Received: 30 August 2022 | Revised: 16 November 2022 | Accepted: 9 December 2022

DOI: 10.1002/cpp.2817

RESEARCH ARTICLE

WILEY

Schema therapy for personality disorders in autistic adults: Results of a multiple case series study

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Funding information
The authors did not receive support from any organization for the submitted work.

Abstract

Background: To our knowledge, treatment of personality disorder (PD) comorbidity in autistic adults is understudied and is still in its infancy. We investigated the effectiveness of schema therapy (ST) for autistic adults with PD.

Method: A multiple case series design with 12 adults (aged 19–62 years) was used with baseline, exploration, ST (with cognitive behavioural and experiential techniques) and follow-up conditions. Participants rated dysfunctional core beliefs (primary outcome) weekly during baseline and treatment and monthly during follow-up. Schema modes, general mental health symptoms, social responsiveness, PD traits and common Axis-I mental disorders were assessed.

Results: Mixed model analyses indicated significant effects of ST with medium to large effect sizes for dysfunctional core beliefs, functional schema modes, PD traits, general mental health symptoms and social responsiveness. Results remained stable during follow-up.

Conclusion: The results of this study indicate that ST might be effective in decreasing dysfunctional core beliefs, PD traits and general mental health symptoms and in increasing functional schema modes and social responsiveness. Improvements persisted over time. ST seems effective in treating PD in autistic individuals.

KEYWORDS
autism, autistic adults, multiple case series design, personality disorder, schema therapy

1 | INTRODUCTION

Autism spectrum disorder (ASD) is considered a neurodevelopmental disorder with an early childhood onset. The core characteristics are persistent challenges in social communication and social interaction over a variety of contexts and a tendency for repetitive patterns of behaviour, interests or activities, and sensory issues including hyper- or hyporeactivity to stimuli or unusual interest in stimuli (American Psychiatric Association [APA], 2013, p. 50). A 1% prevalence of ASD is estimated in the general population (Brugha et al., 2016). The male–female ratio and the male–female differences in ASD presentation have been subject of debate in recent years (Ferri et al., 2018). The cause of ASD is not yet fully understood, but genes influenced by environmental factors are of importance (Waterhouse et al., 2016). High rates of mental health conditions are common in autistic adults, ranging from 59%–69% having mental disorders (Buck et al., 2014; Kentrou et al., 2021) to 79% meeting criteria for a mental disorder at least once in their lives, with depression and anxiety as most common (Lever & Geurts, 2016). The prevalence of meeting personality disorder (PD) criteria ranges from 48% to 62% among autistic adults (Hofvander et al., 2009; Lugnegård et al., 2012). Results of a PD meta-analysis (Vuijk et al., 2018) show that obsessive–compulsive (32%), paranoid (24%), schizoid (24%), avoidant (23%) and schizotypal (14%) PDs are most common in autistic adults.

The increased recognition of personality pathology in autistic adults asks for interventions for personality pathology for them. Until now, it is unknown whether there are effective and evidence-based psychosocial interventions for autistic adults with PD. The National

Institute for Health and Care Excellence (NICE) guideline on recognition, referral, diagnosis and management of adults on the autism spectrum (NICE, 2012) recommends interventions informed by existing NICE guidance for the specific mental health condition with adaptations for ASD. The suggested adaptations are, for example, a more concrete and structured approach, a use of written and visual information and making rules explicit and explaining the context (NICE, 2012). When treating a mental health condition, the therapist, therefore, must have an understanding of the challenges of autistic adults and their possible impact on the treatment of mental disorders (NICE, 2012). Moreover, individually tailored treatment is highly desirable according to a survey exploring the experiences of treatment and support of autistic adults (Camm-Crosbie et al., 2019). The therapist needs to recognize that autistic individuals often think differently as compared to non-autistic people, therefore the therapist needs to be able to translate the concepts and components of the therapy in various ways (Gaus, 2019). Systematic reviews exploring the effectiveness of psychological treatment interventions for autistic adults emerge more and more and demonstrate the effectiveness of treatment especially for comorbid depressive and anxiety disorders. A review of cognitive behavioural therapy (CBT) suggested CBT to be helpful in decreasing comorbid mental health symptoms in autistic adults (Binnie & Blainey, 2013). Moreover, CBT including behavioural, cognitive and mindfulness techniques seems to be moderately effective for comorbid anxiety and depressive symptoms in autistic adults (Spain et al., 2015). Also, a review focusing solely on the effects of mindfulness interventions on autistic adults indicated a reduction in anxiety, depression and rumination and an increased positive affect (Cachia et al., 2016). In yet another review, preliminary evidence for the effectiveness of traditional treatments of posttraumatic stress disorder in autistic individuals (age range 6 to 45 years) was found (Rumball, 2019). A review of psychosocial interventions targeting the specific ASD challenges like applied behaviour analysis and social cognition training provided largely positive results for autistic adults (Bishop-Fitzpatrick et al., 2013). To our knowledge, studies examining PD treatment in autistic adults with PD are lacking. The central question in the current study is whether an established therapy for PD in adults, that is, schema therapy (ST), can also be used as intervention for autistic individuals with PD.

ST, developed by Young (1990), for treating PDs is an integrative model of psychotherapy combining theory and techniques from CBT, psychoanalytic object relations, attachment and other developmental theories and Gestalt and other experiential therapies (Young et al., 2003). Particularly with the more complex and severe PDs, the schema mode model is one of the key elements of ST. Young et al. (2003) identified 10 schema modes each reflecting a constellation of emotions, cognitions and behaviours, resulting from the activation of an early maladaptive schema and a specific way of coping with the schema activation. Whereas schemas are trait-like concepts, schema modes refer to states that people can be in. The goal of ST is to reduce dysfunctional schema modes and to increase functional schema modes and to diminish the strength of the underlying early maladaptive schemas. Schema therapists make use of cognitive behavioural and experiential techniques. There is more and more evidence that ST is an effective treatment for PDs in general (Bamelis et al., 2014; Giesen-Bloo et al., 2006) and also in special populations like older adults with PD (Videler et al., 2018). Will autistic adults with PD be the next special group for which ST can be an effective treatment?

Key Practitioner Message

- Past research suggests the possibility of personality disorder (PD) comorbidity in autistic adults.
- Treatment of PD in autistic adults is understudied.
- PD interventions for autistic adults with PD are needed.
- We propose the value of schema therapy (ST) interventions for autistic adults with PD.
- In our case series, ST interventions were found to be helpful in reducing dysfunctional core beliefs, PD traits and general mental health symptoms and improving functional schema modes and social responsiveness in autistic adults with PD.

Autistic people have a vulnerability to maladaptive schema development because of their challenges processing information about others, self and nonsocial information resulting in struggling with social skills and self-management (Gaus, 2019). A study of Oshima et al. (2015) revealed significantly higher scores for all the early maladaptive schemas, apart from 'self-sacrifice' and 'approval/recognition seeking' in autistic adults compared to non-autistic adults. Early maladaptive schemas appear to account for mental health conditions in adults with autism spectrum conditions (Oshima et al., 2014). A single-arm preliminary study with an open trial design ($N = 10$, aged 20 to 39 years, 5 males, 5 females) indicated that individual ST is applicable as a treatment for young autistic adults with mental health conditions (no PD), showing a significant reduction in early maladaptive schemas and improvement in quality of life and social adjustment (Oshima et al., 2018; Oshima et al., 2021).

For our ST study, ST did not differ substantially for autistic people compared to people with other diagnoses, but some special considerations and modifications for the therapist were (1) setting clear expectations about the role of the therapist and the client: setting a realistic pace, using concrete language, validating the client's experience and providing constructive feedback (Gaus, 2019); (2) repeated psychoeducation of the ST concepts and interventions, explaining or discussing in detail what has been done and what it means for the client's here-and-now situation; and (3) taking into account the autism-specific needs and challenges. For more 'dos and don'ts', we refer to a case study exemplifying the ST approach in an autistic adult with PD (Vuijk et al., 2022).

The aim of the present study was to assess the effectiveness of individual ST as a treatment for PDs in autistic adults. Our hypotheses were that ST would lead to a decrease of dysfunctional core beliefs and schema modes, PD traits, general mental health symptoms and Diagnostic and Statistical Manual of mental disorders (DSM-IV) axis-I mental disorders, an increase of functional schema modes and an improvement in social responsiveness.

2 | METHODS

2.1 | Participants

Participants were 12 clients from Sarr Autism Rotterdam, a mental health institute specialized in ASD in the Netherlands. Inclusion criteria were (1) primary diagnosis of ASD following the DSM-IV or DSM-5, and a PD following DSM-IV. The PD was assessed with the Dutch version of the Structured Clinical Interview for DSM-IV Personality Disorders (SCID-II; Weertman et al., 2000); (2) age 18 to 65 years old; (3) at least a completed primary and secondary education; and (4) having insight into their own personality and recognition of their psychological functioning (based on the impression of the clinical psychologist). Exclusion criteria were (1) schizophrenia or other psychotic disorder; (2) antisocial PD; (3) eating disorder; (4) mental disorder secondary to medical condition; (5) intellectual disability (IQ < 80); (6) addiction requiring clinical detox; and (7) current suicidal ideation. During the ST treatment, no other form of psychotherapy was allowed except supportive therapy if needed. Pharmacotherapy was allowed. The 12 participants were recruited from 15 clients screened for participation; three declined to participate. Figure 1 presents the participant flow recruitment process.

2.2 | Design

We intended to use a multiple baseline case series design (Vuijk & Arntz, 2017), because, just like a randomized controlled trial (RCT), it demonstrates the occurrence of a change over time as being the result of an intervention (Hawkins et al., 2007; Onghena, 2005). But because of a misunderstanding, the 4 to 9 baseline weeks (nonintervention waitlist weeks, during which participants rated their dysfunctional core beliefs without meeting the therapist) were immediately followed by 1 to 6 weeks of supportive sessions (in which rating the dysfunctional core beliefs continued and meeting the therapist started who supported the participant by listening, advising and encouraging) without an assessment between baseline weeks and supportive sessions, finally resulting in a 10-week period of waitlist weeks and supportive sessions for all participants (Vuijk & Arntz, 2017). Further, because of the small numbers of supportive sessions in a considerable subsample of participants, time within supportive sessions' condition could not be reliably estimated for our primary outcome analysis. We therefore combined waitlist weeks and supportive sessions in one condition, which for convenience reasons was labelled as baseline. So, during the 10-week baseline period, six supportive sessions started in week 5 for participants 1 and 2, five supportive sessions started in week 6 for participants 3 and 4 and so on till one supportive session started in week 10 for participants 11 and 12.

Thus, we used a multiple case series design. The design is practical, because it requires fewer participants than an RCT. The loss of power is compensated by the fact that participants serve as their own controls and by the large number of assessments of the primary outcome.

In this study, there were a control condition (baseline), an exploration condition, two ST conditions (cognitive behavioural and experiential techniques) and two follow-up conditions in a within-subject design. This design precludes blinding of treatment. We randomized the order of starting with either cognitive behavioural or experiential techniques to differentiate between time effects and treatment effects.

2.3 | Measures

The primary outcome measure was based on discussion with each participant at screening. Three dysfunctional beliefs were formulated

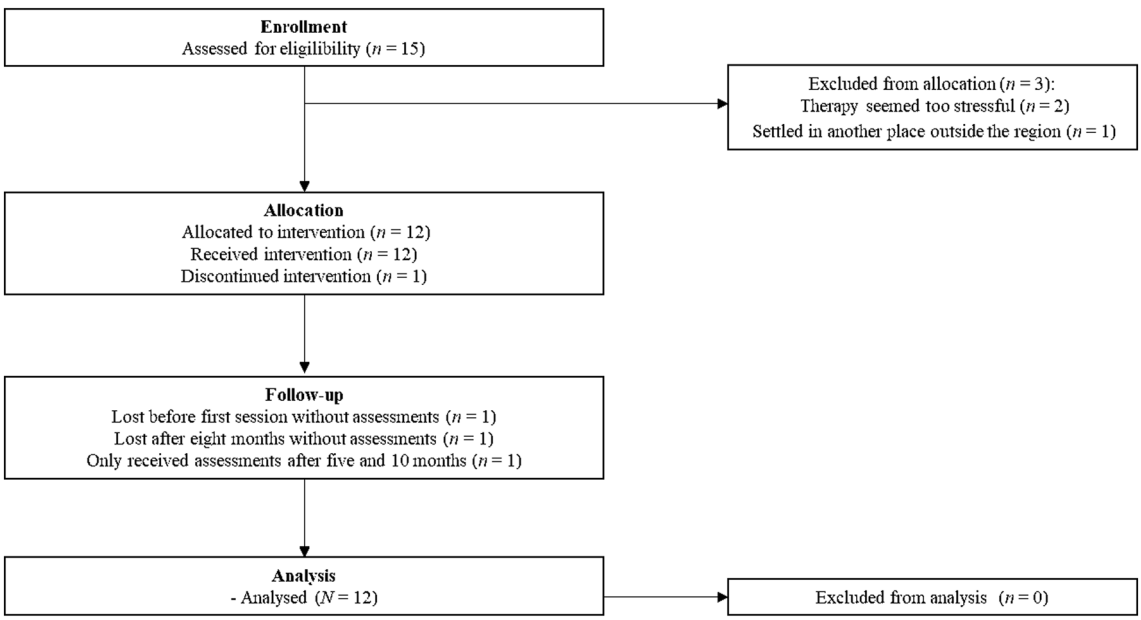

FIGURE 1 Consolidated Standards of Reporting Trails(CONSORT) flow diagram depicting the recruitment process

by the participant with the help of a registered clinical psychologist. Participants rated the degree to which they believed in each statement on 100 mm visual analogue scales (VAS; Freyd, 1923). The average sum score of the three dysfunctional beliefs constituted the final primary outcome.

The first secondary outcome measure was the Schema Mode Inventory (SMI; Young et al., 2007). The SMI contains 118 items with a 1–6 point frequency scale for frequency assessing 14 schema modes. The SMI showed acceptable internal consistencies of the 14 modes, adequate test–retest reliability and moderate construct validity (Lobbestael et al., 2010). We analyzed the dysfunctional, Happy Child (functional) and Healthy Adult (functional) mode scores.

The second secondary outcome measure was the Symptom Check List (SCL-90; Arrindell & Ettema, 2003), a 90-item self-report questionnaire, assessing general mental health symptoms during the last week. Each item is rated on a 1–5 point scale for severity, resulting in a total score of 90 to 450. The SCL-90 showed good reliability and high internal consistency (Holi, 2003). We analyzed the severity of general mental health symptoms.

The third secondary outcome measure was the Social Responsiveness Scale – Adult version (SRS-A; Constantino, 2005; Noens et al., 2012), a 64-item self-report questionnaire, measuring dimensions of interpersonal behaviour, communication and rigid, repetitive behaviour and interests, characteristic for autistic adults. Each item is rated on a 1–4 point scale. The SRS-A showed average to good internal consistency and test–retest reliability, adequate content validity and good congruent validity (Noens et al., 2012). The SRS-A was used to assess improvement in social responsiveness by analyzing total scores.

The fourth secondary outcome measure was PD criteria of the Structured Clinical Interview for Axis-II Personality Disorders (SCID-II; First et al., 1997), assessing the 10 DSM-IV PDs (APA, 2000). Each SCID II criterion has a scoring range of 1 to 3. The SCID-II showed excellent inter-rater agreement (Lobbestael et al., 2011). We used the sum score of the 1–3 ratings.

The fifth secondary outcome measure, added by a post-hoc decision during the trial, was the number of DSM-IV axis-I mental disorders assessed with the Standardized Assessment for Mental disorders (SAM; Hoogduin, 1999), a Dutch semi-structured interview to assess 16 DSM-IV axis-I mental disorders most commonly seen in clinical practice. The SAM consists of 16 questions and subquestions. Psychometric properties of the SAM have not been studied yet. We analyzed the total scores of the 16 questions.

2.4 | Assessments

All participants rated the VAS dysfunctional core beliefs weekly during baseline, exploration and cognitive behavioural and experiential techniques and monthly during follow-up. Participants completed the Dutch SMI, SCL-90 and SRS-A at screening after supportive sessions, exploration, cognitive behavioural techniques, experiential techniques and at 5- and 10-month follow-up. The Dutch SCID-II and the SAM were assessed at screening and 5- and 10-month follow-up.

2.5 | Procedure

Autistic adults with PD, meeting inclusion criteria, were approached by the first author, until the number of 12 participants was reached. The screening procedure consisted of two sessions screening for eligibility to participate, based on the inclusion and exclusion criteria, and in which the dysfunctional core beliefs were formulated. The ASD diagnosis was verified by studying the diagnostic report. ASD had to be diagnosed by a registered psychologist or psychiatrist. PD(s) and common DSM-IV axis-I mental disorders were classified by using the SCID-II and SAM. We further assessed background information like sex, age, level of education, marital status, employment status, health and medication use.

After the screening and 10-week baseline period (including nonintervention waitlist weeks and supportive sessions), a 5-week exploration followed with weekly sessions during which current and past functioning, psychological symptoms, early maladaptive schemas, dysfunctional core beliefs and schema modes were explored, and information about the treatment was provided. Then, 15 weekly sessions with cognitive-behavioural ST techniques were given followed by 15 weekly sessions with experiential ST techniques (or vice versa). Finally, there was a 10-month follow-up with monthly ST booster sessions. Table 1 shows an overview of the assessments and sessions per condition for all participants: The ST was preceded by a 10-week baseline period and a 5-week exploration. Participants started meeting the therapist in the supportive sessions.

2.6 | Therapist training

The screening was conducted by a registered clinical psychologist (RV), qualified for and experienced in diagnostic assessments of ASD in adults. The follow-up assessments were administered by a health care psychologist qualified for and experienced in diagnostic assessments of ASD in adults.

All therapists in this study were trained in ST, registered as a healthcare psychologist, and working with autistic adults for at least 5 years. To optimize treatment integrity, therapists received a four-day ST training. During the study, the therapists were two-weekly supervised by a clinical psychologist, a certified ST supervisor. All sessions were audiotaped, and a random sample of at least three tapes per participant was rated with the Schema Therapy Rating Scale for individual therapy sessions (STRS-I-1; Young & Fosse, 2005) by two judges to formally document treatment integrity. The STRS-I-1 was used to facilitate the blind assessment of the therapists whether they used the type of techniques associated with the phase concerned.

2.7 | Statistical analysis

Data analyses were conducted using SPSS. A mixed model analysis was used with time, condition and interaction as fixed predictors, applied successfully in previous case series studies (Arntz et al., 2013;

TABLE 1 Overview of the assessments and sessions per condition for the participants ($N = 12$)

Participants	A1*	Waitlist weeks**	Supportive sessions**	A2	Exploration sessions	A3	ST sessions 1–15	A4	ST sessions 16–30	A5	Follow-up months 1–5	A6	Follow-up months 6–10	A7
1	x	6	4	x	5	x	15 Exp	x	15 CB	x	5	x	-	-
2	x	5	5	x	5	x	15 Exp	x	15 CB	x	5	x	5	x
3	x	9	1	x	5	x	15 CB	x	15 Exp	x	5	x	5	x
4	x	7	3	x	5	x	15 CB	x	15 Exp	x	5	x	5	x
5	x	8	2	x	5	x	15 Exp	x	9 CB	x	-	x	-	x
6	x	4	6	x	5	x	15 Exp	x	15 CB	x	5	x	5	x
7	x	5	5	x	5	x	15 CB	x	15 Exp	x	5	x	5	x
8	x	9	1	x	5	x	15 CB	x	15 Exp	x	5	x	5	x
9	x	6	4	x	5	x	15 Exp	x	15 CB	x	5	x	5	x
10	x	8	2	x	5	x	15 Exp	x	15 CB	x	5	x	5	x
11	x	7	3	x	5	x	15 CB	x	15 Exp	x	5	x	5	x
12	x	4	6	x	5	x	15 CB	x	15 Exp	x	5	x	3	-

Note: Participant 1 declined after 5-month follow-up. Participant 5 declined after nine sessions of cognitive behavioural therapy (CBT) but still participated in the assessments. Participant 12 declined after 8-month follow-up. Waitlist weeks: nonintervention weeks, during which participants rated their dysfunctional core beliefs without meeting the therapist. Supportive sessions: rating the dysfunctional core beliefs continued, and meeting the therapist started who supported the participant by listening, advising and encouraging. Exploration sessions: rating the dysfunctional core beliefs continued, current and past functioning, psychological symptoms, early maladaptive schemas, dysfunctional core beliefs and schema modes were explored, and information about the treatment was provided.

Abbreviations: A1*, baseline for secondary outcomes; A1–A7, assessment; CB, cognitive-behavioural techniques; Exp, experiential techniques; ST, schema therapy; Waitlist weeks** and supportive sessions**, baseline for primary outcome; x, participant completed assessment; -, participant declined assessment.

Videler et al., 2018). We were not aware of a systematic way to perform power analysis for the design as well as for order of the techniques. As an indication, the study had 80% power to detect a change of Cohen's $d = 1$ or higher at alpha = 0.05, two-tailed, if a paired *t*-test of the pre to post change would be used to evaluate the treatment effect. Effect sizes were calculated as change with baseline SD as denominator (with Cohen's $d = 0.2$ to be considered as a 'small' effect size, 0.5 a 'medium' effect size and 0.8 a 'large' effect size). Change was based on the estimates from the mixed model analysis.

2.7.1 | Primary outcome analysis

For the analysis of the dysfunctional core belief strengths, a model with time, condition (with baseline as reference) and centred time within each condition was run with an AutoRegressive (AR1) structure for the repeated part, and if possible, random intercepts and random slopes for time at the participant level. If the linear time effect became non-significant, and fit of the model did not deteriorate, tested with a Chi-square test on the difference between -2LL values, it was eliminated from the model. Next, non-significant effects with $p \geq 0.10$ were eliminated step by step. We chose for either a random intercept, a random slope for time, or both, based on the best fitting and most parsimonious model, using Chi-square tests on the -2LL value and comparisons of Akaike information criteria (AIC) and Bayesian information criteria (BIC).

We first tested whether cognitive behavioural and experiential conditions differed in effectiveness, controlling for order. If not, the two conditions were combined into one ST condition to simplify the model. Next, we tested whether exploration, ST (or the separate components of ST) and follow-up conditions differed from baseline. We hypothesized that there would be a negative slope of time within ST or within cognitive behavioural and experiential conditions if these conditions would differ, whereas no time effects within baseline condition were expected. We tested the time-within-follow-up effect but did not have a clear hypothesis about it.

2.7.2 | Secondary outcome analysis

For the analysis of SMI, SCL-90 and SRS-A, we used the same approach as for the primary outcome. The reduction in number of symptoms for the initially diagnosed PD using the SCID-II between first (baseline), second (at 5-month follow-up) and last (at 10-month follow-up) measurement was tested using Wilcoxon's signed rank test. For the reduction in number of DSM-IV axis-I disorders assessed with the SAM, we used Friedman's test.

3 | RESULTS

3.1 | Participants

Twelve autistic (nine males and three females) native Dutch speaking individuals participated. Age ranged from 19 to 64 years (mean age: 38 years). Three participants were partnered, nine were single. Level of education ranged from secondary school ($n = 2$), higher vocational education ($n = 7$) to university ($n = 3$). Six participants were employed, three were unemployed and three were student. All participants were diagnosed with at least one PD. We refer to Table 2 for an overview of the demographic data and to Table 3 for an overview of the PD diagnoses and DSM-IV axis-I mental disorder diagnoses of the participants.

TABLE 2 Demographic data of participants ($N = 12$)

Participant	Age	Gender	Marital status	Education	Employment	ASC	Medication
1	62	M	Married	Secondary school	Employed	Asperger's disorder	Antidepressant
2	42	F	Single	Higher vocational education	Unemployed	PDD-NOS	
3	61	M	Single	Higher vocational education	Employed	Asperger's disorder	
4	42	M	Divorced	Higher vocational education	Unemployed	Asperger's disorder	Antidepressant Antipsychotic Anxiolytic
5	23	M	Single	Higher vocational education	Student	Asperger's disorder	
6	30	M	Single	University	Employed	Asperger's disorder	Antidepressant
7	36	F	Married	Secondary school	Employed	PDD-NOS	
8	44	M	Single	University	Employed	Asperger's disorder	
9	27	M	Single	Higher vocational education	Student	ASD	
10	52	M	Single	University	Unemployed	Asperger's disorder	
11	19	M	Single	Higher vocational education	Student	PDD-NOS	Antidepressant
12	29	F	Partnered	Higher vocational education	Employed	PDD-NOS	Antidepressant

Abbreviations: ASC, autism spectrum condition; ASD, autism spectrum disorder; F, female; M, male; PDD-NOS, pervasive developmental disorder-not otherwise specified.

TABLE 3 Personality disorder and DSM-IV axis-I mental disorder diagnoses ($N = 12$) assessed at screening (baseline) and 5- and 10-month follow-up

Participant	Screening	After 5-month follow-up	After 10-month follow-up
1	Avoidant, obsessive–compulsive, depressive PDs; PD-NOS dependent	?	?
	Major depressive disorder		
2	Obsessive–compulsive PD	Obsessive–compulsive PD	PDNOS obsessive–compulsive
	Panic disorder with agoraphobia	Panic disorder with agoraphobia	Panic disorder with agoraphobia
	Social anxiety disorder	Social anxiety disorder	Generalized anxiety disorder
	Generalized anxiety disorder	Generalized anxiety disorder	Somatic symptom disorder
		Persistent depressive disorder	
3	Obsessive–compulsive PD	PDNOS obsessive–compulsive, passive-aggressive	PDNOS obsessive–compulsive
	Adjustment disorder with depressed mood	0	Adjustment disorder with depressed mood
4	Avoidant, depressive, borderline PDs	-	PDNOS avoidant
	Social anxiety disorder	Social anxiety disorder	Social anxiety disorder
	Major depressive disorder	Somatic symptom disorder	Somatic symptom disorder
5	Avoidant, depressive PDs; PDNOS dependent, obsessive–compulsive, paranoid	Obsessive–compulsive PD; PDNOS avoidant, depressive	PDNOS obsessive–compulsive
	Attention-deficit disorder	Attention-deficit disorder	Attention-deficit disorder
	Obsessive–compulsive disorder	Obsessive–compulsive disorder	Obsessive–compulsive disorder
	Generalized anxiety disorder		Generalized anxiety disorder
6	Avoidant, obsessive–compulsive, depressive, schizoid PDs; PDNOS dependent, paranoid	-	-
	Major depressive disorder	0	0
	Social anxiety disorder		
	Panic disorder with agoraphobia		
7	Avoidant PD	PDNOS avoidant	Avoidant PD
	0	Anorexia nervosa	Anorexia nervosa
8	Avoidant, dependent, obsessive–compulsive, depressive PDs	Obsessive–compulsive, depressive PDs	Obsessive–compulsive, depressive PDs
	Major depressive disorder	Major depressive disorder	Major depressive disorder
	Social anxiety disorder	Social anxiety disorder	Social anxiety disorder
	Somatic symptom disorder		
9	Obsessive–compulsive, depressive, narcissistic PDs; PDNOS paranoid	Obsessive–compulsive PD	Obsessive–compulsive PD
	Major depressive disorder	Major depressive disorder	Substance dependence
	Generalized anxiety disorder	Substance dependence	
10	Avoidant, obsessive–compulsive PDs; PDNOS passive-aggressive, depressive	Obsessive–compulsive, paranoid PDs	Obsessive–compulsive PD; PDNOS depressive, paranoid
	Persistent depressive disorder	Persistent depressive disorder	Persistent depressive disorder
	Post-traumatic stress disorder	Post-traumatic stress disorder	Post-traumatic stress disorder
11	Obsessive–compulsive, depressive PDs; PDNOS avoidant	Obsessive–compulsive, borderline PDs; PDNOS depressive	Avoidant PD; PDNOS borderline
	Panic disorder without agoraphobia	Major depressive disorder	0
	Social anxiety disorder	Generalized anxiety disorder	

(Continues)

TABLE 3 (Continued)

Participant	Screening	After 5-month follow-up	After 10-month follow-up
12	Avoidant, obsessive–compulsive, depressive PDs Major depressive disorder Persistent depressive disorder Social anxiety disorder Obsessive–compulsive disorder Generalized anxiety disorder	?	?

Note: PD diagnoses assessed with Structured Clinical Interview for DSM-IV Personality Disorders (SCID-II). DSM-IV axis-I mental disorders assessed with Standardized Assessment for Mental disorders (SAM).
Abbreviations: PD, personality disorder; PDNOS, personality disorder not otherwise specified; ?, assessment was not possible; -, no PD diagnosis; 0, no mental disorder diagnosis.

3.2 | Attrition

Participant 1 declined participation at follow-up because of a sick partner. Participant 5 declined after nine sessions of CBT, because he wanted to start pharmacotherapy not indicated by the psychiatrist. Nevertheless, he was willing to continue to participate in the assessments. Participant 12 declined participation at follow-up because of a new job resulting in severe psychological distress. Table 1 shows an overview of the assessments and sessions per condition for all participants.

3.3 | Treatment integrity

To judge treatment integrity, 43 audio tapes (seven supportive sessions, 12 exploration sessions, 12 cognitive behavioural sessions and 12 experiential sessions) were rated by judging the condition and the techniques associated with the condition concerned. Judge one (RV) rated 43 audio tapes and the other judge, a student psychologist (trained and educated in ASD, PD and ST in adults as part of her master thesis), rated 9 of the 43 audio tapes. With both judges, a score of 91% (RV) and 89% (the other judge) and a strong interrater agreement of 0.83 (Cohen's kappa; $p < 0.001$) were found for using the correct techniques per condition by the therapists.

3.4 | Primary outcome

The individual averaged VAS-scores specifying the credibility of dysfunctional core beliefs during the five conditions are shown in Figure 2. Visual inspection shows large decreases for participants 2, 4, 6, 7 and 9. Participants 1, 3, 10 and 12 have more or less unchanged scores. Participants 8 and 11 have an irregular score pattern.

The differences between time effects and main effects of cognitive behavioural and experiential techniques failed to reach significance, controlled for their order of application. The two conditions were therefore combined into one ST condition. For the analysis of all scores, a random intercept had the best fit. A model with only time as fixed predictor revealed a significant effect of time, with belief scores reducing with time, $p < 0.01$. However, when the conditions and time-within-condition were added, the general time effect became non-significant, and model fit did not significantly deteriorate when time was deleted from the model. Table 4 presents the final results of the mixed model analyses after stepwise deletion of time within condition effects with a significance level ≥0.10. In sum, the results showed that the main effects of ST and follow-up conditions were significant with medium to large effect sizes compared to baseline. Time-within-condition was significant with a large effect size for ST, showing a continuing decrease over time of belief strength during treatment, whereas the time effects within baseline were non-significant and were deleted from the model.

3.5 | Secondary outcomes

For all secondary measures, the cognitive behavioural and the experiential techniques did not differ significantly (Table 5), and therefore, the conditions were combined into one ST condition. For all, a random intercept for participant yielded the best fit. For all, except dysfunctional schema modes, a model with condition fitted better than a model with time or with both time and condition. For dysfunctional schema modes, a model with only time had a superior fit, thus any conclusions from tests of condition remain unclear as time (and non-specific factors) might be a better explanation than condition effects. Table 4 and Figure 3 present the final results of the mixed model analyses for the SMI, SCL-90 and SRS-A after adding random intercepts and with a significance level of <0.05.

1. Change in schema modes (SMI)

For Happy Child mode, changes appeared significant with medium to large effect sizes for all conditions except for the first 15 sessions of ST compared to baseline. For Healthy Adult and dysfunctional modes, changes were non-significant, except for Healthy Adult mode after 10-month follow-up with a medium effect size compared to baseline.

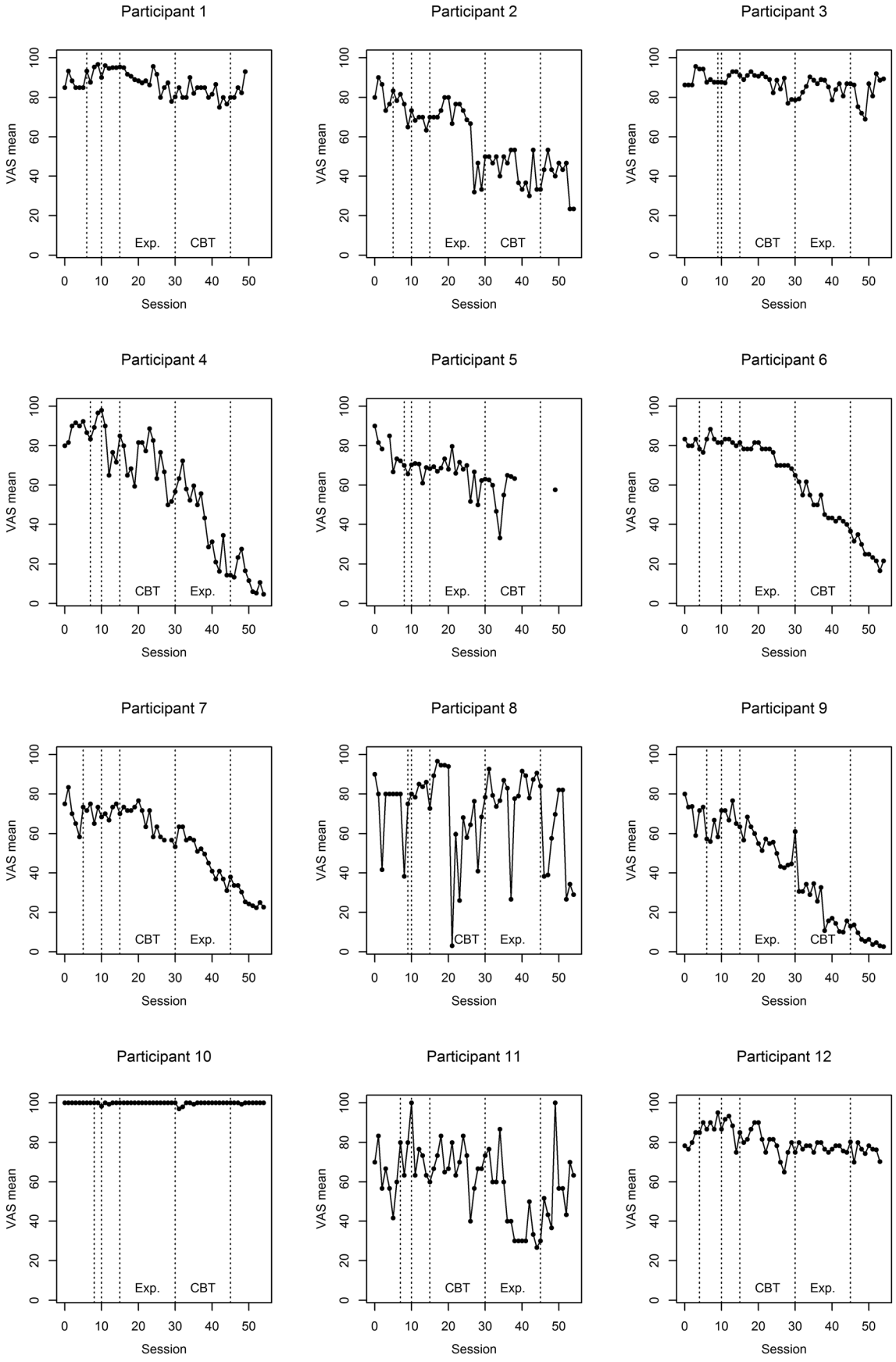

FIGURE 2 Individual average credibility ratings of dysfunctional core beliefs ($N = 12$). Exp, experiential techniques; CBT, cognitive behavioural techniques; VAS, visual analogue scale

TABLE 4 Results of mixed model analyses

	Estimate	SE	df	t	p*	CI	Effect size[a] (Cohen's d)
VAS dysfunctional core beliefs							
Intercept (baseline)	80.78	2.70	14.42	29.97	≤0.001	75.02–86.55	
Exploration	1.16	2.26	240.28	0.51	0.608	−3.29 to −5.61	0.090
ST	−10.92	3.86	20.55	−2.83	0.010	−18.96 to −2.89	0.852
Session in ST	−0.87	0.17	27.09	−5.05	≤0.001	−1.22 to −0.52	2.038
Follow-up	−26.62	6.71	17.17	−3.97	0.001	−40.76 to −12.49	2.078
Session in follow-up	−0.80	0.47	246.57	−1.69	0.092	−1.72–0.13	0.624
SMI Happy Child							
Intercept (baseline)	2.48	0.14	23.10	17.17	≤0.001	2.18–2.77	
After supportive sessions	0.22	0.11	53.08	2.08	0.042	0.01–0.44	0.383
After exploration	0.33	0.13	57.80	2.60	0.012	0.07–0.58	0.574
After 15 ST sessions	0.26	0.14	41.37	1.90	0.066	−0.02–0.53	0.452
After 30 ST sessions	0.61	0.14	30.18	4.28	≤0.001	0.32–0.90	1.061
After 5-month follow-up	0.38	0.15	25.33	2.58	0.016	0.08–0.68	0.661
After 10-month follow-up	0.55	0.15	22.86	3.97	0.001	0.25–0.88	0.956
SMI Healthy Adult							
Intercept (baseline)	3.63	0.17	30.22	22.06	≤0.001	3.29–3.96	
After supportive sessions	−0.03	0.15	50.12	−0.18	0.860	−0.33–0.27	0.042
After exploration	0.08	0.17	59.00	0.49	0.627	−0.26–0.42	0.114
After 15 ST sessions	−0.06	0.18	44.56	−0.32	0.749	−0.42–0.31	0.086
After 30 ST sessions	0.22	0.19	35.23	1.17	0.250	−0.16–0.59	0.315
After 5-month follow-up	0.21	0.19	32.01	1.11	0.275	−0.18–0.60	0.301
After 10-month follow-up	0.49	0.20	30.66	2.47	0.019	0.09–0.89	0.702
SMI Dysfunctional modes							
Intercept (baseline)	2.77	0.13	17.82	20.78	≤0.001	2.49–3.05	
After supportive sessions	0.09	0.09	51.13	0.97	0.335	−0.10–0.28	0.228
After exploration	0.13	0.10	57.84	1.31	0.195	−0.07–0.33	0.329
After 15 ST sessions	0.03	0.10	47.22	0.29	0.770	−0.18–0.24	0.076
After 30 ST sessions	−0.13	0.10	42.18	1.24	0.223	−0.34–0.08	0.228
After 5-month follow-up	−0.13	0.11	41.42	1.27	0.212	−0.35–0.08	0.228
After 10-month follow-up	−0.21	0.11	41.03	1.93	0.061	−0.43–0.01	0.532
SCL-90							
Intercept (baseline)	228.67	13.95	25.57	16.39	≤0.001	199.97–257.37	
After supportive sessions	−9.71	11.89	48.95	−0.82	0.418	−33.61–14.19	0.192
After exploration	−11.80	13.38	56.35	−0.88	0.381	−38.60–15.00	0.233
After 15 ST sessions	−21.00	13.74	39.39	−1.53	0.134	−48.78–6.78	0.415
After 30 ST sessions	−35.17	13.97	31.43	−2.52	0.017	−63.63 to −6.70	0.694
After 5-month follow-up	−28.42	14.35	29.29	−1.98	0.057	−57.77–0.93	0.561
After 10-month follow-up	−53.15	14.80	28.46	−3.59	0.001	−83.45 to −22.85	1.049
SRS-A							
Intercept (baseline)	93.75	6.76	18.31	13.86	≤0.001	79.56–107.94	
After supportive sessions	−3.85	5.47	41.49	−0.71	0.485	−14.89–7.18	0.177
After exploration	−4.03	5.05	54.89	−0.80	0.429	−14.15–6.10	0.185
After 15 ST sessions	−5.92	4.95	59.59	−1.20	0.237	−15.82–3.98	0.272

TABLE 4 (Continued)

	Estimate	SE	df	t	p*	CI	Effect size[a] (Cohen's d)
After 30 ST sessions	−15.75	4.95	59.17	−3.19	0.002	−25.65 to −5.85	0.724
After 5-month follow-up	−14.43	5.08	59.20	−2.84	0.006	−24.59 to −4.27	0.663
After 10-month follow-up	−18.28	5.22	59.02	−3.50	0.001	−28.73 to −7.82	0.840

Abbreviations: CI, confidence interval at 95%; SCL-90, Symptom Check List 90; SMI, Schema Mode Inventory; SRS-A, Social Responsiveness Scale – Adult version; ST, schema therapy; VAS, visual analogue scale.
[a]Effect size calculated as change with respect to baseline, with baseline SD as denominator: VAS SD 12.809; SMI Happy Child SD 0.575; SMI Healthy Adult SD 0.698; SMI Dysfunctional Modes SD 0.395; SCL-90 SD 50.641; SRS-A SD 21.759. Change based on the estimates from the mixed model analysis.
*$p < 0.05$.

TABLE 5 Estimated means on SMI, SCL-90 and SRS-A for the contrast cognitive-behavioural or experiential techniques first

	Contrast cognitive-behavioural versus experiential techniques				
	Estimated means	SE	df	t	p*
SMI Happy Child	0.15	0.08	10.1	1.82	0.10
SMI Healthy Adult	0.06	0.16	10	0.37	0.72
SMI Dysfunctional modes	0.159	0.11	9.97	1.45	0.18
SCL-90	16.2	10.4	9.97	1.56	0.15
SRS-A	6.67	4.97	9.89	1.34	0.21

Abbreviations: SCL-90, Symptom Check List 90; SMI, Schema Mode Inventory; SRS-A, Social Responsiveness Scale – Adult version.
*$p < 0.05$.

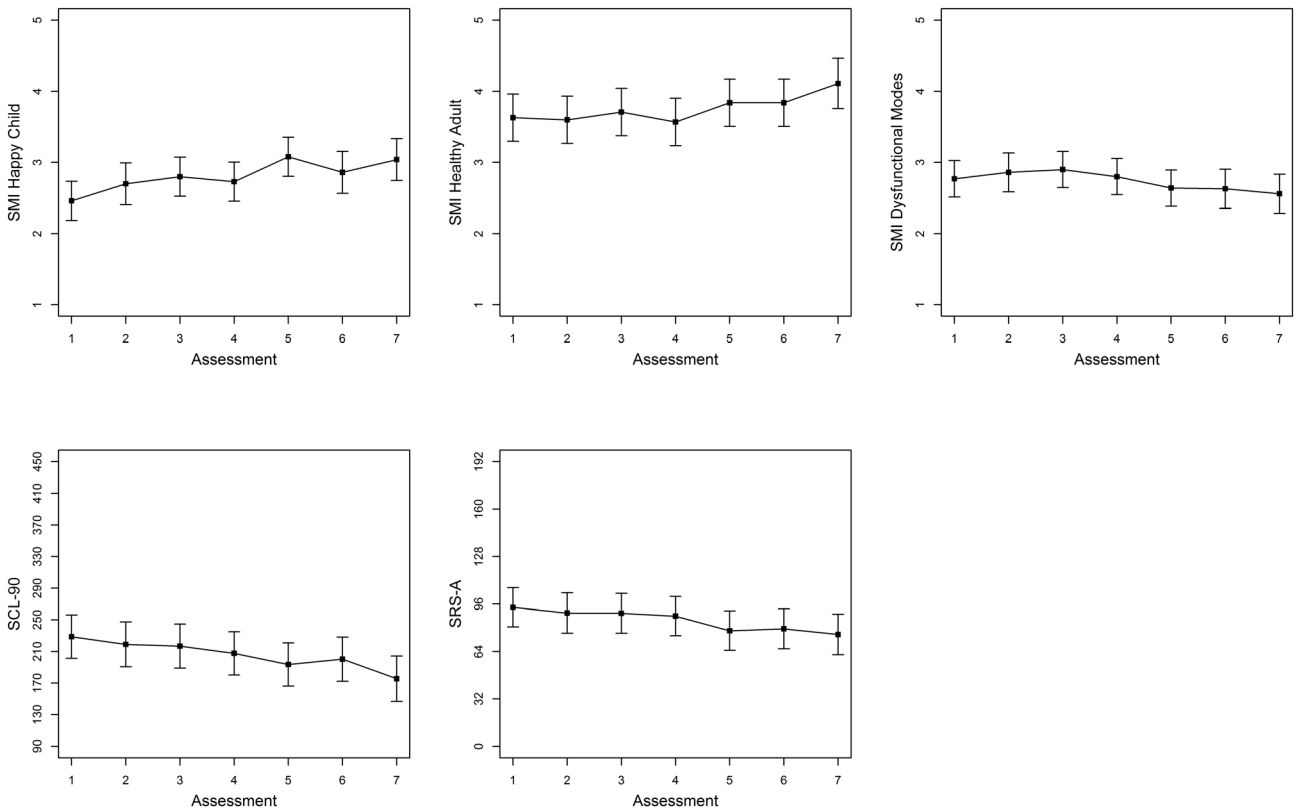

FIGURE 3 Estimated marginal means of total scores of SMI, SCL-90 and SRS-A ($N = 12$). Assessments: 1 = baseline; 2 = after supportive sessions; 3 = after exploration; 4 = after 15 session schema therapy; 5 = after 30 session schema therapy; 6 = after 5-month follow-up; 7 = after 10-month follow-up. SCL-90, Symptom Check List 90; SMI, Schema Mode Inventory; SRS-A, Social Responsiveness Scale – Adult version

2. Change in general mental health symptoms (SCL-90)

Changes in general mental health symptoms assessed with the SCL-90 appeared significant after the second period of ST and after 10-month follow-up with a medium to large effect size compared to baseline.

3. Change in social interaction and communication (SRS-A)

Changes in social interaction and communication assessed with the SRS-A were significant after the second period of ST and at both follow-up conditions compared to baseline. Effect sizes of treatment versus baseline were medium, and follow-up versus baseline were medium to large.

4. Change in personality disorder traits (SCID-II)

The number of SCID-II PD traits decreased significantly with large effect sizes between baseline (median = 26; interquartile range [IQR] = 7.5) and 5-month follow-up (median = 13; IQR = 9; r = −0.68), $p = 0.01$, as well as between baseline and 10-month follow-up (median = 9.5; IQR = 5.5; r = −0.63), $p = 0.005$.

Four participants with PD diagnoses at baseline did not meet full criteria for a specific PD diagnosis anymore at follow-up, now diagnosed as PD not otherwise specified (PDNOS). One participant did not meet any criteria for a specific PD nor PDNOS anymore at follow-up. In one participant, PD diagnosis did not change from baseline to follow-up. One participant assessed with four PD diagnoses including one PDNOS only had one specific PD at follow-up. One participant with four specific PD diagnoses only met full criteria for two specific PDs at follow-up. Two participants assessed with three PDs including one PDNOS only met full criteria for one specific PD and PDNOS at follow-up. Two participants were not reassessed for PD diagnosis because of declining follow-up participation.

5. Change in DSM-IV axis-I disorders (SAM)

For 10 of the 12 participants (two declined participation at follow-up assessment), the number of DSM-IV axis-I mental disorders decreased between baseline (mean = 2.0, SD = 0.94) and 5-month follow-up (mean = 1.8, SD = 1.23), as well as between baseline and 10-month follow-up (mean = 1.8, SD = 1.23). There was no significant difference between the repeated measurements, according to the Friedman's test ($\chi^2(2) = 0.07$, $p = 0.97$).

4 | DISCUSSION

This study explored the effects of ST for autistic adults with PD using a non-concurrent multiple case series design. Our findings confirmed our hypothesis that ST would significantly lead to a decrease of dysfunctional core beliefs, PD traits and psychopathological symptoms, an increase of the functional Happy Child mode and an improvement of social responsiveness. The changes consolidated after treatment, indicated by 5- and 10-month follow-ups. For the functional Healthy Adult mode, we only found a significant increase at 10-month follow-up. The baseline to 10-month follow-up effect sizes were in a range comparable to what other studies reported for ST among patients with PD (Bamelis et al., 2014; Rameckers et al., 2021). Our findings could not confirm that ST would lead to a decrease of dysfunctional schema modes and DSM-IV axis-I mental disorders. Our findings demonstrated no evidence for order in starting with cognitive behavioural or experiential techniques and no superiority for one of these two techniques.

Remarkably, the dysfunctional schema modes did not decrease in our study. Given the focus of ST on schema modes, this finding is hard to interpret. One explanation might be that these modes are persistent due to the vulnerability to maladaptive schema development in autistic people (Gaus, 2019), though the changes in core beliefs contradict this explanation. In a study, examining ST in young autistic adults, a delayed change in schema modes compared to early maladaptive schemas was found, suggesting schema modes to be more stable than early maladaptive schemas (Oshima et al., 2021). Another explanation might be that the ST primarily strengthened functional modes, while not successfully addressing the dysfunctional modes in autistic people. Perhaps the cognitive inflexibility, a common characteristic of ASD, playing a significant role in externalizing and internalizing symptoms in ASD (Ozsivadjian et al., 2021) and a key challenge for therapists (Cooper et al., 2018), might have hindered change in the frequency of dysfunctional schema mode activation in some autistic adults: Adaptations of ST targeting this cognitive process would then be needed. Visual inspection of the individual average credibility of dysfunctional core beliefs (Figure 2) of the participants shows a dichotomy: Some recognize change in their dysfunctional core beliefs and dysfunctional modes, while others (participants 1, 3, 10 and 12) seem to adhere to them. These four participants still benefit from treatment, albeit less than the other participants, because they do show an improvement on the functional modes, which is also reflected in an improvement on the secondary outcome measures. A previous study indicated that the functional Healthy Adult mode and the dysfunctional Vulnerable Child mode are central to the change process in the treatment of PD (Yakın et al., 2020). The four participants seemed to benefit only from one improvement mechanism (i.e., increasing functional modes) and not from the second mechanism (i.e., a decrease in dysfunctional modes).

We do not have an explanation for an unexpected significant increase of functional Happy Child mode after supportive sessions and exploration other than that starting a treatment participants were looking forward to and meeting the therapist contributed to feelings of optimism, understanding, protection and validation. Participant 2 stated at follow-up that this was her first intensive and long-lasting treatment in which she felt really welcome and understood by the therapist, who she described as relaxed, patient, open, clear, with a sense of humour and experienced in ASD. The explanation for the functional Healthy Adult mode only to be increased after 10-month follow-up is that it is a complex and long way dealing with ASD- and PD-related challenges. It takes time to develop positive and functional thoughts and feelings about oneself, and enough ability to deal with

dysfunctional schema modes. A possible explanation for an improvement of social interaction and communication is given by participant 4: ST gave him the opportunity to practice his social skills over a longer period of time with lots of explanation, discussion and recurrence of practicing.

Participants showed great improvement in recovery from PDs, with participant 6 not meeting criteria for any PD anymore: These results are comparable to the results of a study examining clinical effectiveness of ST for PDs in general in which 80% of the participants recovered from PDs after 40 sessions in year 1, followed-up by 10 sessions in year 2 (Bamelis et al., 2014).

Our study did not find superiority for cognitive behavioural techniques over experiential techniques. Both techniques did quite well. Clinicians often doubt whether experiential techniques are suitable for autistic people, because of (assumed) difficulties and challenges in emotional information processing, but contrary to what some may expect, our results show a positive response to experiential techniques in this population. Participant 11 found imagery rescripting very useful, especially the explaining, translating and discussing afterwards leading to meaningful insight in his feelings.

Treatment of mental health conditions research in autistic people is scarce. In line with a survey exploring experiences of treatment and support of autistic adults (Camm-Crosbie et al., 2019), there is an urgent need for validated treatment approaches and therapists trained in autistic people with mental health difficulties and disorders (Adams & Young, 2021). To improve treatment for autistic adults with PD, we advise more specific studies of therapies like ST for them. Our findings emphasize the essence that researchers continue to refine the interventions for PDs in autistic people for optimal effectiveness of the interventions. Future studies should address the issue to what degree ST techniques can be used in autistic adults with PD to reduce the dysfunctional schema modes. Future studies will be needed to further analyze the degree of benefit of ST in autistic people with PD. We suggest future studies examining the benefit of decreasing dysfunctional core beliefs and increasing functional schema modes through ST for comorbid conditions like depressive and anxiety disorders faced by autistic adults. A ST modified for autism spectrum conditions (ST-MASC), developed by Bulluss (2019), already provides a framework and an extension of the regular ST elements in which autism-driven coping responses and autism-specific needs are incorporated and conceptualized. Further, a ST-informed social interaction training (STISI) is suggested for autistic people to reduce the complexity of social interactions by teaching them to identify schema coping behaviour in their non-autistic interaction partners and to learn ways of responding (Parpart et al., 2018).

Strengths of this study are the use of independent study therapists supervised by an independent schema therapist, the assessments at screening and follow-up by two raters, treatment integrity check, randomization of the order of starting with either cognitive behavioural or experiential techniques over participants, the fact that medication was constant and no other treatment was followed by the participants.

There are several limitations of this study. First, as already mentioned, the change of our multiple baseline case series design (Vuijk & Arntz, 2017) into a multiple case series design, especially influenced the possibility to draw causal conclusions from our primary outcome analysis because evidence for the occurrence of change as a result of the intervention instead of time is less strong. Second, 10 participants were already in treatment, unknown if they had already taken more or less some advantage of that treatment when starting to participate in this study. Thereby, in the years before ST, nine participants received CBT for depressive and anxiety disorders, and six participants received ASD treatment (psychoeducation, social skills training and supportive treatment). Given the small sample size, we cannot test the (in-between subject) effects of previous treatment on the effectiveness of ST, let alone effects of a specific previous treatment. Third, participants differed greatly in the number of DSM PD diagnoses and secondary DSM-IV axis-I mental disorder diagnoses, unknown if this variety influenced the results: One can imagine that participants with more diagnoses have had less improvement. Finally, none of the outcome measures, excluding the SRS-A, are validated for autistic people, which means the results should be interpreted with caution.

Overall, as far as we know, our study is the first systematic investigation offering preliminary support for the effectiveness of PD treatment in autistic adults. The results of this study indicate that ST might be effective in decreasing dysfunctional core beliefs, PD traits, general mental health symptoms, increasing functional schema modes and social responsiveness, with achieved improvements persisting over time.

ACKNOWLEDGEMENTS

We greatly appreciate the time and effort devoted by the 12 participants to this study. The authors gratefully acknowledge the study therapists Neeke van den Berg, Tatiana Brandsma and Simone van der Heiden, the assessment therapist Ester van der Sluis and the assistant in data administration Esmee Verhoeks.

CONFLICTS OF INTEREST

Arnoud Arntz was and is project leader of various multicenter randomized controlled trials into the effectiveness and cost-effectiveness of schema therapy. He published several books about schema therapy.

COMPLIANCE WITH ETHICAL STANDARDS AND PATIENT CONSENT STATEMENT

The study was carried out in accordance with the Code of Ethics of the World Medical Association (Declaration of Helsinki, 2000). Approval was granted by the Ethics Committee of the University of Amsterdam (Date: 02 February 2016/No.: 2015-CP-6374).

Informed consent was obtained from all individual participants included in the study.

AUTHOR CONTRIBUTIONS

All authors contributed to the study conception and design. Richard Vuijk and Mathijs Deen performed material preparation, data collection and analysis. Richard Vuijk wrote the first draft of the manuscript, and all authors commented on previous versions of the manuscript. All authors read and approved the final manuscript.

TRIAL REGISTRATION

The Netherlands National Trial Register NTR5788. Registered 01 April 2016. https://www.trialregister.nl/trial/5653.

DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available from the corresponding author upon reasonable request.

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How to cite this article: Vuijk, R., Deen, M., Geurts, H. M., & Arntz, A. (2022). Schema therapy for personality disorders in autistic adults: Results of a multiple case series study. *Clinical Psychology & Psychotherapy*, 1–15. https://doi.org/10.1002/cpp.2817