $g = -0.10$ ). Within-groups effect sizes (i.e. change within the intervention groups) revealed significant effects for outcomes of mental well-being ( $g = 0.40$ ) and psychopathology ( $g = -$

0.70). The findings suggest that we cannot conclude, based on the current evidence, that positive psychology interventions are effective when compared to other active interventions or TAU, but that people with SMI indeed seem to benefit from positive psychology interventions in terms of improvement of well-being and psychological symptoms.

*Chapter 3* presented a systematic review on economic evaluations and cost-of-illness studies in BD. The goal of this study was to review the evidence regarding economic evaluations of non-pharmacological interventions in BD and cost-of-illness studies in BD published since 2012. We identified eight economic evaluations and ten cost-of-illness studies. This shows that the evidence in the field of economic evaluations of non-pharmacological interventions is rather small. High heterogeneity in methodological characteristics and outcomes was found, making it difficult to compare studies and generalize the findings to specific groups or interventions. All non-pharmacological interventions included in the review improved outcomes related with BD, and five treatments additionally reduced the costs associated with BD. Based on data from the cost-of-illness studies, we concluded that BD is associated with substantial direct and indirect costs, but also found that costs strongly varied between studies. In studies that assessed direct and indirect costs, indirect costs (e.g., productivity loss or unemployment) were the main cost drivers. Our findings suggest that non-pharmacological interventions may not only improve clinical outcomes in BD, but also have the potential to be cost-effective treatment alternatives to TAU or other active control conditions. The findings from this study also highlight the high costs BD is associated with, showing the need for cost-effective treatment alternatives for this patient group.

## **Part II: Measurement of relevant constructs**

In *chapter 4* and *5* we conducted two psychometric evaluations of a Dutch translation of the QPR and RPA in a self-selected sample of people with BD. Evaluating psychometric properties of measurement instruments is relevant to determine whether the instrument reliably assesses the construct it intends to measure. Despite the relevance of personal recovery and positive emotion regulation in BD, the QPR and the RPA were not yet validated in people with BD and/or were not translated into Dutch before. Although the QPR has been validated before (Law et al., 2014; Williams et al., 2015) and has been translated from English into several languages (Argentzell et al., 2017; Chien & Chan, 2013; Kanehara et al., 2017), this study was the first to translate the QPR into Dutch and to evaluate the psychometric properties in people with BD. We found excellent internal consistency for the QPR and high correlations with validation measures, including mental well-being, social participation and measures of psychopathology, which indicates convergent validity. In addition, our findings indicate that the QPR has incremental validity, as it explained variance in psychopathology above and



beyond mental well-being, despite the fact that personal recovery and mental well-being are highly correlated. This suggests that assessing personal recovery in BD besides mental well-being might be of added value. The fact that a Dutch translation of the QPR showed such promising results in the study in chapter 4, also means that that a validated Dutch version of the QPR is now available and can be used to assess personal recovery in patients with BD in the Netherlands.

*Chapter 5* of this thesis presented a psychometric evaluation of the Dutch Responses to Positive Affect questionnaire (RPA) in a sample of people with BD. The RPA assesses three different strategies of positive emotion regulation, namely dampening, emotion-focused and self-focused positive rumination (Feldman et al., 2008). While *dampening* is described as the suppression of positive affect to reduce the intensity of a positive mood (Quoidbach et al., 2010), *positive rumination* can be defined as the tendency to respond to positive affective states with recurrent thoughts about positive experiences or feelings (Feldman et al., 2008). Although prior studies already validated the RPA (Feldman et al., 2008; Olofsson et al., 2014; Voss et al., 2019; Yang & Guo, 2014) and translated the RPA into Dutch (Raes et al., 2010), all of these studies used community or student samples. The study provided in *chapter 5* was the first to examine the psychometric properties of the RPA in people with BD. Our findings showed that the subscales had adequate to good internal consistency. Also, both dampening and positive rumination were related with criterion measures of mental well-being, personal recovery, social role participation and anxiety and depressive symptoms, indicating construct validity. The RPA also showed incremental validity, as the RPA explained variance in personal recovery above and beyond mental well-being, social role participation and measures of depression and anxiety symptoms. Altogether, the findings from *chapter 4 and 5* suggest that the QPR and the RPA are reliable and valid instruments to assess personal recovery and positive emotion regulation in BD.

### **Part III: Determinants of personal recovery**

Studies exploring factors contributing to personal recovery remain relatively scarce. Although some previous studies investigated determinants of personal recovery, none of the previous studies specifically examined the contribution of social role participation and positive emotion regulation in BD. Therefore, in *chapter 6* of this dissertation we combined data from the survey study (*chapter 4 and 5*) and baseline data from the RCT (*chapter 8*) to explore factors associated with personal recovery in BD. The sample was split into a training and test set and explored a set of sociodemographic, social and psychological factors as potential predictors of personal recovery using block-wise multiple regression. Afterwards, a parsimonious model was created using model training and cross-validation. The final parsimonious model contained the

four variables satisfaction with social roles, anxiety symptoms, manic symptoms and emotion-focused positive rumination and explained 57.3% variance in personal recovery (adjusted  $R^2 = .56$ ). The final parsimonious model was then used to predict personal recovery in the test set to evaluate its ability to predict personal recovery in an independent sample. The model performed comparably well in predicting personal recovery in the test set (adjusted  $R^2 = .49$ ). These findings highlight the importance of symptomatology, social participation and positive rumination as independent predictors of personal recovery in BD and suggest that they might represent relevant treatment targets when aiming to improve personal recovery.

## Part IV: Intervention development and evaluation

As outlined earlier, studies investigating the effect of specific interventions aimed at personal recovery in BD are scarce, as well as studies that examine the effect of positive psychology interventions to improve mental well-being or personal recovery in BD. Therefore, in *chapter 7* we described the development of a novel multicomponent positive psychology treatment for patients with BD as well as the study design and procedure for the RCT presented *chapter 8* of this thesis. The intervention *Living well with bipolar disorder* is based on the self-help book *Dit is jouw leven* (English translation: *This is your life*) (Bohlmeijer & Hulsbergen, 2013). In *chapter 7*, steps that were taken to customize the original intervention for patients with BD. Initial steps to evaluate positive psychology exercises in patients with BD using a formative user evaluation in a small group of patients are also described. The final intervention consists of eight group sessions containing eight modules, for example (self)-compassion, positive emotions, optimism and personal strengths. The intervention is given by trained therapists. One session takes 2 hours and each sessions is accompanied by homework exercises.

In *chapter 8* we presented outcomes from a pragmatic RCT in which we evaluated the effect of the positive psychology treatment as an adjunct to TAU compared to TAU only. The primary outcome was mental well-being measured with Mental Health Continuum-Short Form (Keyes et al., 2008; Lamers et al., 2011). Personal recovery, social participation, symptomatology and different positive psychological processes were also assessed. We included 97 euthymic BD patients in the trial, which were allocated to receive either the positive psychology treatment in addition to TAU ( $n = 54$ ) or TAU alone ( $n = 43$ ). Participants were followed until six months after baseline. Significant medium to large between-group effects at posttest were found for mental well-being ( $d = 0.79$ ) and personal recovery ( $d = 0.83$ ). The effects for mental well-being were sustained until 6-month follow-up. Significant between-group effects at posttest were also found for emotion-focused positive rumination and a significant interaction effect was found for positive emotions. Effects on outcomes of depression, anxiety and (hypo)mania were nonsignificant. The findings show that an 8-week positive psychology group treatment is

effective in improving mental well-being until at least six months after baseline. This suggests that positive psychology can contribute to leading a joyful and meaningful life in patients with BD, also in the presence of a SMI.

## DISCUSSION OF MAIN FINDINGS

### Positive psychology in severe mental illness: still a long way to go

In the systematic review and meta-analysis conducted in *chapter 2*, it was striking that the literature search merely identified 16 studies examining the effect of positive psychology interventions for SMI. Considering that thousands of positive psychology studies were published in the past two decades (Seligman, 2019), this is a remarkable finding that shows that the evidence-base of positive psychology interventions in SMI is still scarce. Also, only two small studies were identified that specifically included samples of patients with BD (Celano et al., 2020; Painter et al., 2019), of which only one study was a controlled study (Celano et al., 2020). In comparison, a systematic review of all types of psychological interventions in BD identified 55 trials (Oud et al., 2016), showing the comparably small evidence-base for positive psychology for BD compared to other psychological interventions. It is also notable that the 16 studies included in our review included 36 different outcome measures related to well-being. These measures also assessed substantially different facets of well-being, such as positive emotions, satisfaction with life, optimism, gratitude or hope. This is also in line with previous meta-analyses of positive psychology interventions, that also included a vast of array of different outcomes and grouped them together under the umbrella term well-being (Bolier et al., 2013; Chakhssi et al., 2018; Hendriks et al., 2020; Sin & Lyubomirsky, 2009). This leaves an ambiguous picture of the effectiveness of the interventions, since these outcomes assess different processes related to well-being. In addition, grouping together all these outcomes under the broad concept of well-being makes it difficult to determine the effect of interventions on specific parts aspects of well-being. A relevant implication of this finding is that a stronger consensus is needed on how well-being should be operationalized and assessed more consistently across studies. This would increase comparability across studies and allow us to determine the effect of interventions on specific aspects of well-being.

Although we did not find significant between-group effects for positive psychology interventions compared with TAU or other active interventions (e.g., CBT or mindfulness), we did find significant within-group improvements. This is a promising finding and suggests that patients from these high-risk populations indeed do benefit from positive psychology interventions in terms of improvement of mental well-being and psychopathology. Therefore, it might be interesting for future studies to consider positive psychology treatments as a viable

treatment option for people with SMI. This is also in line with previous accounts arguing for the potential of positive psychology for people with mental disorders and a synergy of positive psychology and recovery approaches (Resnick & Rosenheck, 2006; Slade, 2010). In *chapter 8* of this thesis we could convincingly show in a full RCT that a positive psychology treatment as an addition to TAU indeed is effective in improving well-being and personal recovery in euthymic BD patients. Overall, the findings in this thesis are promising and suggest that people with SMI can benefit from positive psychology interventions in general (*chapter 2*) and that a new multicomponent positive psychology treatment is effective in improving mental well-being and personal recovery in euthymic BD patients (*chapter 8*). These are promising conclusions for future studies aiming to improve mental well-being and personal recovery in people with SMI, yet there is still a long way to go in examining the potential of positive psychology and recovery-approaches to enhance mental well-being and personal recovery in SMI. Most previous studies used weak methodological designs or were underpowered. Also, specific working mechanisms of these interventions in patients with SMI remain largely unknown, as well our understanding of what works for whom and in which phase of a specific disorder. Since SMI is associated with high societal burden (Trautmann et al., 2016), further comparisons with treatment as usual (TAU) would also be important for future research to gain a better understanding of the (cost)-effectiveness and added value of positive psychology treatments for current TAU.

## **No evidence for the cost-effectiveness of recent psychotherapeutic approaches in bipolar disorder**

In *chapter 3* we found that that BD is associated with substantial economic costs and that indirect costs are the main driver in the costs associated with BD. This in line with previously published reviews on the costs of BD (Jin & McCrone, 2015; Kleine-Budde et al., 2014). It is also not surprising considering the strong functional and cognitive impairments BD is associated with, also leading to strong impairments in being able to work. Compared to a previous review on economic evaluations (Abdul Pari et al., 2014), four additional economic evaluations were identified in our review, showing that research in this field has been conducted since 2013. This is promising and suggests that the evidence-base regarding cost-effectiveness of non-pharmacological interventions in BD is slowly increasing, yet the total number of economic evaluations that were identified was still rather small. In comparison, a systematic review of economic evaluations in anxiety disorders identified 42 economic evaluations, of which more than 25 studies were economic evaluations of non-pharmacological interventions (Ophuis et al., 2017). This suggests that – similar to the conclusion in *chapter 2* – the evidence-base of economic evaluations for BD is small compared with other mental disorders. However, it is important to mention that prevalence estimates for anxiety disorders are higher than for BD

(Baxter et al., 2013). Also, anxiety disorders are estimated to be associated with higher societal costs than BD (Olesen et al., 2012). Relatively seen, the current number of cost-effectiveness studies of non-pharmacological interventions for BD is therefore not per se small. Still, the small number of studies makes it difficult to generalize findings or prioritize treatments for specific groups of BD patients.

Importantly, we found that all included interventions were effective in improving clinical outcomes (e.g., fewer weeks in any episode, hospital admissions or days spent in relapse) compared with TAU or other treatment alternatives. In addition, five studies also reported decreased total costs, i.e. the interventions in these studies dominated their comparator. This suggests that non-pharmacological interventions in BD might not only help the individual patient, but also reduce the societal costs associated with BD. Examples of these dominant interventions are cognitive behavioral therapy (Lam et al., 2005), structured group psychoeducation (Scott et al., 2009) or a joint crisis plan (Flood et al., 2006). These non-pharmacological interventions are already incorporated in the multidisciplinary guideline for BD in the Netherlands (Kupka et al., 2015). It is important to stress that none of the included interventions were based on more recent psychotherapeutic approaches, such as mindfulness (Segal et al., 2018), acceptance and commitment therapy (Hayes, 2004), positive psychology (Seligman et al., 2006) or recovery-focused approaches (Jones et al., 2015). This suggests that not only the evidence-base regarding the effect of positive psychology treatments on mental well-being and psychological symptoms for BD is small (*chapter 2*), but also that there is no evidence for the cost-effectiveness of these psychotherapeutic approaches in BD. This underlines the importance for further research examining the (cost)-effectiveness of recent psychotherapeutic approaches for BD, such as positive psychology treatments. Although the cost-effectiveness of the intervention presented in this thesis still needs to be studied, the RCT presented in *chapter 8* of this dissertation can be seen as one important step to fill this gap.

## **The QPR as a promising measurement instrument for personal recovery**

In *chapter 4* we found that a Dutch translation of the QPR is a reliable and valid self-reported measurement instrument to assess personal recovery in BD, but the QPR is also a promising instrument for other reasons. When developing the questionnaire, the patient-perspective was taken into account, as the items were developed together with patients with SMI (Neil et al., 2009). This means that the content of the QPR is not only theory-driven, but also based on experiences of patients themselves. With 15 items, the QPR is also a relatively short instrument. Personal recovery is a concept that is especially included in studies including patients with mental disorders, and burden for participants should be kept as small as possible.

The QPR therefore represents a feasible, less burdensome alternative to other widely-used personal recovery questionnaires, such as the 41-item RAS (Corrigan et al., 1999) or 50-item Stages of Recovery Instrument (Andresen et al., 2006).

Furthermore, the QPR covers all dimensions of the CHIME-framework of recovery from mental illness (Shanks et al., 2013), namely connectedness, hope and optimism, identity, meaning and empowerment. The CHIME-framework is a leading framework to conceptualize personal recovery, and is theoretically based on a narrative synthesis of 97 papers including descriptions and models of personal recovery (Leamy et al., 2011). Other widely used questionnaires of personal recovery do not exclusively assess processes related to personal recovery. A systematic review (Shanks et al., 2013) found that the RAS and the 15-item Illness Management and Recovery Scale (IMR; Salyers et al., 2007) are the two most widely used questionnaires in studies on personal recovery. Yet, the RAS also contains items related to symptomatic recovery and about one third of the items does not map to one of the dimensions of the CHIME-framework. About half of the items of the IMR do not match with dimensions of the CHIME-framework, but assesses processes such as impairment in functioning, symptoms, stress, hospitalizations and medication. Also, the IMR only assesses the personal recovery processes connectedness and empowerment (Shanks et al., 2013). Other widely used measures of personal recovery seem to not exclusively assess personal recovery processes and/or only appear to assess specific parts of personal recovery. This should be kept in mind when choosing a personal recovery instrument and makes the QPR a content-valid and feasible choice if one is interested in exclusively and comprehensively assessing personal recovery.

Despite the strengths of the QPR as a measurement instrument for personal recovery, one potential drawback of the questionnaire is that it was not specifically developed for people with BD, but was developed in patients with psychosis (Neil et al., 2009). Therefore, it remains uncertain whether items included in the QPR actually represent personal recovery from the perspective of patients with BD. With the Bipolar Recovery Questionnaire a psychometrically sound alternative exists that was specifically designed in cooperation with patients with BD (Jones et al., 2013). However, the BRQ contains 36 questions and therefore is a more burdensome alternative to the QPR. This was also the main reason why the QPR was chosen for this dissertation. In addition, similar processes as described in the CHIME-framework are also indicators for increased personal recovery in BD (Mansell et al., 2010; Tse et al., 2014). It can thus be assumed that personal recovery as operationalized with QPR also largely represents personal recovery for patients with BD. Nevertheless, this potential limitation should be kept in mind when considering the QPR as measurement instrument for personal recovery. For future research it could be interesting to study to what extent personal recovery as operationalized with the QPR represents recovery from the perspective of patients with BD.

This might help to determine whether the QPR adequately represents personal recovery for patients with BD.

The QPR may have several valuable applications in practice and research. Practitioners can use the QPR as an individual treatment tool to identify recovery gaps, tailor the recovery process and set individual treatment goals (Neil et al., 2009). This makes the QPR a potentially interesting instrument for clinicians aiming to improve personal recovery in mutual collaboration with their patient. The QPR can also help therapists and patients to individualize the recovery process, which might be relevant to increase treatment motivation and engagement since recovery from mental illness is consistently described as a unique and individual process by patients (Leamy et al., 2011). From a research-perspective, the QPR may be a comprehensive and theory-driven instrument to evaluate the effect of interventions on personal recovery. In this context, a recent study aimed to determine the minimal important difference of the QPR using an anchor-based approach. The authors suggested that a within-person improvement of 5 and a between-group difference of 4 on the total QPR score can be seen as meaningful (Dehmahdi et al., 2021). Neil et al. (2009) suggested that a medium effect size is equivalent to a change of 4.63 on the QPR. This provides relevant additional information for researchers who want to use the QPR for intervention research or routine outcome monitoring. In *chapter 8* of this thesis, we could also show that the QPR appears to be sensitive to change, since significant improvements in personal recovery were found in the intervention group of our RCT. This indicates that the QPR is able to detect change in personal recovery and might therefore be useful to detect intervention effects. The evidence provided in this thesis is not yet definite though. Future research might want to examine additional psychometric properties of the Dutch QPR and also examine the psychometric properties in groups of patients with other psychiatric diagnoses. Additional psychometric properties such as test-retest reliability and more research on its sensitivity to change would be interesting to investigate.

## **Measurement of responses to positive affect and its role in bipolar disorder**

In *chapter 5* we provided a psychometric evaluation of the RPA in people with BD and found that the RPA appears to be a reliable and valid instrument to assess response styles to positive affect in people with BD. Evidence is accumulating suggesting that not only the way people react to negative emotions is important for the onset and maintenance and course of mood disorders, but also the way an individual reacts to positive emotions (Carl et al., 2013). Previous studies also specifically emphasized the importance of these processes for people with BD (Edge et al., 2013; Gilbert et al., 2013; Gruber et al., 2011; Johnson et al., 2008). We found

additional support for this original 3-factor, as the model with three factors (i.e. self-focused and emotion-focused positive rumination were modelled as separate factors) showed a better fit compared to a two-factor structure (i.e. self-focused and emotion-focused positive rumination were modelled as one single factor). This is in line with previous psychometric evaluations (Feldman et al., 2008; Raes et al., 2010; Yang & Guo, 2014), yet there is an ongoing debate about the actual factor structure of the RPA (Nelis et al., 2016). However, support for the three-factor structure of the RPA was also found in the RCT in *chapter 8* of this thesis. We found that the positive psychology intervention did improve emotion-focused positive rumination, while self-focused positive rumination remained largely unchanged. This suggests that these two factors indeed represent relatively independent dimensions of positive rumination, at least in the BD patients selected for our RCT.

Assessing response styles to positive affect using the RPA in patients with BD might also be relevant to foster the recovery process, since improving these positive emotion regulation strategies might help to increase recovery in BD. Research suggests that dampening might be a particularly maladaptive strategy in BD (Edge et al., 2013; Gilbert et al., 2013) and may represent a potential barrier for personal recovery (Mansell, 2016). In addition, there are indications that higher positive rumination is associated with higher lifetime frequency of mania (Gruber et al., 2011) and lifetime diagnoses of mania or hypomania (Johnson et al., 2008). In this context, especially extreme appraisals of positive emotions are linked to worsened illness course in BD (Ford et al., 2015). Yet, positive rumination is also associated with increased positive emotions (Quoidbach et al., 2010) and positive emotions, in turn, have been found to be associated with improved functioning in BD (Johnson et al., 2016). This suggests that positive rumination, depending on the context and level of rumination, might also be an adaptive emotion regulation strategy in BD that might be associated with increased functioning. Therefore, positive rumination may also have the potential to contribute to mental well-being and personal recovery. The study conducted in *chapter 6* provides additional support for the important contribution of positive rumination to personal recovery. In this study, we found that emotion-focused positive rumination was uniquely associated with personal recovery in BD, independent of symptomatology and social role participation. Clinicians aiming to improve mental well-being and personal recovery in BD could use the RPA to determine levels of positive emotion regulation in their patients and try to specifically enhance positive rumination in patients scoring low in these strategies. Exercises from positive psychology, including the best-possible-self exercise (Sheldon & Lyubomirsky, 2006) or savoring strategies such as capitalizing (Langston, 1994) and mental time travel (Quoidbach et al., 2009) might represent promising interventions to enhance the experience of positive emotions and foster positive rumination (Quoidbach et al., 2010). For researchers, the RPA



could be a valuable addition for intervention studies to assess the effect of psychological interventions in improving responses to positive affect.

## **Social participation strongly contributes to personal recovery**

The study presented in *chapter 6* widens our knowledge of what contributes to personal recovery in BD. Although some previous studies already examined factors related to personal recovery in BD (Dodd et al., 2017; Mezes et al., 2020), studies examining what factors contributed to personal recovery specifically in BD remain relatively scarce. Prior studies also did not explicitly examine the role of social participation and positive emotion regulation. Our findings showed that anxiety symptoms, social participation and emotion-focused positive rumination were uniquely related with personal recovery. The finding that these processes were independently related with personal recovery shows that not only the absence of symptoms should be taken into account when aiming for recovery, but that also processes related to social functioning and responses to positive emotion should be improved, as they seem to uniquely contribute to personal recovery.

Social participation was the strongest predictor in our final model in the study presented in *chapter 6*. This finding highlights the unique contribution of social functioning to personal recovery. Nonetheless, social participation can still be seen as relatively distinct from personal recovery, and might rather be seen as being part of functional recovery, as it entails the ability to work, to function in relationships or to follow educational activities (Oude Voshaar et al., 2016). The ability to function in these social roles might be one important prerequisite for personal recovery, as relationships, work or education might pave the way towards a more joyful, meaningful and engaged life. The assumption that social participation is still relatively independent from personal recovery, means that we might also need different interventions to improve social participation than we would need to improve personal recovery. Interventions that might be effective to improve social role functioning are cognitive therapy (Lam et al., 2005) or collaborative care (van der Voort et al., 2015). In sum, we still know relatively little about what contributes to personal recovery in BD. Future research might want to further examine factors contributing to personal recovery. This could also be combined with idiographic or experience sampling research to examine within-person processes and examine what influences recovery in which context. It might for example be interesting to examine resources for resilience such as emotion regulation, self-compassion or gratitude in daily life and how they contribute to the regulation of mental health in BD.

## Effectiveness and added value of Living well with bipolar disorder

The finding from our RCT in *chapter 8* that the positive psychology treatment was effective in improving mental well-being and personal recovery in BD provides strong evidence for the effectiveness of the positive psychology group intervention. This also shows that mental well-being and personal recovery indeed can be improved in this high-risk population. Previous studies already suggested that mental well-being and personal recovery can be improved in BD (Deckersbach et al., 2012; Eisner et al., 2017; Jones et al., 2015; Todd et al., 2012) and also evaluated the effect of positive psychology interventions for BD, but were not sufficiently powered and/or did not use a control group (Celano et al., 2020; Painter et al., 2019). To our knowledge, the current study was the first to show in a full RCT that mental well-being and personal recovery can be improved in patients with BD using a positive psychology treatment.

These findings have implications for current treatment of BD, as the positive psychology treatment might supplement current TAU of BD with a treatment that enhances recovery beyond clinical and functional recovery. Current TAU, also in the Netherlands, primarily consists of pharmacotherapy combined with supportive and psychoeducational interventions and the enhancement of self-management skills for the long-term management of BD. If indicated, this can be supplemented with psychological interventions such as social rhythm therapy, cognitive behavioral therapy or family-focused therapy (Grande et al., 2015; Kupka et al., 2015). The intervention we presented in this dissertation seems to be an effective treatment and could supplement TAU with an intervention that specifically enhances mental well-being and personal recovery. A pragmatic benefit of the intervention is that it is relatively easy to deliver. Therapists that gave the intervention only needed a training of one day, making the intervention relatively easy to learn. Personal feedback we received from patients and therapists in the course of our study was largely positive, and patients and therapists both indicated that they see added value in the intervention. Furthermore, findings from our RCT in *chapter 8* suggest that current TAU does not explicitly enhance mental well-being and personal recovery yet, since well-being and personal recovery remained largely unchanged in the TAU group over the whole study period of six months. In contrast, outcomes of depression and anxiety also decreased similarly in the TAU group. This suggests that current TAU already focuses on the reduction of symptoms, while outcomes related to mental well-being or personal recovery were not impacted as strongly. This underlines the relevance of augmenting current TAU with an intervention specifically aimed at mental well-being and personal recovery. The positive psychology treatment described in this thesis seems to be effective in doing so and might thus provide a valuable addition to current TAU.

## **The importance of improving mental well-being and personal recovery in bipolar disorder**

One might ask the question why we should actually invest in the improvement of mental well-being and personal recovery. Importantly, patients with SMI find outcomes related to personal recovery important (de Vos et al., 2017; Pitt et al., 2007; Zimmerman et al., 2006). From the perspective of patients with BD, improving personal recovery is relevant since patients with BD themselves describe recovery as not only being symptom-free (Todd et al., 2012) and indicate that reduction of symptoms alone does misrepresent treatment success (Mezes et al., 2020). Patients with BD often experience severe consequences because of their disorder, often describing life as disrupted or “derailed” by the illness (Sajatovic et al., 2008). Personal accounts of people living with BD show the severe impact BD has on personal aspects of life (Warwick et al., 2019). These negative consequences include loss of control and autonomy, hope, purpose, social connections or identity (Fernandez et al., 2014; Inder et al., 2008; Jönsson et al., 2008; Mortensen et al., 2015; Warwick et al., 2019). Others describe the fear of relapse (Mansell et al., 2010), as well as the intentional suppression of pleasure to avoid relapse as negative consequences (Russell & Moss, 2013). The positive psychology treatment presented in this thesis could therefore help to alleviate some the negative consequences of the disorders by specifically improving outcomes related to mental well-being personal recovery. In addition, mental well-being has been shown to be longitudinally related with less mental illness (Lamers et al., 2015; Schotanus-Dijkstra et al., 2017) and to increase the likelihood of recovery from mental disorders (Iasiello et al., 2019; Schotanus-Dijkstra et al., 2019). Improving mental well-being could thus also have positive effects on the prevention of mood episodes in BD. Since BD is also associated with high societal burden (Cloutier et al., 2018; Kraiss et al., 2020), improving mental well-being could not only be of importance from a patient-perspective, but also from a healthcare provider and societal perspective if relapse can be prevented, although it remains unclear whether this will actually be the case considering that BD is recurrent and has a strong biological basis (Kato, 2019). The telephone interviews at 12-month follow-up will provide additional information regarding the ability of the intervention to prevent relapse.

The treatment presented in this dissertation might also equip patients with important strategies to better handle adversities they experience. As described earlier, BD is a chronic and recurrent mood disorder that leads to significant impairments in many patients. In the face of these difficult circumstances, living a satisfying and joyful life with limitations caused by the illness (Anthony, 1993) can become a difficult to accomplish goal. In order to successfully regulate mental health, patients are asked to show adaptive responses in the face of adversity, i.e. being resilient (Zautra et al., 2010). There is increasing attention that also

more long-term positive responses to adversity or negative life-events as one dimension of resilience are relevant. These long-term positive responses can for example include self-compassion, optimism, positive relationships, positive emotions or personal strengths (Trompetter et al., 2017; Zautra et al., 2010; Zautra et al., 2005). Positive long-term responses to adversity might be crucial for successful regulation of mental health and specifically of mental well-being (Bohlmeijer & Westerhof, 2020; Zautra et al., 2010). Improving outcomes such as positive emotions and positive relationships with a positive psychology treatment as presented in this dissertation might thus also improve mental resilience in BD patients. These strategies might support patients to lead a meaningful and joyful life, even with the difficult circumstances they may experience.

## IMPLICATIONS

Several implications for research and practice arise from the findings in this thesis. These implications can be summarized as follows. First, although the evidence base of positive psychology interventions for SMI is still small, previous findings are promising and suggest that patients from these high-risk populations indeed do benefit from positive psychology in terms of improvement of mental well-being and psychopathology. The findings from this dissertation suggests that positive psychology interventions represent a promising treatment approach for patients from these groups. Merely two studies investigated the effect of positive psychology for BD. Similarly, the evidence-base regarding the cost-effectiveness of non-pharmacological interventions in BD is small, but suggests that non-pharmacological treatments have the potential to be cost-effective. No evidence-base regarding the cost-effectiveness of recent psychotherapeutic approaches such as positive psychology, ACT or mindfulness-based therapies exists, indicating the need to further examine the potential of such interventions to reduce the societal burden associated with BD.

Second, the QPR is a reliable and valid questionnaire to assess personal recovery in BD and represents a relatively feasible and theory-driven measurement instrument. The QPR is now available in Dutch and can therefore also be used in the Netherlands in practice and research. Practitioners can use the QPR to individualize the recovery process and researchers might use it to assess the effect of interventions to improve personal recovery. The QPR also seems to be sensitive to change in participants with BD, since we were able to detect changes in personal recovery in the RCT described in the *chapter 8*. We also found that the RPA is a reliable and valid instrument to assess response styles to positive affect in people with BD. Practitioners might find it valuable to assess response styles to positive affect in their clients, as these strategies might represent barriers or facilitators for recovery.

Third, positive rumination, social participation and anxiety symptoms are uniquely related with personal recovery in BD and might therefore represent relevant treatment targets when aiming to improve personal recovery. We still know relatively little about what contributes to personal recovery in BD, and future studies might want widen our knowledge of determinants of personal recovery in this group of patients.

Fourth, a specific positive psychology treatment is effective in improving mental well-being and personal recovery in BD. This positive psychology treatment could be a valuable addition to current TAU for patients with BD, since it specifically improves mental well-being and personal recovery. Personal recovery and mental well-being may not only be relevant from the perspective of patients, but might also be relevant from a societal perspective, as improving positive resources associated with mental well-being and personal recovery might improve resilience and could prevent relapse. However, this still needs to be shown by analyzing the 12-month follow-up data including the telephone interviews that are aimed to determine whether patients experienced relapsed.

## **STRENGTHS AND LIMITATIONS**

The current thesis provides a comprehensive overview of the current state of the art of positive psychology interventions for SMI and economic studies for BD. We also validated two relevant measurement instruments in BD, which are now available in Dutch and can be used to assess processes of positive emotion regulation and personal recovery. We also explored factors associated with personal recovery in BD. To our knowledge, this was the first study examining the association of positive emotion regulation and social role participation with personal recovery in BD. Furthermore, in this thesis a novel positive psychology treatment to improve mental well-being and personal recovery was developed and its effectiveness was evaluated in a group of euthymic patients with BD. To our knowledge, this is the first full RCT that examined the effect of positive psychology for patients with BD. The RCT study therefore represents an important step towards a more holistic treatment of BD, also taking into account the improvement of mental well-being and personal recovery.

Three overarching limitations of the studies in this dissertation need to be mentioned. First, all studies were conducted on a group-level, therefore no conclusions can be made about within-person effects of the intervention or relationships between processes and outcomes within individuals. In addition, the studies examining psychometric properties and determinants of personal recovery were based on cross-sectional data, which makes inferences about causality or the longitudinal nature of associations impossible. Specific psychometric properties of the

QPR and RPA could also not be determined, for example sensitivity to change or test-retest reliability.

Second, sample sizes of the empirical studies were relatively small and we did not reach the originally planned sample size for our RCT. Although we were still able to detect significant differences in the primary outcome in the RCT, it is possible that differences in some secondary outcomes were not detected due to low power. Also, the sample in our RCT is likely to be underpowered for analyses that could help to understand for which patient with which characteristics the intervention works or to determine specific working mechanisms of the intervention. The current thesis does therefore not provide an answer to the questions what specific characteristics are associated with increased treatment outcomes or through which pathways the intervention unfolds its effect on mental well-being and personal recovery.

Third, this dissertation does not yet include longer follow-up data on the sustained effectiveness of the positive psychology treatment. Data on the outcome at 12-month follow-up is still being collected. These results will be published at a later moment together with the 6-month outcomes provided in this thesis. Similarly, the telephone interviews with participants assessing relapse are not yet included in this thesis, since they are also conducted at 12-month follow-up. This additional information will provide valuable information on the long-term (cost-)effectiveness of the intervention and its potential to prevent relapse, but is not yet available.

## FUTURE DIRECTIONS

### Implementation in the current treatment landscape

One important question that remained unaddressed so far is at which point in the current treatment landscape the positive psychology intervention presented in this thesis can be implemented. We are currently still conducting interviews with important stakeholders to answer this question, and also to determine potential facilitators and barriers of implementation. In general, as described earlier, we think that *Living well with bipolar disorder* might be a valuable addition to current TAU. The intervention should therefore also not be seen as replacement of treatments that already exist, but as a supplement. We think that it is not appropriate to use this intervention during acute manic or depressive episodes, since mood symptoms will be too severe and impairing and safety of the patient and its environment should always have the first priority. Stabilization of acute mood symptoms should remain the primary goal in this phase of treatment. In the RCT described in *chapter 8* we therefore deliberately only included relatively euthymic BD patients. The positive psychology treatment

might rather be a valuable addition for maintenance treatment, in phases when patients are relatively stable and may show subsyndromal symptoms. The intervention might thus ideally be given in phases in which the symptoms have largely cooled off and the patient is stable for a relatively long time. This may give the patient sufficient flexibility and resources to work on mental well-being. However, functional impairment including the limitations in the ability to work, maintain family relationships or home management can still be high in phases of remission (Fagiolini et al., 2005). It might thus be advisable to first aim to enhance this part of functional recovery before aiming to enhance mental well-being and personal recovery. In the study in *chapter 6* we could also show that social role participation was uniquely associated with personal recovery in BD. This shows the importance of processes such as being able to work and being able to function in relationships, suggesting that functional recovery also significantly contributes to personal recovery. In this context, a certain level of social functioning might represent an important prerequisite for personal recovery, since joy, meaning and engagement are possibly only created by being able to fulfill social roles such as being able to work, having intimate relationships or following educational activities.

If a patient is relatively stable and functionally restored, it might be a suitable moment to follow the positive psychology treatment to enhance mental well-being and personal recovery. However, recovery often is an iterative and non-linear process (Leamy et al., 2011). The course of recovery assumed here (i.e. first symptomatic, second functional recovery and third personal recovery), therefore probably often does not work in practice and patients might often bounce backwards and forwards in their recovery process. This also means that the intervention might be given more than once if patients experience setbacks in their recovery process. This might be especially true in patients with BD, considering the recurrent course of the disorder. Importantly, the decision on whether a patient should take part in the positive psychology treatment should always be made on an individual basis and should always be based on the needs of the patient. Some patients actually indicated in the course of our study that they were already further in their recovery and stopped to participate in the course. In this context, utilizing the MHC-SF or QPR to assess levels of mental well-being and personal recovery might also help to determine whether a patient actually has an explicit need for the positive psychology treatment.

## **Bringing back the individual into psychological research**

One issue that remained unaddressed in this dissertation is the question in how far the constructs we measured vary and interact on a within-person level. In other words, all studies presented in this dissertation were conducted on a between-person or group-level, while within-person variability was not explicitly assessed. While analyses on a between-person level

determine what happens across a set of individuals or a group of people, within-person analyses examine what happens within individuals. Between-person analyses would, for example, examine the question whether people with higher levels of well-being also show higher levels of depression on average (Curran & Bauer, 2011). Within-person analyses would examine whether a person who reports higher well-being than his or her own average also reports lower depression at the same time point. In other words, between-person analyses compare means of individuals, while within-person analyses examine fluctuations around means of individuals (Hamaker et al., 2007). Interestingly, psychological theories often aim to make inferences about processes that happen within individuals, but they often conduct analyses on a between-person or group-level. Often, this is done using cross-sectional data (Curran & Bauer, 2011). However, processes or relationship between constructs do not necessarily function similarly on these two levels of analyses. To generalize processes from a between-person to a within-person level, constructs need to be similar across individuals and stable over time (Molenaar, 2004). It is very unlikely though that processes observed in psychological research fulfill these criteria (Johnston & Johnston, 2013; Molenaar, 2004). Therefore, inferences made about relationships and changes in individuals can hardly ever be generalized from the between-person to the within-person level. In fact, it is even highly unlikely that the average person found on a between-person level even exist at all, since the average person found on a between-person level merely represents an aggregate of individual means. The false assumption that processes are the same on both levels of analyses is described as ecological fallacy (Piantadosi et al., 1988).

Of course, this does not mean that group-level research should be discarded. It should still be acknowledged for its potential to gain an overall understanding of the effectiveness of interventions or relationships between constructs. However, if we want to increase our understanding of how processes unfold within individuals, future research should supplement group-based research with idiographic studies to understand processes within individuals. This would also help to individualize care offered to patients. Experience sampling might be particularly suitable for this (Larson & Csikszentmihalyi, 2014) and promising research designs for this purpose might be N-of-1 trials (Lillie et al., 2011). This could also have implications for how we conduct research for patients with BD in the future. Only utilizing knowledge that is aggregated across a set of BD patients to provide mental health care does not account for highly individual differences in patients and their individual context. In fact, this thought also aligns well with the overall concept of recovery from mental illness, since recovery is consistently described as individual process (Leamy et al., 2011). While some factors might be related to personal recovery for some patients in some situations, they might not contribute to personal recovery for other patients in other situations. Assessing processes within persons would help us to understand how processes change within individuals and what influences



these changes. It would also help us to determine the temporal order of relationships and whether certain processes occur at the same time. Resources of mental resilience could for example be examined and what role they play in the regulation of mental health. This would refine our understanding of psychological processes as they unfold within individuals and might pave the way towards more personalized mental health models and interventions. Experience sampling or N-of-1 trials would then ideally be combined with group-level RCTs to determine individual trajectories of change or to study individual change processes.

## **Data science approaches in mental health research**

Data science approaches might play an increasingly important role in mental health research due to the increasing use of big data and accumulation of data, for example through routine outcome monitoring. The data analysis approach we presented in *chapter 6* of this thesis also contained methods derived from data science. We split our sample into a training and test sample and used model training to build a parsimonious model. The fit of the parsimonious model was evaluated using k-fold cross-validation. This helped us to evaluate how robust our model is and to determine how well it generalizes to an independent data set before even testing the model on the independent test set. This also helps to avoid overfitting a model (Hastie et al., 2009). All these techniques are not part of the traditional statistical repertoire social scientists use to analyse their data, yet these techniques are likely to become more and more important in the future. The increasing use of big data for prediction calls for techniques that can be used to analyze these data sets, also in mental health research. For example, the use of technology and experience sampling methods in mental health research to assess daily behavior and mood is becoming increasingly popular. This data may also include passive data such as GPS or sensor data. This can lead to fairly complex data sets, containing many variables and assessment points. Separating signal from noise and avoid overfitting will become increasingly important in order to make robust predictions that generalize to other data sets and other settings.

One project we are currently working on is the use of different supervised machine learning techniques to predict favorable treatment outcomes in patients with mental disorders based on routine outcome monitoring data. This real-world data is routinely collected in all mental health care centers in the Netherlands and might represent a valuable source of information, since this data represent real-world naturalistic data from many patients. However, these data sets are also fairly complex and cluttered, making it potentially difficult for traditional statistical techniques to build robust models based that are also generalizable. Data science approaches might support us in using this data. Another application of data science techniques in mental health research might be to use data mining to detect patterns in unstructured data

sets such as counselling transcripts to derive relevant information from this data. Although the study presented in *chapter 6* of this dissertation cannot be considered an example of big data, it is still a good example that data science can be of added value for mental health research (Imel et al., 2015). To bridge mental health research with big data in the future, data science approaches might play an increasingly important role.

## Personalizing treatment

Similar to the importance of examining within-person variability and processes within individuals, another important direction for the future is to develop personalized mental health care solutions (Ng & Weisz, 2016). Data science techniques, as described before, might again play an important role to determine what works best for whom. Machine learning techniques align well with the increasing importance of tailoring psychological interventions for individuals. They can help to examine which interventions offer the optimal benefit for which patient, by extracting information on what patient characteristics are associated with favorable outcomes in psychotherapy (Aafjes-van Doorn et al., 2021). Determining individual process-outcome associations in psychotherapy research are one example, and might help to indicate which particular processes in psychotherapy are associated with improvement for which patient (Rubel et al., 2020). Identifying specific subgroups of patients that poorly respond to current treatments and find alternative ways to improve mental health outcomes in these groups might be another way to increase personalization (Bernal et al., 2009). This also relates to our findings in our RCT in *chapter 8*, in which we found that the intervention was effective in improving mental well-being and personal on a group-level, but there were also patients with BD in the intervention group that did not benefit. Identifying their characteristics, including the stages of a disorder and individual profiles, and finding alternative solutions for these groups might also enhance personalization of psychological interventions. Another promising approach to tailor psychological interventions might be individualized metrics (DeRubeis et al., 2014). Personalized advantage indices might also help to determine patient characteristics that are associated with optimal treatment outcomes of a specific intervention, albeit this requires the use of head-to-head RCTs comparing two active interventions. This could help to provide individualized advice regarding which treatment should be given to which individual, for example CBT or psychopharmacotherapy (DeRubeis et al., 2014) or CBT or psychodynamic therapy (Cohen et al., 2020).

Combining experience sampling with mobile technologies and data science techniques might also represent a promising approach to develop adaptive personalized and timed interventions and bring therapy into the daily life of patients (Clough & Casey, 2015). An experience sampling application could collect data in the daily life of patients. This technology could be

programmed to deliver just-in-time interventions in moments when patients are indicating that they might be struggling. The collected data could also be harnessed to build models that predict certain behavior before this behavior even occurs and deliver timed interventions to prevent certain behavior. This could also be combined with the collection of passive data such as GPS or sensor data, which might increase the accuracy of the models and decrease the burden of participants. Data science techniques of reinforcement learning might even hold bigger potential for the personalization of psychological interventions, as they might be able to learn based on individual input what specific interventions are effective for a specific person in a specific state (Kelly et al., 2012). These personalized and timed interventions might also support people in their mental resilience, as they can offer support to regulate mental health in daily life and generalize psychological treatment outside the treatment chamber. These applications may continuously support people in regulating their mental health and might revolutionize the way we provide mental health care in the future.

## **OVERALL CONCLUSION**

The objective of this thesis was to contribute to the integration of positive psychology and personal recovery into the treatment of BD. We could show in a full RCT, that the positive psychology group treatment developed and evaluated in this thesis represents an effective treatment to enhance mental well-being and personal recovery in euthymic patients with BD. This intervention may therefore represent a valuable addition for TAU for BD, since it complements current treatment with an intervention that contributes to live a joyful, meaningful and engaged life, also in the presence of a severe mental illness. Furthermore, we found that a Dutch translation of the QPR represents a reliable and valid instrument to measure personal recovery in BD. The QPR is now available to be used in the Netherlands. We could also show that the RPA is a psychometrically sound instrument to assess positive emotion regulation in BD. The RPA can be used to assess positive emotion regulation strategies in BD, which may represent important facilitators or barriers for recovery. Furthermore, we found that positive rumination, anxiety symptoms and social participation are independently associated with personal recovery in BD and might therefore represent valuable treatment targets when aiming to improve personal recovery. Possible future directions for practice and research include the implementation of this intervention in the current treatment landscape, the integration of idiographic research and data science approaches in mental health research and the development of personalized treatments to advance the care we provide for patients with mental disorders.

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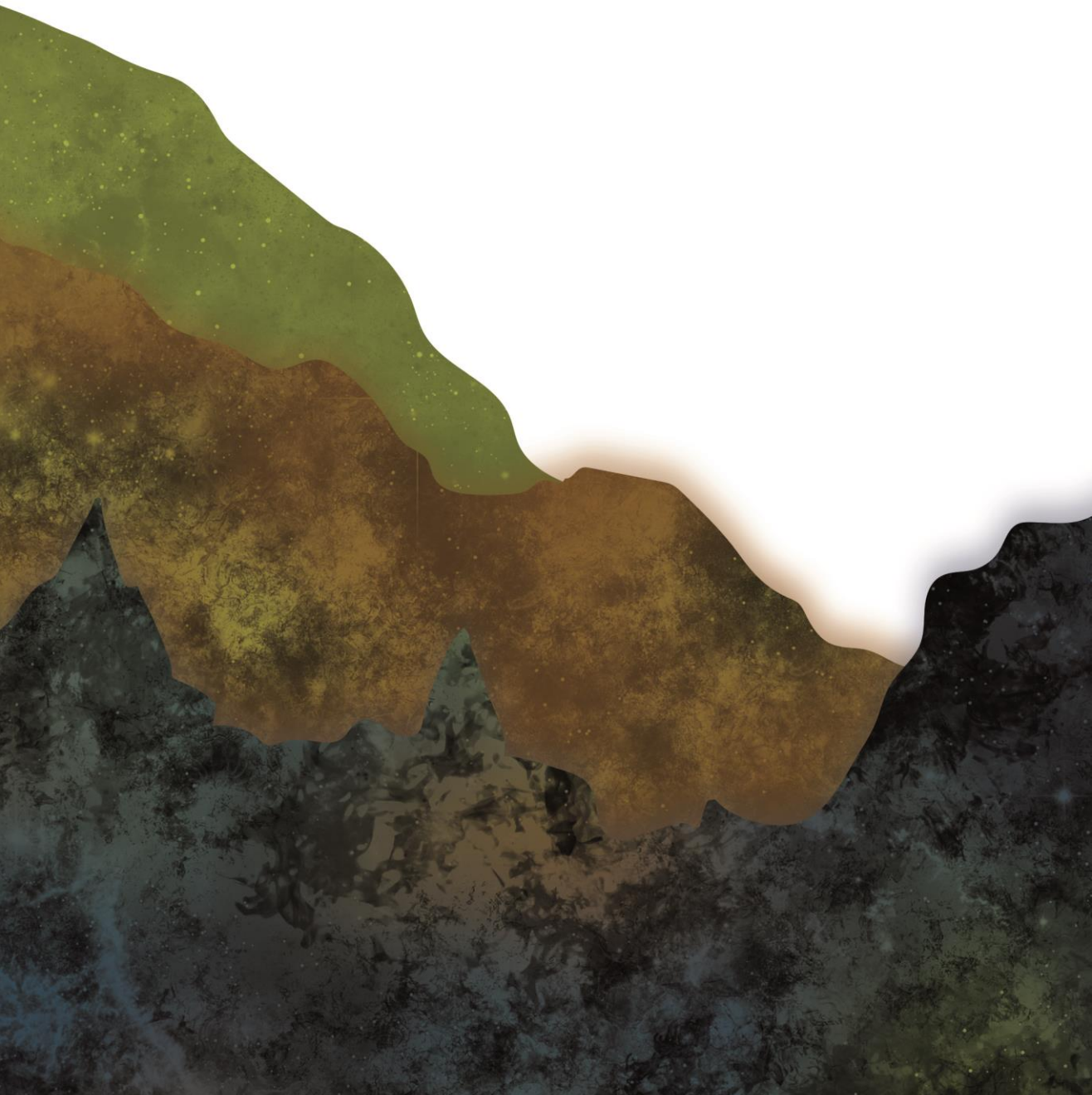
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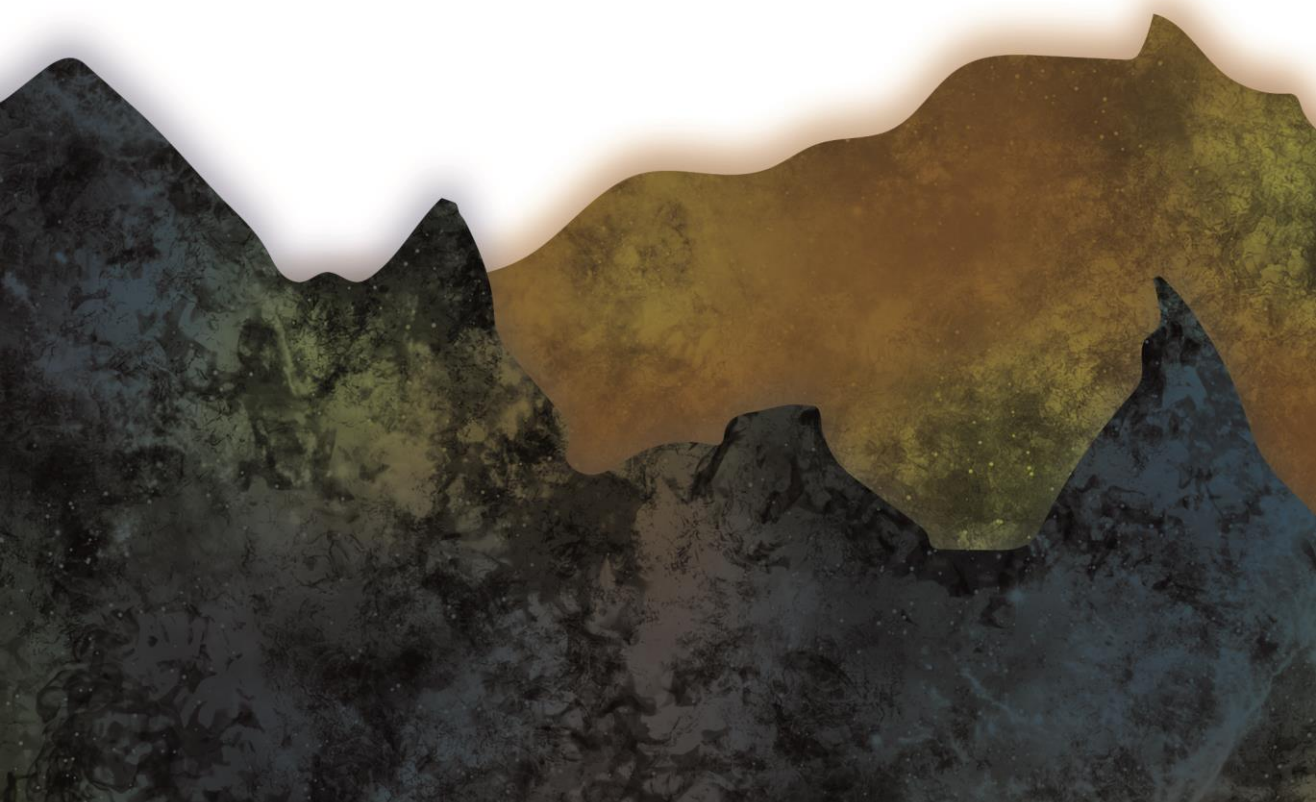


# Appendices

Dutch summary

Acknowledgements

About the author





## DUTCH SUMMARY

Bipolaire stoornissen (BS) zijn recidiverende stemmingsstoornissen die gepaard kunnen gaan met substantiële negatieve gevolgen voor het individu en met hoge maatschappelijke lasten. De huidige behandelrichtlijnen voor BS zijn primair gericht op symptomatisch en functioneel herstel. Behandelresultaten gerelateerd aan mentaal welbevinden en persoonlijk herstel worden echter steeds vaker gezien als een belangrijk onderdeel van het herstel van psychische stoornissen, ook bij patiënten met BS. Mentaal welbevinden en psychopathologie kunnen worden gezien als gerelateerde, maar verschillende dimensies van geestelijke gezondheid. Mentaal welbevinden omvat de aanwezigheid van emotioneel, sociaal en psychologisch welbevinden. Persoonlijk herstel kan worden beschreven aan de hand van vijf componenten, namelijk verbondenheid, hoop, identiteit, betekenis en empowerment. Een gebied van de psychologie dat zich specifiek richt op de verbetering van mentaal welbevinden en persoonlijk herstel, is de positieve psychologie. Patiënten met BS beschouwen uitkomsten met betrekking tot mentaal welbevinden en persoonlijk herstel als belangrijk en studies laten zien dat zowel symptomen als ook mentaal welbevinden verbeterd zouden moeten worden bij mensen met psychische stoornissen wanneer het doel is algehele mentale gezondheid te bevorderen. Ondanks het belang van mentaal welbevinden en persoonlijk herstel voor BS, zijn studies naar het effect van (positieve psychologie) interventies gericht op persoonlijk herstel en mentaal welbevinden bij BS schaars.

Dit proefschrift beoogt een bijdrage te leveren aan de integratie van positieve psychologie en persoonlijk herstel in de behandeling van BS. Vier doelen zijn hiervoor gesteld in dit proefschrift. Ten eerste is een overzicht gegeven van het onderzoeksgebied van positieve psychologische interventies voor ernstige psychische aandoeningen (EPA) en van economische studies voor BS. Hierdoor is de huidige stand van zaken van positieve psychologische interventies voor EPA in het algemeen en de kosteneffectiviteit van niet-farmacologische interventies voor BS in het bijzonder samengevat. Dit is gedaan door middel van twee systematische reviews van de literatuur, waarvan één studie ook gebruik maakte van meta-analysetechnieken. Ten tweede zijn twee instrumenten psychometrisch gevalideerd die persoonlijk herstel en positieve emotie-regulatie strategieën meten, namelijk de Questionnaire about the Process of Recovery (QPR) en de Responses to Positive Affect questionnaire (RPA). Deze constructen zijn relevant voor BS, maar de QPR en RPA zijn niet eerder in het Nederlands vertaald en/of gevalideerd in mensen met BS. Ten derde onderzoekt dit proefschrift determinanten van persoonlijk herstel bij BS, door factoren te bepalen die samenhangen met persoonlijk herstel. Ten vierde is er een nieuwe groepsbehandeling op basis van de positieve psychologie ontwikkeld die gericht is op het verbeteren van mentaal welbevinden en persoonlijk herstel bij euthyme patiënten met BS. De effectiviteit van deze

behandeling is onderzocht in een pragmatische gerandomiseerde gecontroleerde studie. Daarmee overeenkomend zijn de volgende vier doelen behandeld in dit proefschrift:

- (1) Een overzicht geven van het onderzoeksveld met betrekking tot positieve psychologische interventies voor patiënten met EPA en economische studies bij BS.
- (2) Het onderzoeken van de psychometrische eigenschappen van een Nederlandse vertaling van de Questionnaire about the Process of Recovery (QPR) en de Responses to Positive Affect questionnaire (RPA) bij mensen met BS.
- (3) Het onderzoeken van determinanten van persoonlijk herstel bij BS.
- (4) Het ontwikkelen van een positieve psychologische interventie voor euthyme patiënten met BS gericht op mentaal welbevinden en persoonlijk herstel en het evalueren van de effectiviteit van deze interventie in een pragmatische, multicenter gerandomiseerde gecontroleerde studie.

Deze vier doelen komen aan de orde in de volgende vier delen van dit proefschrift: (I) overzicht van het onderzoeksveld, (II) meten van relevante constructen, (III) determinanten van persoonlijk herstel, en (IV) interventie-ontwikkeling en evaluatie.

## **Deel 1: Overzicht van het onderzoeksveld**

In *Hoofdstuk 2* van dit proefschrift gaven we een overzicht van het onderzoeksveld met betrekking tot positieve psychologie interventies voor mensen met EPA. De effecten van deze interventies zijn samengevat met behulp van meta-analyse technieken. EPA kan worden gedefinieerd als een psychiatrische stoornis met ernstige functionele problemen, waarbij de beperkingen oorzakelijk en consequent zijn en niet tijdelijk van aard, en waarbij behoefte is aan gecoördineerde professionele zorg. Voorbeelden van EPA zijn schizofrenie, schizoaffectieve stoornis, persoonlijkheidsstoornis, BS en depressieve stoornis. We identificeerden 16 studies in onze review ( $N = 729$ ), waarvan negen studies gerandomiseerde gecontroleerde studies waren. De meta-analyses lieten niet-significante effecten tussen groepen zien voor uitkomsten gerelateerd aan welbevinden ( $g = 0.16$ ) en psychopathologie ( $g = -0.10$ ). Effectgroottes binnen groepen (d.w.z. verandering in mentaal welbevinden of psychologische symptomen binnen de interventiegroepen) waren significant voor uitkomsten van mentaal welbevinden ( $g = 0.40$ ) en psychopathologie ( $g = -0.70$ ). De bevindingen suggereren dat we op basis van de huidige studies niet kunnen concluderen dat positieve psychologische interventies effectief zijn in vergelijking met andere actieve interventies of TAU, maar dat mensen met EPA wel lijken te profiteren van positief psychologische interventies in termen van verbetering van welbevinden en psychologische symptomen.

In *Hoofdstuk 3* presenteerden we een systematische review van economische evaluaties en ziektekostenstudies bij BS. Het doel van deze review was om economische evaluaties van niet-farmacologische interventies in BS en ziektekostenonderzoeken in BS die sinds 2012 zijn gepubliceerd samen te vatten. We identificeerden acht economische evaluaties en tien ziektekostenstudies. Dit laat zien dat de evidentie op het gebied van economische evaluaties van niet-farmacologische interventies vrij klein is. We vonden ook een hoge heterogeniteit in methodologische kenmerken en uitkomsten, waardoor het moeilijk was om studies te vergelijken en de bevindingen te generaliseren naar specifieke groepen of interventies. Alle niet-farmacologische interventies konden de klinische uitkomsten met betrekking tot BS verbeteren, en bovendien verlaagden vijf interventies de kosten die verbonden zijn met BS. Op basis van de ziektekostenstudies konden wij concluderen dat BS geassocieerd is met substantiële directe en indirecte kosten, maar het was ook te zien dat de geschatte kosten sterk varieerden tussen studies. In onderzoeken waarin directe en indirecte kosten werden beoordeeld, waren indirecte kosten (bijv. productiviteitsverlies of werkloosheid) de belangrijkste kostenfactoren. De bevindingen van deze review tonen aan dat niet-farmacologische interventies niet alleen de klinische resultaten bij BS kunnen verbeteren, maar mogelijk ook kosteneffectieve behandelingsalternatieven zijn vergeleken met TAU of met andere actieve controlecondities. De bevindingen van deze studie ondersteunen ook de hoge kosten waarmee BS verbonden is. Dit laat het belang zien van kosteneffectieve behandelingsalternatieven voor deze patiëntengroep.

## **Deel 2: Meten van relevante constructen**

In *hoofdstuk 4 en 5* hebben we twee psychometrische evaluaties uitgevoerd van een Nederlandse vertaling van de QPR en RPA in een gelegenheidssteekproef van mensen met BS. Het evalueren van psychometrische eigenschappen van meetinstrumenten is relevant om te bepalen of het instrument consistent het construct meet dat het beoogt te meten. Ondanks de relevantie van persoonlijk herstel en positieve emotieregulatie bij BS zijn psychometrische evaluaties van mogelijke meetinstrumenten hiervoor in BS schaars. De QPR is eerder gevalideerd en vanuit het Engels in verschillende talen vertaald. De studie in *hoofdstuk 4* was de eerste die de QPR in het Nederlands heeft vertaald en de psychometrische eigenschappen bij mensen met BS heeft geëvalueerd. We vonden uitstekende interne consistentie voor de QPR en hoge correlaties met alle validatie instrumenten, waaronder mentaal welbevinden, sociale participatie en uitkomsten van psychopathologie. Dit ondersteunt de convergente validiteit van de QPR. De resultaten laten ook zien dat de QPR incrementele validiteit heeft, omdat persoonlijk herstel gemeten met de QPR variantie in psychopathologie verklaarde bovenop mentaal welbevinden. Dit suggereert ook dat het meten van persoonlijk herstel naast mentaal welbevinden van toegevoegde waarde is. Het feit dat een Nederlandse vertaling van

de QPR veelbelovende resultaten liet zien in het onderzoek in *hoofdstuk 4*, betekent ook dat er nu een gevalideerde Nederlandse versie van de QPR beschikbaar is die kan worden gebruikt om persoonlijk herstel bij patiënten met BS in Nederland te meten.

In *Hoofdstuk 5* van dit proefschrift werd een psychometrische evaluatie van de Nederlandse RPA in mensen met BS gepresenteerd. De RPA meet drie verschillende strategieën van positieve emotieregulatie, namelijk *dempen* en emotiegerichte en zelfgerichte *positieve ruminatie*. Dempen wordt beschreven als het onderdrukken van positieve emoties om de intensiteit van een positieve stemming te verminderen. Positieve ruminatie kan worden gedefinieerd als de neiging om op positieve affectieve toestanden te reageren met terugkerende gedachten over positieve ervaringen of gevoelens. Hoewel eerdere studies de RPA al hebben gevalideerd of de RPA al naar het Nederlands hebben vertaald, gebruikten eerdere onderzoeken uitsluitend steekproeven uit de algemene bevolking of studenten. De studie in *hoofdstuk 5* was de eerste die de psychometrische eigenschappen van de RPA bij mensen met BS heeft onderzocht. Onze bevindingen laten zien dat de subschalen voldoende tot goede interne consistentie hadden en dat zowel dempen als positieve ruminatie correleerden met validatie instrumenten, wat duidt op constructvaliditeit van de RPA subschalen. Wij concludeerden ook dat de RPA incrementele validiteit liet zien, aangezien de RPA variantie in persoonlijk herstel verklaarde bovenop sociale rolparticipatie, als ook bovenop symptomen van depressie of angst. Samengevat laten de resultaten uit *hoofdstuk 4 en 5* zien dat de QPR en de RPA betrouwbare en valide instrumenten zijn om persoonlijk herstel en positieve emotieregulatie in BS te meten.

### **Deel 3: Determinanten van persoonlijk herstel**

Onderzoek naar factoren die bijdragen aan persoonlijk herstel is relatief schaars. Hoewel sommige eerdere studies determinanten van persoonlijk herstel hebben onderzocht, heeft geen van de eerdere studies specifiek de bijdrage van sociale rolparticipatie en positieve emotieregulatie bij BS in kaart gebracht. Daarom hebben wij in *hoofdstuk 6* van dit proefschrift gegevens uit het vragenlijstonderzoek (*hoofdstuk 4 en 5*) gecombineerd met baseline gegevens van de gecontroleerde gerandomiseerde studie (*hoofdstuk 8*) om factoren te onderzoeken die gerelateerd zijn aan persoonlijk herstel bij BS. De steekproef werd opgesplitst in een training en test dataset. Vervolgens hebben wij met behulp van bloksgewijze meervoudige regressies een reeks sociodemografische, sociale en psychologische factoren als mogelijke voorspellers van persoonlijk herstel onderzocht. Daarna werd een spaarzaam model ontwikkeld met behulp van modeltraining en kruisvalidatie. Het uiteindelijke spaarzame model bevatte de vier variabelen tevredenheid met sociale rollen, angstsymptomen, manische symptomen en emotiegerichte positieve ruminatie en verklaarde 57.3% van de variantie in persoonlijk herstel

(gecorrigeerde  $R^2 = .56$ ). Het spaarzame model werd vervolgens toegepast in de test dataset om het vermogen van het model te evalueren om persoonlijk herstel in een onafhankelijke steekproef te voorspellen. Het model presteerde vergelijkbaar goed bij het voorspellen van persoonlijk herstel in de test dataset (gecorrigeerde  $R^2 = .49$ ). De bevindingen ondersteunen het belang van sociale participatie, psychologische symptomen en positieve ruminatie als onafhankelijke correlaten van persoonlijk herstel bij BS. Dit suggereert dat deze factoren ook relevante behandeldoelen zouden kunnen zijn als het doel is persoonlijk herstel te bevorderen.

## Deel 4: interventie-ontwikkeling en evaluatie

Zoals eerder beschreven, zijn studies die het effect van specifieke (positieve psychologie) interventies gericht op mentaal welbevinden en persoonlijk herstel bij BS schaars. Daarom presenteren we in *hoofdstuk 7* de ontwikkeling van een nieuwe positieve psychologische groepsbehandeling voor euthyme patiënten met BS. Ook is de onderzoeksopzet en procedure voor de gerandomiseerd gecontroleerde trial beschreven dat in *hoofdstuk 8* is gepresenteerd. De interventie *Goed leven met een bipolaire stoornis* is gebaseerd op het zelfhulpboek *Dit is jouw leven*. De stappen die zijn genomen om de oorspronkelijke interventie aan te passen voor euthyme patiënten met BS werden beschreven. Verder werd beschreven hoe een eerste formatieve gebruikersevaluatie bij een kleine groep patiënten is doorgevoerd, en hoe de resultaten van deze evaluatie zijn gebruikt om de interventie verder aan te passen. De uiteindelijke interventie *Goed leven met een bipolaire stoornis* bestaat uit acht groepsessies met acht modules, bijvoorbeeld (zelf)compassie, positieve emoties, optimisme en persoonlijke sterktes. De interventie wordt gegeven door getrainde behandelaren. Eén sessie duurt 2 uur en elke sessie wordt aangevuld met thuiswerk opdrachten.

In hoofdstuk 8 presenteren we de resultaten van een pragmatische RCT waarin we het effect van de positieve psychologie behandeling evalueerden. Het effect van de interventie als aanvulling op gebruikelijke zorg (TAU) om mentaal welbevinden en persoonlijk herstel te bevorderen werd vergeleken met alleen TAU. De primaire uitkomstmaat was mentaal welbevinden gemeten met de Mental Health Continuum-Short Form. Persoonlijk herstel, sociale participatie, symptomatologie en verschillende positief psychologische processen werden ook gemeten. We includeerden 97 euthymische patiënten met BS in het onderzoek, die vervolgens gerandomiseerd zijn in de positieve psychologie groep ( $n = 54$ ) of de controlegroep ( $n = 43$ ). Participanten werden gevolgd tot zes maanden na baseline. Significante matige tot grote verschillen tussen de groepen op de posttest werden gevonden voor mentaal welbevinden ( $d = 0.79$ ) en persoonlijk herstel ( $d = 0.83$ ). De effecten op mentaal welbevinden hielden aan tot 6 maanden na baseline. Significante effecten tussen de groepen bij de posttest werden ook gevonden voor emotiegerichte positieve ruminatie en een



significant interactie-effect werd gevonden voor positieve emoties. Verschillen tussen groepen voor uitkomsten van depressie, angst en (hypo)manie waren niet significant. De bevindingen tonen aan dat een 8-weekse positieve psychologie groepsbehandeling effectief is in het verbeteren van het mentaal welbevinden. Dit laat zien dat positieve psychologie kan bijdragen aan het leiden van een vreugdevol en zinvol leven bij patiënten met BS, ook in de aanwezigheid van een ernstige psychische aandoening.

## Implicaties

Een aantal belangrijke implicaties komen voort uit de studies in dit proefschrift. Deze kunnen als volgt worden samengevat: (1) Hoewel de evidentie van positief psychologische interventies voor EPA nog klein is, zijn eerdere bevindingen veelbelovend. De resultaten laten zien dat patiënten uit deze populaties inderdaad baat hebben bij positieve psychologie in termen van verbetering van mentaal welbevinden en psychopathologie. Positieve psychologie lijkt dus een veelbelovende behandelaanpak te zijn voor patiënten uit deze groepen. Slechts twee eerdere kleine studies hebben het effect van positieve psychologie specifiek bij BS onderzocht. Hoewel de evidentie met betrekking tot de kosteneffectiviteit van niet-farmacologische interventies bij BS vrij klein is, laten huidige bevindingen zien dat niet-farmacologische interventies potentieel kosteneffectief zijn. Er zijn geen studies beschikbaar die de kosteneffectiviteit van nieuwe psychotherapeutische benaderingen hebben onderzocht, zoals positieve psychologie, ACT of op mindfulness gebaseerde therapieën. Dit onderstreept het belang om de kosteneffectiviteit van dergelijke interventies te onderzoeken. (2) De QPR blijkt een betrouwbare en valide vragenlijst om persoonlijk herstel bij BS te meten. Verder is de QPR een relatief eenvoudig bruikbaar en theorie-gedreven meetinstrument. De QPR is nu beschikbaar in het Nederlands en kan dus door behandelaren of onderzoekers gebruikt worden om persoonlijk herstel te meten. Behandelaren kunnen de QPR gebruiken om het herstelproces te individualiseren en onderzoekers kunnen het instrument gebruiken om het effect van interventies ter verbetering van persoonlijk herstel te beoordelen. De QPR lijkt ook gevoelig te zijn voor veranderingen bij patiënten met BS, aangezien we veranderingen in persoonlijk herstel konden detecteren in de studie beschreven in *hoofdstuk 8*. Verder hebben wij gevonden dat de RPA een betrouwbaar en valide meetinstrument lijkt om positieve emotieregulatie te meten bij mensen met BS. Behandelaren vinden het mogelijk een waardevolle aanvulling om reactiestijlen op positief affect te meten bij hun cliënten, aangezien deze strategieën belemmerende of bevorderende factoren voor herstel in BS kunnen zijn. (3) Sociale participatie, positieve ruminatie en angstsymptomen zijn onafhankelijk gerelateerd aan persoonlijk herstel bij BS. Deze uitkomsten kunnen daarom relevante behandeldoelen zijn als het doel is persoonlijk herstel te bevorderen. (4) Een specifieke nieuwe positieve psychologie groepsbehandeling is effectief om mentaal welbevinden en persoonlijk herstel te

bevorderen in euthyme patiënten met BS. De positieve psychologie behandeling kan een waardevolle aanvulling zijn op de huidige TAU voor patiënten met BS, aangezien de interventie specifiek het mentaal welbevinden en persoonlijk herstel verbetert.

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## ABOUT THE AUTHOR

Jannis Kraiss was born in Bielefeld, Germany, on the 4th of December 1991. He graduated from high school in 2011. In the same year, he moved to the Netherlands and started to study



psychology at the University of Twente. He finished the bachelor program in 2014, and completed his master in Positive Psychology & Technology in 2016. During his master, he also worked as student assistant for the department of Psychology, Health and Technology. After one year as research fellow at the University of Münster, Germany, Jannis began his PhD research on positive psychology and personal recovery for people with bipolar disorders at the department of Psychology, Health and

Technology at the University of Twente in 2017. He also supported the department in various educational tasks, including the supervision of students and coordination of the course Applied Positive Psychology. He also gave multiple lectures for the psychology master program. In 2019, he organized the track Health & Happiness for the summer school CuriousU in Enschede. Jannis was also part of the PhDs for PhDs initiative at the Faculty of Behavioral, Management and Social sciences. Here he helped to organize social events and to improve the experience of PhDs studying at the faculty. Early 2021, he started a postdoc position at the University Medical Center Radboud in Nijmegen. In September 2021, Jannis was appointed an assistant professor position at the department of Psychology, Health and Technology at the University of Twente. In this role, he will be able to combine his passion for teaching and research, and aims to initiate projects that bridge innovative research methods and data analyses with clinical psychology and mental health research.