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Title:

Social management training in individuals with 47,XXY (Klinefelter Syndrome): a neurocognitive-behavioral treatment targeting social, emotional and behavioral problems.

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Abstract:

Klinefelter syndrome (47,XXY) is associated with problems in social interaction and behavioral adaptation. Sixteen adolescents and adult men with 47,XXY enrolled in a pilot-study evaluating the effectiveness of Social Management Training, a novel neurocognitive-behavioral based treatment program targeted at improving social, emotional and behavioral functioning. Participants reported improved emotional stability from pre- to post-test (5 months). Informants reported reductions in internalizing and externalizing symptoms, including improvement in self-regulation. Although informants did not report changes in autism-like symptoms, increased awareness of social challenges was found. The Social Management Training may improve emotional stability, self-regulation and self-reflection in individuals with Klinefelter syndrome. This potentially efficacious treatment approach may prove to be a promising psychosocial therapeutic intervention for this population.

Keywords:

Sex chromosome aneuploidy, social functioning, intervention, psychosocial treatment.

Social management training in individuals with 47,XXY (Klinefelter Syndrome): A pilot study of a neurocognitive-behavioral treatment targeting social, emotional and behavioral problems

Abstract

Klinefelter syndrome (47,XXY) is associated with problems in social interaction and behavioral adaptation. Sixteen adolescents and adult men with 47,XXY enrolled in a pilot-study evaluating the effectiveness of Social Management Training, a novel neurocognitive-behavioral treatment program targeted at improving social, emotional, and behavioral functioning. Participants reported improved emotional stability from pre- to post-test (5 months). Informants reported reductions in internalizing and externalizing symptoms, including improvement in self-regulation. Although informants did not report changes in autism-like symptoms, increased awareness of social challenges was found. Social Management Training may improve emotional stability, self-regulation, and self-reflection in individuals with Klinefelter syndrome. This potentially efficacious treatment approach may prove to be a promising psychosocial therapeutic intervention for this population.

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Introduction

Klinefelter syndrome (47, XXY) is the most common chromosomal aberration among men. The presence of an extra X chromosome is estimated to occur in 1 in 500-600 males (Bojesen, Juul, & Gravholt, 2003; Boada, Janusz, Hutaff-Lee & Tartaglia, 2009). The physical and cognitive phenotypes associated with XXY are highly variable. However, certain cognitive-behavioral vulnerabilities are commonly reported, including language-based learning disabilities, executive functioning problems, and social disabilities (Geschwind, Boone, Miller & Swerdloff, 2000; Boone, Swerdloff, Miller, Geschwind, Razani, Lee, Gonzalo, Haddal, Rankin, Lu, & Paul, 2001; Rovet, Netley, Bailey, Keenan & Stewart, 1995; Graham, Bashir, Stark, Silbert & Walzer, 1988). Because of the increased likelihood of language, executive and social cognitive weaknesses, boys and men with Klinefelter syndrome are at risk for developing problems in social interaction and social adjustment. Many males with Klinefelter syndrome find social interaction difficult, appear introverted, anxious, impulsive, unassertive or socially withdrawn (Geschwind et al., 2000). On average, they report less participation in social interaction and more feelings of tension and stress in social situations than typically developing individuals (van Rijn, Swaab, Aleman & Kahn, 2006; van Rijn, Swaab, Aleman & Kahn, 2008b).

Men with Klinefelter syndrome sometimes report few friendships, low energy level and passivity, low participation in work and leisure activities, and limited contact with family (Nielson, Johnsen & Sorensen, 1980). If present, these problems clearly have a great impact on daily functioning and on self-esteem and consequently on quality of life. These social difficulties may be impactful and even carry the risk of developing serious psychopathology such as depression and social anxiety for which psychological counseling is indicated (Boone et al., 2001; Boada et al., 2009; Leggett, Jacobs, Nation, Scerif & Bishop, 2010; Van Rijn, Aleman, De Sonneville & Swaab, 2009).

Recent studies have shown that poor awareness of social competence and difficulties in executive functioning and emotion regulation were underlying mechanisms of social impairment in men with Klinefelter syndrome (Van Rijn et al., 2006; Van 't Wout, van Rijn, Jellema, Kahn & Aleman, 2009; Van Rijn, Stockmann, Borghgraef, Bruining, van Ravenswaaij, Govaerts, Hansson & Swaab, 2014; Van Rijn, Bierman, Bruining & Swaab, 2012). To date, no systematic studies have been published on the effects of interventions that target psychosocial problems in males with Klinefelter syndrome. It is important, however, to evaluate the effect of clinical care that is tailored to the neurocognitive profile of social cognitive vulnerability that is often found in males with Klinefelter syndrome. Evaluating specific interventions for Klinefelter syndrome is particularly important since prior research has demonstrated that the underlying mechanisms of social adaptation problems may be different in XXY as compared to other conditions with social dysfunction key to the disorder, such as autism (Brandenburg-Goddard, van Rijn, Rombouts, Veer, & Swaab, 2014; Goddard, Swaab, Rombouts, & van Rijn, 2015; van Rijn, Stockmann, van Buggenhout, van Ravenswaaij-Arts, & Swaab, 2014).

In order to improve clinical care for individuals with Klinefelter syndrome, we developed and evaluated a neurocognitive-behavioral self-management group treatment aimed at improving social, emotional, and behavioral functioning in individuals with Klinefelter syndrome seeking psychosocial support. Self-management refers to the ability of an individual to regulate their emotions and resulting behaviors in ways that are socially adaptive. This includes how the individual copes with unmet wants or needs, perseveres when faced with obstacles, and sets goals for oneself (Bandy & Moore, 2010). The goal of this social management training (SMT) is to increase the ability of individuals to regulate their emotions and resulting social behaviors in ways that are adaptive, by enhancing awareness of social competence, coping skills, self-confidence, and emotion regulation skills.

The Socio-Cognitive-Integration of Abilities (SOCIAL) Model was used as a theoretical framework for the training. This model defines the core dimensions of social skills on a biological, psychological, and social level (Beauchamp and Anderson, 2010). In the model, the biological and cognitive underpinnings of social competence are articulated; in addition, the influences of interactions with the environment on brain and behavior are described. The cognitive functions in the SOCIAL Model include perceptual functions, attention and executive functioning, social cognitive functions, and communication abilities. SMT, the treatment evaluated in the current study, was set up to train the aforementioned cognitive functions with a specific focus on executive functioning, given that executive functions are crucial to self-management of social behavior (Vohs & Baumeister, 2011). Executive functions (EF) are essential for flexible adaptive functioning in complex situations which have a high load of information, for inhibition of irrelevant thoughts and actions, for responding to changing environmental demands, and for the organization of thoughts and actions in a goal-directed way. Several components of this regulatory system can be distinguished, such as strategic planning, organized search, inhibition, focused and sustained attention, monitoring, holding a mental representation “on-line” in working memory and flexibility of thought and action (Anderson, 2001). This domain of cognitive function has been shown to be vulnerable in individuals with Klinefelter syndrome (Van Rijn & Swaab, 2015; Tartaglia, Cordeiro, Howell, Wilson & Janusz, 2010; DeLisi, Maurizio, Svetina, Ardekani, Szulc, Nierenberg, Leonard & Harvey, 2005; Ross, Zeger, Kushner, Zinn & Roeltgen, 2009; Lee, Wallace, Clasen, Lenroot, Blumenthal, White, Celano & Giedd, 2011; Skakkebak, Gravholt, Rasmussen, Bojesen, Jensen, Fedder, Laurberg, Hertz, Ostergaard, Pedersen & Wallentin, 2014).

In the current study, SMT group training involved four components: 1) pre-treatment individual neuropsychological assessment of known cognitive vulnerabilities in Klinefelter

syndrome that permitted the development of personalized goals and individual neurocognitive 'ID-cards', 2) psychoeducation based both on the profile of strengths and weaknesses that is often seen in Klinefelter syndrome, and personalized ID cards to enhance awareness of the social competence profile, 3) skills and strategy training, including role play and relaxation exercises in order to decrease social distress and to strengthen coping and emotion regulation skills, and 4) weekly exercises in a personalized workbook to increase transfer of social management skills to situations in daily life.

The effectiveness of the training was assessed in terms of changes in behavior in three behavioral domains: 1) emotional and behavioral problems, 2) social competence, and 3) autism-like behaviors. We evaluated this by questionnaires based on self-report as well as by informant questionnaires to disentangle subjective experiences and perspectives on behavior adaptation as evaluated by others. No specific a priori hypothesis were generated about treatment outcome or improvement of quality of life. However, the expectation was that a group training program should have a positive effect on wellbeing and social competence due to the effects of peer interactions, feelings of acceptance, recognition of corresponding problems, and the possibility to practice social skills in a group.

This study is the first to examine the effectiveness of a neurocognitive-behavioral treatment in males with Klinefelter syndrome. Because treatment studies that target psychological functioning and social behavior in individuals with sex chromosome anomalies are lacking, this study is unique and may prove to have important implications for clinical care and future research aimed at fine-tuning psychosocial treatments for individuals with Klinefelter syndrome.

Methods

Participants

Participants were individuals with Klinefelter syndrome referred to an academic outpatient department who were seeking psychosocial support due to problems in everyday or professional functioning. 16 males with Klinefelter syndrome agreed to participate in the SMT intervention. Psychosocial and behavioral problems in the clinical range were reported at the time of referral by the men themselves and by significant others on the ASEBA questionnaires (The Achenbach System of Empirically Based Assessment) (Achenbach & Rescorla, 2003). These occurred in the following domains: anxiety and feelings of depression, withdrawal, somatic complaints, aggressive behavior, thought problems and attention problems (table 1).

Exclusion criteria for the study were intellectual disability ($IQ < 70$) or inability to participate in the training (for example, because of extensive travel distance). One participant dropped out of the group treatment program after the first session because he preferred individual treatment.

The participants were 16 to 56 years old (mean age 37,5 years, SD 11,7). All but two men with Klinefelter syndrome were using testosterone supplements. Participants had intellectual abilities between 77 and 123 (mean IQ 96.7, SD 15.8).

Approval for the study was obtained from the medical ethics committee from the University Medical Center and all patients gave written informed consent.

Table 1

Therapeutic setting of the Social Management Training

SMT was delivered to the participants in ten, 90-minute sessions, every other week over a period of five months. The groups contained 4 to 8 participants and were chaired by

licensed clinical neuropsychologists. All trainers were experienced (group) therapists. Written material and weekly exercises were distributed to the participants before and after each session and were compiled into a participant's workbook. Taking into account language difficulties that men with Klinefelter syndrome may experience (Legett et al., 2010; Boada et al., 2009), schematic visual support of the psychoeducation, by means of PowerPoint presentation, was given during each session. Other adjustments included avoidance of psychological jargon, simplifying explanations and information, elaborately explaining the take home assignments as well as discussing the results of the take home assignment, ending each session with a summary and starting each session with a summary of the previous session.

Treatment protocol

Based on those cognitive functions from the SOCIAL-model that are relevant for social competence, as well as the typical profile of neuropsychological strengths and weaknesses in men with Klinefelter syndrome, the SMT included the following topics: social information processing, attention in a social environment, inhibition and emotion in a social environment, flexibility and planning in a social environment, and working memory in a social environment. It included a session with parents or partner (see table 2). It was thought that improving knowledge on these topics, practicing skills, and receiving feedback would help the participants train their executive functions which are important for social functioning. For example, increased working memory and attention may positively contribute to recognizing and processing social signals. Similarly, decreased impulsivity and increased flexibility may contribute to the appropriate selection and control of emotional reactions (Van Rijn & Swaab, 2015).

Each session was conducted following the same structure, starting with a summary and evaluation of the previous session, an introduction of the new topic, psychoeducation,

cognitive-behavioral exercises, and finally instructions for the take home assignment. All sessions ended with a relaxation exercise helpful to use in daily life to reduce stress and anxiety.

At the end of each session, the group evaluated a set of topics, such as relevance of the session subjects, quality and quantity of information (psychoeducation), (sub)goals of the individual sessions, involvement of the participants and the therapists, difficulty level, relevance of the weekly exercises, and current personal well-being. At the end of the treatment program, there was an individual session to discuss the results of the assessment, to hand over the participant's personalized ID card and workbook, and to individually evaluate the SMT.

Table 2

Assessment

Assessment design.

The effectiveness of the training was assessed in terms of changes in behavior in three domains: 1) emotional and behavioral problems, 2) social competence, and 3) autism-like behaviors. These changes were assessed not only via self-report, but also by use of informant report. The informant report questionnaires were completed by parents or partners. Pretest assessment occurred within a three-week period following referral to the academic outpatient department. After the intake and neuropsychological assessment, the participants were waitlisted for a period ranging from three weeks to three months to start SMT. Posttest assessment was conducted three weeks following the last SMT session.

Intellectual functioning.

Intellectual abilities were assessed using the Vocabulary and Block Design subtests of the Wechsler Adult Intelligence Scale or the Wechsler Intelligence Scale for Children, the V-BD short form. The V-BD short form is often used to estimate full scale intelligence (FSIQ)

according to the algorithm (2.9 (sum of normed scores) +42). The V-BD short form correlates highly with full scale IQ ($r=.88$) and the V-BD short form has been found valid for the estimation of intelligence, with good psychometric properties.

Emotional and behavioral problems.

The Achenbach System of Empirically Based Assessment (ASEBA) is a standardized measure of emotional and behavioral problems as well as social competence. It is normed for adults ages 18 to 59 years. The system includes report forms for multiple informants, including Adult Self-Report (ASR) and Adult Behavior Checklist forms (ABCL) (Achenbach & Rescorla, 2003). The child equivalent of the ASEBA form, Child Behavior Checklist has been used in several studies on boys with Klinefelter syndrome (Samango-Sprouse, Stapleton, Chea, Lawson, Sadeghin, Cappello, de Sonnevile & van Rijn S., 2018; Van Rijn & Swaab, 2015; Ross, Kushner, Kowal, Bardsley, Davis, Reiss, Tartaglia & Roeltgen, 2017; Ross, Roeltgen, Kushner, Zinn, Reiss, Bardsley, McCauley & Tartaglia, 2012). Reliability and validity for the ASEBA and the CBCL is well-established. For the ASEBA, the norms are based on an American national sample in the general population.

Self-report: ASR.

The ASR is a self-administered instrument that examines diverse aspects of adaptive functioning and problems. Profiles display scale scores in relation to norms for each gender at ages 18-35 years and 36-59 years. Eight subscales cover diverse aspects of adaptive, emotional and behavioral functioning; anxious/depressed behavior, withdrawn behavior, somatic complaints, thought problems, attention problems, aggressive behavior, rule-breaking behavior and intrusive behavior. Both forms have parallel scales for internalizing, externalizing, and total problems. Raw scores for each scale are converted to norm-referenced T-scores ($M = 50$, $SD = 10$). Higher scores indicate more problems, with the cutoff for the clinical range at a T-score of ≥ 67.8 .

Informant report: ABCL.

The ABCL contains scales parallel to the ASR and can be used to obtain information about the individual assessed by others who know the individual well, such as a spouse, partner, family member, or friend. The ABCL contains items on diverse aspects of adaptive functioning and problems.

Social competence.

Self-report: SIB.

The Scales of Independent Behavior - Revised (Bruininks, Woodcock, Weatherman & Bradley, 1996) is a self-administered questionnaire that assesses the frequency of participation in social interactions and intensity of distress during social interactions. In addition to an overall total score, there are four factorially-derived subscales: (I) Display of negative feelings (negative assertion), such as refusing a request or standing up for one's rights in a public situation, (II) Expression of and dealing with personal limitations, such as ability to deal with criticism or requesting attention/help, (III) Initiating assertiveness, such as starting a conversation with strangers or expressing one's own opinion, and (IV) Praising others and the ability to deal with compliments/praise of others (positive assertion), such as giving and receiving compliments. Scores that are obtained from the SIB represent mean item-scores for each dimension of social behavior, on a scale from one (high frequency or low intensity of distress) to five (low frequency or high intensity of distress). In this study, the Dutch version of the SIB was used. The SIB has been well validated in the general population as well as in individuals with social phobia and both psychiatric outpatients and inpatients (Arrindell, Bridges, van der Ende, St Lawrence, Gray-Shellberg, Harnish, Rogers & Sanderman, 2001; Arrindell, De Groot & Walburg, 1984). In a previous study with 31 men with Klinefelter syndrome, the SIB was proven to be a reliable instrument to assess social distress (van Rijn et al 2008) in men with Klinefelter syndrome compared to a control group

of men from the general population. XXY men reported increased distress during social interactions and less engagement in specific social behaviors.

Informant report: SSRS.

The Social Skills Rating System (SSRS) (Gresham & Elliot, 1990) is an informant questionnaire that assesses social skills across four subscales: Cooperation (e.g. 'Helps you with household tasks without being asked'), Assertion (e.g. 'Starts conversations spontaneously rather than waiting for others to talk first'), Self-control (e.g. 'Ends disagreements with you calmly'), and Responsibility (e.g. 'Requests permission before leaving the house'). Each of the four subscales consists of 10 items, which are rated on a 3-point Likert scale. Higher scores indicate better social skills (Gresham & Elliott, 1990).

The Dutch version of the SSRS has been used in a previous study with XXY boys (van Rijn, Stockmann, Borghgraef, Bruining, van Ravenswaaij, Govaerts, Hansson & Swaab, 2014).

Autism-like traits.

Self-report: AQ.

The Autism Spectrum Quotient (AQ) (Baron-Cohen, Weelwright, Skinner, Martin & Clubley, 2001) is a self-administered questionnaire that assesses the degree to which any individual adult of normal intelligence might have features of the core autistic phenotype. Five subscales cover personality traits associated with the autistic spectrum: social skills, communication, imagination, attention to detail, and attention switching. Scores on the AQ are reported to be normally distributed in the general population. Higher scores on the AQ indicate higher levels of autism traits.

Informant report: SRS.

The Social Responsiveness Scale (Constantino & Gruber, 2012) distinguishes autism spectrum conditions by identifying the presence and extent of autistic social impairment. It is

an informant-report questionnaire that assesses the degree of autism spectrum symptoms as they occur in natural social settings. The SRS includes items that ascertain social awareness, social cognition, social communication, social motivation and autistic mannerisms. Higher scores indicate higher levels of autism traits. The Dutch version of the SRS (Noens, De la Marche & Scholte, 2012) was used in this study.

The SRS has been developed as a diagnostic survey to assess social impairments associated with autism spectrum disorders and to quantify its severity. However, the SRS has been shown to be useful in conceptualizing general social impairment non-specific to autism, and can be used to identify social difficulties in other populations as well, such as individuals diagnosed with ADHD (Reiersen et al., 2007).

The SRS has proven to be a reliable instrument in studies concerning social challenges and autistic traits in individuals with Klinefelter syndrome (Tartaglia et al., 2010).

Statistical analysis

Data were analyzed using SPSS (Statistical Package for the Social Sciences) version 17.0. Paired t-tests were used to assess within subject effects of treatment on level of symptoms at baseline (pre-test T1) as compared to follow-up (post-test T2). In order to control for multiple comparisons, paired t-tests of specific subdomains for each measure were evaluated only in the case of significant effects on the instrument's total score. Level of significance was set at $p < 0.05$.

To determine if a significant effect was clinically meaningful, the distribution-based method according to the concept of the Minimal Clinically Important Difference was used (Stefanovics, Rosenheck, Jones, Huang, & Krystal, 2018). A minimal clinically important difference was operationalized as a change in score on an outcome measure that was more than one-half of the measure's standard deviation or a change in score of Cohen's $d > 0.5$.

Results

Emotional and behavioral problems**Self-report: ASR.**

Paired samples t-test revealed a borderline significant change in total ASR score from pretest to posttest ($t(9) = 2.0, p = .07$). Further examination of syndrome scales showed a significant decrease of anxious/depressed behavior from pretest to posttest ($t(9) = 2.7, p = .02$). Scores are presented in table 3.

Informant report: ABCL.

Paired samples t-test revealed significant improvements on total ABCL score from pretest to posttest ($t(8) = 2.27, p = .02$). Paired T tests for specific broadband scales, showed a significant decrease in internalizing behavior from baseline to follow up ($t(8) = 2.27, p = .05$). No significant effects on the subscales of internalizing behavior were found. A significant decrease in externalizing behavior from pretest to posttest ($t(8) = 3.21, p = .01$) was found. Further specification on syndrome scales of externalizing behavior showed a significant decrease in aggressive behavior from pretest to posttest ($t(8) = 2.6, p = .032$) and a significant decrease in rule-breaking behavior from pretest to posttest ($t(8) = 2.9, p = .021$). The subscale attention problems, which is also part of the total score but not contributing to internalizing or externalizing sum scores, showed a significant decrease from pretest to posttest ($t(8) = 2.8, p = .024$). Scores are presented in table 3.

Social competence**Self-report: SIB.**

Overall distress during social interaction was not significantly decreased after participation in the training program ($M = 116.1, SD = 35.17$) compared to distress in social situations before training ($M = 129.5, SD = 39.52$); $t(13) = 1.89, p = 0.082$. Scores are presented in table 3. Overall frequency of engagement in social behavior did not significantly

differ ($t(13) = -0.83, p = 0.421$) between pretest ($M = 139.9, SD = 20.95$) and posttest ($M = 145.2, SD = 25.14$).

Informant report: SSRS.

A proxy paired samples t-test revealed no significant difference in social competence on the SSRS from pre-test ($M = 49.7, SD = 11.1$) to post-test ($M = 53.3, SD = 14.6; t(8) = -0.64, p = 0.54$).

Autism-like traits

Self-report: AQ.

A paired samples t-test revealed a significant increase on total AQ score from pretest to posttest ($t(13) = -2.36, p = 0.03$). Subsequent analyses of the AQ subscales showed that on the Social skills subscale there was an increase of autistic traits from pretest to posttest ($t(13) = -2.72, p = .02$). On the Communication subscale, an increase of autistic traits from baseline to follow up ($t(13) = -2.43, p = .03$) was found. Scores are presented in table 3.

Informant report: SRS.

A paired samples t-test revealed no significant change in total SRS scores from pretest to posttest ($t(11) = -0.15, p = .88$). Scores are presented in table 3.

Table 3

Discussion

There is a great need for evidence-based psychosocial treatment programs for individuals with Klinefelter syndrome. This pilot study is one of the first to assess the effectiveness of a treatment program specifically tailored to the typical cognitive-behavioral profile found in individuals with Klinefelter syndrome. In this study, clinically-referred adolescents and adults with Klinefelter syndrome participated in the newly developed Social Management Training (SMT), which was aimed at increasing overall social adaptability in daily life through increased self-management of social behavior.

Promising results were found regarding the effectiveness of SMT. In general, the participants and their significant others reported decreased scores on negative behaviors and improved scores on positive behaviors on the various questionnaires which indicates a general experience of improvement. Informant reports showed a significant decrease in attention problems (effect size 0.93), aggression (effect size 0.95), rule breaking behavior (effect size 0.87), and internalizing problems (effect size 0.76). These changes in scores exceed the recommended cutoff for a Minimal Clinically Important Difference (Stefanovics et al., 2018), suggesting that these findings are clinically meaningful. The improvement in attention reflects a difference between mean scores in the borderline range to scores in the normal range. The scores in aggression and rule breaking behavior both improved, although the mean scores were already in the non-clinical range pre and post treatment. The scores on internalizing problems improved from the clinical range to the borderline range. Self-reports showed a significant decrease in anxiety and depression (effect size 0.87) and a trend for reduced social distress (effect size 0.50). Interestingly, although informants did not report changes in autism-like behaviors, following the intervention there was a significantly and clinically relevant increased awareness of such autism like behaviors according to self-report assessment. Effect sizes on the AQ subscales ranged from 0.65 to 0.73. There was no significant change in social skills and adaptive behavior reported by informants.

The main conclusion of this study is that SMT may lead to improvements in awareness (insight), coping, and behavioral adaptation. The participants became more self-aware of their autism-like behaviors and areas of social challenge following intervention. Whether this change in awareness of autism-like behavior is the outcome of an actual increased understanding of one's limitations or due to the effect of being more comfortable in reporting one's challenges after treatment remains unclear. That the participants reported increases in the awareness of their limitations in social functioning and adaptation after participating in

SMT is an important result, because awareness of shortcomings in social flexibility and communication and increased confidence in reporting them is a prerequisite for learning compensatory strategies. Being aware of limitations of social adaptive skills, including communication and flexibility, is fundamental to coping with emotional feelings and managing related behavioral impulses. According to studies on emotion regulation, conscious awareness of emotions in relation to one's behavior is essential for the use of adaptive emotion regulation strategies (Subic-Wrana, Beutel, Brähler, Stöbel-Richter, Knebel, Lane & Wiltink, 2014; Koole, 2009). The finding that others, however, did not report changes in autism-like behaviors, may illustrate that learning compensatory and alternative strategies may be a valuable avenue of support rather than focusing on social skills training alone. Increase in awareness and self-reflection may be the first step in improving coping and behavior adaptation. Being aware of problems, feelings, thoughts, and behavior may lead to a reduction in anxiety and distress. Subic-Wrana et al. (2014), for example, found that the use of reappraisal of one's situation as an emotion regulation strategy to reduce physiological arousal had a positive effect on symptoms of anxiety and depression.

Following SMT, participants reported that their feelings and symptoms of (social) anxiety and depression were reduced. This reduction in overall distress may, in time, result in opportunities for increased assertiveness, such as starting a conversation with a stranger or expressing one's own opinion. Furthermore, these reductions in anxiety may allow individuals to be better able to express positive emotions to others and have positive social interactions as a result.

Reductions in externalizing behaviors, such as aggression and offensive behavior, were also reported by informants. Decreases in such behaviors lead to increased self-control, which is an important factor supporting adequate interaction with other people. Improvements in attention were reported by informants (but not on self-report). The finding that attention

problems seem to decrease (although still in the clinical range of problem behavior), may help explain this improved behavioral control. It is possible that improved attention could help individuals to stay focused in social interactions and prevent impulsive reactions to others.

Taken together, the results of the current study show that participants engaging in SMT improved in several important areas of functioning, including self-awareness and behavior adaptation. The preliminary finding that SMT may indirectly contribute to reduced anxiety through improved self-regulation (Subic-Wrana et al., 2014) is especially significant, given that these are areas of vulnerability for men with Klinefelter syndrome (Bender, Harmon, Linden & Robinson, 1995).

There were also areas of functioning that did not change following the SMT. Specifically, no changes in social skills as expressed in frequency of social participation were found. Although this may indicate that SMT is not suitable to improve actual social skills, we cannot exclude that reduced anxiety and improved self-control may over time lead to more positive social learning experiences, and possibly increased social participation in the long term. Further research, in the form of long term follow-up studies, is necessary to assess this. This finding does however suggest that the SMT should not be considered a 'social skills training', but rather a training to improve self-management of social awareness and emotional control.

Regarding the clinical utility of the SMT, according to subjective evaluation, participants indicated that they experienced the training as being helpful and effective. The participants reported that the group therapy format involving interactions with other men with Klinefelter syndrome, the psychoeducation, and the relaxation exercises were the most helpful components of the therapeutic process. Participants were also asked for recommendations for improvements to the treatment. They reported that the take home assignments were sometimes difficult to understand and to apply. To improve generalization and applicability to

everyday situations, we recommend including a coach or to use partner participation in the implementation of behavioral exercises in daily life. Step-by-step explanation of the exercises by adding elements and demonstrations may also further contribute to the intended behavioral change. Despite the concerns about the complexity and feasibility of the take home assignments and applicability of the exercises in daily life for some participants, all participants showed great motivation to complete the homework assignment forms. All forms were completed, and there was almost one hundred percent attendance throughout the treatment sessions. The motivation, determination, and dedication of the men with Klinefelter syndrome to participate in the training and to complete the treatment program suggests that SMT has promise as a treatment for this group. In order to quantify feasibility and acceptability of the training, to measure useful elements of the training, and to increase commitment and involvement from the participants and the trainers, future research may benefit from the use of a questionnaire to measure the treatment outcome, such as the Outcome Rating Scale, the Session Rating Scale, or the Group Rating Scale (Miller, Duncan, Brown, Sparks & Cloud, 2003; Duncan, Miller, Sparks, Cloud, Reynolds, Brown, & Johnson, 2003; Norcross & Wampold, 2011; Harmon, Lambert, Smart, Hawkins, Nielson, Slade, & Lutz, 2007).

While the results of this study are encouraging, future research is needed to address its limitations. First, the small sample size limits the generalizability of the results, and replication is therefore necessary. The results of this preliminary study may be used to formulate specific hypotheses about treatment outcome and encourage larger controlled studies. Second, a control group who received care as usual was not included in the study. A controlled clinical study is warranted to further investigate which elements of the group training were most effective. Third, the multidimensional approach of SMT (i.e. focusing on specific neurocognitive topics, and including partner sessions, setting personal targets,

training exercises, psychoeducation, and relaxation exercises) makes it difficult to identify which particular aspects of the training are necessary or sufficient for improving adaptability in daily life. Fourth, it is important to mention that the outcome measures used in this study may not adequately capture some positive treatment effects, as some areas of experienced improvement may be more subtle or not tapped by the items included on the scales. Additional use of performance-based neurocognitive measures could also be an interesting line of inquiry. Fifth, the current study only examined post-treatment outcome, and did not have a follow-up period to investigate maintenance and reinforcement of treatment gains. Finally, it is important to stress that the group of men with Klinefelter syndrome included in the current study represented a more severely affected subpopulation, as they were all referred to an academic outpatient department. Although this limits generalizability for the Klinefelter group as a whole, it appears appropriate for the subgroup of those with Klinefelter syndrome for which SMT was developed, i.e. adolescents and adults with Klinefelter syndrome who experience difficulties in adaptation to daily life with emotional and behavioral problems as a result. Thus, the findings appear representative for the group of men who seek cognitive behavioral interventions in the clinical setting.

Given the negative psychosocial effects of growing up and living with Klinefelter syndrome on everyday functioning, it is important that effective and evidence-based psychological treatment protocols become available in order to improve tailored care for this group. The findings of the current study suggest that a multidimensional, neurocognitive-behavioral group intervention (SMT) may be an effective way of enhancing the self-management of social behavior, including increased awareness of social competence, coping and adaptive functioning, in adolescents and adult males with Klinefelter syndrome.

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Table 1

Percentage of participants in the clinical range on the ASEBA questionnaires (N =16) at baseline of the study.

ASEBA	Self-report (ASR)	Informant report (ABCL)
Total problems	20.1 %	8.3%
Internalizing problems	46.2%	41.7%
Anxious/depressed	30.8%	25.0%
Withdrawn behavior	38.5%	33.0%
Somatic complaints	38.5%	25.0%
Externalizing problems	7.7%	8.3%
Intrusive behavior	0.0%	8.3%
Aggressive behavior	20.1%	8.3%
Rule-breaking behavior	0.0%	0.0%
Other problems		
Thought problems	15.4%	8.3%
Attention problems	46.2%	50.0%

Table 2

Overview: contents of the Social Management Training.

Session no.	Topics
1	Introduction and learning targets
2	Information process in a social context
3	Attention and emotions in a social context
4	Inhibition and emotions in a social context
5	Flexibility and planning in a social context
6	Working memory in a social context
7	Parent/partner session
8	Individual learning targets
9	Individual learning targets
10	Summary/integration

Table 3

Outcome measures from pretest to posttest.

Outcome measure:	Baseline	Follow-up	Statistics	Cohen's d
	M (SD)	M (SD)		
Emotional and behavioral problems				
<i>Self-report</i>				
ASR-Total score	65.7 (± 12.9)	61.5 (± 14.4)	$p = .07^*$	$d = 0.64^*$
Internalizing problems	69.4 (± 12.4)	64.8 (± 13.8)	$p = .15$	
Anxious/depressed	67.2 (± 12.7)	62.4 (± 11.2)	$p = .02^*$	$d = 0.87^{**}$
Withdrawn/depressed	68.7 (± 11.2)	66.6 (± 12.1)	$p = .43$	
Somatic complaints	67.8 (± 11.5)	63.8 (± 10.6)	$p = .12$	
Externalizing problems	58.3 (± 9.1)	57.2 (± 10.6)	$p = .49$	
Aggression	63.0 (± 9.4)	61.0 (± 9.5)	$p = .43$	
Rule breaking behavior	56.2 (± 5.0)	55.2 (± 6.3)	$p = .47$	
Intrusive behavior	54.0 (± 5.9)	50.9 (± 8.1)	$p = .26$	
Other problems				
Thought problems	62.3 (± 11.1)	61.3 (± 9.9)	$p = .49$	
Attention problems	68.7 (± 9.1)	65.8 (± 9.6)	$p = .19$	
<i>Informant report</i>				
ABCL-Total score	64.1 (± 3.7)	61.7 (± 3.8)	$p = .02^*$	$d = 0.92^{**}$
Internalizing problems	70.1 (± 6.6)	65.9 (± 5.6)	$p = .05^*$	$d = 0.76^*$
Anxious/depressed	65.6 (± 6.5)	63.1 (± 5.5)	$p = .06^{**}$	$d = 0.72^*$
Withdrawn/depressed	67.8 (± 10.5)	67.7 (± 11.3)	$p = .95$	
Somatic complaints	67.9 (± 7.9)	62.2 (± 8.6)	$p = .11$	
Externalizing problems	60.7 (± 5.1)	58.8 (± 4.6)	$p = .01^*$	$d = 1.07^{**}$
Aggression	63.7 (± 4.5)	61.4 (± 4.3)	$p = .02^*$	$d = 0.95^{**}$
Rule breaking behavior	57.3 (± 4.7)	55.0 (± 3.8)	$p = .03^*$	$d = 0.87^{**}$
Intrusive behavior	54.0 (± 5.9)	50.9 (± 8.1)	$p = .26$	
Other problems				
Thought problems	61.2 (± 6.7)	57.9 (± 7.2)	$p = .13$	
Attention problems	66.8 (± 5.1)	63.8 (± 3.5)	$p = .02^*$	$d = 0.93^{**}$
Social competence				
<i>Self-report</i>				
SIB-Total distress	129.5 (± 39.5)	116.1 (± 35.2)	$p = .08^{**}$	$d = 0.50^*$
SIB-Total frequency	139.9 (± 20.9)	145.2 (± 25.1)	$p = .42$	
<i>Informant report</i>				
SSRS-Total score	49.7 (± 11.1)	53.3 (± 14.6)	$p = .54$	
Cooperation	12.3 (± 4.7)	14.9 (± 2.3)	$p = .17$	
Assertion	12.3 (± 2.7)	11.6 (± 4.8)	$p = .64$	
Responsibility	13.3 (± 3.0)	14.2 (± 5.0)	$p = .59$	
Self-control	11.7 (± 2.4)	12.7 (± 4.1)	$p = .49$	
Autism-like traits				
<i>Self-report</i>				
AQ-Total score	107.4 (± 25.0)	126.8 (± 16.8)	$p = .03^*$	$d = 0.63^*$
Social skills	21.3 (± 8.1)	27.7 (± 6.0)	$p = .02^*$	$d = 0.73^*$
Attention switching	24.4 (± 7.7)	28.8 (± 5.0)	$p = .08$	
Attention to detail	21.0 (± 5.2)	21.4 (± 5.4)	$p = .83$	
Communication	19.9 (± 5.7)	25.3 (± 5.0)	$p = .03^*$	$d = 0.65^*$
Imagination	20.9 (± 3.7)	23.6 (± 4.6)	$p = .08$	
<i>Informant report</i>				
SRS-Total score	64.9 (± 21.9)	65.9 (± 22.7)	$p = .88$	
Awareness	19.3 (± 7.1)	19.3 (± 6.3)	$p = 1.0$	
Communication	22.3 (± 7.2)	23.6 (± 10.1)	$p = .64$	
Motivation	14.3 (± 7.6)	14.3 (± 5.9)	$p = .97$	
Autistic mannerisms	9.0 (± 3.5)	7.6 (± 5.2)	$p = .14$	

Statistics: *significant effect, **borderline effect

Cohen's d: *medium effect, **large effect

ASR = ASEBA Adult Self report (high scores indicate more problems), ABCL = ASEBA Adult Behavior Checklist (high scores indicate more problems), SIB = Scale for Interpersonal Behavior (high scores indicate high intensity of distress and low frequency of social interaction), SSRS = Social Skills Rating System (high scores indicate better social skills), AQ = Autism Questionnaire (high scores indicate higher level of autism traits), SRS = Social Responsive Scale (high scores indicate higher level of autism traits)