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Supplementing stuttering treatment with online cognitive behavior therapy: An experimental trial

Authors

Ross Menzies a,1, Sue O’Brian a,1, Ann Packman a,1, Mark Jones b,2, Fjóla Dógg Helgadóttir a,c,3, Mark Onslow a,1*,

a Australian Stuttering Research Centre, The University of Sydney, Australia

b School of Public Health, The University of Queensland, Australia

c The Vancouver CBT Centre, Canada (current location for author)

*Corresponding Author: Mark Onslow. Australian Stuttering Research Centre, University of Technology Sydney, Building 1, 15 Broadway, Ultimo, NSW, 2007, AUSTRALIA. E-mail: Mark.Onslow@uts.edu.au

1 Present address: Australian Stuttering Research Centre, University of Technology Sydney, Building 1, 15 Broadway, Ultimo, NSW, 2007, Australia

2 Present address: Bond University, Gold Coast, Australia

3 Present address: private practice, Vancouver, Canada
SUPPLEMENTING STUTTERING TREATMENT WITH ONLINE CBT

E-mail addresses: Ross.Menzies@uts.edu.au (R.G. Menzies), Susan.Obrian@uts.edu.au (S. O’Brien), Ann.Packman@uts.edu.au (A. Packman), majones@bond.edu.au (M. Jones), fjola@ai-therapy.com (F. Dógg Helgadóttir), Mark.Onslow@uts.edu.au (M. Onslow)
SUPPLEMENTING STUTTERING TREATMENT WITH ONLINE CBT

Abstract

Purpose: It is now well established that adults who present to speech clinics for help with stuttering will have an increased risk of having an anxiety disorder, particularly social anxiety disorder. Concomitant psychological problems are known to interfere with the maintenance of the benefits of behavioral speech treatments for stuttering. The current team has developed and trialed a cognitive behavior therapy (CBT) program designed specifically to reduce anxiety in adults who stutter, and trials have shown promise for both an in-clinic version and a standalone internet-based version. The aim of the present study is to determine whether iGlebe, the internet-based version of the team’s internet CBT treatment (previously known as CBTPsych), enhances the benefits of behavioral stuttering treatment.

Method: Participants were 32 adults seeking treatment for stuttering. The design was a two-arm randomized experimental trial with blinded outcome assessments at 6 and 12 months post-randomization. Both arms received basic speech-restructuring training to reduce stuttering, without any anxiolytic (anxiety reducing) components. The experimental arm also received 5 months access to iGlebe.

Results: There was evidence that, at 12 months post-randomization, iGlebe added clinically significant improvements to self-reported stuttering severity and quality of life. The present experimental trial provides the first evidence that the addition of CBT to speech restructuring improves speech outcomes.

Conclusions: The present results will be the basis for the development of a comprehensive, internet-based treatment program for anxiety associated with stuttering. Ultimately, it may be possible for such an economical, scalable, and translatable comprehensive treatment model to supplement standard speech-language pathology treatment practices for those who stutter.

Keywords: Stuttering, Adult, Speech, Speech restructuring, CBT
1. Introduction

1.1 Stuttering and social anxiety

Social anxiety disorder (also known as social phobia) is a condition that centers on intense fear of negative evaluation in social situations (American Psychiatric Association, 2013). Avoidance of activities that may involve scrutiny from others is commonplace among those with social anxiety disorder. The Diagnostic and Statistical Manual of Mental Disorders (DSM-5) lists a variety of situations that present difficulties for people with social anxiety disorder (American Psychiatric Association, 2013), including public speaking, meeting new people, leading a group discussion, and conversations with potential to involve conflict or confrontation. Social anxiety disorder is a particularly disabling condition because so many aspects of modern life involve verbal interaction. Social anxiety disorder is associated with reduced educational and occupational achievement and lower socioeconomic status (Stein & Kean, 2000).

Adults who stutter and who are seeking help for their stuttering have increased risk of social anxiety disorder. Data from several independent research teams suggest that up to 60% of adults seeking treatment for stuttering have clinically significant levels of social-evaluative fear, the core construct of social anxiety disorder (e.g., Blumgart, Tran, & Craig, 2010; Iverach et al., 2009; McAllister et al., 2017; Menzies, O’Brien, Onslow, Packman, St. Clare, & Block, 2008; Stein, Baird, & Walker, 1996). In a comprehensive demonstration of the increased risk of social anxiety disorder associated with adults who stutter, Iverach et al. (2009) used a structured psychiatric interview to compare 92 treatment-seeking adults who stuttered with 920 age- and sex-matched non-stuttering community controls. Participants who stuttered were shown to have 16- to 34-fold increased odds of meeting established diagnostic criteria for social anxiety disorder.
Children who stutter are also at risk of developing social anxiety disorder (Iverach et al., 2016). In a study of 75 children aged 7–12 years who were stuttering, Iverach et al. (2016) found the stuttering children to have a six-fold increased risk of having social anxiety disorder compared to controls, with a prevalence of 24%. However, the majority of the children (80%) who were stuttering were receiving or had received treatment for stuttering. Hence, the children selected in this study may have been from a severely affected population of children who stutter. This could be the reason that those findings were not confirmed by Smith et al. (2017) for a non-clinical, community sample of 20 school-age children who were stuttering.

Many social situations are known to arouse fear for many who stutter, and avoiding situations is a common way to cope with that fear. The high prevalence of social anxiety disorder among those who stutter, and the potentially debilitating effects of the condition, indicate there is a need to screen those seeking stuttering treatment for social anxiety. A scale developed specifically for people who stutter by the present team and colleagues, the Unhelpful Thoughts and Beliefs About Stuttering (UTBAS), can be used for this purpose (see Iverach et al., 2011). The call for screening is given more weight by the likely effect of social anxiety disorder on treatment outcomes for speech-restructuring programs. Speech restructuring involves the person adopting a novel speech pattern such as smooth speech or prolonged speech, which are known to reduce stuttering (Onslow & Menzies, 2010). Iverach et al., (2009) found that the presence of mental health conditions—any anxiety, personality, or mood disorder—was associated with failure to maintain the benefits of speech-restructuring treatment programs. Only those without a mental health diagnosis before treatment were able to control their stuttering 6 months after treatment. At that assessment, those with a mental health disorder were unable to effectively demonstrate a restructured
SUPPLEMENTING STUTTERING TREATMENT WITH ONLINE CBT

speech technique in beyond-clinic phone calls (Iverach et al., 2009). These results are consistent with client reports in a study by Craig and Hancock (1995).

1.2 Social anxiety intervention for adults who stutter

Given the increased rates of anxiety-related mental health conditions associated with stuttering and the effect of those conditions on outcomes of behavioral treatments for stuttering, it is imperative that effective treatment for anxiety-related disorders be available for those who stutter. Cognitive behavior therapy (CBT) is the gold standard psychological intervention for social anxiety, with controlled effect sizes in the range of $d = 0.70$ (Acarturk, Cuijpers, van Straten, & de Graaf, 2009) to $d = 0.86$ (Powers, Sigmarsson, & Emmelkamp, 2008). CBT includes strategies to encourage the unlearning of fear associated with social situations. These strategies are based on cognitive restructuring and include exposure to feared activities, behavioral experiments to challenge negative expectations about social encounters, and elimination of safety behaviors, checking behaviors, and avoidance. Safety behaviors are thoughts and behaviors that a socially anxious person continues to engage in, believing erroneously that they enhance their performance in social situations (Clarke, & Wells, 1995; for safety behaviors and stuttering see Helgadottir, Menzies, Onslow, Packman, & O’Brien, 2014a). The use of safety behaviors is known to minimize the chances of social anxiety decreasing.

Early research into CBT for stuttering (e.g., Blood, 1995; Maxwell, 1982) combined the cognitive and behavioral procedures of CBT with speech procedures designed to decrease stuttering. However, in this research the contribution of CBT to outcomes is difficult to establish. For example, Maxwell (1982) combined speech treatment elements with the (now discredited) thought-stopping technique of suppressing maladaptive thoughts. With a group of 23 adults who stuttered, Maxwell reported significant reductions in stuttering severity. Similarly, Blood (1995) combined a computer-assisted biofeedback program for reducing
SUPPLEMENTING STUTTERING TREATMENT WITH ONLINE CBT

stuttering with CBT based on Bandura’s self-efficacy model (Cameron & Meichenbaum, 1980) and the Donovan and Marlatt (1980) relapse prevention model. Although reductions of stuttering severity were observed and maintained after 1 year, the relative contributions of the speech treatment and the psychological interventions cannot be isolated.

More recently, Ezrati-Vinacour, Gilboa-Schechtman, Anholt, Weizman, and Hermesh (2007) reported on a group CBT package for people who stutter with comorbid social anxiety disorder. Participants received 18 weekly group cognitive-behavioral sessions of 1.5 hours duration. The treatment program included psychoeducation about the nature of social anxiety, cognitive restructuring, behavioral experiments, and procedures to encourage the elimination of safety behaviors and avoidance strategies. Post-treatment scores were statistically significantly lower than pre-treatment scores for a range of psychological and emotional measures, but not for percentage syllables stuttered. The authors concluded that CBT may produce improvements of everyday functioning, anxiety, and emotional reactivity to stuttering. However, there was no control group and no long-term outcomes. Hence, only tentative conclusions about the efficacy of CBT for those who stutter can be made from that study.

The present authors and colleagues have developed a CBT program designed specifically for adults who stutter (Menzies et al., 2008). Menzies et al. (2008) randomly allocated 30 adults who stuttered to either 14 hours of speech-restructuring treatment alone or 14 hours of speech restructuring-treatment preceded by 12 one-hour weekly in-clinic sessions of this CBT program. CBT sessions centered on restructuring unhelpful thoughts (e.g., “people will think I’m stupid if I stutter”), exposure to feared situations (e.g., making phone calls), removal of avoidance behaviours (e.g., avoiding particular syllables), and safety behaviors (e.g., asking open-ended questions to keep the conversational partner talking). The speech-restructuring treatment, “was modified specifically to ensure that it contained only
SUPPLEMENTING STUTTERING TREATMENT WITH ONLINE CBT

techniques for the control of stuttered speech and was free of the cognitive and exposure interventions typically incorporated into the treatment” (Menzies et al., 2008, p. 1455). The majority of participants in both groups were identified as having social anxiety disorder, with no significant difference between the groups.

Immediately after the speech treatment, and at 12 months follow-up, those who had received CBT (1) had significantly higher mean Global Assessment of Functioning (American Psychiatric Association, 2000) scores, (2) were able to complete more tasks in individualized fear hierarchies, and (3) were less likely to be diagnosed with social anxiety disorder. In fact, no participant who received CBT was diagnosed with social phobia in blinded clinical interviews at follow-up. This report offers the strongest support to date for the claim that CBT may be a useful adjunct to speech restructuring for adults who stutter. Notably, although the addition of CBT significantly improved psychological outcomes, it did not enhance speech outcomes of participants.

1.3 Internet-delivered CBT for those who stutter

Recently, there has been considerable interest in providing CBT treatments, and indeed many mental health interventions, on the internet. The advantages of this mode of delivery include (1) low cost of delivery with no or minimal clinician expenses, (2) access in remote areas where community treatment services may be limited, (3) convenience for the user of flexible consultation times, and (4) consistency of treatment delivery that excludes the possibility of therapist drift (Waller, 2009). In several randomized controlled trials with non-stuttering cohorts, internet-delivered CBT (iCBT) for social anxiety disorder has been shown to lead to rapid improvements in anxiety-related symptoms (Berger, Hohl, & Caspar, 2009; Furmark et al., 2009; Tillfors et al., 2008; Titov, Andrews, Schwencke, Drobny, & Einstein, 2008). The extensive follow-up periods in these studies indicate that gains made in iCBT programs may be maintained for periods as long as 5 years. Further, well-controlled
SUPPLEMENTING STUTTERING TREATMENT WITH ONLINE CBT

Noninferiority trials suggest that iCBT and face-to-face CBT may be equally effective (e.g., Hedman et al., 2011).

One of the earliest iCBT programs developed for social anxiety disorder was designed specifically for adults who stutter (see Helgadottir, Menzies, Onslow, Packman, & O’Brien, 2009; 2014b). The program was developed by the present authors and colleagues at the Australian Stuttering Research Centre and was an adaptation of the face-to-face CBT program that had previously been shown to eliminate social anxiety disorder diagnoses in adults who stutter (see Menzies et al., 2008). This internet-based CBT program was named CBTPsych. There was no clinician contact in CBTPsych, but the pre-recorded voices and faces of two clinical psychologists were used throughout. The program is highly interactive (see Helgadottir et al. (2009), for a full description of the program).

The Phase I trial of CBTPsych was conducted with two adults who were stuttering and had social anxiety disorder (Helgadottir et al., 2009). Both experienced clinically significant reductions of social fears, their quality of life improved, and their social anxiety disorder diagnosis was no longer present at post-treatment assessment. A Phase II trial of the program (Helgadottir et al., 2014b) achieved similar results with 14 participants in a nonrandomized design. Seven participants were diagnosed pre-treatment with social anxiety disorder and all, with the exception of two who did not complete all modules, lost that diagnosis at the post-treatment assessment. Significant improvements were reported for the Fear of Negative Evaluation scale (FNE), the Depression Anxiety Stress Scales (DASS), and the Unhelpful Thoughts About Stuttering scales (UTBAS). In general, the results replicated the earlier positive findings of Menzies et al. (2008) with therapist-driven CBT. However, given the uncontrolled nature of the study, only tentative claims could be made for the efficacy of CBTPsych. In addition, these Phase I and II trials involved pre- and post-treatment assessment at a speech clinic. This contact may have increased compliance with,
SUPPLEMENTING STUTTERING TREATMENT WITH ONLINE CBT

and commitment to, the program, hence increasing adherence and raising questions about the external validity of these trials.

Given this, Menzies, O’Brian, Lowe, Packman, and Onslow (2016) conducted a large international trial of this CBT program with no contact of any kind from researchers or clinicians. Two hundred and sixty-seven stuttering participants, recruited through speech services in 23 countries, were given a maximum of 5 months access to CBTPsych. Forty-nine participants (18%) completed all seven modules and all on-line assessments. This compliance rate is much superior to trials of similar on-line program for mental health in the general community (see Fleming et al., 2018 for a systematic review). Completion of the program was associated with large, statistically and clinically significant, reductions for all measures. The reductions were similar to those obtained in earlier trials and those obtained in trials of the face-to-face version of the program with an expert clinician. The mean post-test DASS scores were within the normal community range on all scales. Again, however, the study was uncontrolled and does not allow strong claims to be made about the improvements in psychological functioning experienced by the participants.

1.4 The present study

A CBT program developed by the authors specifically for adults who stutter has been shown to eliminate social anxiety disorder among treatment-seeking adults who stutter, and an internet version of this program (CBTPsych) has been successfully trialed in Phase I and Phase II studies. Since these trials, CBTPsych has been re-named iGlebe in order to align it with the suite of treatments for stuttering already developed by the authors at the Australian Stuttering Research Centre. The present randomized trial used a primary outcome of stuttering severity to compare outcomes for (1) modified speech-restructuring treatment for adults alone with outcomes for (2) iGlebe when added to speech restructuring treatment. The study was approved by the Human Ethics Committee of The University of Sydney.
2. Method

2.1 Research design

The design was a two-arm experimental clinical trial with blinded outcome assessments at 6 and 12 months post-randomization. The control arm received instruction and practice in speech restructuring without any anxiolytic (anxiety reducing) component. The experimental arm received the speech restructuring plus 5 months access to iGlebe, the internet treatment program for adults who stutter. An external, independent, randomization service was used to assign participants to their respective arms.

2.2 Participants

Participants were 32 adults (26 men; 6 women) aged 18 years and over (mean age=35.4 years; range=18-68 years) recruited in order from the stuttering treatment waiting list of a university research clinic. The waiting list draws from the greater Sydney metropolitan area. The mean age of participants in the control arm was 35.8 years and the mean age of participants in the experimental arm was 34.9 years. For the control arm, 10 participants reported previous speech treatment; and in the experimental arm, 12 participants reported previous speech treatment. For the control arm, 12 participants reported a positive family history of stuttering; and in the experimental arm, nine participants reported a positive family history of stuttering. There were 14 men and two women in the control arm; and 12 men and four women in the experimental arm.

Diagnosis of stuttering was by consensus between the assessing speech-language pathologist (SLP) and participant, but there was no minimum stuttering severity criterion for entry into the study. To be eligible, participants needed uninterrupted access to a computer with internet, at least on a weekly basis, and functional English for reading, writing, and speaking. The latter was determined by the assessing SLP. Exclusion criteria were (1) onset of stuttering after 12 years of age or due to a known psychological or neurological disorder;
(2) speech or CBT treatment in the previous 6 months; unwillingness to stabilize dosage of any psychological medication for the duration of the trial; or (3) risk of self-harm as assessed by the Beck Depression Inventory (Beck, 1996). Pre-treatment speech and psychological measures for the participants can be found in Table 1.

2.3 Speech restructuring program

The speech restructuring program was conducted by SLPs, assisted by SLP students, at a university clinic under the direction of qualified SLPs. Each program was implemented over three consecutive days for approximately eight hours per day, with a 1-hour group follow-up session each month for five months. The treatment focused exclusively on learning and practicing a speech restructuring technique within the clinic, for 3 days, without any anxiolytic (anxiety reducing) components. The procedures were based on Stages 1 and 2 of the Camperdown Program (O’Brian, Onslow, Cream, & Packman, 2003). The Camperdown Program is a model of speech-restructuring instruction that does not involve programmed instruction. At the end of the third day, participants were instructed to use their speech technique in their everyday talking. The monthly follow-up sessions focused on practicing their speech technique again within the clinic environment only. Students were told to refrain from discussing anxiety or teaching clients any anxiety-management strategies. For example, there were no opportunities for group speaking which may have provided desensitization, and there was no discussion or problem solving about using the new speech pattern in feared situations.

2.4 Internet CBT program

The iGlebe program (Helgadottir et al., 2009, 2014b) does not require clients to have personal contact with a clinician. However, the program incorporates the faces and voices of a man and woman clinical psychologist who communicate to the users throughout the treatment.
SUPPLEMENTING STUTTERING TREATMENT WITH ONLINE CBT

The program is individualized for each user, designed around on-line pre-treatment assessments, and is highly interactive based on subsequent data that are uploaded regularly by users throughout the program. There are seven sections covering the following: an introduction to the program, challenging negative thoughts about stuttering, the construction by the user of their own social anxiety disorder formulation (selected from situation avoidance, safety behaviors, mental imagery, and physical symptoms of anxiety), and behavioral experiments based on the user’s uploaded responses. The strength of the program is that it can produce thousands of specifically designed interventions involving cognitive challenges, behavioral experiments, and homework exercises that are specific to the individual needs of each user, based on the user’s individual data acquired at assessment. Treatment progression and the selection of homework procedures depend on the individual’s presentation and rate of improvement in response to prior homework. Users receive emails congratulating them for completion of each section and reminding them to log on if they have failed to do so for predetermined periods.

2.5 Procedure

All participants visited the clinic for assessment by a SLP before beginning treatment. At this assessment stuttering was confirmed, eligibility was determined, and each participant completed a computerized self-administered interview designed to identify mental health disorders (see description below). Participants were informed of the telephone recording procedure to assess percentage of syllables stuttered (see description below). Participants were also given three speech-related questionnaires (self-report severity, avoidance of speaking situations, Overall Assessment of the Speaker’s Experience of Stuttering) and three psychological assessments (Unhelpful Thoughts and Beliefs About Stuttering, Social Phobia Anxiety Inventory, Brief Fear of Negative Evaluation). These six forms were combined into a booklet that participants were instructed to complete at home at their leisure and return to the
SUPPLEMENTING STUTTERING TREATMENT WITH ONLINE CBT

clinic on the first day of the speech restructuring program. Details of these assessments are provided below.

All participants then attended the 3-day intensive speech training program. At the end of the third day, they were randomized by an independent, remotely-located person to either the experimental arm or the control arm. The experimental arm participants were given access to iGlebe for 5 months. They worked through this on their own while they continued with their monthly follow-up speech sessions. The control arm participants continued with their five monthly follow-up appointments without access to iGlebe or assistance from the students or clinicians with any anxiety-related strategies. All students and SLPs remained blind to the randomization status of participants and were instructed not to mention the iGlebe program. Participants were also told not to disclose their randomization status to students, SLPs, or other participants. All participants were assessed at 6 months and 12 months post-randomization. For these assessments, the booklets were mailed to participants with reply paid envelopes for return. After the 12-month assessment, those in the control group were given access to the iGlebe program if they wished to access it. A flowchart of the protocol is shown in Figure 1.

INSERT FIGURE 1 ABOUT HERE

2.6 Primary outcome

2.6.1 Percentage syllables stuttered (%SS)

Because the purpose of the experimental trial was to determine whether iGlebe enhances the benefits of behavioral stuttering treatment, this standard measure of behavioral stuttering treatment outcome was chosen as a primary outcome. For each participant, %SS was calculated from two 10-minute audio recordings of the participant conversing with a
SUPPLEMENTING STUTTERING TREATMENT WITH ONLINE CBT

stranger by telephone. These telephone calls were clinic-initiated and unscheduled; hence, participants could neither predict nor prepare for the calls. The strangers were research assistants who were not involved with the experimental procedures and who had no previous contact with the participants. After all recordings had been collected they were de-identified and presented in random order to a SLP independent of the trial. This person counted unambiguous stutters and syllables in real time to determine %SS using a two-button counting device. Unambiguous stutters were defined as what the listener believed would be perceived as stutters by the majority of SLPs and did not include normal disfluencies. The mean of the two recordings was used in all analyses.

To assess intrajudge agreement, 10% of all samples were presented to the same SLP at least 2 weeks later for reanalysis. These samples were chosen from different participants and across different assessment occasions. Intrajudge correlation was \( r = 0.99 \) (mean difference 0.4 %SS). The same samples were also presented to a second judge independent of the present trial for analysis. Interjudge correlation was \( r = 0.86 \) (mean difference 1.6 %SS).

2.6 Secondary outcomes

2.6.1 Self-reported stuttering severity

At each assessment, participants documented a typical stuttering severity score and a worst stuttering severity score for each of eight standard speaking situations: (1) talking with a family member; (2) talking with a familiar person, not a family member; (3) talking in a group of people; (4) talking with a stranger; (5) talking with an authority figure; (6) talking on the phone; (7) ordering food or drink; and (8) giving name and address. Scores were made on a 9-point scale where 1 = no stuttering and 9 = extremely severe stuttering (see O’Brian, Onslow, Cream, & Packman, 2003). The mean score for all eight situations for each participant was used in the analyses.
2.6.2 Avoidance of speaking situations

For each of the above eight standard speaking situations, participants were also asked to report how often they avoided each situation or avoided speaking in each situation. Allowable responses were: never (scored as 1), sometimes (scored as 2) or usually (scored as 3). The cumulative score for all eight situations for each participant was used in the analyses, resulting in a maximum possible score of 24 and a minimum possible score of eight for any participant.

2.6.3 Overall Assessment of the Speaker’s Experience of Stuttering (OASES)

All participants completed the OASES (Yaruss & Quesal, 2010) at each assessment. The OASES is a self-report questionnaire that measures the impact of stuttering on a person’s life. It has four sections each of which produces an impact score, which can be added to produce an overall impact score. Overall impact scores range from 1 to 5 and can be used to determine impact ratings as follows: mild, mild-to-moderate, moderate, moderate-to-severe, and severe. In this analysis only the overall score was used.

2.6.4 Composite International Diagnostic Interview (CIDI)

The CIDI-Auto-2.1 (World Health Organisation, 1997) is a standardized, computerized, self-administered interview designed to assess mental health disorders according to the criteria of the Diagnostic and Statistical Manual of Mental Disorders, Fourth Edition (DSM-IV) (American Psychiatric Association, 2000) and the International Classification of Disorders, Tenth Revision (ICD-10) (World Health Organisation, 1993). At each assessment, the CIDI was completed alone in a clinic room at the centre. On completion, the CIDI generates a score for the total number and type of mental health disorders for each participant. For this study, the number of social phobia diagnoses during the previous month were used.
2.6.5 Unhelpful Thoughts and Beliefs About Stuttering (UTBAS)

The UTBAS is a self-report measure that assesses unhelpful cognitions about stuttering that are related to social anxiety (Iverach et al., 2011; St Clare et al., 2009). It consists of three scales each containing 66 items dealing with a thought. Participants are required to respond to each item according to how often they have this thought (Scale I), how much they believe the thought (Scale II) and how anxious the thought makes them feel (Scale III) on a scale of 1-5. The minimum total score is 168 and the maximum total score is 330.

2.6.6 Social Phobia Anxiety Inventory (SPAI)

The SPAI (Turner, Beidel, & Dancu, 1986) is a comprehensive and widely used measure of social anxiety. The SPAI yields a total score and a difference score, both of which control for the presence of agoraphobia symptoms. The difference score was used in the analyses for this study as it is generally considered a more valid measure of social anxiety. Difference scores above 80 indicate probable social anxiety disorder (social phobia), scores between 60-70 indicate possible social anxiety disorder, scores between 34-59 indicate possible mild social anxiety disorder, and scores less than 34 indicate that a diagnosis of social anxiety disorder is unlikely.

2.7.7 Brief Fear of Negative Evaluation scale (BFNE)

The BFNE (Leary, 1983) is a self-report public domain test that assesses concern about negative evaluation by others. It provides a reasonable indication of the severity of the cognitive and behavioral symptoms of social anxiety. It contains 12 items that are rated on a 1-5 scale (1 = not at all characteristic of me; 5 = extremely characteristic of me). Examples of items include: “I worry about what kind of impression I make on people” and “I often worry that I will say or do wrong things”. The minimum total score is 12 and the maximum total score is 60.
SUPPLEMENTING STUTTERING TREATMENT WITH ONLINE CBT

2.8 Statistical analyses

Statistical analyses were conducted using SAS version 9.4 for Windows (SAS Institute Inc., Cary, NC, USA). Between group comparisons were made using analysis of covariance (ANCOVA), which was conducted at 6 months and 12 months. ANCOVA in this case means linear regression of each follow up outcome variable (dependent variable) with adjustment for baseline outcome variable (independent variable). In all analyses, missing data were ignored. However, in a sensitivity analysis we assessed the potential impact of ignoring missing data using multiple imputation (Sterne et al., 2009) to impute missing variables prior to conducting ANCOVA. Multiple imputation replaces missing data with substituted values. This was done to investigate the sensitivity of our main findings to assumptions about the missing data at 12 months. We did not adjust p-values for multiple comparisons hence statistically significant results should be interpreted with caution.

3. Results

3.1 Compliance

The iGlebe program tracks participant usage through the program as they progress. Two participants completed the entire program including the within-program questionnaires; three others completed the program but did not complete the questionnaires. In total just under a third of the participants completed the program. This is a superior compliance rate to the 18% reported for the program by Menzies et al. (2016).

3.2 Descriptive statistics

Figure 2 shows the group mean scores for each of the speech-related and psychological variables for each group at each assessment. The vertical axes indicate the scores for each assessment procedure. The graphs show trends over time suggesting the control and experimental groups had similar improvements at 6 months on speech-related and
SUPPLEMENTING STUTTERING TREATMENT WITH ONLINE CBT

psychological outcomes; however, at 12 months the experimental group continued to
improve while the control group generally worsened. An exception to this trend was %SS
which showed similar worsening. Table 1 shows the means, standard deviations, and
participant numbers at each assessment shown in Figure 1.

3.3 Analysis of covariance

Table 2 shows the result of the ANCOVAs (linear regression analysis adjusting for
baseline values) at 6 months and 12 months. The ANCOVAs tested for differences between
the two groups for each of the variables at 6 and 12 months. At 6 months there were no
significant differences between the two groups for any of the outcome variables. However, at
12 months the experimental group scored significantly better than the control group for the
speech measures typical ($p=.04$) and worst ($p=.02$) self-reported severity. At 12 months the
experimental group also scored significantly better for the psychological measures OASES
($p<.01$) and FNE ($p=.05$). Differences between groups for the UTBAS neared significance
($p=.07$) at 12 months. A limitation of these analyses is the missing data for most measures
that were apparent for nine participants at 12 months (five in the control group and four in the
experimental group). There were insufficient participants with a CIDI diagnosis of social
anxiety disorder for statistical analysis.
3.4 Sensitivity analysis

The sensitivity analysis was conducted for all outcomes at 12 months using multiple imputation to impute missing values prior to conducting ANCOVA. This analysis indicates the stability of findings when missing data are taken into account by imputation. For the imputation procedure, a multivariate normal distribution was assumed for the speech and psychological variables, and 20 imputed datasets were generated. The results suggest that the self-report severity scores, %SS, and OASES outcomes are similar whether or not imputation of missing data is conducted prior to ANCOVA. However, the psychological variable outcomes (FNE and UTBAS) are not similar. The $p$-values for these outcomes increased to $p=.34$ and $p=.13$, respectively, due to the increased variation in the estimate of effect size. The results of the Sensitivity Analyses are shown in Table 3.

4. Discussion

Adults who stutter have an increased risk of having anxiety disorders, particularly social anxiety disorder, and this has been detected as early as age 7–12 years (Iverach et al., 2016). Consequently, CBT treatment is essential for effective management of adults who stutter and clinical trials to date have shown this to be feasible. In particular, trials to date of a standalone internet CBT treatment developed specifically for adults who stutter—CBTPsych (known as iGlebe in the present study)—showed results equivalent to face-to-face treatment with a clinical psychologist.

The aim of the present experimental clinical study, then, was to determine the effects of adding 5 months access to iGlebe to speech treatment involving restructuring skills. Little can be concluded from the CIDI scores because the number of participants with diagnosis of
SUPPLEMENTING STUTTERING TREATMENT WITH ONLINE CBT

social anxiety disorder (social phobia) was small; three participants in the control group and two in the experimental group. However, with sensitivity analyses taking account of missing data, the experimental group (iGlebe) at 12 months showed significant improvement in self-reported stuttering severity scores and in quality of life, as measured by OASES, compared to the control group. The improved self-reported stuttering severity scores were not reflected with improvement of %SS scores at 12 months post-randomization. This result could be due to self-reported severity ratings averaged over eight speaking situations being a more valid measure of stuttering severity than %SS measured during 10-minute phone calls. Alternatively, it must be acknowledged that improvements in self-reported severity may be a reflection of participants feeling better about themselves after CBT, rather than due to explicit changes in speech.

These results of the benefits of the iGlebe program for adults who stutter need to be interpreted cautiously for a number of reasons. The reductions in stuttering from pre-randomization to 12 months post-randomization in this study were modest, compared to results obtained in standard pragmatic trials of speech restructuring treatment. This result is not surprising as the speech restructuring training in this experiment was not a complete speech treatment. For the purpose of identifying the benefits of isolating the effects of iGlebe, the conventional speech treatment was stripped of all typical anxiolytic components and problem-solving guidance that are usually included. Such components typically include discussion of anxiety during treatment; opportunities for anxiety desensitization such as group speaking during speech training; as well as discussion about, and hierarchical planning for, everyday speaking situations.

It is also a caveat to these findings that improvements at 12-months randomization were constrained to speech related measures of stuttering severity and OASES quality of life, and did not include improvements for psychometric measures of BFNE, SPAI, and UTBAS,
SUPPLEMENTING STUTTERING TREATMENT WITH ONLINE CBT

which involve fear of negative evaluation, social phobia, and unhelpful thoughts and beliefs about the disorder. However, there are two plausible explanations for effects not being detected with psychometric measures. First, compared to a previous trial of this internet CBT treatment (Helgadottir et al., 2014b), the present participants were less anxious at pre-treatment. In the former trial, around half were diagnosed with social anxiety disorder but less than a quarter of the present participants received that diagnosis. Second, the inevitable drop-outs from internet-based treatments likely reduced the statistical power to detect any psychological effects of iGlebe.

In short, the present results show the potential benefits of iGlebe as an add-on to conventional speech treatments for adults who stutter to the extent that they are likely to enhance their effects. Interestingly, the findings suggest that a diagnosis of social anxiety disorder is not a pre-requisite for benefitting from CBT, especially a CBT program designed specifically for those who stutter. An important advantage of internet-based programs such as iGlebe is that there is no therapist drift (Waller, 2009). Therapist drift is the tendency for practitioners to stray from evidence-based treatment.

Treatment of adults who stutter lies within the domain of SLP practitioners, yet the treatment of anxiety is typically within the professional domain of clinical psychologists and psychiatrists. Speech-language pathology professional preparation programs around the world vary in the extent to which they incorporate anxiety management training and, despite SLPs having an understanding of CBT (see Menzies, Onslow, Packman, & O’Brian, 2009), a SLP qualification alone is not sufficient for the formal management of anxiety disorders. The present study, however, provides a potential solution to this problem. iGlebe was developed by clinical psychologists with extensive experience in CBT, leading a team of SLPs experienced clinically with stuttering. The findings suggest that when iGlebe is publicly available, it can be accessed as an add-on to speech treatment alone.
SUPPLEMENTING STUTTERING TREATMENT WITH ONLINE CBT

For the longer term, the present results lend support to the prospect of developing a suite of internet-based treatment programs for adults, and possibly adolescents, who are seeking help to manage their stuttering. This prospect seems realistic with the current development of standalone internet-based speech restructuring treatment (Erickson et al., 2012; Erickson et al., 2016). Ultimately, it may be possible to attain an economical, scalable, and translatable comprehensive stuttering treatment model to supplement standard speech-language pathology treatment practices for the disorder. The future development of this line of research could incorporate the benefits of a full, pragmatic, speech restructuring treatment in combination with access to iGlebe. It is not anticipated that all people who stutter will benefit from internet-based treatments, and research will be needed to establish predictors of success.

Acknowledgements:

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SUPPLEMENTING STUTTERING TREATMENT WITH ONLINE CBT

References


SUPPLEMENTING STUTTERING TREATMENT WITH ONLINE CBT


SUPPLEMENTING STUTTERING TREATMENT WITH ONLINE CBT


SUPPLEMENTING STUTTERING TREATMENT WITH ONLINE CBT


SUPPLEMENTING STUTTERING TREATMENT WITH ONLINE CBT


SUPPLEMENTING STUTTERING TREATMENT WITH ONLINE CBT


SUPPLEMENTING STUTTERING TREATMENT WITH ONLINE CBT

Design of the trial (see text for details)

- Recruitment
  - Assessment
  - Speech Restructuring Practice
    - Randomization
      - Control
      - iGlebe
  - 6 Months Post-randomization
  - 12 Months Post-randomization
Figure 2
Graphs showing outcomes at each assessment. %SS=percent syllables stuttered, OASES= Overall Assessment of the Speaker’s Experience of Stuttering, CIDI=Composite International Diagnostic Interview (social phobia diagnoses), BFNE=Brief Fear of Negative Evaluation scale, SPAI=Social Phobia Anxiety Inventory, UTBAS=Unhelpful Thoughts and Beliefs About Stuttering. The vertical axes show the scoring for each assessment (see sections 2.7.1 and 2.7.2 in the text for explanations of these).
Table 1

Means, standard deviations, and participant numbers at each assessment. %SS=percent syllables stuttered, OASES= Overall Assessment of the Speaker’s Experience of Stuttering, BFNE=Brief Fear of Negative Evaluation scale, SPAI=Social Phobia Anxiety Inventory, UTBAS=Unhelpful Thoughts and Beliefs About Stuttering.

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Analysis of Covariance (ANCOVA) for differences between groups at 6 month and 12 months for the 3 speech-related variables and the 5 psychological variables. %SS = percentage syllables stuttered, OASES = Overall Assessment of the Speaker’s Experience of Stuttering, UTBAS = Unhelpful Thoughts and Beliefs About Stuttering, SPAI = Social Phobia Anxiety Inventory, FNE = Fear of Negative Evaluation.

<table>
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Supplementing Stuttering Treatment with Online CBT

Table 3

Sensitivity Analyses. \%SS = percentage syllables stuttered, OASES = Overall Assessment of the Speaker’s Experience of Stuttering, UTBAS = Unhelpful Thoughts and Beliefs About Stuttering, SPAI = Social Phobia Anxiety Inventory, FNE = Fear of Negative Evaluation.

<table>
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<th>p-value</th>
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CRediT Author Statement

ROSS MENZIES
Conceputalization
Methodology
Software
Validation
Investigation
Writing–review and editing
Supervision
Funding acquisition

SUE O’BRIAN
Conceputalization
Methodology
Validation
Investigation
Writing–review and editing
Funding acquisition

ANN PACKMAN
Conceputalization
Methodology
Validation
Writing–review and editing
Funding acquisition

MARK JONES
Conceputalization
Methodology
Formal analysis
Resources
Data curation
Writing–review and editing
Funding acquisition
Fjóla Dögg Helgadóttir
Software
Writing–review and editing

MARK ONSLOW
Conceptualization
Methodology
Validation
Writing–original draft
Visualisation
Project administration
Funding acquisition