Therapeutic intervention for internalized stigma of severe mental illness: A systematic review and meta-analysis

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Abstract

Objective.

Internalized stigma can lead to pervasive negative effects among people with severe mental illness (SMI). Although prevalence of internalized stigma is high, there is a dearth of interventions and meanwhile a lack of evidence as to their effectiveness. This study aims at unravelling the existence of different therapeutic interventions and the effectiveness internalized stigma reduction in people with SMI via a systematic review and meta-analysis. Methods.

Five electronic databases were searched. Studies were included if they (1) involved community or hospital based interventions on internalized stigma, (2) included participants who were given a diagnosis of SMI>50%, and (3) were empirical and quantitative in nature. Results.

Fourteen articles were selected for extensive review and five for meta-analysis. Nine studies showed significant decrease in internalized stigma and two showed sustainable effects. Meta-analysis showed that there was a small to moderate significant effect in therapeutic interventions (SMD=-0.43; p=0.003). Among the intervention elements, four studies suggested a favorable effect of psychoeducation. Meta-analysis showed that there was small to moderate significant effect (SMD=-0.40; p=0.001).

Conclusion.

Most internalized stigma reduction programs appear to be effective. This systematic review cannot make any recommendation on which intervention is more effective although psychoeducation seems most promising. More Randomized Controlled Trials (RCT) on particular intervention components using standard outcome measures are recommended in future studies.

Keywords: psychoeducation, severe mental illness, self-stigma, internalized stigma, psychosocial intervention

1. Introduction

Internalized stigma, also called self-stigma, is the process of adopting the public's stigmatizing opinions into own thoughts. Its prevalence among people with severe mental illness (SMI) stands at 41.7% (Gerlinger et al., 2013). Meta-analysis (Livingston and Boyd, 2010) found that high levels of internalized stigma were significantly correlated with hopelessness, poorer self-esteem, reduced empowerment/ mastery, and reduced self-efficacy. Internalized stigma significantly predicted poor social functioning over time (Fung et al., 2008; Tsang et al., 2010; Yanos et al., 2012). Higher internalized stigma was also associated with poorer quality of life on all the domains of WHOQOL-Brief (Mosanya et al., 2014). In addition, higher levels of internalized stigma were significantly related to more severe psychiatric symptoms, poorer treatment adherence, and lower utilization of mental health services (Rüsch et al., 2009). Up to 20% of people may even discontinue treatment prematurely due to internalized stigma (Corrigan et al., 2014). Unfortunately, poorer treatment adherence was related to poorer treatment outcomes, more re-hospitalization, and increased health costs (Lacro et al., 2002). All of the above studies suggest that internalized stigma has tremendous negative impact on functional outcomes of people with SMI and increases societal burden in taking care of these individuals.

Given the close negative relationship between internalized stigma and recovery (Yanos et al., 2008), interventions that attempt to reduce it among those with SMI so as to improve their

prospect for recovery are important both in psychiatric rehabilitation and community integration.

Although a conceptual framework (Hayward and Bright, 1997) on internalized stigma reduction is available, there is a dearth of interventions and meanwhile a lack of evidence as to their effectiveness. The programs usually consist of a combination of different elements of intervention such as psychoeducation, cognitive behavioral therapy, and social skills training. Currently there are no widely accepted treatment protocols on internalized stigma reduction. Numerous studies on internalized stigma were published in the past few years. There are a few reviews (e.g., Mittal, et al., 2012) on this area. But no attempt on meta-analysis has ever been conducted. To fill the knowledge gap, this systematic review and meta-analytical study focuses on studies which are randomized control trials, clinical trials, or experimental trials in nature and reported therapeutic interventions to reduce internalized stigma in people with SMI.

2. Methods

The PRISMA Statement Criteria (Liberati et al., 2009) was adopted throughout the review to report our results.

2.1 Data Sources

Systematic search was done to locate studies from *PubMed, PsychInfo* (1806-Present), *SCI* (1970-Present), *SSCI* (1970-Present), and *Scopus* on October 3rd, 2014. The search terms were

generated based on the four types of eligibility criteria mentioned above. They are presented in Supplementary Appendix 1. In addition, emails were sent to a few prominent researchers on mental illness stigma that had close collaborations with the first author. One replied and suggested an in press article that we missed in the systematic search and eventually included in the systematic review.

2.2 Study Selection

Studies were included if the following criteria were met: (1) studies: randomized clinical trials, clinical trials, and experimental studies studying internalized stigma reduction in people with SMI which was operationally defined as mental illness having a chronic course and leading to significant social and occupational dysfunction such as Schizophrenia, Psychotic disorder, Psychosis, Delusional disorder, Schizoaffective, Bipolar disorder and Personality disorder; (2) participants: more than 50% studies participants were given a diagnosis of SMI,; (3) intervention: studies that compared community or hospital based therapeutic interventions with conventional treatment; (4) outcome measures: studies that used validated instruments for screening and assessing the severity of internalized stigma. Qualitative studies and literature reviews were excluded.

There were no limitations in the follow up period. Search was restricted to publications in English but there was no limit to the years of publication.

Two reviewers independently conducted the search in five electronic databases. The results

were screened on titles and abstracts by all reviewers independently. Disagreements were resolved via discussion between reviewers with the facilitation of the corresponding author.

2.3 Data Extraction and Analysis

Data extracted from selected studies included: (1) characteristics of the study (e.g., aims, study design, intervention setting, participants' characteristics, randomization procedures, inclusion and exclusion criteria, etc.); (2) outcome measures (e.g., outcome measure instruments, assessment period, dropout rate, etc.); (3) interventions (e.g., intervention approach, trainers' quality, etc.); and (4) study results (e.g., significant effects, sustainability, etc.). To ensure accuracy, five reviewers assessed two to three studies independently and the data were cross-checked by another reviewer. Also, emails were sent to the authors for clarifying missing or unclear data.

2.5 Assessment of Methodological Quality

The methodological risk of bias for each trial was evaluated using the risk of bias table (Higgins and Green, 2011). Five reviewers worked independently to determine the adequacy of randomization sequence, blinding of patients and assessors, and the extent of follow up. Disagreements were resolved in discussion.

2.6. Data Synthesis and Analysis.

Review Manager 5.3, developed by the Cochrane Collaboration (2014), was used for meta-analysis of the results of included studies. The Outcomes across the trials were

expressed in *Cohen's D*. Standardized mean differences (*SMDs*) were calculated for the pooled effects. *SMDs* were interpreted using the "rule of thumb": 0.2 represents a small effect, 0.5 represents a moderate effect, and 0.8 represents a large effect (Cohen, 1988). Heterogeneity was tested with an X^2 test. I^2 was also reported. I^2 statistic >75% was considered to have high degree of heterogeneity while I^2 statistic of 25%-50% was considered to have a low degree of heterogeneity (Higgins et al., 2003). We used random-effects model for heterogeneity (p<0.05) and a fixed effects model for heterogeneity (p>0.05). Sensitivity analyses were conducted with psychoeducation group and professional-led or peer-led intervention. Publication bias was examined using funnel plot. A value of p<0.05 was considered statistically significant.

Since the outcome of internalized stigma reduction program was assessed by more than one tool in a trial, only the primary outcome (i.e., internalized stigma reduction) was included in the meta-analysis. Emails were sent to corresponding authors for clarifying missing data.

3. Results

3.1. Results of Literature Search

Eight hundred nineteen articles were retrieved. Three hundred eighty duplicated and 421 irrelevant articles were excluded after initial screening of title and abstract. Full reports of 22 studies were acquired and eight were further excluded for the following reasons: (1) not a clinical trial, (2) <50% participants given diagnoses of SMI, (3) exploratory research, and (4)

duplicated studies (Fig. 1).

3.2. Description of Included Studies

Fourteen studies, including seven RCTs (Corrigan et al., 2015; Çuhadar and Çam, 2014; Fung et al., 2011; McCay et al., 2007; Russinova et al., 2014; Rüsch et al., 2014; Yanos et al., 2012), three controlled clinical trials (Roe et al., 2014; Sibitz et al., 2013; Uchino et al., 2012), and four uncontrolled studies without a control group (Costainet al., 2014; Lucksted et al., 2011; Lysaker et al., 2012; Staring et al., 2013) met inclusion criteria. These studies were originated from nine countries across Americas, Europe, and Asia. Most programs adopted psychoeducation approach with inclusion of a combination of other components such as CBT, social skills training, goal attainment program, and narrative therapy. The duration of these programs ranged from 10 to 40 sessions (Russinova et al., 2014; Sibitz et al., 2013). Other than the above more conventional therapeutic elements, there were two evolving innovative interventions. Both of them were peer led group interventions. One was Coming Out Proud (Corrigan et al., 2015; Rüsch et al., 2014). It consisted of group discussion focusing on topics of secrecy and disclosure of own mental illness. The other was Photovoice (Russinova et al., 2014). In the group, individuals photograph objects or events in their daily lives were used to generate narratives for group discussion. The characteristics of included studies are summarized in Table 1.

Participants were given a diagnosis of schizophrenia, schizophrenia spectrum disorder, bipolar

disorder, or major mood disorder. One study included patients with schizophrenia only (Fung et al., 2011) and one included patients with bipolar disorder only (Çuhadar and Çam, 2014). Participants were mostly given a diagnosis by psychiatrists according to DSM IV or ICD-10 (Yanos et al., 2012). Two studies recruited participants according to their self-reported diagnosis. Only one study reported the use of the structured interview procedure in verifying the diagnosis of the research participants (Lysakeret al., 2012).

Insert Figure 1 here

Sample size of the studies varied from 21 to 205 participants, with a total of 1,131 participants including 879 participants in the experimental groups and 452 participants in the control groups.

Controlled group design was employed in ten studies. Treatment as usual (TAU) was applied in seven studies (McCayet al.; 2007, Roe et al., 2014; Russinova et al., 2014; Rüsch et al., 2014; Sibitz et al., 2013; Uchino et al., 2012; Yanos et al., 2012), conventional treatment group (i.e., newspaper reading) in one study (Fung et al., 2011), no active treatment in one study (Çuhadar and Çam, 2014), and waitlist control in one study (Corrigan et al., 2015). All studies provided information on the duration of treatment in patient intervention groups.

Thirteen included studies examined the effect of internalized stigma reduction and one examined the effect of negative symptoms immediately following the internalized stigma reduction intervention. In addition, seven studies (Corrigan et al., 2015; Costain et al.,

2014; Fung et al., 2011; Lysakeret al., 2012; Russinova et al., 2014; Rüsch et al., 2014; Yanos et al., 2012) examined the sustainability of intervention effect after follow-up periods ranging from three weeks to six months.

Regarding outcome measures, internalized stigma was assessed as the primary outcome in all the included studies except one which treated internalized stigma as a mediator of dysfunctional beliefs (Staringet al., 2013). The internalized stigma scales applied in the studies included Internalized Stigma of Mental Illness (ISMI) (Costain et al., 2014; Çuhadar and Çam, 2014; Lucksted et al., 2011; Lysaker et al., 2012; Roe et al., 2014; Russinova et al., 2014; Rüschet al., 2014; Sibitzet al., 2013; Yanos et al., 2012), Short form of Self-stigma of Mental Illness Scale (SSMIS) (Corrigan et al., 2015), Chinese Self-stigma of Mental Illness Scale (CSSMIS) (Fung et al., 2011), Link Perceived Stigma Questionnaire (LPSQ) (McCayet al., 2007), and Japanese version of Social Distance Scale (SDS-J) (Uchinoet al., 2012). Other outcome measurements were also applied to assess the intervention effect, for examples, the Positive and Negative Syndrome Scale (PANSS) and the Rosenberg Self-Esteem Scale (RSES). These outcome measures varied substantially across studies.

3.3 Results of Individual Studies

Nine studies reported significant internalized stigma reduction. Studies that reported significant effects of therapeutic intervention other than internalized stigma reduction are shown in Table.1. Seven studies had follow-up periods but only two studies showed

sustainable effects for one to three months (Corrigan et al., 2015; Russinova et al., 2014).

3.4. Effects of Intervention Approaches on Internalized stigma.

Meta-analysis was performed among studies which used similar treatment approaches (e.g., psychoeducation, CBT, etc.) and similar outcome measures. Following this, six controlled studies were identified to use ISMI as outcome measure. However, one of the studies was eventually excluded (Çuhadar andÇam, 2014) because it used the Turkish version of ISMI which was not comparable to the original version. Consequently, five trials were included in meta-analysis (Roe et al., 2014;Russinova et al., 2014; Rüschet al., 2014;Sibitz et al., 2013; Yanos et al., 2012).

Comparison 1: Psychoeducation Element. Four out of seven trials which contained psychoeducation were RCTs. Two trials indicated significant effects when compared with controlled group with no active treatment (Çuhadar andÇam, 2014) and TAU (Russinova et al.,2014). The former focused on patients with bipolar disorder and the latter focused on patients with schizophrenia spectrum disorder and bipolar disorder. ISMI total scores from two RCTs and one controlled trial were pooled and the pooled SMD was -0.40 [-0.64, -0.16], indicating a small-moderate effect (p=0.001). There was a low degree of heterogeneity (I²=17%).

Insert Figure 2 here

Comparison 2: Cognitive Behavioral Therapy Element. Two out of five trials containing CBT

element were RCTs. Both trials suggested no intergroup difference when compared to newspaper reading group (Funget al., 2011) and TAU (Yanos et al., 2012). The former focused on patients with schizophrenia and the latter focused on patients with schizophrenia, schizoaffective, and bipolar disorder.

Comparison 3: Group Discussion Element. Three out of the four trials which had group discussion element were RCTs. These RCTs included innovative interventions, namely photovoice (Russinova et al., 2014) and Coming Out Proud (COP) (Corrigan et al., 2015; Russinova et al.,2014). Photovoice was a discussion group on the selected photographs regarding to stigma while COP was a peer-led group intervention which empowered people with mental illness to decide on secrecy and disclosure of illness. When compared to TAU, the trial on photovoice indicated significant effect (Russinova et al., 2014). For COP, one of them suggested significant effect on reduction in internalized stigma when compared to waitlist control (Corrigan et al., 2015). Another trial, however, suggested no group x time effect (Rüsch et al., 2014). All of them focused on patients with serious mental illness. Comparison 4: Social Skills Training Element. One out of two trials that contained social skills training element was RCT. It suggested no intergroup difference on patients with schizophrenia when compared to newspaper reading group (Funget al., 2011). Comparison 5: Narrative Enhancement and Cognitive Therapy (NECT) Element. This approach focused on the process of telling and sharing personal narratives about their

experience of mental illness. Two out of three trials containing NECT element were RCTs. When compared to TAU, one trial indicated significant effect (Russinova et al.,2014) while another suggested no intergroup difference (Yanos et al., 2012). Both focused on patients with schizophrenia spectrum disorder and bipolar disorder. Moreover, the former focused on depressive disorder.

Other Intervention Approaches. There was one RCT which reported the effect of group psychotherapy on internalized stigma reduction among individuals with schizophrenia spectrum disorder (McCay et al., 2007). No intergroup difference was reported. The other two trials were uncontrolled clinical trials involving genetic counseling (Costain et al., 2014) and vocational rehabilitation (Lysaker et al., 2012).

3.5. Meta-analysis.

Five studies were eligible for meta-analysis (Roe et al., 2014; Russinova et al., 2014; Rüschet al., 2014; Sibitz et al., 2013; Yanos et al., 2012). ISMI included 5 subscales: alienation, stereotype endorsement, discrimination experience, social withdrawal, and stigma resistance. The total scores from three psychosocial interventions were pooled and the pooled SMD was -0.43 [-0.72, -0.14] which indicated a small-moderate effect (p=0.003) of psychosocial interventions. There was a low degree of heterogeneity (I^2 =22%). As two studies excluded the 5th subscale (i.e., stigma resistance) of ISMI score (Roe et al., 2014; Rüschet al., 2014), we pooled the ISMI scores by taking average of the sum of sub-scales 1-4 from four studies.

Similar to the result of total ISMI score, it indicated a small to moderate effect (SMD=-0.33 [-0.54, -0.12]; p=0.002). The results from both professional led and peer led psychosocial intervention were pooled. Result of meta-analysis showed moderate significant effect professional led intervention (SMD=-0.51 [-0.79, -0.23]; p<0.001) but not peer led intervention.

Insert Figure 3 and 4 here

Quality assessment for each trial is presented in Table 2. None of the studies met all of the criteria for high quality. Inclusion and exclusion criteria were clearly defined except one trial (Roe et al., 2014). Seven studies were RCTs with randomization method reported clearly except one study (Corrigan et al., 2015). Common problems included inadequate blinding of patients and outcome assessors, lack of information on sample size estimation, and intention-to-treat analyses not applied.

4. Discussion

This paper presents the most updated systematic review and meta-analysis on internalized stigma reduction. Half of the reviewed studies were published from 2013 to 2015 which were not included in earlier reviews. The studies were conducted in nine countries across America, Europe, and Asia which suggest cross-cultural some degree of generalizability of the results. As pointed out in the introduction, programs to reduce internalized stigma consist of many

components. Our review reveals the beneficial effects of the psychoeducation component in internalized stigma reduction when compared to TAU. For COP and NECT, effects were inconsistent. For CBT and social skills training, RCTs reported no significant effects. Interpretation and generalization of results should be cautiously treated due to limited number and methodological problems of studies. Because of bias among studies, the results of meta-analysis should also be read with caution. The studies used different diagnostic criteria for participants, including self-reported and diagnoses given by psychiatrists. Moreover, the proportion of participants with SMI was slightly less than 50% in the control group in one of the studies (Rüsch et al., 2014). Both of these factors limit the comparability between results for the participants with SMI in the studies. The baseline internalized stigma level among participants varied, with mean total scores of ISMI at baseline ranging from 1.63 to 2.23 out of 4.0. Although there does not have any evidence, this variation in baseline level of internalized stigma might affect the effectiveness of programs and hence the comparability of effectiveness between different studies. Some of the studies only presented group x time effect or main effect. As intergroup difference was not presented in all studies, the significance of between group differences was unclear. Randomization was involved only in seven studies. Twelve trials did not adhere to the intention-to-treat principle. These two factors lead to possible over-estimation of the treatment effect. There is also a possibility of publication bias. Among the limited number of studies, different combinations of intervention

approaches and outcome measurements were adopted. It is therefore hard to draw an accurate conclusion based on our meta-analysis. Finally, as few trials involved post-intervention follow up or the follow-up period was short, it is difficult to make conclusion on the sustainability of the interventions.

To date, a variety of interventions are available but there is a lack of commonly accepted framework on internalized stigma reduction. The framework proposed by Hayward and Bright (1997) adopted CBT as the main intervention with the role of psychosocial factors and self-management skills also taken into consideration.

There are some implications for further research. Our meta-analysis shows that psychoeducation is common among the studies and seems to have significant effect. However, among these three studies, only two (Roe et al., 2014; Russinova et al., 2014) mentioned clearly the content of psychoeducation, which included understanding of stigma and strategies to cope with stigma. Given that psychoeducation was not the sole intervention in these studies and the follow-up was short, it is difficult to make a solid recommendation as to the optimal duration of psychoeducation program. It is however suggested that future studies should focus on studying the effectiveness of psychoeducation as the only intervention with longer follow up period to evaluate the sustainability of its outcome. As the content of psychoeducation varies, it is also necessary to study the specific content that actually contributes to it effectiveness. Our systematic review found that there were only limited studies meeting our

inclusion criteria. Most studies had numerous limitations in measurement tools and methodological qualities. These added to our difficulties in making conclusion based on our results. Following this, we suggest the use of common outcome measures among studies such as ISMI, and improved methodological qualities of future studies to facilitate similar reviews in the future.

Two evolving innovative interventions, Coming Out Proud and Photovoice, deserve more attention. The results from trials of COP were inconclusive. Yet the study on Photovoice had a significant effect in reduction of internalized stigma and was shown sustainable in a period of three months. As new and promising interventions, more research on their effectiveness is needed. As both of these two innovative interventions involve peer-led activities, the therapeutic values of this type of activities should be more carefully discerned.

Other than research, this study has some implications for clinical practice. First, the serious consequences of leaving internalized stigma untreated should be brought to the awareness of clinicians. Second, internalized stigma reduction programs have to be delivered by health and rehabilitation professionals. Relevant training courses should be made available to those professionals who show interest in putting these programs into their clinical and rehabilitation practice. Third, if recourses are limited, practitioners should be recommended to select those programs with a psychoeducation element because it is found to be the most evidence-based in reducing internalized stigma based on existing evidence. Finally, clinicians should be

encouraged to explore more innovative approaches to counteract internalized stigma because existing interventions are limited and their evidence is not entirely compelling.

5. Conclusion

Most programs we reviewed showed significant effects in reducing internalized stigma. As an emerging area, studies were still limited. The programs are implemented by professionals including clinicians, psychiatric nurse, social worker, and non-professionals who are trained according the program manuals. Among different intervention psychoeducation seems to be more promising and two novel techniques (i.e., coming out proud and photovoice) deserve more attention. In future, more innovative approaches to reducing internalized stigma should be developed and more RCTs on particular intervention components using standard outcome measure should be conducted so that meta-analysis could be conducted and effects of the intervention could be compared. All of the above adds to evidence-based practice in internalized stigma reduction.

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- Yanos, P. T., Lucksted, A., Drapalski, A. L., Roe, D., & Lysaker, P., 2014. Interventions targeting mental health self-stigma: A review and comparison. Psychiatr Rehabil J. Advance online publication. doi:10.1037/prj0000100

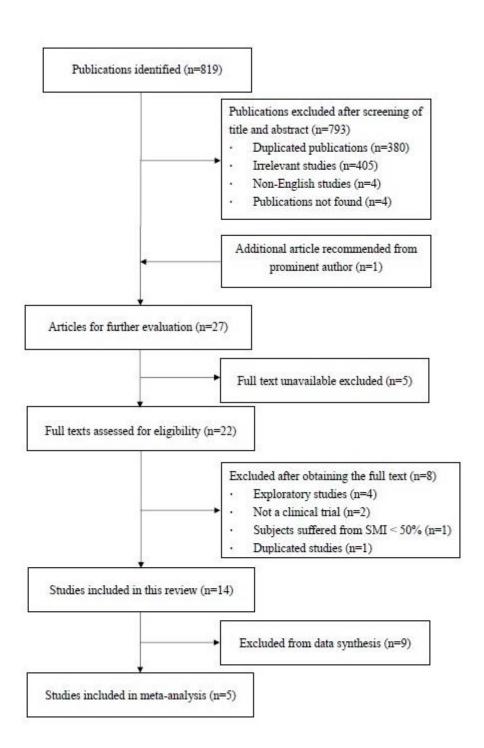


Fig. 1. Selection process of included studies

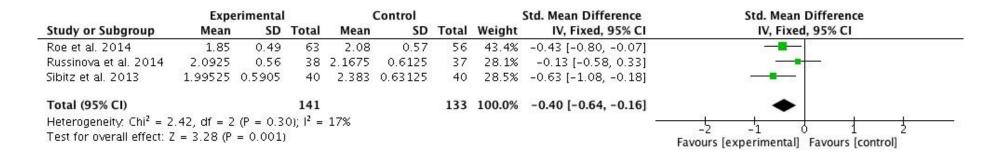


Fig. 2. A forest plot of the meta-analysis of three studies comparing psychoeducation to TAU for changes in ISMI score.

	Exp	perimental			Control			Std. Mean Difference	Std. Mean Difference
Study or Subgroup	Mean SD Tot		Total	Mean	Mean SD		Weight	IV, Fixed, 95% CI	IV, Fixed, 95% CI
Russinova et al. 2014	2.04	0.42	38	2.13	0.47	37	40.8%	-0.20 [-0.65, 0.25]	
Sibitz et al. 2013	2.049	0.465	40	2.399	0.508	40	41.0%	-0.71 [-1.16, -0.26]	
Yanos et al. 2012	1.4337	0.42112	18	1.5895	0.49187	16	18.2%	-0.33 [-1.01, 0.34]	
Total (95% CI)			96			93	100.0%	-0.43 [-0.72, -0.14]	•
Heterogeneity: $Chi^2 = 2$	2.55, df =	2 (P = 0.2)	8); 12 =	22%				•	- 5 5 7 7 7
Test for overall effect: 2	Z = 2.94 (P = 0.003	ř.						Favours [experimental] Favours [control]

Fig. 3. A forest plot of the meta-analysis of three studies comparing self-stigma reduction program to TAU for changes in ISMI total score.

	Expe	erimental			Control			Std. Mean Difference	Std. Mean Difference
Study or Subgroup	Mean SD Total		Mean	Mean SD		Weight	IV, Fixed, 95% CI	IV, Fixed, 95% CI	
Roe et al. 2014	1.85	0.49	63	2.08	0.57	56	32.9%	-0.43 [-0.80, -0.07]	a de la companya de l
Russinova et al. 2014	2.0925	0.56	38	2.1675	0.6125	37	21.3%	-0.13 [-0.58, 0.33]	-
Rüsch et al. 2014	2.12	0.53	39	2.18	0.53	47	24.2%	-0.11 [-0.54, 0.31]	
Sibitz et al. 2013	1.99525	0.5905	40	2.383	0.63125	40	21.6%	-0.63 [-1.08, -0.18]	19 B. C. C.
Total (95% CI)			180			180	100.0%	-0.33 [-0.54, -0.12]	•
Heterogeneity: Chi ² = 3	3.78, df = 3	P = 0.2	9); 12 =	21%				-	5 5 5
Test for overall effect: I	Z = 3.11 (P)	= 0.002	Ŕ						Favours [experimental] Favours [control]

Fig. 4. A forest plot of the meta-analysis of four studies comparing self-stigma reduction program to TAU for changes in average of the sum of sub-scales 1-4 of ISMI.

Table 1: Summary of the effects of self-stigma reduction program

	Study	Participants'		Intervention			Outcomes	
		Diagnosis						
Author,	Design; Country	Inclusion	Sample	Approach	Control	Duration	Outcome	Result
year			Size	(Frequency)		of	Measures	
			(Pre-/Post			follow-up		
)					
Corrigan et	RCT; U.S.	People with	EG:	Coming Out	Waitlist	1 month	1. (Short Form)	Effect in subscales under SSMIS:
al, 2015		serious mental	107/51	Proud (COP)			SSMIS	1. Group x Time: No significant effect
		illness who have	CG:	(2 hours			2. Stigma	in change in 'harm" comparing COP
		life disabilities	98/75	session, 3			Stress Scale	group to control group. Significant
				sessions, over			3. CES-D	reduction in "harm" from pre to
				3 days or 1				posttest (p < 0.01) and pre to
				daylong group)				follow-up for COP group. $(p < 0.05)$
								2. Group x Time: Significant
								interaction for change in applying
								stereotypes to self across groups
								(F(2,98)=3.74, p<.05). Significant
								reduction in applying stereotypes to
								self from pre to posttest (p < 0.05)
								and at one month follow-up (p < 0.05)
								3. Group x Time: Significant
								interaction for agreeing on stereotype
								4. Group x Time: No significant
								change in "awareness"
								Stigma Stress Scale
								1. Group x Time: Significant in
								stigma-related harm

					29	1		_
								2. Group x Time: No significant for
								resources to cope with stigma
								Depression
								1. Group x Gender x Time:
								Significant difference in CES-D
Costain et	Uncontrolled clinical	Schizophrenia,	25/22	Genetic	NA	7 weeks	1. ISMI	1. Decrease in self-stigma (p=0.0111)
al., 2012	trial; Canada	Schizoaffective	(21 for	Counseling				2. Reduction in self-blame (p=0.0401)
		disorder	FU)	(1 session with				3. Improvement in understanding of
				mean 46.4 [SD				the empiric recurrence risk
				11.1] minutes)				(p=0.0090)
								4. Reduction in associated concern
								(p=0.0020)
								5. Significant increase in subjective
								(p=0.0007) and objective (p=0.0103)
								knowledge
Çuhadar &	RCT; Turkey	Biploar disorder	63/47	Psychoeducati	No	NA	1. ISMI	1. Significant decrease in total ISMI
Çam, 2014				on program	treatme		(Turkish	score (p < 0.001) and sub-scale 1-4 (p
				(90 minutes	nt		Version)	< 0.001)
				session; once a			2. BDFQ	2. Significant gain in domestic
				week; 7				relationship subscales (p = 0.03)
				sessions)				

Fung et al.,	RCT; Hong Kong	Schizophrenia	CG:	Self-stigma	Conven	3 months	1.CSSMIS	1. Group x time: significant
2010			32/30	reduction	tional		2.CAQ-SPMI	improvement in the self-esteem
			EG:	program	Therap		3.PTCS	decrement subscale of CSSMIS
			34/34	(Twice a week,	y		4.BPRS	*others subscale: stereotype
				6 weeks; New			5.GAF	agreement and self-concurrence
				life: once 1			6.SUMD	show trend of improvement
				week, 12			7.CGSS	2. Group x Time: significant changes
				weeks)				in stages of change continuous score
								of CAQ-SPMI and the participation
								subscale of PTCS
								3. Group x Time: no siginificant
								changes found in SUMD and CGSS
Lucksted et	Pilot study; U.S.	Self Reported:	EG:	Ending	NA	NA	1. ISMI	1. Group x Time: Significance in self
al., 2011		Schizophrenia,	50/34	Self-Stigma			2. MHRM	stigma reduction.(p<0.01)
[26]		Schizoaffective		(ESS) with			3. MSPSS	2. Group x Time: Recovery
		disorder,		lectures and			4.	orientation (MHRM) and social
		Major mood		group			Empowerment	support (MSPSS) significantly
		disorder		discussions.			Scale	increase
				(90 mins			(10 out of 28	3. Group x Time: Mean
				session; once a			items)	Empowerment increased but
				week, 9 week)				insignificantly (p= $0.0556 > 0.05$)
								4. Group x Time: All ISMI subscale
								scores increased significantly except
								Stigma Resistance

Lysaker et	Quasi-experimental;U.	Schizophrenia,	EG:	Vocational	NA	5 months	1. ISMI (5th	1. Group x Time: Modest but
al., 2012	S.	Schizoaffective	70/70	Rehabilitation:			scale excluded)	significant decrease in internalized
		disorder		part-time paid			2. MSEI	stigma (p = 0.007)
				work			3. PANSS	2. Group effect: Significant for both
				placement				emotional discomfort and self-esteem
				(Non-specified				3. Time effect: Significant for
				frequency)				emotional discomfort and self-esteem
								4. Group x Time: Significant for
								self-esteem
								5. No Group or Time effect for
								positive symptoms
McCay et	RCT; Canada	Schizophrenia,	EG:	Psychotherapy	TAU	NA	1.MES	1. Group x time: No significance
al., 2007		Schizophrenifor	67/47	group			2.TSCS	improvement in self concept, self
		m Disorder,		(1.5 hours			3.RSES	esteem, self efficacy, and stigma
		Schizoaffective		session; once a			4.SES	2. Group x time: Significant changes
		Disorder		week;12			5.LPSQ	on measure of engulfment, hope and
				weeks)			6.QLS	quality of life measures compared
							7.MHS	with control
							8.PANSS	
							9.GAF	
Russinova	RCT; US	Schizophrenia	EG:	Antistigma	TAU	3 months	1. ISMI	1. Significant decrease in overall
et al., 2014		spectrum	40/38	Photovoice (90			2. ACSS	ISMI score (p=0.03) in experimental
		disorder,	CG:	mins per			3. PGRS	group
		Bipolar	42/37	session, 10			4.	2. Significant more likely to report
		disorder,		weeks)			Empowerment	using proactive strategies: challenging
		Depressive					Scale	(p=0.04), educating others (p=0.01) to
		disorder,					5. CES-D	cope with perceived stigma.
		Other diagonsis					6. GPSES	3. Significantly greater increases in

					J2			Citit
								Community activism and autonomy
								(p=0.02) on Empowerment Scale
								4. Significant increases in overall
								level of PRGS (p=0.04)
Roe et al.,	Controlled clinical	SMI ≥ 50%	EG:	NECT	TAU	NA	1. ISMI	1. A significant group x time
2013	trails; Israel		222/119	(1-hour			2. MANSA	interaction $(F(4,114) = 2.68, p < 0.04)$
				session; once a			3. ADHS	and with higher improvement in
				week; 20			4. RSE	self-stigma in the treatment group.
				sessions)				2. A significant group x time
								interaction ($p = 0.043$) and with
								higher improvement in quality of life
								in the treatment group.
								3. No significant group x time effect
								4. A significant group x time
								interaction ($p = 0.003$) and with
								higher improvement in self-esteem in
								the treatment group.
Rüsch et	Pilot RCT;	Self Reported:	CG:	Coming Out	TAU	3 weeks	1. ISMI (5th	1. Group x Time: No significant effect
al., 2014	Switzerland	Schizophrenia	50/47	Proud (COP):			scale excluded)	detected on self-stigma as primary
		spectrum	EG:50/40	with peer-led			2.	outcome.
		disorder,		group			Empowerment	2. Group x Time: No significant effect
		Bipolar		discussion.			Scale	of treatment on empowerment or on
		disorder,		(2 hours			3. Link's 5-item	disclosure-related self-efficacy.
		Depressive		session, once a			secrecy scale	3. Group x Time: Significant pre-post
		disorder		week, 3			4. COMIS (7	effect in some secondary outcome
				sessions)			Items)	including decreasing stigma stress and
							5. CAS (8 item)	secrecy as compared to the control
							6. Stress	group, with a medium-effect size for

					33			,
							Appraisal Score	stigma stress reduction
							7. CES-D	4. The effect is only partly
							(15-item	sustainable. Disclosure-related
							German	distress decreased significantly in the
							version)	COP group from baseline to
								post-intervention and follow up.
								Perceived benefits of being out
								increased at post-intervention and
								remained stable for follow up.
Sibitz et al.,	Controlled Clinical	Schizophrenia,	EG:	Recovery	TAU	NA	1. ISMI	1. Significant group effect on ISMI
2013	Trial; Austria	Schizoaffective	46/40	Oriented Day			2.	subscale, discrimination experience
		disorder, other	CG:	Clinic			WHOQOL-BR	and social withdrawl, which
		F2 disorders	51/40	Treatment			EF	internalized stigma within control
				(8am-330pm,				group at both baseline and follow-up
				Mon - Friday,				were higher.
				2 months)				2. Significant group, time, and group
								by time effect in QoL of Experimental
								group and improved in greater extent
								over time.
								3. Significant time, group by time
								interaction in which signifcant
								reduction in psychopathology over
								time in both group.

Starin et al.,	Uncontrolled clinical	Schizophrenia,		CBT-n	NA	NA	1. PANSS	1. Time effect: the improvement on
2013	trial; Netherlands	Schizoaffective		(45-min			2. A combined	the primary outcome measure was
		disorder,		session, once a			score of BDI-4,	highly significant and clinically
		Schizophrenifor		week, 20			10, 12, 13, 15,	relevant.
		m disorder,		sessions)			19, 20, 21;	
		Psychotic					BHS-2, 6	
		disorder NOS					inverted, 17;	
							DAS-DPA-6,	
							14, 15; ISMI	
Uchino et	Controlled clinical	Schizophrenia,	EG:	Weekly	TAU	NA	1. SDS-J	1. Group effect: significant change
al., 2012	trial; Japan	Schizoaffective	29/29	psychoeducatio			2. KIDI	between 2 groups for the SDS-J, KIDI
		disorder	CG:	n program			3. DAI-10	and BPIS.
			27/27	(Once a week,			4. BPIS	2. Correlation: Significant correlation
				6 weeks)			5. GAF	found between SDS-J and KIDI,
								BPIS and DAI-10.
								3. No significant difference for
								DAI-10, GAF, age, and duration of
								treatment.
Yanos et	RCT; U.S.	Schizoaffective,	EG:	NECT	TAU	3 months	1. ISMI	1. No significant differences between
al., 2012		Schizophrenia,	21/15	(1-hour			2. BHS	the NECT and TAU groups.
		Bipolar I,	CG:	session, once a			3. RSES	
		Bipolar II,	18/NR	week, 20			4. CSC	
		Major		sessions)			5. QLS	
		depression					6. PANSS	
							7. SUMD	

CG: Control Group, EG: Experimental Group, NA: Non Applicable; ACSS: Approaches to Coping with Stigma Scale, ADHS: Adult Dispositional Hope Scale, BDI: Beck Depression Inventory, BHS: The Beck Hopelessness Scale, BPIS: Birchwood's Psychosis Insight Scale, BPRS: The Brief Psychiatric Rating Scale, CAS: Cognitive Appraisal of Stigma, CAQ-SPMI: The Change Assessment Questionnaire for People with Severe and Persistent Mental Illness, CBT-n: Cognitive Behavioral Therapy for

negative symptoms, CES-D: Center for Epidemiological Studies Depression scale, CGSS:The Chinese General Self-efficacy Scale, COMIS: Coming Out with Mental Illness Scale, CSC: The Coping with Symptoms Checklist, CSSMIS: The Chinese Self-stigma of Mental Illness Scale, DAI-10: Drug Attitude Inventory, DAS-DPA: Dysfunctional Attitude Scale - Defeatist Performance Attitude, GAF: Global Assessment of Functioning Scale, GPSES: Generalized Perceived Self-Efficacy Scale, ISMI: Internalized Stigma of Mental Illness Scale, KIDI: Knowledge of Illness and Drugs Inventory, LPSQ: Link Perceived Stigma Questionnaire, MANSA: The Manchester Short Assessment of Quality of Life, MES: Modified Engulfment Scale, MHRM: The Mental Health Recovery Measure, MSEI: Multidimensional Self-Esteem Inventory, MSPSS: Multidimensional Scale of Perceived Social Support, NECT: Narrative Enhancement/ Cognitive Therapy, PANSS: The Positive and Negative Syndrome Scale, PGRS: Personal Growth and Recovery Scale, PTCS: The Psychosocial Treatment Compliance Scale, QLS: The Quality of Life Scale, RSES: Rosenberg Self-Esteem Scale, SDS-J: Japanese version of Social Distance Scale, SES: Self-Efficacy Scale, SUMD: The Scale for Assessing Unawareness of Mental Disorder, TAU: Treatment as usual, TSCS: Tennessee Self-Concept Scale

Table 1: Summary of the effects of self-stigma reduction program

	Study	Participants' Diagnosis		Intervention		
			Sample Size			Duration
Author, year	Design; Country	Inclusion	(Pre-/Post)	Approach (Frequency)	Control	follow-u
Corrigan et al, 2015 (?)	RCT; U.S.	People with serious mental illness who have life disabilities	EG: 107/51 CG: 98/75	Coming Out Proud (COP) (2 hours session, 3 sessions, over 3 days or 1 day long group)	waitlist (=TAU??)	1 month
Costain et al., 2012 [25]	Uncontrolled clinical trial; Canada	Schizophrenia, Schizoaffective disorder	25/22 (21 for FU)	Genetic Counseling (1 session with mean 46.4 [SD 11.1] minutes)	NA	7 weeks
Çuhadar & Çam, 2014 [16]	RCT; Turkey	Biploar disorder	63/47	Psychoeducation program (90 minutes session; once a week; 7 sessions)		NA
Fung et al., 2010 [18]	RCT; Hong Kong	Schizophrenia	CG: 32/30 EG: 34/34	Self-stigma reduction program (Twice a week, 6 weeks; New life: once 1 week, 12 weeks)	Conventional Therapy	3 months

Lucksted et al., 2011 [26]	Pilot study; U.S.	Self Reported: Schizophrenia, Schizoaffective disorder, Major mood disorder	EG: 50/34	Ending Self-Stigma (ESS) with lectures and group discussions. (90 mins session; once a week, 9 week)	NA	NA
Lysaker et al., 2012 [28]	Quasi-experimental;U.S.	Schizophrenia, Schizoaffective disorder	EG: 70/70	Vocational Rehabilitation: part-time paid work placement (Non-specified frequency)	NA	5 months
McCay et al., 2007 [17]	RCT; Canada	Schizophrenia, Schizophreniform Disorder, Schizoaffective Disorder	EG: 67/47	Psychotherapy group (1.5 hours session; once a week;12 weeks)	TAU	NA
Russinova et al., 2014 [21]	RCT; US	Schizophrenia spectrum disorder, Bipolar disorder, Depressive disorder, Other diagonsis	EG: 40/38 CG: 42/37	Antistigma Photovoice (90 mins per session, 10 weeks)	TAU	3 months

Roe et al., 2013 [22]	Controlled clinical trails; Israel	SMI ≥ 50%	EG: 222/119	NECT (1-hour session; once a week; 20 sessions)	TAU	NA
Rüsch et al., 2014 [19]	Pilot RCT; Switzerland	Self Reported: Schizophrenia spectrum disorder, Bipolar disorder, Depressive disorder	CG: 50/47 EG:50/40	Coming Out Proud (COP): with peer-led group discussion. (2 hours session, once a week, 3 sessions)	TAU	3 weeks
Sibitz et al., 2013 [23]	Controlled Clinical Trial; Austria	Schizophrenia, Schizoaffective disorder, other F2 disorders	EG: 46/40 CG: 51/40	Recovery Oriented Day Clinic Treatment (8am-330pm, Mon - Friday, 2 months)	TAU	NA

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Starin et al., 2013 Uncontrolled clinical trial; [27] Netherlands		Schizophrenia, Schizoaffective disorder, Schizophreniform disorder, Psychotic disorder NOS	CBT-n (45-min session, once a week, 20 sessions)		NA	NA
Uchino et al., 2012 [24]	Controlled clinical trial; Japan	Schizophrenia, Schizoaffective disorder	EG: 29/29 CG: 27/27	Weekly psychoeducation program (Once a week, 6 weeks)	TAU	NA
Yanos et al., 2012 [20]	RCT; U.S.	Schizoaffective, Schizophrenia, Bipolar I, Bipolar II, Major depression	EG: 21/15 CG: 18/NR	NECT (1-hour session, once a week, 20 sessions)	TAU	3 months

CG: Control Group, EG: Experimental Group, NA: Non Applicable;

ACSS: Approaches to Coping with Stigma Scale, ADHS: Adult Dispositional Hope Scale, BDI: Beck Depression Inventory, BHS: The Beck Hopelessness Scale, BPIS: CAQ-SPMI:The Change Assessment Questionnaire for People with Severe and Persistent Mental Illness, CBT-n: Cognitive Behavioral Therapy for negative symptoms, out with Mental Illness Scale, CSC: The Coping with Symptoms Checklist, CSSMIS: The Chinese Self-stigma of Mental Illness Scale, DAI-10: Drug Attitude Inventory GPSES: Generalized Perceived Self-Efficacy Scale, ISMI: Internalized Stigma of Mental Illness Scale, KIDI: Knowledge of Illness and Drugs Inventory, LPSQ: Link Personal Composition of Mental Illness Scale, MHRM: The Mental Health Recovery Measure, MSEI: Multidimensional Self-Esteem Inventory, MSPSS: Multidimensional Scale of Perceived Social Support, Nand Recovery Scale, PTCS:The Psychosocial Treatment Compliance Scale, QLS: The Quality of Life Scale, RSES: Rosenberg Self-Esteem Scale, SDS-J: Japanese version Treatment as usual, TSCS: Tennessee Self-Concept Scale

Table.2 Quality Assessment of the included studies

Studies	Randomization	Randomization	Inclusion and	Patient	Outcome	Withdrawal	Sample	Intention-to-treat	Intervention	Qualification
	employed	methods	exclusion	Blinded	assessors	and	size	analysis	described	of the trainer
			criteria		blinded	dropouts	estimation			
Corrigan et	Y	N	Y	N	N	Y	Y	N	Y	N
al, 2015			(Inclusion							
			only)							
Costain et al., 2012	NA	NA	Y	N	N	Y	N	N	Y	N
Çuhadar & Çam, 2014	Y	Random digit number table	Y	N	N	Y	Y	N	Y	Y
Fung et al., 2010	Y	Generation of of random number by SPSS	Y	N	Y	Y	N	N	Y	N
Lucksted et	NA	NA	Y	N	N	Y	N	N	Y	NA
al.,2011			(Inclusion							
			only)							
Lysaker et al., 2012	NA	NA	Y	N	N	Y	N	N	Y	NA
McCay et al., 2007	Y	By randomly pulling group	Y	N	N	Y	N	N	N	Y
		assignment								
		(group vs								
		control from an								
		envelope)								

Russinova et	Y	Computer-gene	Y	N	N	Y	N	Y	Y	N
al., 2014		rated program	•	11	11					11
un, 2011		stratified on								
		gender &								
		racial-ethnic								
		minority status								
Roe et al.,	N	N	N	N	N	Y	N	N	Y	Y
2013				1	2 1					-
Rüsch et al.,	Y	Random	Y	N	N	Y	Y	N	Y	N
2014		number table,								
		and envelopes								
		with numbers								
		were drawn by								
		research								
		assistants who								
		had no								
		knowledge								
		which number								
		was in each								
		envelope								
Sibitz et al., 2013	N	NA	Y	N	Unblinded	Y	N	N	Y	Y
	NA	NA	Y	Unblinded	N	Y	N	Y	Y	Y
Staring et al., 2013	NA	NA	I	Unblinded	IN	I	IN	I	1	ĭ
Uchino et al.,	No	NA	Y	N	N	N	N	N	Y	N
2012										

Yanos et al.,	Y	Separated	Y	N	N	Y	N	N	Y	Y
2012		randomization								
		for two settings								
		based on								
		computerized								
		number								
		generating								
		system.								

N: not reported; NA: not applicable; Y: yes

Supplementary Appendix 1

Search strategy

- 1. "Internalized Stigma"
- 2. OR "Internalised Stigma"
- 3. OR "Stigma"
- 4. OR "Self-Stigma"
- 5. 1 or 2 or 3 or 4
- 6. "Severe mental illness*"
- 7. "Mental illness*"
- 8. "Schizophrenia"
- 9. "Psychotic disorder*"
- 10. "Psychosis"
- 11. "Delusional disorder*"
- 12. "Schizoaffective"
- 13. "Bipolar disorder"
- 14. "Personality disorder*"
- 15. 6 or 7 or 8 or 9 or 10 or 11 or 12 or 13 or 14
- 16. "Intervention*"
- 17. "Management"
- 18. "Motivational interviewing"
- 19. "Psychosocial"
- 20. "Treatment"
- 21. "Group*"
- 22. "Program*"
- 23. "Module*"
- 24. "Course*"
- 25. "CBT"
- 26. "Psychoeducation"
- 27. 16 or 17 or 18 or 19 or 20 or 21 or 22 or 23 or 24 or 25 or 26
- 28. "Clinical Trial"
- 29. "RCT"
- 30. "Controlled Trial"
- 31. "Experimental Studies"
- 32. 28 or 29 or 30 or 31
- 33. 5 and 15 and 27 and 32