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Evaluation of Family- Focused-Cognitive- Behavioral Therapy ”RAINBOW” for Pediatric Bipolar Disorders Adapted to Adolescents in Sweden: A Case Series

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Abstract

The RAINBOW program, a Child and Family-Focused Cognitive Behavioral Therapy program (CFF-CBT) for Pediatric Bipolar Disorder (PBD) designed for children aged 8-12 years and their parents, in a parallel group format, has been adapted to adolescents aged 14-17. The aim of this study was to investigate if CFF-CBT is effective and feasible for adolescents and their parents in Sweden**.** *Method:* The sample of seven adolescents with PBD and eleven parents participated in the Rainbow-program consisting of 12 sessions over a three month period. The adolescents and parents were assessed pre- and post treatment with focus on symptoms of mania and depression, psychosocial functioning, expressed emotion and increased knowledge and skills to cope with the disorder. For the adolescents a case series design was used and clinical significance applied as statistical method. A quasi-experimental design with paired t-test was also used. *Results:* Psychosocial functioning improved when rated by parents and clinicians. Parent self rated Expressed Emotions improved. No changes were observed in symptoms of mania and depression. *Conclusions*: The RAINBOW program seems feasible and possibly effective for adolescents and their families in Sweden with minor adaptations.

Keywords: pediatric bipolar disorder, family treatment, adolescent, cognitive-behavioral therapy, group treatment, psychosocial therapy, expressed emotion, case series design, clinical significance, quasi experimental.

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Introduction

Bipolar Spectrum Disorders in the Pediatric Population

Bipolar disorder (BD), also known as manic-depressive illness, is a brain disorder that causes unusual shifts in mood, behaviour and energy (Pandey, Rizavi, Dwivedi, &Pavuluri, 2008). BD is a disorder with high heritability. Stressful events, relational or social problems, drugs or lack of sleep can be triggers of a depressive or manic episode. The four syndromatic presentations described in the DSM-IV criteria - BD type I, BD type II, BD Not Otherwise Specified (BD-NOS) and Cyclothymia are accepted for use in the diagnosis of PBD with the additional emphasis on two age-adjusted phenomena: child-specific symptom manifestation of the criteria and severely fluctuating mood within the episodes (Birmaher & Axelson, 2006). Youths with bipolar disorder experience unusually intense emotional states that occur in distinct periods called mood episodes. Signs of PBD that clinicians look for based on DSM-IV are presented in table 1. The frequency, depth and duration of mood episodes can vary a lot from child to child. No child-specific criteria for bipolar disorder are provided in the current Diagnostic and Statistical Manual of Mental Disorders (DSM-IV) and debate remains in the literature over what characterizes PBD. The disagreements typically fall into two camps: the “narrow” and “broad” phenotypes of PBD. Even assuming a consistent and agreed-upon definition of PBD, diagnosis remains challenging. The “narrow” criteria for PBD focus on symptoms highly specific to bipolar disorder, clear episodicity of mania requiring euphoria or severely irritable mood with subsequent less need for sleep, grandiosity and unstable self-esteem, hypersexuality, pressured speech, racing thoughts and goal-directed activity and loss of function. The mood changes are primary and causing the behavioral changes. In addition to symptoms there should be evidence for mood episodes including depressive and elevated episodes often requiring retrospective assessments to identify. According to degree and lengths of symptoms in combination with loss of function, the disorder is called Bipolar Disorder I (BD-I), Bipolar Disorder II (BD-II), Bipolar Disorder Not Otherwise Specified (BD-NOS) or Cyclothymia (DSM-IV 2002). A manic episode lasting seven days or more is required for a diagnosis of BD-I. A manic episode lasting less than seven days but more than four days in combination with at least one depressive episode of at least 14-days long is diagnosed as BD-II. If manic and depressive episodes occur but do

not fulfil the criteria for length of time, the disorder is called BD-NOS. Cycling between low grade mania and low grade depression not meeting full diagnostic criteria for mania or depression is called cyclothymia.

Developmental differences can make separation from normative behavior and symptoms of BPD difficult. PBD can be characterized by less clearly defined mood episodes, shorter duration of these episodes and different hallmark symptoms than in adults. The experienced clinician define what is “abnormal” by comparing the severity and persistence of a child´s symptoms to the average, given the child´s age, stage of development as well as the situation and psychosocial setting in which the behaviour occurs. The prevalence of PBD in children and adolescents is today estimated at 0.1-1.8 % of the general population (Lewinsohn, Klein, & Seeley, 2000; Merikangas et al., 2007). However, the actual prevalence of various subtypes of bipolar spectrum disorders in the clinical and general population has not been determined.

There is a high degree of comorbid disorders in PBD, however little agreement among researchers as to how common the comorbid conditions are. Comorbidity widely range between 10-75 % for ADHD, 45-75 % for oppositional defiant disorder (ODD), 5-40 % for Conduct disorder (CD), 12-60 % for anxiety disorders and up to 40 % for substance abuse disorders, depending on age of the child, social setting, research methodology and diagnostic criteria (Pavuluri, 2008)

Table 1

Signs of PBD based on DSM-IV

Symptoms of a manic episode	Symptoms of a depressive episode
Mood Changes • Overly silly or joyful mood • Severely irritable mood with high energy. • Grandiosity and inflated self-esteem	**Mood Changes** • Sad , tearful, and having low self esteem • Losing interest in activities once enjoyed • Feelings of hopelessness, worthlessness or exaggerated guilt. • Increased anxiety
Behavioral Changes • Sleeping less but not feeling tired • Being more active in an exaggerated and unusual way for the child. • Talking a lot and having racing thoughts, not distractable • Having difficulties to concentrate, attention jumping from one thing to the next in an unusual way • Talking and thinking about sex more often • Behaving in risky ways more often, seeking pleasure, and showing poor judgement.	**Behavioral Changes** • Complaining about pain more often, such as headache, stomach-ache, and muscle pains • Eating a lot more or less and gaining or losing a lot of weight • Sleeping less or oversleeping when these were not problems before • Being more irritable or aggressive • Losing energy, less active, losing interest in friends and activities usually liked • Recurring thoughts of death or suicide.

BD is the 6:th most costly disease in the western world. Quite often it is a lifetime disorder leading to a low functional capacity. 25-50% of the BD-population attempt suicide at least once in their lifetime, with a mortality rate due to suicide of 8.6% to 18.9% (Chen & Dilsaver, 1996). Longitudinal studies have pointed at high rates of hospitalization, psychotic features, substance abuse, family and legal problems as well as low psychosocial function. Around 60 % of adults with a diagnosis of BD experienced their first affective episode before the age 18 (Egeland, Hostetter, Pauls & Sussex, 2000). Early-onset bipolar populations show more

violent behavior, more use of drugs, poor insight, poor medication adherence, higher rates of suicidal behavior, self harm and anxiety, rapid cycling, and longer time to recovery (Carlson, Bromet, & Sievers, 2000). Having PBD results in a higher risk for developing a more serious course of the illness and a higher comorbidity. The often progressive nature of bipolar disorder further supports the concept that the first episode is a period that requires energetic broad-based treatment. Prompt treatment may be neuroprotective and perhaps attenuates or even prevents the neurostructural and neurocognitive changes seen to emerge with chronicity of PBD (Berk et al., 2010). This highlights the need for early identification and the necessity of implementing treatments and services at a stage of the illness where prognosis is optimal. Therefore, identifying early intervention strategies that can change the course of the disorder and understanding the impact of such interventions is of great importance. The primary target in BD treatment is mood stabilization. To find effective treatments that in combination with medication can stabilise and prevent or reduce new affective episodes in PBD is an urgent focus. Treatment of PBD requires a multimodal approach that incorporates pharmacological and psychosocial interventions. Though medications are the cornerstone for achieving mood stabilization, family and psychosocial approaches are critical to achieve long term stabilization (McClellan, Kowatch, & Findling, 2007

Expressed emotions and family focused treatment.

High levels of expressed emotions (EE) defined as high levels of criticism, hostility, or emotional overinvolvement among caregivers have repeatedly been found to predict relapse among patients with bipolar, depressive, and schizophrenic disorders (Butzlaff & Hooley 1998; Jarbin, Gråve & Hansson 2000; Hooley, 2007). For example Kim and Miklowitz (2004) found that a higher frequency of critical comments among caregivers predicted higher levels of mania and depression among bipolar patients at 2-year follow up. Although most of this work has been accomplished in adults, one study established EE as a predictor of time to recovery in depressed children and as a predictor of symptom resolution among BD adolescents in a 2-year open trial of family focused treatment for adolescents (Miklowitz, Biuckians, & Richards, 2006).

Family focused therapy seeks to reduce the high levels of stress and conflict in the families of bipolar patients. Rea and coworkers (Rea et al., 2003) compared individual therapy with family-focused treatment in a randomized controlled study. Patients in family focused treatment were less likely than patients in individual therapy to be rehospitalized during the 2-year study period. Moreover, patients in family treatment also experienced fewer

mood disorder relapses over the 2 years, although they did not differ in their likelihood of a first relapse from patients in individual treatment. Results suggest that family psychoeducational treatment is a useful adjunct to pharmacotherapy in decreasing the risk of relapse and hospitalization frequently associated with bipolar disorder. In a study by Miklowitz et al. (2009) adolescents with PBD in high-EE families showed greater reduction in depressive and manic symptoms after Family Focused Treatment compared to a brief psychoeducational treatment, while these differential effects were not found among adolescents in low EE families. The authors suggested assessment of EE could be used to help decide which families should have the more extensive family focused treatment.

Psychosocial treatments for children and adolescents with BD

A challenge in developing psychosocial treatments is to find disorder specific interventions for symptoms and functioning along with promotion of sustained remission. Given the complex presentation of symptoms in PBD, the likelihood of comorbid disorders and the variable efficacy of psychopharmacological interventions, psychotherapeutic treatments can be critical to improvement. Despite the clear need for psychosocial and psychotherapeutic interventions for PBD, treatment programs have just recently been developed. There are today a few adjunctive psychosocial treatments described for PBD in different stages of empirical validation.

Fristad and colleagues as one of the first groups developed and studied multi-family psychoeducation groups (MFPG), adjunctive group treatment for parents and school-aged children with PBD and depressive spectrum disorders. The goal of MFPG includes psychoeducation, symptom management, improved skills in problem-solving, communication and how to get support from others. The initial pilot study, a randomized clinical trial of 35 children, indicated that families increased their knowledge about mood disorders, improved family interactions and increased their perceived social support compared with those of a wait-list control group. However, children's mood symptom severity did not decrease significantly (Fristad, Goldberg-Arnold, & Gavazzi, 2002). In a larger study by Fristad, Verducci, Walters and Young (2009), one hundred and sixty-five children were studied in a randomized controlled trial of multifamily psychoeducational psychotherapy plus treatment as usual (n = 78) compared with a wait-list control condition plus treatment as usual (n = 87). Children and parents participated in eight 90-minute multifamily psychoeducational psychotherapy sessions. Parent and child groups met separately but began and ended sessions

together. The results showed that mood symptoms decreased significantly. The researchers concluded that brief, adjunctive psychoeducational group psychotherapy is associated with improved outcome for children aged 8 to 12 years with major mood disorders.

Miklowitz and colleagues is the first group that has developed a family-focused treatment (FFT-A) for adolescents with BD. FFT-A has the goal of reducing symptoms through increased awareness of how to cope with the disorder, decreased levels of expressed emotion from caregivers and improved family problem-solving and communication skills. The model was originally developed for adults with BD. In the first randomized controlled trial of a psychosocial intervention for adolescent bipolar disorder, Miklowitz et al. (2008) examined the effectiveness of medication and FFT-A in 58 adolescents with bipolar disorder followed for 2 years after an illness episode. The results of the study showed that patients in FFT-A had significantly shorter times to recovery from depression, less time in depressive episodes, and lower depression severity scores for 2 years compared to adolescents treated with medication and 3 sessions of family psychoeducation, called enhanced care.

Hlastala and colleagues (Hlastala, Kotler, McClellan, & McCauley, 2010) are developing an adapted version of interpersonal and social rhythm therapy (IPSRT-A) for adolescents with BD. The IPSRT interventions seek to stabilize social and sleep routines. It is primarily an individual treatment, however the adaptation also includes brief family psychotherapy. In an open trial of IPSRT-A, the participants experienced significant decreases in manic, depressive, and general psychiatric symptoms over the 20 weeks of treatment. Participants' global functioning increased significantly as well. Effect sizes ranged from medium-large to large. A randomized controlled study is underway.

Goldstein and colleagues (Goldstein, Axelson, Birmaher, & Brent, 2007) are adapting dialectical behavior therapy (DBT) for adolescents with BD. This treatment targets emotion dysregulation. The intervention is delivered over the course of one year. It is composed of two modalities: family skills training and individual psychotherapy with the adolescent. A 1-year open trial of DBT in 10 adolescents with BD found significant decreases in suicidality, emotional dysregulation and depression after intervention.

The child and family-focused cognitive- behavioral therapy program for PBD, called "RAINBOW".

Pavuluri and colleagues (Pavuluri et al., 2004) developed the child and family-focused cognitive-behavioral therapy program (CFF-CBT) for PBD, also called RAINBOW. CFF-

CBT was adapted from FFT and developed as an adjunctive intervention for children 8-12 years old with BD and their families. The intervention integrates psychoeducation, CBT and IPSRT techniques, tailored to the unique needs of these children, to augment the effects of medication. The theoretical framework is based on (1) the specific problems of children and families coping with bipolar disorder, (2) a biological theory of excessive reactivity, and (3) the role of environmental stressors. It is a 12-session protocol driven treatment program meant to be delivered over the course of three months. CFF-CBT was initially a single family treatment with joint parent and child sessions, but was later adapted to a multi-family group format with parallel parent and child groups taking 1.5 hours for each session. In the exploratory study, Pavuluri et al. (2004) examined the feasibility of CFF-CBT in 34 families with children and adolescents with PBD ranging from 5 to 17 years old. Treatment integrity, adherence, and parent satisfaction were assessed and showed positive effects. Patients with PBD showed significant reductions in severity scores on all the severity scales of the Clinical Global Impression Scales for Bipolar Disorder scales (CGI-BP) and significantly higher Children's Global Assessment Scale (CGAS) scores compared to pre-treatment results. West and colleagues (West et al., 2009) adapted RAINBOW to a multi-family group treatment format. In the pilot study conducted with 26 families and children with PBD, West et al. (2009) concluded that CFF-CBT was feasible and acceptable to families. The results of the study showed significant improvement in manic but not in depressive symptoms and in children's psychosocial functioning as compared to pre-treatment results. Parents also reported an increased, however not statistically significant, ability to cope with their child's illness. Furthermore, West and colleagues (West, Henry, & Pavuluri, 2007) developed a maintenance model to follow the acute phase of PBD therapy. This second phase consists of psychosocial booster sessions along with medication management, delivered in a systematic way. Thirty-four patients aged 5 to 17 years who underwent CFF-CBT were delivered the maintenance model of treatment over a 3-year period and were assessed for symptom changes as measured by CGI-BP and global functioning, measured by CGAS. Results indicated that participation in the maintenance model of CFF-CBT treatment was associated with positive effects in symptoms and functioning over the 3-year follow-up period. There were no statistically significant differences in post-acute-phase treatment scores and scores at years 1, 2, or 3 on any study measures, indicating the maintenance of clinically significant improvements. A randomized controlled trial of CFF-CBT is currently being conducted by Amy West and her colleagues.

Current Study- Aims and Hypotheses

The CFF-CBT program is being implemented as the first manualized treatment for children and adolescents with BD and their families in Sweden. This program was developed in the US for children between 8-12 years. Very few children in Sweden under the age of 12 are diagnosed with PBD, most patients with PBD in clinical work in Swedish Child and Adolescent Psychiatry are between 13- 18 years old. The CFF-CBT is therefore being adapted to this age group. The CFF-CBT (RAINBOW) manual has been translated into Swedish at the department for PBD in Lund and has been offered in the multi-family format with parallel groups for parents and adolescents. No data of psychosocial treatments for adolescents with PBD in Europe is published as far as we know.

The general purpose of this study was to evaluate if CFF-CBT is effective and feasible for adolescents and their parents in Sweden. Our hypotheses are that after the CFF-CBT program adolescents will show improvements in 1) symptoms of depression and mania and 2) psychosocial functioning; that adolescents and their parents will improve regarding 3) self rated Expressed Emotions in the family and 4) knowledge and skills to cope with the disorder.

Method

Study Design

The aims of this study was to investigate the feasibility and effectiveness of a given treatment in a "real-life" setting with a relative small number of patients with PBD, therefore a case series design was applied. A case series, also known as a clinical series, is a medical research descriptive design that follows a group of patients who have a similar diagnosis or who are undergoing the same procedure over a certain period of time (Kooistra, Dijkman, Einhorn, & Bhandari, 2009). The purpose of case series is to describe patients, interventions and primarily results and is regarded as a sensitive research method to make hypothesis about treatment effects and to describe new interventions (Kooistra et al., 2009).

In addition, paired sampled t- tests were carried out to investigate pre-post changes in parent's given EE, knowledge and skills about the disorder and their assessment of their child's psychosocial function. At the last session feedback was requested from the parents in form of a group interview.

Recruitment and Diagnosis

Patients were recruited from the department of Psychoses and Bipolar disorders at the clinic for child and adolescent psychiatry in Lund, Sweden. Patients are referred to this specialized department from the region of Skane, a region of 1.253 000 inhabitants. All patients 13-17 years old referred to the team who are diagnosed with BD I, BD II, BD-NOS and cyclothymia according to DSM IV criteria, were offered participation in the RAINBOW program. Exclusion criteria were defined as severe episodes that makes it impossible to participate, autistic syndrome and moderate or severe mental retardation.

Diagnoses were made through clinical interviews and clinical assessments with adolescents and their parents. The clinical interviews were conducted by a child and adolescent psychiatrists specialized in pediatric bipolar disorders. The criteria for BD in DSM-IV were used.

Sample

Two consecutive parallel parent-adolescent groups were studied. The first parallel parent and adolescent groups started in September 2011. The second parallel groups started in January

2012. In the first groups seven parents and six adolescents participated. Two of the adolescents were siblings. One of the siblings left the group after two sessions. One parent participated without having a child in the adolescent group. This adolescent, treated for PBD, ADHD, ODD and social anxiety disorder could not be motivated to participate. In the second group, two patients participated in the adolescent group, and all four of their parents participated in the parent group. The patients had had their diagnoses and been under treatment for different length of time, from three to thirty months. Three of the patients had had psychotic symptoms of delusions during previous affective episodes. In total, eight adolescents and eleven parents approved to the treatment. Seven adolescents and eleven parents completed treatment and their results are presented and discussed in the present study. Adherence to treatment was high both in the adolescent and the parent groups. Rate of participation was 89 % and attended sessions mean 10,7 for both adolescents and parents. Patients are presented in table 2.

Table 2

Patient Characteristics and Attendance

Patient	Diagnosis	Comorbidity	Medicine	Parent participated in treatment	Attendance N (%) of sessions	
					Adolescent	Mother Father
P1	BP NOS	ADHD PDD	Lamotrigin Metylphenidate Olanzapine	Mother Father	12 (100%)	12 (100%) 12 (100%)
P2	BP I	ADHD ODD	Lithium Valproic acid Metylphenidate	Mother	11 (92%)	11 (92%)
P3	BP II	ADHD GAD	Lithium Mirtazapin Amfetamin	Mother	12 (100%)	12 (100%)
P4	BP I	ADHD GAD	Lamotrigin Quetapine Sertraline Metylphenidate	Mother	9 (75%)	9 (75%)
P5	BP I	ADHD	Valproic acid Olanzapin Metylphenidate	Father	9 (75%)	8 (67%)
P6	BP-I	OCD AD NOS	Lithium Seroquel Sertraline	Mother Father	10 (83%)	11 (92%) 11 (92%)
P7	BP-NOS	AD NOS	Lithium Seroquel	Mother Father	12 (100%)	12 (100%) 12 (100%)
P8	BP-NOS	ADHD SAD ODD	Lamotrigine Sertraline Metylphenidate	Mother	0 (0%)	8 (67%)

Note. PDD = Pervasive Developmental Disorder; ODD = Oppositional Defiant Disorder; GAD = Generalized Anxiety Disorder; SAD = Social Anxiety Disorder; AD = Anxiety Disorder; OCD = Obsessive Compulsory Disorder; NOS = Not otherwise specified.

Questionnaires

Symptoms of mania, depression and global illness.

Changes in symptoms were assessed with the Child Mania Rating Scale- Parent Version (CMRS-P; Pavuluri, Henry, Devineni, Carbray, & Birmaher, 2006), the Clinical Global Impressions Scale for use in Bipolar Disorder (CGI-BP; Spearing, Post, Leverich, Brandt, & Nolen, 1997), and the Montgomery-Asberg Depression Rating Scale (MADRS-S; Montgomery & Åsberg, 1979; Svanborg & Åsberg 2001).

CMRS-P (Pavuluri et al., 2006) is a 21-item screening tool for pediatric mania symptoms based on DSM-IV criteria for a manic episode. It was scored by the parents. Items are age-specific; each item is considered to be a problem only if it is causing impairment, a deviation from what is normative for that child's age, and has been causing a problem in the last month. The items are answered on a four-point Likert-type scale. Each item is scored between 0-3. The alternative responses are never/rarely, sometimes, often and very often. Higher scores indicate increased impairment .The total score is calculated by summing the answers of the 21 items, the total sum ranging between 0 and 63. The CMRS-P has an internal consistency reliability of .96 reported by Pavuluri et al. (2006). It has proved to be a valid assessment of mania when compared to clinician-rated scales, and shows sensitivity and specificity for differentiating pediatric mania from other disorders and no disorder (Pavuluri et al., 2006). A score of 20 is considered to differentiate children with PBD from children with ADHD and healthy controls and to indicate remission from mania symptoms (West, Celio, Henry, & Pavuluri, 2011).

MADRS-S (Montgomery & Åsberg, 1979; Svanborg & Åsberg, 2001) is a widely used instrument to measure depression. It was scored by the adolescents. The scale consists of 9 items assessing patient´s mood, feelings of unease, sleep, appetite, ability to concentrate, initiative, emotional involvement, pessimism and zest for life. Each item is scored between 0-6 by the patient. Higher scores indicate increased impairment. The total score is calculated by adding the answers of the nine items, the total sum ranging between 0 and 54. Cronbach's alpha is reported to be between 0.85 and 0.94 (Bondolfi et al., 2010). CGI-BP (Spearing et al., 1997) was rated after each group session by the therapist of the group treatment. The clinicians rated current severity of mania, depression, and overall psychiatric illness including comorbidity as presented during the group session, on a 7 point scale. A range of responses is used from 1 (normal) 2 (minimally ill), 3 (mildly ill) and so

forth up to 7 (very severely ill). The CGI-BP was designed as a primary measure of improvement; it only takes a minute to complete.

Psychosocial function.

To measure the patient's global psychosocial health and function, both parents and adolescents scored the Strength and Difficulties Questionnaire (SDQ; Goodman, 1997), and a child psychiatrist rated the adolescents together with the therapist leading the adolescent group on the Children's Global Assessment Scale (C-GAS; Shaffer et al., 1983).
SDQ (Goodman, 1997) is a widely used reliable and valid screening instrument to assess children's overall psychosocial functioning. It consists of 25 questions about different psychological attributes, some positive and others negative, which are responded to by the alternatives; "not true", somewhat true" and "certainly true". The questions address emotional symptoms, conduct problems, hyperactivity/inattention, peer relationship problems, and prosocial behavior. Based on 20 of the items a score of "total difficulties" is generated ranging from 0 to 40 points. SDQ exists in several versions. Parents scored the SDQ parental version (P 4-16) and the adolescents scored the self-completion version for adolescents (S11-17). Both versions ask about the same 25 traits, though the wording is slightly different in the self-completion version (Goodman, Meltzer, & Bailey, 1998).

CGAS (Shaffer et al., 1983) is a single item scored 1–100 to rate functioning and degree of impairment due to symptoms for children under the age of 18. Descriptors appropriate for children and adolescents are provided for each 10- point range. Clinicians are instructed to rate the CGAS, taking into account clinical interviews with both parents and children and to record the lowest level of general functioning over the past month. Reports on interrater reliability on CGAS vary. In research settings reliability is high ranging from 0.8-0.9 (Bird, Canino, Rubio-Stipec, & Ribera, 1987; Shaffer et al., 1983). In typical clinical settings, only moderate agreement has been demonstrated (0.53–0.66) (Rey, Starling, Wever, Dossetor, & Plapp, 1995). In a recent study, intra class correlation coefficient among 703 health care professionals in Swedish CAMHS was 0.73. (Lundh, Kowalski, Sundberg, Gumpert, & Landén, 2010)

Expressed emotions.

Expressed emotions were assessed with Questions about Family members (QAFM; Hansson & Jarbin, 1997). QAFM is a self rating instrument that aims to describe a dyadic relationship with another family member. The questionnaire consists of 30 items which are rated on a 5 point Likert scale. The answers range from "almost always" to "almost never". The adolescents scored one QAFM each about their relation to their mother and to their father. The parents individually scored one QAFM about their relation to the adolescent/patient. The questionnaire has been homogenized by factor analysis, resulting in four factors; two factors about "given" EE: critical comments and emotional over-involvement and two factors about "perceived EE": perceived criticism and perceived emotional over-involvement. Cronbach's alpha is reported to be 0.87 for critical comments, 0.81 for emotional over-involvement, 0.73 for perceived criticism and 0.69 for perceived emotional over-involvement (Hansson & Jarbin, 1997). In this study, only given EE, that is critical comments and emotional over-involvement are reported and analyzed. Conventionally EE is assessed with the Camberwell Family Interview (CFI). Unfortunately training in CFI is difficult to obtain and the instrument is time-consuming to administer and rate. As the QAFM has shown good reliability and validity and was chosen as a reasonable alternative (Hansson & Jarbin, 1997).

Adolescents and parents knowledge and skills to cope with the disorder.

Parent's feelings and perceptions regarding their child's bipolar disorder, including their knowledge about the disorder and sense of efficacy in coping with it, was assessed with Treatment Outcomes Parents Scale (TOPS; West et al., 2009). The scale was translated to Swedish, then translated back to English and accepted as valid by the constructors of the scale. To assess the teenager's sense of knowledge, coping and perceptions of their bipolar condition, a similar scale, Treatment Outcomes Teenagers Scale (TOTS) was developed and clinically used here for the first time. The items in TOTS are almost identical to the items in TOPS but are changed in wording to be addressed to the adolescents. Both patients and parents rate 20 items on a 5-point Likert scale ranging from strongly disagree to strongly agree. Higher scores indicate greater knowledge and perceived self-efficacy in coping with the disorder.

Procedure

Data collection.

Data was collected at the first and last session of the program in the form of questionnaires filled out by adolescents and parents. According to the manual, time for questionnaires at these sessions is a planned activity. A therapist assisted in case of any questions from the parents and adolescents. Clinicians rated CGAS before and after treatment and CGI-BP was rated after each group session by the therapist of the group treatment

Treatment.

Treatment was given weekly for twelve consecutive weeks. The treatment was driven by a manual where each session contained certain interventions that should be worked through. In case, an intervention for some reason was not covered during the session, the therapist was instructed by the manual to cover it in the following session. Each session lasted for 90-100 minutes with a break of 15-20 minutes at halftime. Both the parent and adolescent group were led by two therapists. At the end of each session the adolescents joined the parents for about ten minutes and the groups summarized what they had been working on. The homework for the next session was clarified both for adolescents and the parents together. The acronym "RAINBOW" was formed to help parents and children to remember the key components of CFF-CBT. The essential components of CFF-CBT "RAINBOW" covered throughout the treatment sessions are as follows: *R: Routine*. The goal of this component is to increase affect regulation and decrease symptom exacerbation by establishing predictable routines around sleep, diet, medication and making transitions. Parents are also urged to integrate pleasurable activities into their own and their child's routine. *A: Affect regulation.* The goal of this component is to provide psychoeducation about symptoms of PBD. Parents are educated about the biological basis of BD and the nature of symptoms. Children are educated about recognizing and responding to affective states and consistently self-monitoring moods. *I:I can do it.* The goal of this component is to increase parents' and children's beliefs in their ability to cope with the disorder. *N: No negative thoughts and live in the now*. The goals of this component are twofold. The first goal is to decrease negativistic thinking and thought distortions associated with depression. The second goal is to encourage children and parents to focus on the present moment. Mindfulness techniques such as the use of positive mantras are incorporated. *B: Be a good friend and balanced lifestyle for parents*. The goals of this component are also twofold. The first is to improve social functioning in children. Children

are taught the skills necessary to be a good friend and are provided opportunities to practice the skills. The major goal is to help the children establish and maintain friendships. The second goal is to help parents develop a balanced lifestyle that involves finding ways to rest, replenish their energy and enjoy life. *O: Oh, how can we solve this problem?* The goal of this component is to engage parents and children in a collaborative and effective problem-solving process. Parents and children are encouraged to try creative ways to approach problem solving to minimize reactivity and the exacerbation of negative emotions. *W: Ways to get support.* The goal of this component is to increase social support. Techniques used emphasize the identification and active seeking out of people who can help the child and the parents through difficult situations. School advocacy is also a part of this component.

Ethics.

The study was approved by the regional ethics committee in Lund. All participants gave their written consent to participate in the study. They were informed that they could cease participation at any time they wished and that giving consent to participate would not affect access to treatment. The risk of a patient being identified in a case series study is considerable high. To limit this risk, it was decided not to report the age of the patients and present them in a random order. For the same reason, leaving out reports of comorbidity was also considered, however it is of high value for understanding the complexity of unique cases and therefore it was decided to report fully in all but two cases. To protect the integrity and decrease risk of identification the diagnosis of dyslexia with expressive language disorder for one of the patients and self harm for another are not reported in table 2.

Data analysis.

Both clinical and statistical significance is reported in the present study. When evaluating a treatment, statistical significance is important in order to analyze if changes in patients symptoms were due to the treatment itself or due to confounding factors. The risk with only relying on statistical significance though, is that significant improvements might be clinically insignificant on an individual level. Even if a small sample does not have the ability to reliably detect statistically significant changes on a group level, it can give information about individual changes which in other designs, for example large RCTs are easily masked by group variance (Atkins, Bedics, McGlinchey, & Beauchaine, 2005).

Clinical significance.

Assessing individual variance can give important information regarding clinically significant and meaningful changes. The "clinical significance" of a treatment refers to its ability to meet standards of efficacy set by consumers, clinicians, and researchers (Jacobson & Truax, 1991). There are several definitions of clinical significant change, for example that the problem in question is eliminated, the magnitude of change, if there is change in the individuals daily functioning, if these changes are visible to friends and significant others or that the individual has reached a level of functioning which is no longer different from the "normal" population (Lambert & Ogle, 2009). The definition of clinical significance used in this study is the magnitude of the change after treatment and if the treatment has moved the patient outside the range of the dysfunctional population into the range of the functional population.

The most frequently used method for evaluating the reliability of changes in patients is the Jacobson and Truax model for clinical significance (Jacobson & Truax, 1991; Lambert & Ogles, 2009). According to Jacobson and Truax model, the assessment of clinical significance is made in two steps. First, a Reliable Change Index (RCI) for each individual is calculated to ensure that a difference before and after an intervention is not caused by measurement error of the outcome instrument. The RCI for each individual is based on the pre-treatment score (X_{pre}), the post treatment score (X_{post}) and the standard error of the difference between two test scores (S_{diff}). The formulas below are from Lambert and Ogles (2009):

$$RCI = \frac{X_{post} - X_{pre}}{S_{diff}}$$

S_{diff} in the denominator is calculated by the formula: $S_{diff} = \sqrt{2}(SE)^2$ and $SE = SD\sqrt{1-r}$. "SE" stands for the standard error of measurement, "SD" for the standard deviation of the instrument and "r" for the reliability coefficient. Thus, the standard error of the difference between two test scores depends on the standard deviation and the reliability of the outcome measure. This means that the more reliable the instrument, the smaller the resulting standard error and thus the smaller changes are required between pre- and post-test scores to achieve a statistically reliable change. The change is considered reliable ($p<0.05$), or unlikely to be the product of measurement error, if the RCI is greater than 1.96 (Jacobson & Truax, 1991). When the individual has a change score greater than 1.96, one can reasonably assume that the individual has improved. Similarly, a score beyond - 1.96 in the opposite direction would indicate that the individual reliably deteriorated.

The second step in the Jacobson-Truax model is to decide if the individual has moved from a theoretical dysfunctional (patient) population to a functional (non-patient) population as a result of the intervention. This is done by determining a cut-off value. There are different ways to choose cut-off point depending on if there is information available about norms for the clinical and non-clinical populations. Either one can use the mean and standard deviation of the functional or the dysfunctional group or one can use existing suggested norms in the research literature (Lambert & Ogles, 2009). Using the RCI and the cut off value, each individual can be classified as (1) recovered (passed both criteria), (2) improved (passed only the RCI criterion in the positive direction), (3) unchanged (did not pass the RCI criterion), or (4) deteriorated (passed the RCI criterion in the negative direction) (Lambert & Ogles, 2009). In this study, reliability coefficients, standard deviations and cut off points were collected from the research literature (see Table 3). The only exception is the QAFM questionnaire, where the cut off values were calculated to be one standard deviation from the mean in a non-clinical group.

Table 3

The Data Used in the Assessment of Clinical Significance

Questionnaire	Cronbach's alpha	SD	SE	Cut-off	Reference
MADRS-S	0.84 [1]	4.5[1]	1.8	<9	Fantino & Moore (2009) Hawley, Gale, & Sivakumaran, (2002)
CMRS-P	0.96	9.20	1.84	<20	West, Celio, Henry, & Pavuluri,(2011)
SDQ-S	0.80[1]	4.59[2]	2.05	14[1]	[2]West et al. (2009) Malmberg, Rydell, & Smedje (2003)
SDQ-P	0.80	5.29	2.37	14	West et al. (2009)
CGAS	0.92[1]	5.2[1]	[1]1.47	>70[2]	[1]Lundh et al. (2010) [2]Bird et al. (1987)
QAFM CR	0.87	0.76	0.27	<2.16*	Hansson & Jarbin (1997)
QAFM EO	0.81	0.64	0.33	<2.47*	Hansson & Jarbin (1997)
TOPS	0.86	15.30			West et al. (2009)
CGI-BP Overall		0.81		< 3	West et al. (2009)
CGI-BP Depression		1.69		< 3	West et al. (2009)
CGI-BP Mania		1.52		< 3	West et al. (2009)

Note. *Cut off values for QAFM were set to be 1 standard deviation from the mean in a non clinical group

Statistical significance.

In order to investigate pre-post changes in parent´s scores on TOPS, QAFM CR, QAFM EO, SDQ-P and CMRS-P paired samples t-test was used. Pre-post effect sizes (Cohen's d) were calculated using the formula suggested by Rosenthal (1984) for matched-pairs data ($d= t/\sqrt{df}$). Cohen's d conventions for mean differences are as follows: an effect size around 0.20 is a small effect, around 0.50 a medium effect and 0.80 and larger, a large effect.

Results

Clinical significance

The results regarding clinical significance are reported for each case in four consecutive figures numbered one to four.

Figure 1 illustrates symptoms of depression and mania measured pre and post treatment by MADRS-S scored by the patient and CMRS-P scored by mother (M) and father (F). Figure 2 shows severity of mania, depression and overall bipolar illness rated by clinicians after each session during treatment, using parts of CGI-BP. Figure 3 shows psychosocial function measured by SDQ-S (self-rating) and SDQ-P (parental rating) by mother (SDQP-M) and father (SDQP-F). Clinician's measure of patient's psychosocial function is shown by C-GAS. Figure 3 also shows patient's and parent's knowledge and skills to cope with the disorder measured by TOPS that was filled out by mother (TOPS-M), father (TOPS-F) and by adolescent (TOTS). Figure 4 shows results from QAFM illustrated by the two factors assessing "given" EE: critical remarks (CR) and emotional over-involvement (EOI). S-M refers to self-rating, patient in relation to mother. S-F refers to self-rating, patient in relation to father**.** P-M refers to parental rating, mother in relation to patient. P-F refers to parental rating, father in relation to patient as follows;

CR S-M Critical Remarks Selfrated by adolescent in relation to the Mother
CR S-F Critical Remarks Selfrated by adolescent in relation to the Father
CR P-M Critical Remarks Parental rating by Mother in relation to the adolescent
CR P-F Critical Remarks Parental rating by Father in relation to the adolescent
EOI S-M Emotional Over-Involvement Selfrated by adolescent in relation to the Mother
EOI S-F Emotional Over-Involvement Selfrated by adolescent in relation to the Father
EOI P-M Emotional Over-Involvement Parental rating by Mother in relation to the adolescent

EOI P-F Emotional Over-Involvement Parental rating by Father in relation to the adolescent

According to Jacobson and Truax model using the RCI and the cut off value, each individual was classified in one of four categories , as recovered (passed both criteria), improved (passed only the RCI criterion in the positive direction), unchanged (did not pass the RCI criterion), or deteriorated (passed the RCI criterion in the negative direction). RCI for each measurement is presented in figure 1-4.

Patient 1

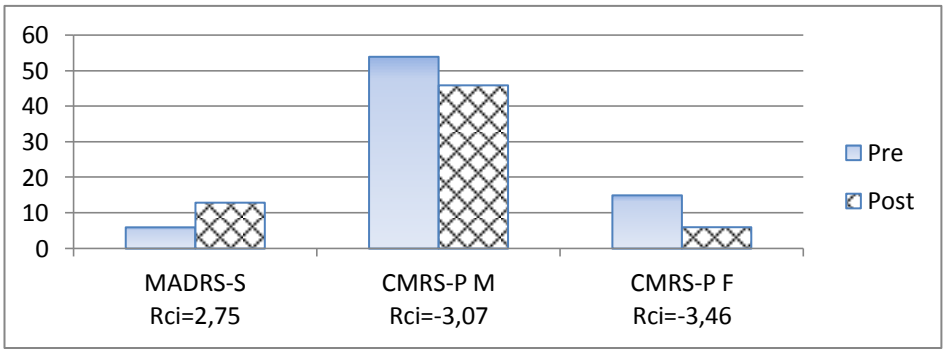

Figure 1.1. Patient 1's symptoms of depression and mania measured pre and post treatment.

There was deterioration in MADRS-S. Both parents scored an improvement in CMRS-P, the mother above cut off level and the father below cut off level both pre and post-treatment.

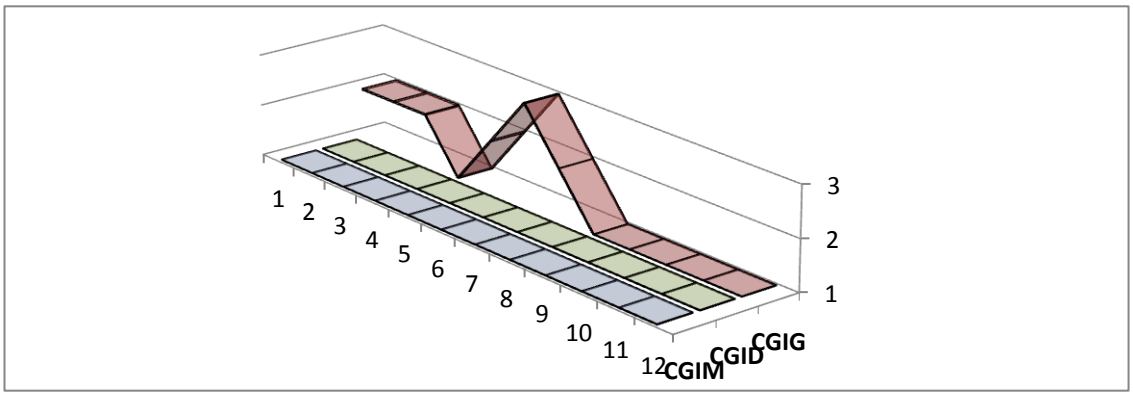

Figure 1.2. Patient 1's severity of mania, depression and overall psychiatric illness rated by clinicians after each session during treatment.

Clinician's rating of severity regarding mania, depression and overall bipolar illness did not record any change in manic or depressive symtoms. Between session four and eight there was a period of detoriation regarding severity of overall illness, up to a mildly severity level.

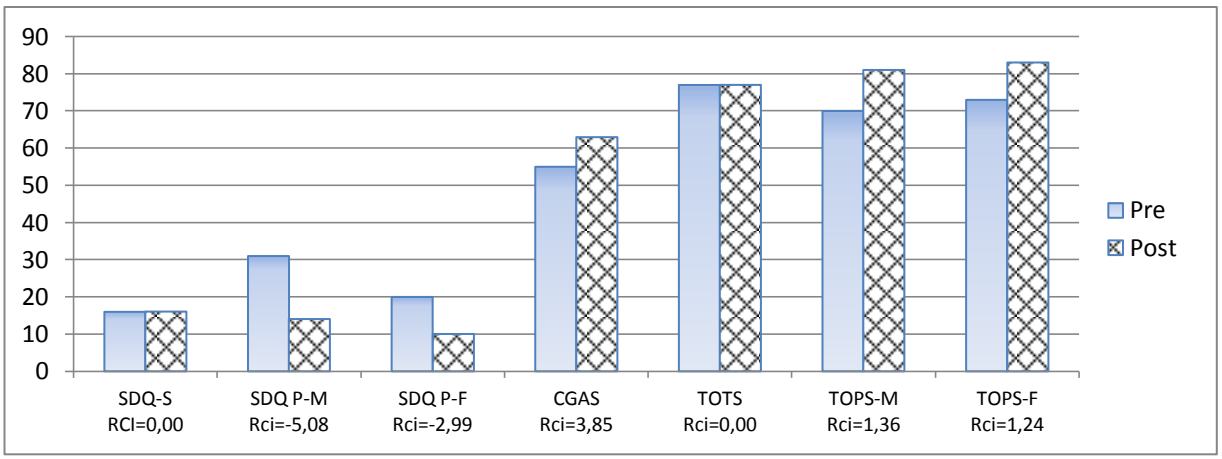

Figure 1.3. Psychosocial function, knowledge and skills measured pre and post treatment (Patient 1).

The patient did not score any change in SDQ-S while both parents scored a recovery in SDQ-P and clinicians scored improvement in CGAS. There was no change in TOTS and TOPS.

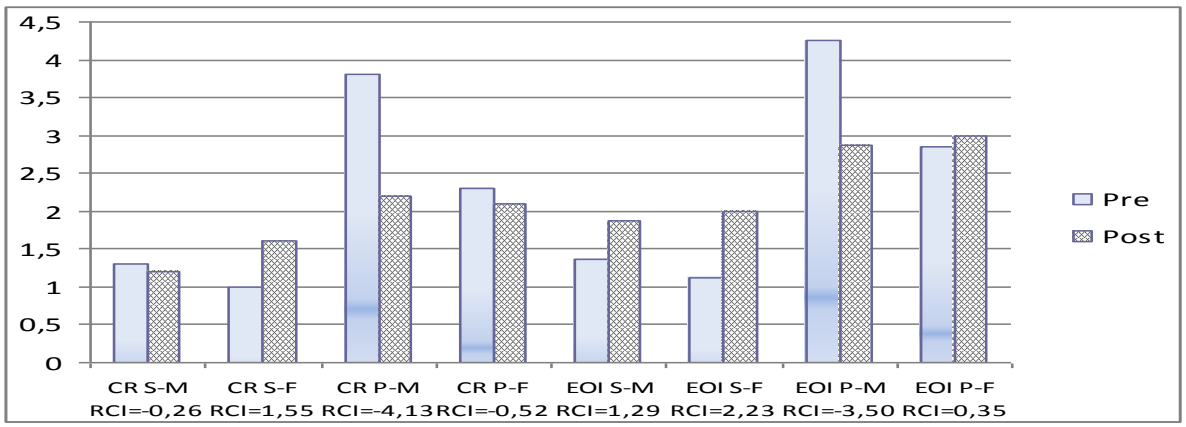

Figure 1.4. Scores on expressed emotions regarding Patient 1's family.

The patient scored no change and under cut off level for both critical remarks and emotional over involvment in relation to the parents. The mother scored an improvement both regarding critical remarks and emotional overinvolvment. The father scored no change regarding emotional overinvolvement or critical remarks. Post-treatment both parents have values above cut off for emotional overinvolvment and the mother also for critical remarks indicating high a EE in the family both pre and post treatment.

Patient 2

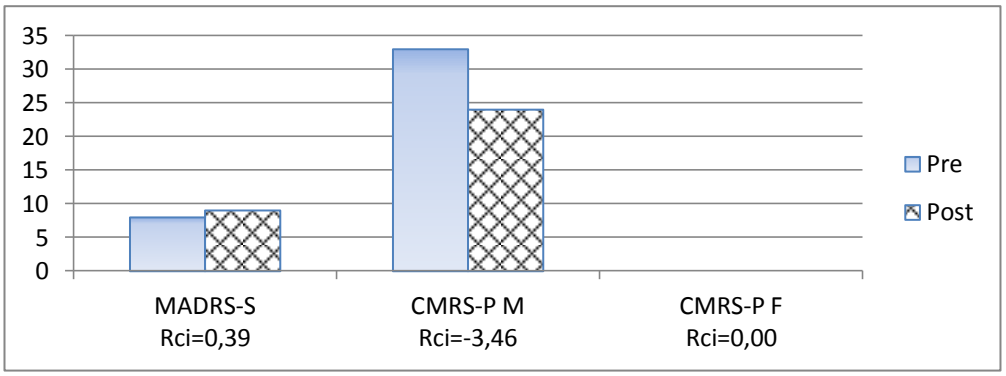

Figure 2.1. Patient 2's symptoms of depression and mania measured pre and post treatment.

The patient scored no change in MADRS-S. The mother scored improvement in CMRS-P.

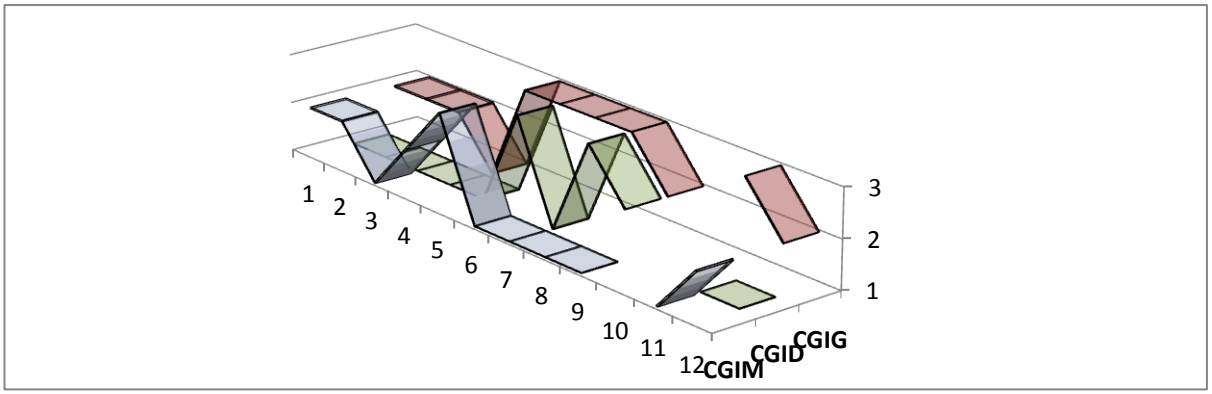

Figure 2.2. Patient 2's severity of mania, depression and overall illness rated by clinicians after each session during treatment.

There is an instability noticed from week three with mildly manic symtoms for a period of three weeks and after week six mildly depressive symtoms . The severity of the illness during these weeks was increased minimally to mildly.

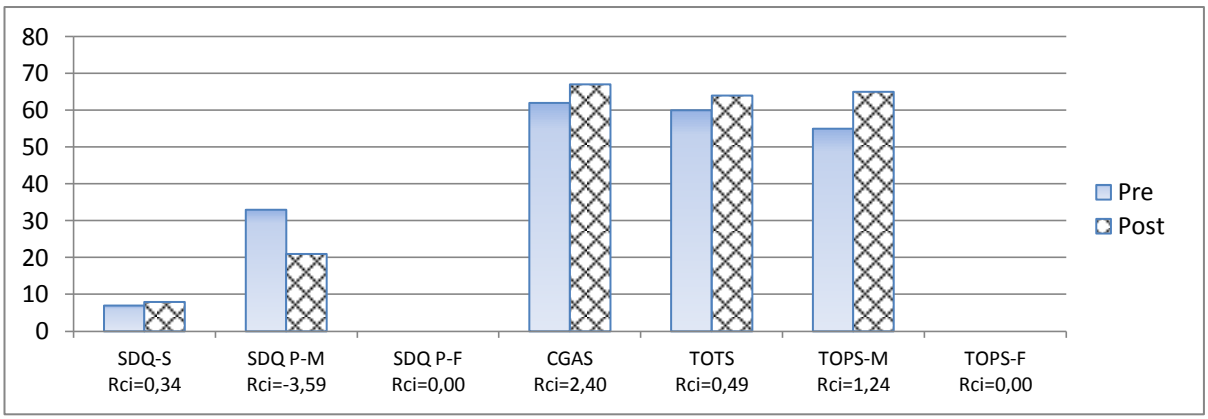

Figure 2.3. Psychosocial function, knowledge and skills measured pre and post treatment (Patient 2).

The patient scored no change in SDQ. Mother rated an improvement at SDQ-P. CGAS improved. Both patient and mother scored no change in TOTS and TOPS.

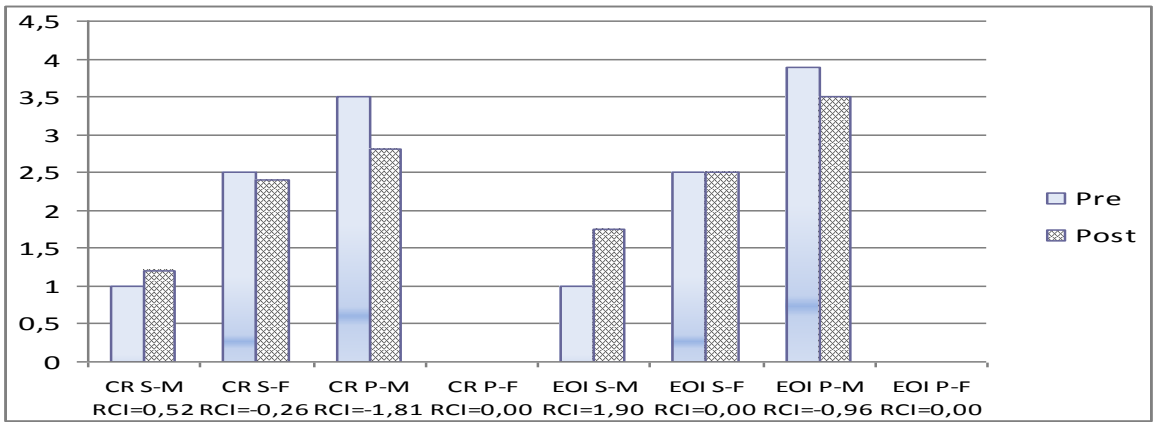

Figure 2.4. Scores on expressed emotions regarding Patient 2's family.

The patient scored no change and higher than cut off on critical remarks in relation to the father and below cut off in relation to the mother. The mother scored no change and higher than cut off for both critical remarks and emotional over-involvement pre and post treatment indicating a high EE.

Patient 3

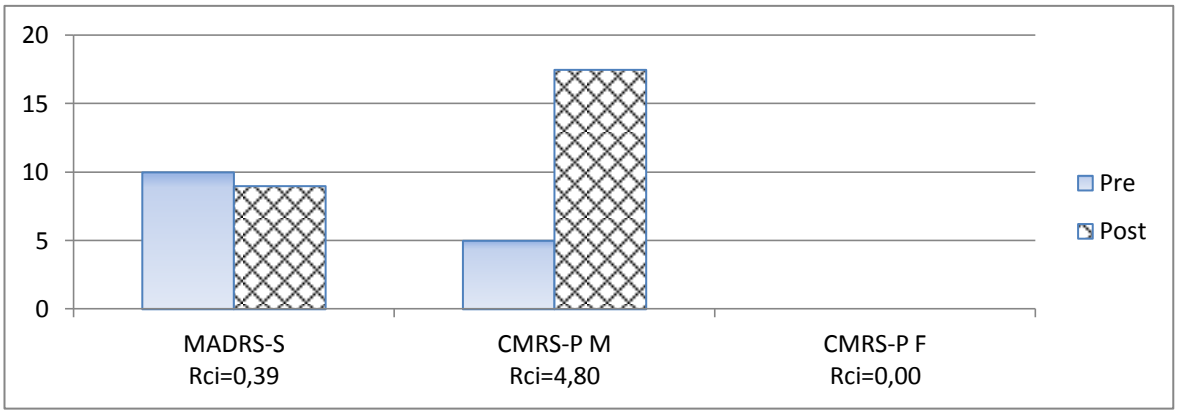

Figure 3.1. Patient 3's symptoms of depression and mania measured pre and post treatment.

The patient scored no change in MADRS-S. The mother scored deterioration in CMRS-P post treatment.

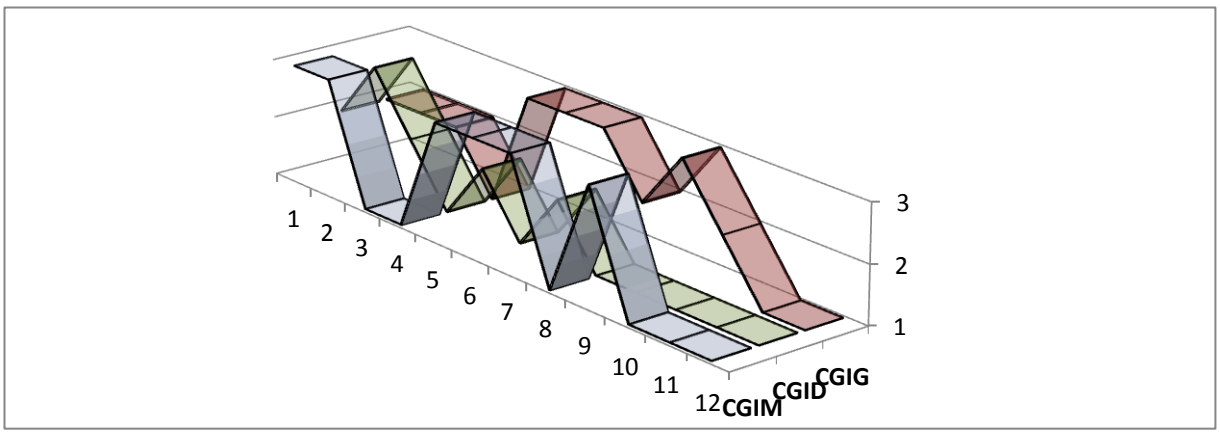

Figure 3.2. Patient 3's severity of mania, depression and overall psychiatric illness rated by clinicians after each session during treatment.

Weekly CGI-BP shows an instability both regarding severity of mania, depression and overall illness with highest score 3, mildly severity. The last two group sessions the patient was scored as having minimally symtoms

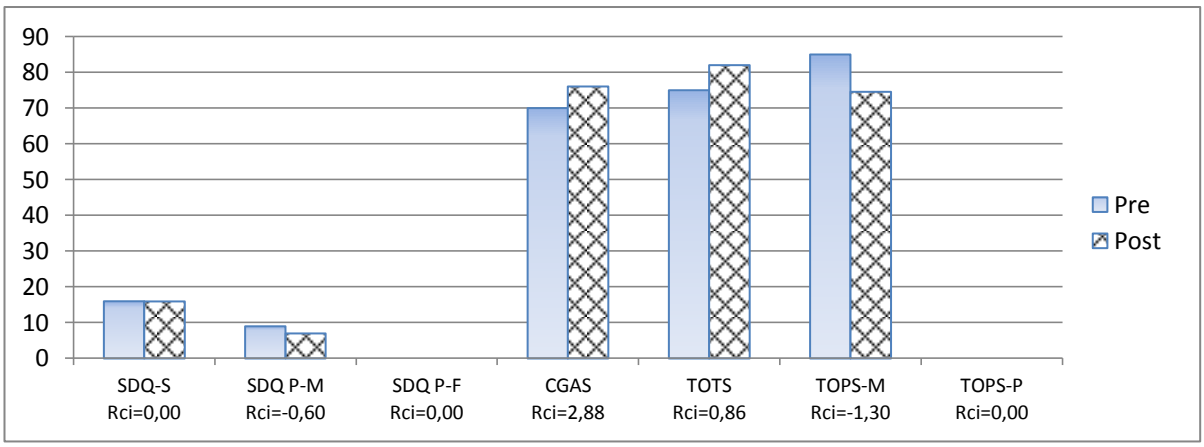

Figure 3.3. Psychosocial function, knowledge and skills measured pre and post treatment (Patient 3).

This was an already well functioning patient, under cut off pre treatment in SDQ-P, and no change was scored in self or parental rating of psychosocial functioning. Clinician's rating of global functioning show recovery. In the case of knowledge and skills the patient and the mother scored no change.

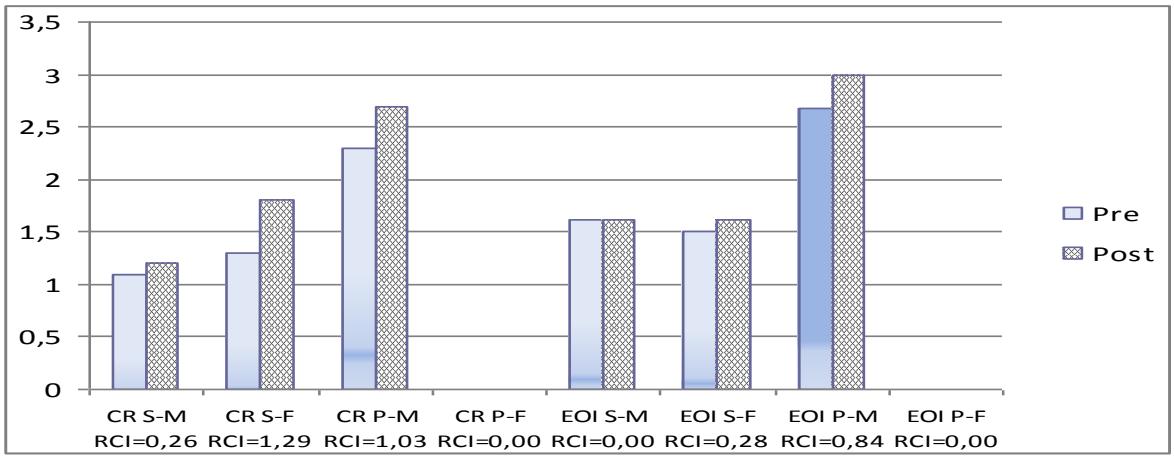

Figure 3.4. Scores on expressed emotions regarding Patient 3's family.

Both patient and mother scored no change in critical remarks and emotional over-involvement. Mother scoring above cut off for both critical remarks and emotional over-involvement pre and post-treatment indicating a high EE.

Patient 4

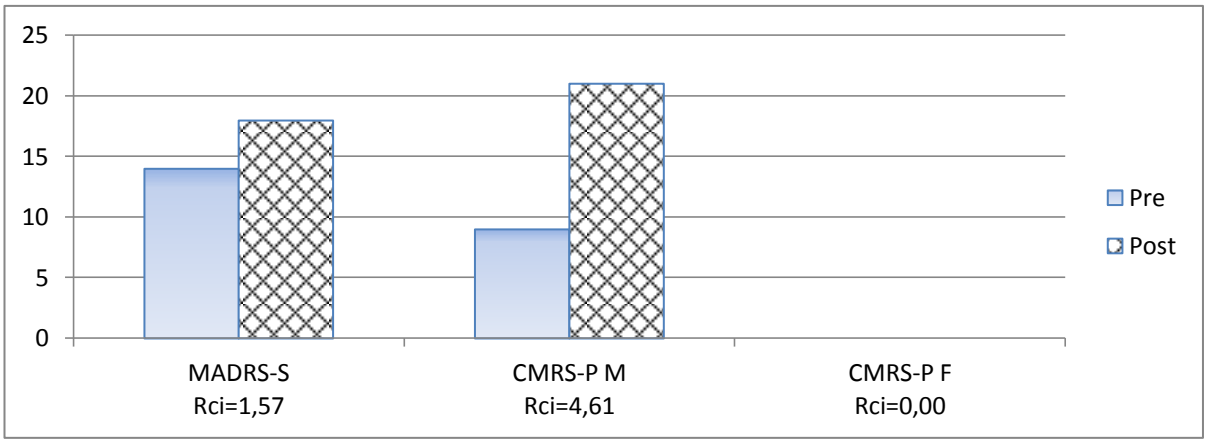

Figure 4.1. Patient 4's symptoms of depression and mania measured pre and post treatment.

The patient scored no change in MADRS-S. The mother scored deterioration in CMRS-P passing cut off.

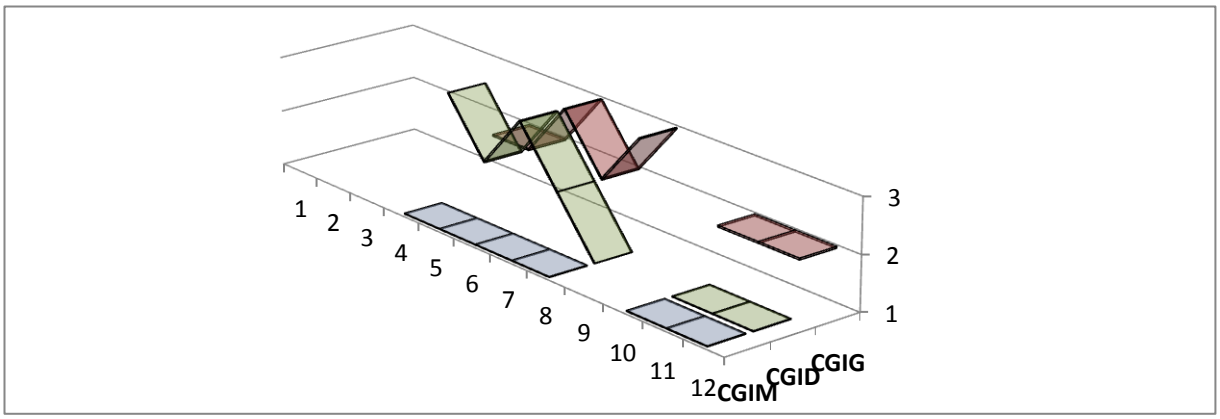

Figure 4.2. Patient 4's severity of mania, depression and overall psychiatric illness rated by clinicians after each session during treatment.

Mild instability was recorded week three to seven, and thereafter a stabilization on a non clinical level reagding depressive and manic symtoms when rated by clinician.

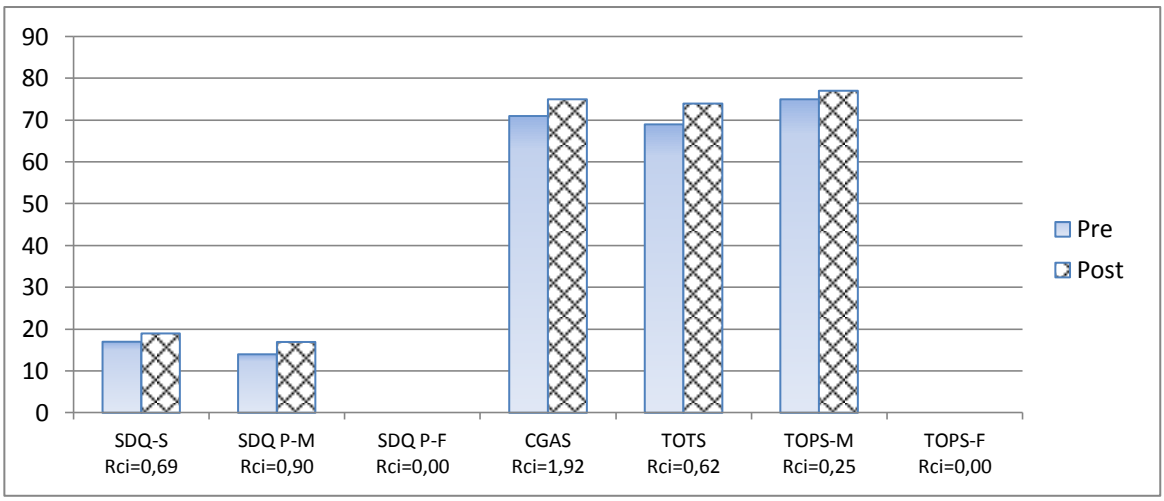

Figure 4.3. Psychosocial function, knowledge and skills measured pre and post treatment (Patient 4).

There was no change in SDQ-S or SDQ-P. No change in CGAS. No change in TOTS or TOPS.

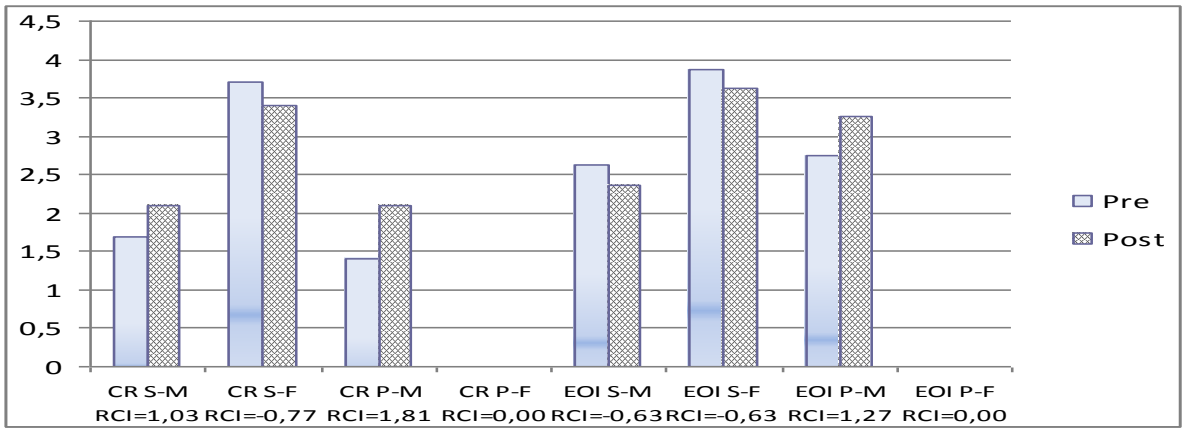

Figure 4.4. Scores on expressed emotions regarding Patient 4's family.

There was no change in critical remarks or emtional overinvolvement scored by the patient and the mother. Scores for critical remarks and emotional overinvolvment in patient's relation towards the father are higher than cut off both pre and post-treatment. Also the mother scored higher than cut off regarding emotional overinvolvment both pre and post treatment. The results indicate a high EE in the family.

Patient 5

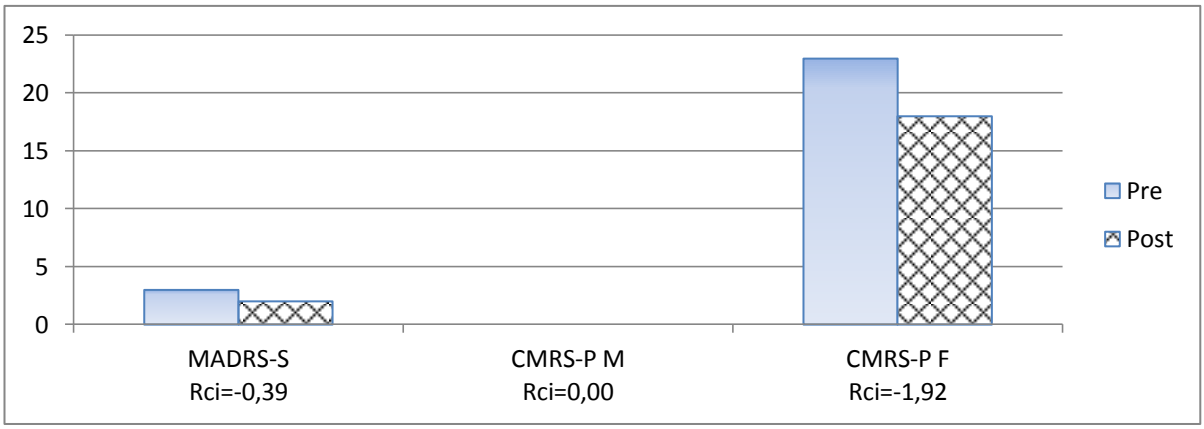

Figure 5.1. Patient 5's symptoms of depression and mania measured pre and post treatment.

The patient scored no change in depressive symptoms. The father scored no change in CMRS-P.

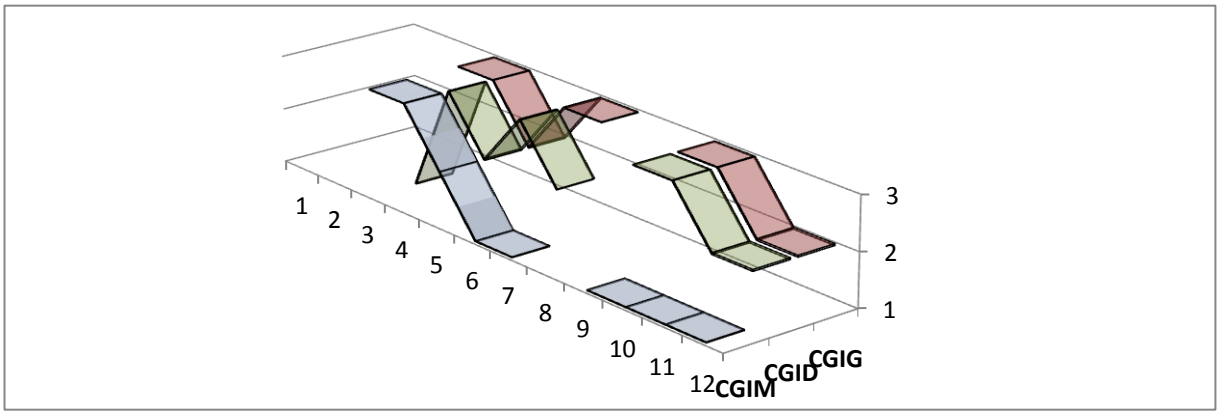

Figure 5.2. Patient 5's severity of mania, depression and overall illness rated by clinicians after each session during treatment.

CGI indicates a rather high level of instability and a decrease of symptoms at the last two sessions.

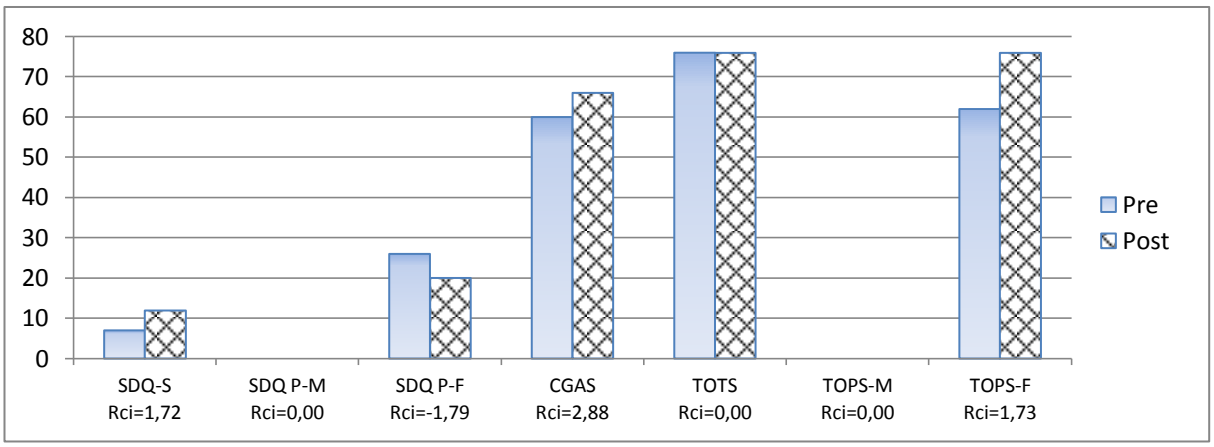

Figure 5.3. Psychosocial function, knowledge and skills measured pre and post treatment (Patient 5).

Patient and mother scored no change in SDQ. CGAS showed improvement. The patient and mother scored no change in TOTS or TOPS.

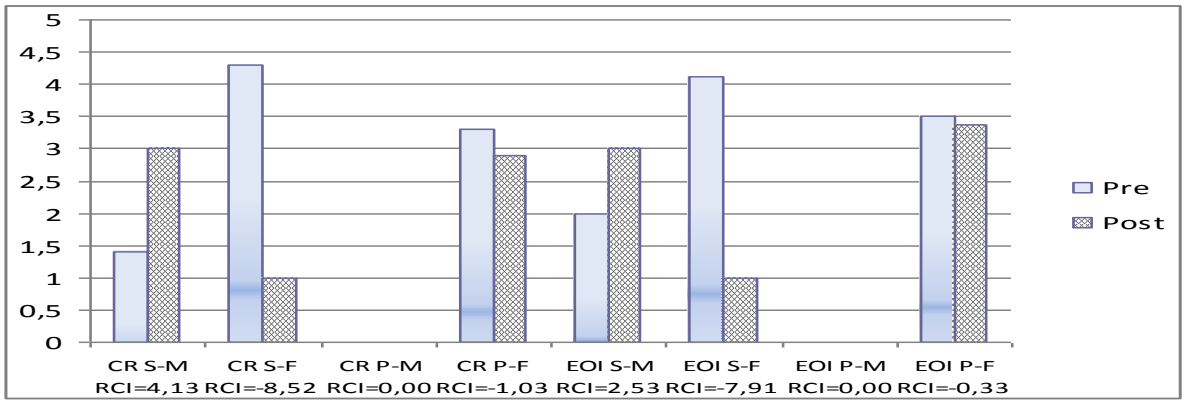

Figure 5.4. Scores on expressed emotions regarding Patient 5's family.

The patient scored deterioration both in critical remarks and emotional over-involvement in relation to the mother. In the relation to the father the patient scored recovery in critical remarks and emotional over-involvement. The father scored no change in critical remarks and emotional over-involvement.

Patient 6

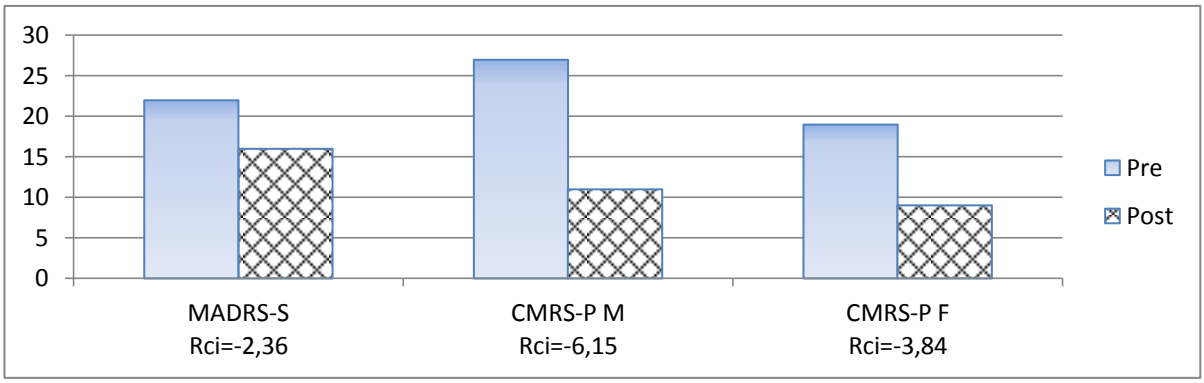

Figure 6.1. Patient 6's symptoms of depression and mania measured pre and post treatment.

This patients scored improvement on MADRS-S. Pre-treatment the mother scored CMRS-P above cut off and the father just below cut off. Post-treatment both parents scored improvement and the mother passing cut off reaching recovery.

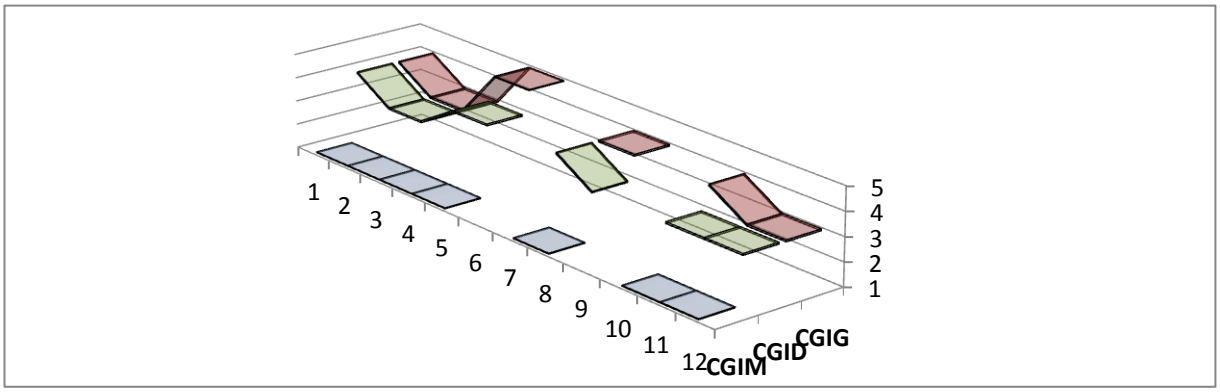

Figure 6.2. Patient 6's severity of mania, depression and overall illness rated by clinicians after each session during treatment.

This patient showed no manic symptoms according to the clinicans who led the group. Regarding severity of depression and overall bipolar illness the patient scores went from moderately ill to mildly ill.

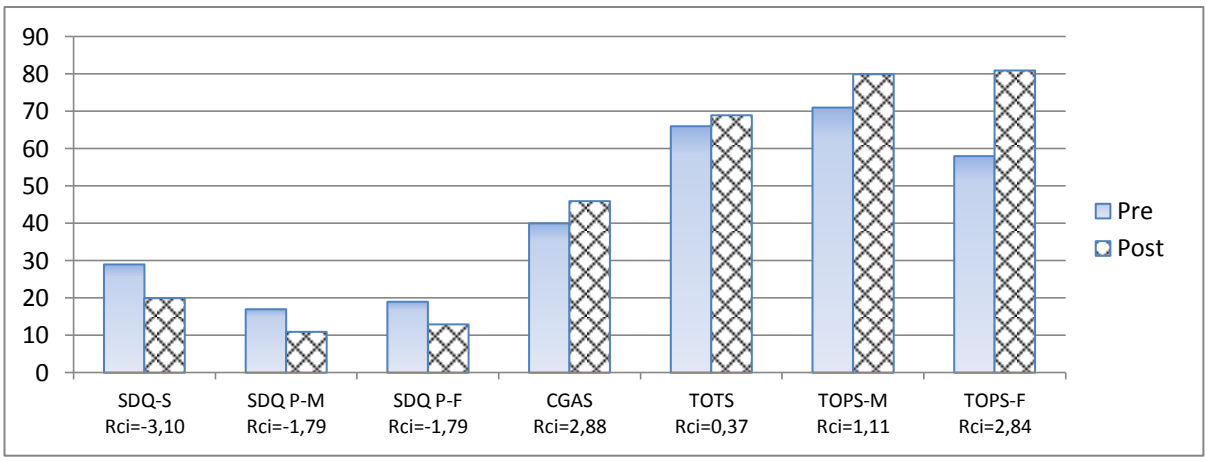

Figure 6.3. Psychosocial function, knowledge and skills measured pre and post treatment (Patient 6).

The patient scored improvement on SDQ-S. The parents scored no change in SDQ-P. CGAS improved, scores both pre and post-treatment were below cut off. The father scored improvement and mother no change on TOPS. The patient scored no change on TOTS.

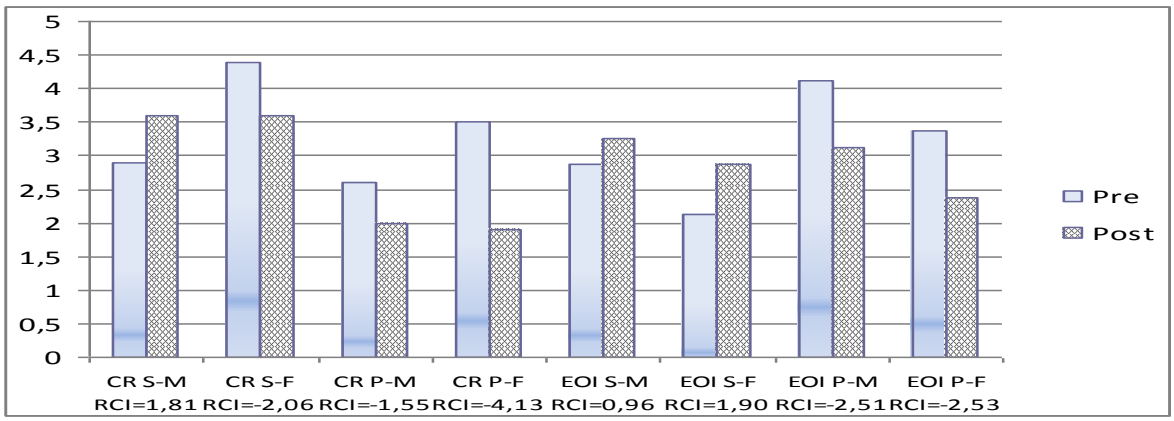

Figure 6.4. Scores on expressed emotions regarding Patient 6's family.

Critical remarks rated by adolescent in relation to the father improved but were still above cut off. Critical remarks rated by the father in relation to the adolescent showed recovery. The parental over-involvement decreased at the level of improvement for the mother and at the level of recovery for the father.

Patient 7

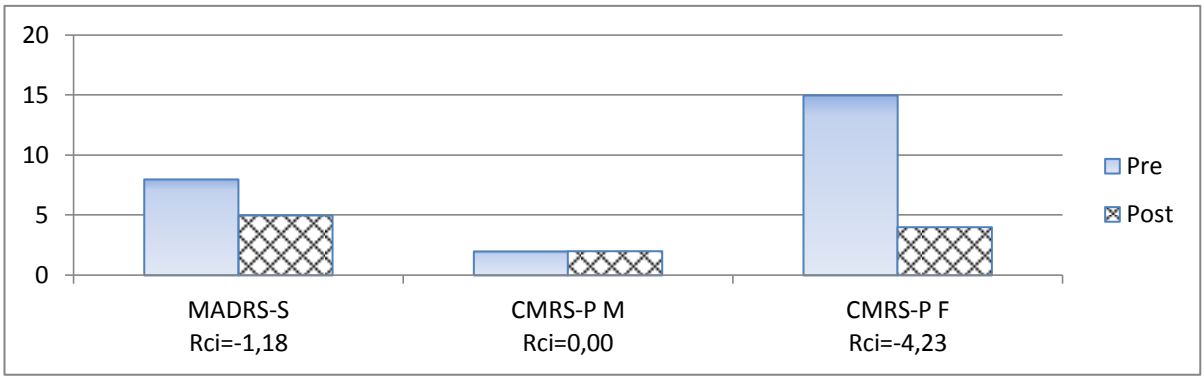

Figure 7.1. Patient 7's symptoms of depression and mania measured pre and post treatment.

The patients scored no change in MADR-S. Mother scored no change in CMRS-P. The father scored improvement in CMRS-P.

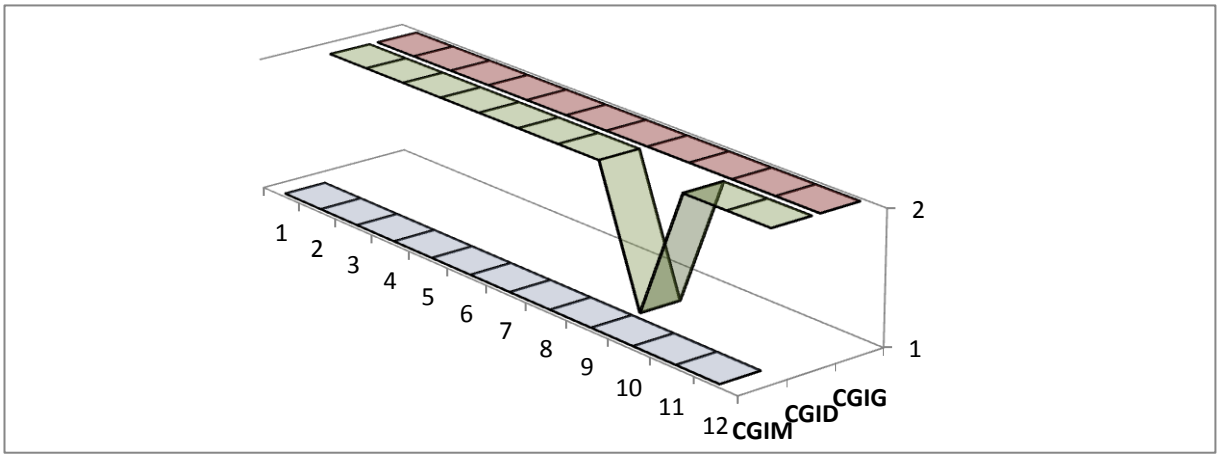

Figure 7.2. Patient 7's severity of mania, depression and overall illness rated by clinicians after each session during treatment.

The group leaders ratings of manic symptoms was low. The ratings of the patients severity of depression and overall bipolar illness was "borderline"

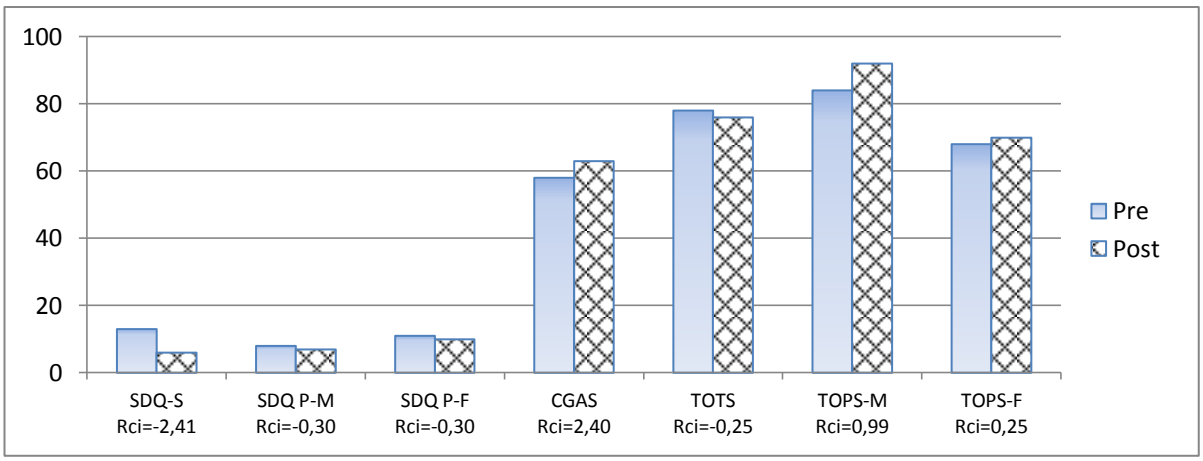

Figure 7.3. Psychosocial function, knowledge and skills measured pre and post treatment (Patient 7).

The patient scored improvement in SDQ-S however pre-treatment scores was below cut off. The parents scored below cut off pre-treatment and no change in SDQ-P. CGAS improved below cut off. The patient and parents scored no change in TOPS and TOTS.

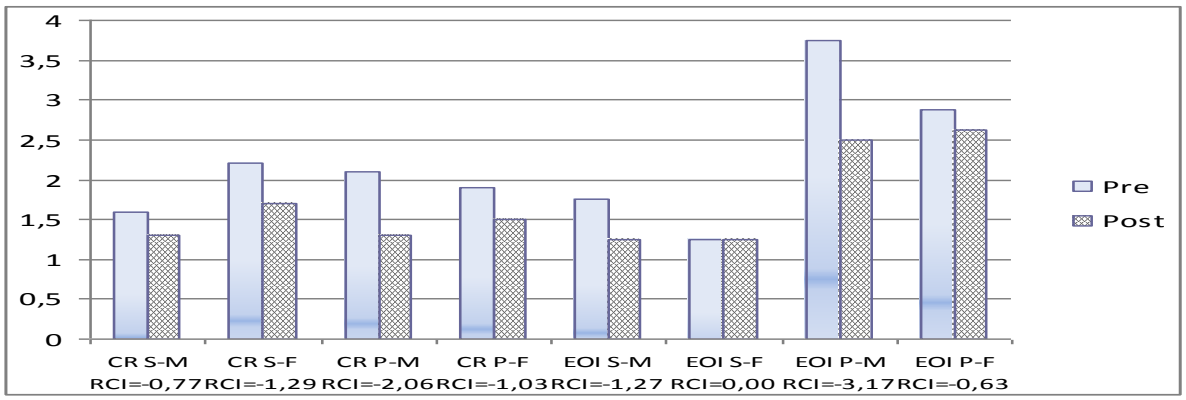

Figure 7.4. Scores on expressed emotions regarding Patient 7's family.

The patient scored no change in critical remarks and emotional overinvolvement in relation to the parents. Critical remarks scored by the mother schowed recovery. The mother scored improvement in emotional over-involvement. The father scored no change in critical remarks or emotional overinvolvement.

Patient 8

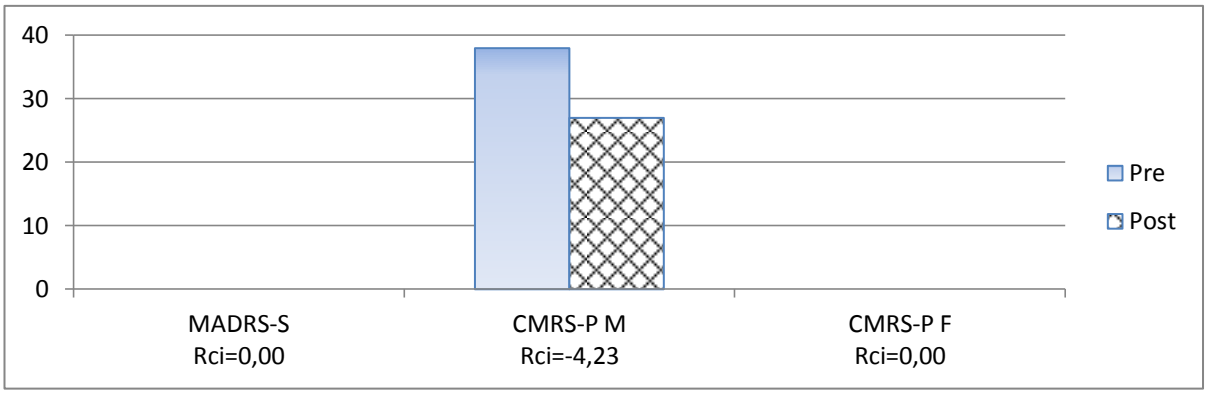

Figure 8.1. Patient 8's symptoms of depression and mania measured pre and post treatment.

The CMRS scored by the mother was improved above the level off cut off.

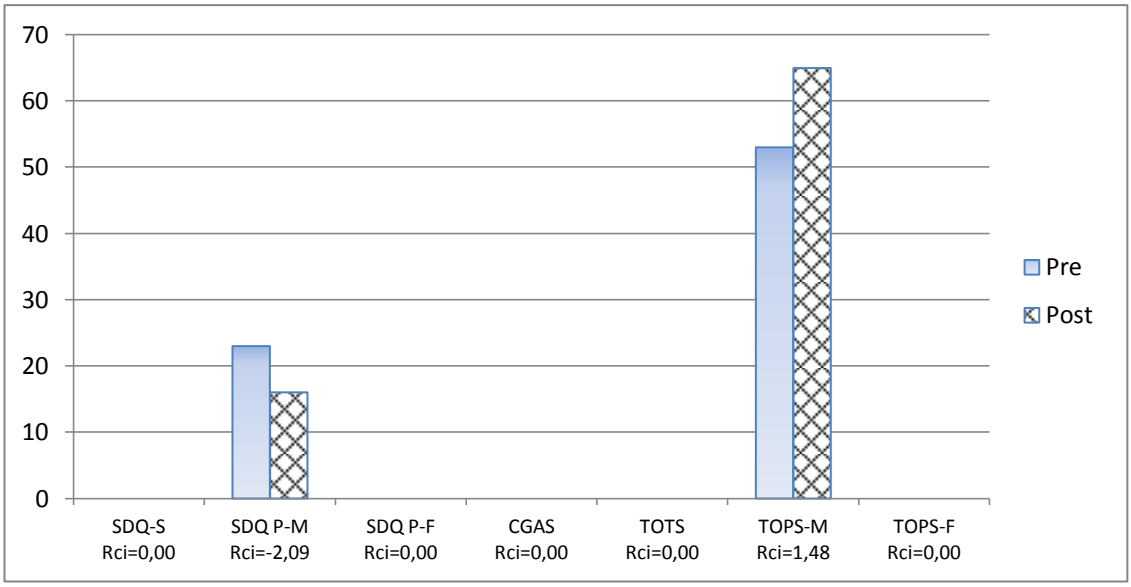

Figure 8.3. Psychosocial function, knowledge and skills measured pre and post treatment (Patient 8).

The mother scored improvement in SDQ, above level cut off . The mother scored no change in TOPS.

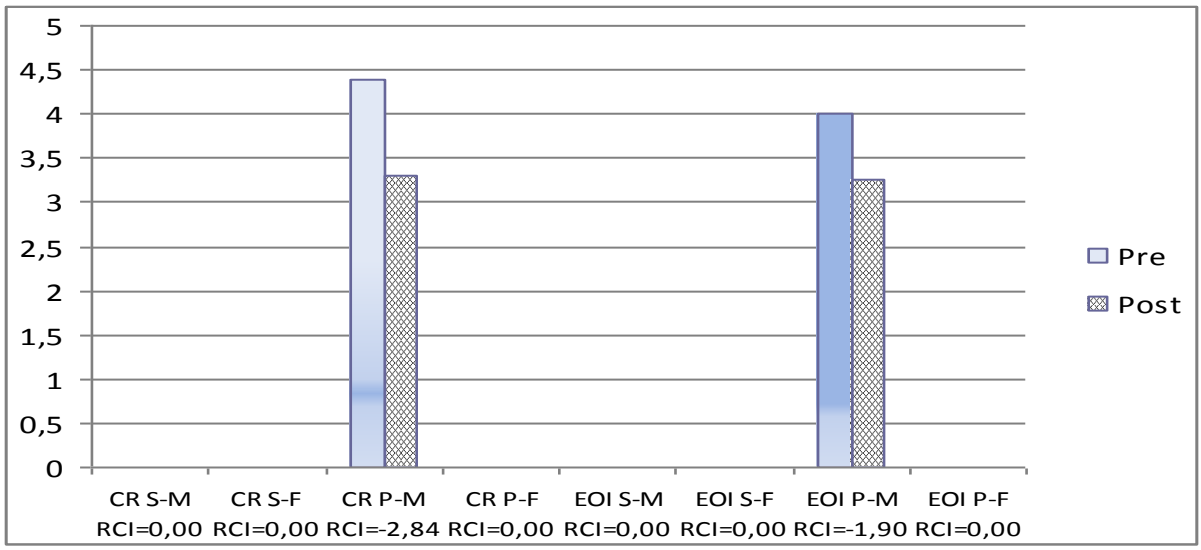

Figure 8.4. Scores on expressed emotions regarding Patient 8's family.

The mother scored improvement in critical remarks, and no change in emotional over-involvement. Both measurements above cut off pre and post-treatment indicating a high EE.

For each patient and parent, the summaries of their categorization are presented in Table 4, 5 and 6.

Table 4

Summary of Classification after Treatment of Patients' Measurements

Patient	MADRS-S	SDQ-S	QAFM CR S-M	QAFM CR S-F	QAFM- EOI S-M	QAFM EOIS -F	TOTS
1	Deteriorated	UC	UC	UC	UC	Deteriorated	UC
2	UC	UC	UC	UC	UC	UC	UC
3	Deteriorated	UC	UC	UC	UC	UC	UC
4	UC	UC	UC	UC	UC	UC	UC
5	UC	UC	Deteriorated	Recovered	Deteriorated	Recovered	UC
6	Improved	Improved	UC	Improved	UC	UC	UC
7	UC	Improved	UC	UC	UC	UC	UC

Note. UC = unchanged

Table 5

Summary of Classification after Treatment of Parents' Measurements

Parent	CMRS-P	SDQ-P	QAFM CR	QAFM- EOI	TOPS
1 M	Improved	Recovered	Improved	Improved	UC
1 F	Improved	Recovered	UC	UC	UC
2 M	UC	Improved	UC	UC	UC
3 M	Deteriorated	UC	UC	UC	UC
4 M	Deteriorated	UC	UC	UC	UC
5 F	UC	UC	UC	UC	UC
6 M	Recovered	UC	UC	Improved	UC
6 F	Improved	UC	Recovered	Recovered	Improved
7 M	UC	UC	Recovered	Improved	UC
7 F	Improved	UC	UC	UC	UC
8 M	Improved	Improved	Improved	UC	UC

Note. UC = unchanged

Table 6

Summary of Classification after Treatment of Clinicians' Measurement

Patient	**CGAS**
1	Improved
2	Improved
3	Improved
4	Improved
5	Improved
6	Improved
7	Improved

Statistical significance

Paired t-tests were conducted to determine changes in a number of variables for the adolescent- and parent groups separately (see Table 7). Results show that parents reported significant improvement of their children's overall psychosocial functioning as measured by SDQ (p = .006). Parents also reported greater knowledge and skills in coping with their child's disorder as measured by TOPS (p = .009), improved relationships with their child regarding critical comments (p = .025) and emotional over-involvement (p = .037). No significant change was found between pre-treatment and post-treatment on measure of manic symptoms by CMRS-P (p = .110).

Table 7

Clinical Measures Pre and Post-treatment

	Mean (SD)				
Measure	Pre-treatment	Post-treatment	t	p	Cohen's d
Parents (N=11)					
SDQ-P	19.18 (8.43)	13.27 (4.82)	3.46	.006	1.09
QAFM CR-P	2.83 (0.93)	2.25 (0.91)	2.63	.025	0.83
QAFM EOI-P	3.46 (0.59)	2.99 (0.36)	2.40	.037	0.76
CMRS-P	21.82 (15.46)	16.86 (12.74)	1.75	.110	0.55
TOPS	68.55 (10.76)	76.77 (8.03)	-3.24	.009	1.03
Adolescents (N=7)					
TOTS	71.57 (6.75)	74 (5.86)	-2.00	.092	0.82
SDQ-A	15.00 (7.46)	13.86 (5.37)	0.60	.569	0.25

Discussion

Results of the present study show that after the RAINBOW program parents and clinicians rated the adolescents' psychosocial functioning as improved. The parent rated their given Expressed Emotions as lower and their knowledge and skills to cope with BD as higher. Ratings of manic and depressive symptoms after treatment varied from deterioration to recovery. Attendance was high both in the adolescent and the parent groups. Feedback from clinicians and parents were mainly positive, suggesting that CFF-CBT seems feasible for parents, adolescents and clinicians in Sweden. Except for minor changes in phrases and wording, the manual does not seem to require "cultural" adaption to Swedish clinicians, parents and patients aged 13-18 years old.

The results of this study show great similarity to the pilot study of the RAINBOW-program in a group treatment format (West et al., 2009), concerning improvement on parental knowledge, skills to cope with the disorder, and parental ratings of their children's psychosocial functioning. The same large variance in the sample data was also observed. In line with this study there was no significant improvement in the patients own ratings of their psychosocial function measured by SDQ. The high ratings of Emotional Expressions among the families in this study are similar to what has been found in other studies of BD (Miklowitz et al., 2009) and psychotic disorders (Hansson & Jarbin, 1997).

The RAINBOW treatment is composed of many different interventions. What mediate the treatment effects is not clear. One can suppose that increased knowledge about the disorder and skills to communicate, cope with problem-solving, increased ability to regulate affects all lead to a lower experience of stress in everyday life both for the individual but also for the family interactions. This stress reduction could be one, among others, important mediator of treatment effects.

An observation made in this study was that the three parental couples participating together rated their child's manic symptoms surprisingly different. In two of the couples the mothers scored higher and in one couple the father scored higher. It could be that a parent being closer to the child in everyday life handling more of the conflicts and distress might score higher symptoms because of a greater knowledge and experience. Or it could be that an exhausted or depressed parent exaggerates the symptoms out of personal sensitivity.

Another surprising observation was that the adolescents´ critical remarks in relation to their mothers increased in most cases, while their critical remarks decreased in most cases

in relation to the fathers. This seems independent on which parent participated in the treatment. Interesting to note is that the two cases (3 and 4) which deteriorated in symptoms of mania were the only cases where parental rated Expressed Emotions showed a tendency to be increased after treatment. Commonly high Expressed Emotions is considered to cause symptoms but the effect could perhaps also be bi-directional.

Most parents and all clinicians scored the psychosocial functioning of the adolescent higher after treatment, while most of the adolescents did not rate themselves improved in this aspect. One could interpret this in a positive way, as an increased self knowledge and insight in their difficulties related to the PBD. Another explanation could be that behavioural changes are noted by parents and clinicians before the adolescents are aware of them.

Limitations

There are several limitations to this study. First of all there is no control group which makes it difficult to draw causal inferences about the treatment. The sample is small which makes it harder to detect statistical differences on a group level, which is delicate when working with a patient population with high comorbidity and fluctuations in symptoms as in bipolar disorder, making generalizations more difficult. The small size of the sample might be the reason that no statistical significant change was detected concerning parents´ ratings of mania on CMRS-P, a possible type II error. Another possible source of type II errors is that the criteria of clinical significance are conservative. If a patient has a low degree of problems, a great change is needed in order to classify him/her as improved. And for a very troubled patient with a chronic condition, even a large and clinically meaningful change does not make the patient go over the cut off to the nonpatient population, making it impossible to be categorized as "recovered". All patients CGAS ratings improved, but this result should be interpreted with caution. A source of bias might be that CGAS pre and post treatment was scored on the same sheet of paper, the first rating could have influenced the clinicians' second rating. Also when the therapists scored CGI, the previous scores were visible, perhaps producing a bias. The feedback discussions held with parents and clinicians were also prone to be biased for social reasons. Another procedure could have been to use anonymous evaluation forms.

Other limitations are that the manual has not been backtranslated. The group leaders were experienced but not specifically trained in this method. Adherence to the treatment manual was not controlled for, except in the feedback discussion after treatment finished.
A general weakness with self reports is that they require a good self-consciousness and insight in your own difficulties, not always present among bipolar adolescents. Parents are probably more reliable in most cases, however in some instances highly emotional interactions with a child can colour the parents report and give "halo" effects. It would have been beneficial for the study to have relied more on independent raters and perhaps different, more objective methods, such as biocorrelates or behavioral measurements. That the adolescent's didn´t self report their manic symptoms is also a limitation of the study. It could also be of interest to have parent observations of depressive symptoms.

A confounding factor that could be of importance is that the treatment starts in the beginning of a semester, September and January and is ending in December and in May. After holidays with opportunity to rest and low academic stress, overall symptoms of PBD could be expected to be lower. The stressors from school and activities normally peek in December and May which are known triggers for new episodes and mood instability. A final limitation is the lack of follow up data. This however is being collected and is planned to be reported later.

One of the contributions of this study is that it is the first evaluation of the program with focus on adolescents with PBD. The case series design helps illustrate the individuality of change. Using informants with different perspective; clinicians, parent, adolescents gives a broad picture of what happened in treatment and may increase the understanding of the response to the treatment program for each unique case. The symptoms were followed weekly in order to capture any mood swings in between pre- and post-treatment. The combination of clinical significance on an individual level with statistical methods on a group level makes it possible to describe and further interpret the cases that did not improve or even deteriorated. This can open for new questions concerning for whom the treatment is effective and why. It could also be mentioned that this study design and approach on adolescents with BD has not been published before.

Implications for Future Research

Future studies on CFF-DBT should explore mediators and moderating factors, for example parent level of education and patient cognitive abilities. A larger sample and an experimental design could make it possible to differentiate patients by comorbidity and and study differential treatments effects. An important issue is to understand more about correlations and causal effects between bipolar disorder and expressed emotions in the family. It would also be of importance with future longitudinal designs in order to evaluate the maintenance of treatment effects, with and without booster sessions. In order to develope the treatment program further, it could be useful to use a qualitative design to study how the program is experienced by patients and parents and how it is effecting their quality of life.

Conclusion

The general purpose of this study was to evaluate if CFF-CBT is effective and feasible for adolescents with PBD and their parents in Sweden. The results of this study show that Expressed emotions, psychosocial function and knowledge and skills to cope with the disorder as rated by the parents were improved. An effective psychosocial intervention including parents to complement medication seems crucial in treatment of PBD. With minor adaptations the RAINBOW program is feasible and to some extent effective also for adolescents and their parents in a North European cultural context.

Feedback from parents

(Parents were encouraged to voice both positive and negative feedback. Parents were also encouraged to tell if they found any particular strategies helpful)

Very valuable to meet likeminded. And share experiences To get new thoughts and ideas. To get confirmation that you´re not alone To unburden your heart to others who can listen. Encouragement and more energy. To get an understanding of what it´s all about. A neutral place to discuss. You get a feeling for the other families. Get to forget your own troubles for a while. Felt safer in the group as time got by. Had the courage to share more and more. Got strategies. Have thought about “backing off”, (anger management). Good for you to meet others who are in the same situation. You recognize yourself in others. Everyone has been

very active in the group. A help to get courage to tell others. Good to learn about giving positive feedback and praise. Would have wanted sessions to last a little bit longer. I would want them to start a bit later, 4 pm instead of 3 pm in order for our children not to miss school and have a chance for a short break. Good to learn about active listening. -"I manage that at work but I have to try hard at home". The group had the indirect effect that my husband who didn´t want to come here, now has accepted that our child has bipolar disorder. And I got my husband to visit a psychologist.

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