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Re: manuscript submitted to NeuroRehabilitation

Dear Dr. Nejati,

I am pleased to inform you that your manuscript, *Attention Training in rehabilitation of children with developmental stuttering* has been accepted for publication in NeuroRehabilitation. It is tentatively scheduled to be published in March 2013 issue of NeuroRehabilitation. Free to contact me if you have questions or require additional information.

Sincerely,

Melissa Oliver
Managing Editor, NeuroRehabilitation

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Attention Training in rehabilitation of children with developmental stuttering

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Attention Training in rehabilitation of children with developmental stuttering

Abstract:

Attention and executive function play an important role in fluent speaking. The aim of the present study is to evaluate effect of attention training in rehabilitation of stuttering.

In this random clinical trial 30 children with developmental stuttering participate in a random allocation sampling in case or control group. In case group, we trained patient with NEurocognitive Joyful Attention Training Implement (NEJATI) in 12 sessions. Riley Stuttering severity instrument-3(SSI-3), Wisconsin Card Sorting Test (WCST), and Stroop Color Word Test (SCWT) are used for evaluation of executive function before and after intervention. Dependent T-Test was used for analysis. Comparing case group before and after intervention showed a significant enhancement in executive function and reduced stuttering severity.

We can conclude that neurocognitive joyful attention training implement can improve basic cognitive functions of older adults.

Key Words: NEurocognitive Joyful Attention Training Implement (NEJATI), cognitive rehabilitation, Developmental Stuttering
Introduction:

Stuttering is the most common fluency disorder with defined genetic (Lan et al., 2009; Kang et al., 2010; Dworzynski et al, 2007), physiologic (Sato et al, 2011 ) and psychologic etiology (Coleman, Yaruss, & Quesal, 2004; Murphy, Yaruss, & Quesal, 2007).

Stuttering is an inability to talk fluently under self-imposed or external demands such as time limited respond or complex sentences formulation (Starkweather, 2002).

Fluent speaking requires some component such as motor abilities (speech muscle motor control), linguistic abilities (formulation and planning of speech), socioemotional abilities (speech planning and execution under emotional or communicative stress) (Boshardt, 2006). One missing part of this model is attention in a supervisory role. Attention is requires for making coordination between all component of fluent speaking and allocation of attention resource to them.

Recently several studies state that there are some attentional difficulties in stuttering (Anderson, Pellowski, Conture, & Kelly, 2003; Guitar, 2006; Karrass et al., 2006). Some attention-related problem behaviors in children with stuttering (CWS) are high distractibility and difficulties in shifting attention from one task to another one (Embrecths, Ebben, Franke, & van de Poel, 2000; Karrass et al., 2006; Monfrais-Pfauwadel & Lacombe, 2002; Riley & Riley, 2000; Schwenk, Conture, & Walden, 2007). Similarly dividing attention as an ability of attention resource allocation to several tasks is problematic for children with stutter so that concurrent tasks with speech increase severity of stuttering for them (Bosshardt, 1999).
Bajaj (2007) shows that central executive part of working memory impaired in stuttering. The central executive or supervisory attentional system of working memory is responsible for managing information and regulating attention. Actually attentional problem in CWS reduced working memory performance in them. (Anderson et al., 2006; Bakhtiar et al., 2007; Hakim & Ratner, 2004). Furthermore clinical and epidemiological studies show that stuttering is often co-morbid with Attention Deficit Hyperactivity Disorder (ADHD) (Alm & Risberg, 2007; Biederman et al., 1993). These studies provide evidence for a relationship between speech fluency and attentional performance. In contrast, children with ADHD have some difficulties in expressive and receptive language.

Stuttering can be studied in three levels; a cognitive level (central neural processes), a motor level (observable behavior) and a social (contextual or environmental) level. Subsequently intervention might be based on three levels.

Social level intervention postulates on emotional coping strategies for reducing stress in communicative conditions. Motor level intervention implies pausing, physical alterations of articulators and providing some feedback for correcting motor component of fluent speaking. (Lincoln, Packman, & Onslow, 2006) In cognitive level intervention defines in training cognitive demand of fluent speaking such as attention, working memory and planning.

There are several behavioural approaches for the treatment of stuttering. The most applied behavioural approach are habit reversal or regulated breathing (Miltenberger et al., 1998), time-out punishment (Onslow et al., 1997) or Lidcombe program for parents (Onslow et al., 2001).

These behavioural methods typically have not identified the antecedents which are associated with, or promote dysfluent speech (Reed and Howell, 2000).

The problems of behavioural technique in rehabilitation of stuttering are theoretical base of them. This technique based on topological properties of stuttering so that refers to the type of verbal response emitted by the subject.
It seems that a successful technique in treatment of stuttering should be based on involved cognitive function and brain structure in stuttering.

In proposed neurocognitive technique for rehabilitation of stuttering attention as a central core of all cognitive demand of fluent speech is trained.

In present study, cognitive demand of fluent speech is trained and severity of stuttering evaluate in single and dual task.

Method:
Subjects:
Thirty children participated in study available sampling method. Participants assign randomly to case and control group. Inclusion criteria were starting stuttering before school age. None of the subjects were taking psychoactive medication and they did not report any neurological or psychiatric impairment and history of head injury, learning disorder and seizure.

Procedures:
Riley Stuttering severity instrument-3(SSI-3), Wisconsin Card Sorting Test (WCST) and Stroop Color and Word Interference Test (SCWT) are used for evaluation of executive function before and after intervention.

Riley Stuttering severity instrument-3(SSI-3):
During the assessment, interviews of about 20 min duration were recorded in a quiet room using a recorder. The recording included a reading of a text and a sample of spontaneous speech containing a minimum of 200 syllables. The interview was subsequently used to assess the frequency and duration of stuttering and any associated physical concomitants. These were scored according to the guidelines specified in Riley (1994). Although SSI-3 is a measure of severity rather than a way of differentiating fluent speakers from speakers who stutter, it has been used for contrasting the fluency of speakers who stutter with fluent speakers in
other studies (Arnold, Conture, & Ohde, 2005). In present study SSI-3 is used isolate as a single task and with a tapping task as a dual task.

The WCST is a neuropsychological test assumed to be sensitive to frontal lobe damage, especially dorsolateral prefrontal cortex dysfunction (Demakis, 2003). It was administered and scored according to Heaton's standardized criteria (Heaton et al., 1993). In summary, subjects were given four stimulus cards with symbols differing in color, form, and number in front and were instructed to match 128 response cards with different colors, shapes, and number combinations to one of the stimulus cards according to a specific criterion (color, form, or number). Subjects were not informed of the criterion, but were told after each trial whether the match was correct. The criterion was shifted in order of color, form, and number after 10 consecutive correct selections. This procedure was repeated until six criteria were passed. For our purpose, we considered only the total numbers of perseveration and the number of categories completed. These two variables on the WCST are generally the most sensitive to bilateral DLPFC damage (Heaton et al., 1993). Perseveration involves the subject sorting the cards consecutively in the same way or repeating the previous principle. The number of categories completed is the number of categories sorted with 10 consecutive correct responses.

The Stroop Color-Word Test with Three sets of stimuli are employed: First, a set of color names (e.g., red, green, blue, yellow) is presented on screen and participant should press corresponding colored key on keyboard, time is registered by program that presented stimuli by milliseconds. In the second stage, a set of varying color hues (e.g., red, blue, green, yellow) is presented and each hue is identified by the participant, who is timed. Third, a set of color words is presented wherein each color word is printed incongruently in a contrary color hue (e.g., the word, red, is printed in blue; the word, green, is printed in yellow, etc.) and the color hues are named by the participant (whose task is to inhibit the well-learned, now-ingrained tendency to blurt the printed word), and timed.

Intervention: NEurocognitive Joyful Attention Training Implement (NEJATI)
For intervention, we present four joyful computer based tasks to participants. These tasks were graded and increased in level of difficulty based on response of user. Grading of tasks was based on amount of flanker stimuli, velocity of stimuli presenting, number of goal stimuli and changing task rule. For example in one of these tasks, the user should arrange faces in different categorizes based on three properties; emotional status (sad, angry and neutral), hair color (green, white and black) and skin color (yellow, white and black). Each faces have one property of each categories and participant should act only based on presented rule. In each set of tasks user should inhibit two properties of face and act based on one of them. Cognitive demand of this task is inhibition of unrelated properties of stimuli and selective attention to related one.

In case group after the first evaluation session we trained participants in 10 sessions in one hour three time per week and reassessed them in 12th session. Control group were evaluated in 2 sessions with same time period of case group.

Analysis: Data were analyzed using SPSS18. Paired Sample T- Test is used for evaluation of case group before and after evaluation and for control group in first and second session. Independent Sample T- Test is used for comparing case and control group before and after intervention. For exact evaluation of both case and control group, grade of post test subtract from pretest in all measures and Independent T Test used for comparing this data in case and control group.

**Results**

Demographic data such as age, education, gender and Intelligence Quotient that assessed by Weschler Child Intelligence Scale III, were shown in Table 1 for the subjects. Findings show that all demographic measures are similar in case and control group (P> 0.05) that confirm random group allocation to case and control groups.
Table 1: Demographic Variable and Independent T Test Between Them

<table>
<thead>
<tr>
<th>Variable</th>
<th>Case Group (n=15)</th>
<th>Control Group (n=15)</th>
<th>T- Ratio</th>
<th>P Value</th>
</tr>
</thead>
<tbody>
<tr>
<td>Age (years)</td>
<td>12 ± 1.74</td>
<td>12 ± 1.74</td>
<td>0.00</td>
<td>1.00</td>
</tr>
<tr>
<td>Education (Years)</td>
<td>6.20 ± 1.56</td>
<td>6.20 ± 1.56</td>
<td>0.00</td>
<td>1.00</td>
</tr>
<tr>
<td>Non Verbal IQ</td>
<td>100.74 ± 3.18</td>
<td>99.8 ± 3.82</td>
<td>0.51</td>
<td>0.6</td>
</tr>
<tr>
<td>Gender</td>
<td>2 female/ 13 male</td>
<td>2 female/ 13 male</td>
<td>--</td>
<td>--</td>
</tr>
</tbody>
</table>

Stroop color word test shows significantly lower reaction time after intervention (p<0.01) and case group have lower reaction time than control group in post intervention (p<0.01). Furthermore, error rate was significantly lower after intervention in case group (p<0.01) and case group have lower error in post test than control group (p< 0.01). Case and control group have similar performance in error and reaction time before intervention (P >0.05) Both criteria of stroop test have better performance in control group in before and after intervention difference (p<0.01).

Table 2: Cognitive measure before and after intervention

<table>
<thead>
<tr>
<th>Cognitive Measures</th>
<th>Before Intervention</th>
<th>After Intervention</th>
<th>T- Ratio (P- Value)</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Color Word Stroop Test</strong></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Reaction time</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Case</td>
<td>2.02±0.482</td>
<td>1.32±0.371</td>
<td>5.460 (0.000)**</td>
</tr>
<tr>
<td>Control</td>
<td>1.95±0.518</td>
<td>1.94±0.510</td>
<td>0.337 (0.741)</td>
</tr>
<tr>
<td>T (P-Value)</td>
<td>0.398 (0.694)</td>
<td>-3.833(0.001)**</td>
<td>-5.418 (0.000)**</td>
</tr>
<tr>
<td>Error Rate</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Case</td>
<td>6.07±4.992</td>
<td>0.87±1.125</td>
<td>4.096 (0.001)**</td>
</tr>
<tr>
<td>Control</td>
<td>5.07±4.183</td>
<td>6±3.402</td>
<td>-1.825 (0.089)</td>
</tr>
<tr>
<td>T (P-Value)</td>
<td>0.595 (0.557)</td>
<td>-5.549(0.000)**</td>
<td>-4.482 (0.000)**</td>
</tr>
<tr>
<td><strong>Wisconsin Card Sorting Test</strong></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Completed Cluster</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Case</td>
<td>1.53±0.743</td>
<td>4±0.655</td>
<td>-8.487(0.000)**</td>
</tr>
<tr>
<td>Control</td>
<td>1.8± 0.862</td>
<td>1.6±0.632</td>
<td>1 (0.334)</td>
</tr>
<tr>
<td>T (P-Value)</td>
<td>-0.907 (0.372)</td>
<td>10.212 (0.000)**</td>
<td>7.559 (0.000)**</td>
</tr>
<tr>
<td>Preservation Error</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Case</td>
<td>14.87±4.734</td>
<td>11.93±4.906</td>
<td>5.537 (0.000)**</td>
</tr>
<tr>
<td>Control</td>
<td>14.53±4.240</td>
<td>14.87±4.068</td>
<td>-1.099 (0.290)</td>
</tr>
<tr>
<td>T (P-Value)</td>
<td>0.203(0.840)</td>
<td>-1.783(0.086)</td>
<td>-5.351 (0.000)**</td>
</tr>
<tr>
<td>Non Preservation Error</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Case</td>
<td>39.53±6.739</td>
<td>15.20±5.158</td>
<td>9.237 (0.000)**</td>
</tr>
<tr>
<td>Control</td>
<td>39.07±6.924</td>
<td>40.47±6.937</td>
<td>-1.245 (0.234)</td>
</tr>
<tr>
<td>T (P-Value)</td>
<td>0.187(0.853)</td>
<td>-11.320(0.000)**</td>
<td>-8.984 (0.000)**</td>
</tr>
</tbody>
</table>

Significancy in level 0.01 shown with two stars (**) and in level 0.05 shown with one star (*)
T ratio and P value in rows show Independent T- test between case and control before or after intervention. T ratio and P value in column show Pair T- test between initial and final evaluation
Wisconsin Card Sorting Test shows significantly higher completed cluster after intervention (p<0.01) and case group have higher completed cluster than control group in post test evaluation (p<0.01). WCST shows significantly lower non preservation error in case group than control after intervention (p<0.01) and case group have lower reaction time than control group (p<0.01). Furthermore, preservation error was significantly lower after intervention (p<0.01) in case group and in initial and final evaluation of case group (p< 0.05). Case group significantly have lower error in post test evaluation than control group (p<0.01).

Riley stuttering severity instrument criteria show lower severity after intervention in single task (P< 0.01) and dual task (P< 0.01). Case and control group have similar stuttering severity before intervention (p> 0.05) Nonetheless case group have lower stutter in dual task situation. Difference of stuttering severity before and after intervention was better in case group in both single and dual task (P< 0.01).

Table3: Stuttering severity measure before and after intervention

<table>
<thead>
<tr>
<th>Stuttering Severity</th>
<th>Before Intervention Mean (SD)</th>
<th>After Intervention Mean (SD)</th>
<th>T- Ratio (P-Value)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Riley Stuttering severity instrument-3</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Single task</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Case</td>
<td>17.47±5.543</td>
<td>12.916±5.693</td>
<td>3.806 (0.002)**</td>
</tr>
<tr>
<td>Control</td>
<td>16.39± 7.120</td>
<td>16.44±6.523</td>
<td>-0.146 (0.886)</td>
</tr>
<tr>
<td>T (P-Value)</td>
<td>0.503 (0.619)</td>
<td>-1.915 (0.066)</td>
<td>-3.189 (0.004)**</td>
</tr>
<tr>
<td>Dual task</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Case</td>
<td>18.30±6.447</td>
<td>13.76±6.452</td>
<td>3.576 (0.003)**</td>
</tr>
<tr>
<td>Control</td>
<td>19.69± 7.069</td>
<td>19.76±6.298</td>
<td>0.054 (0.958)</td>
</tr>
<tr>
<td>T (P-Value)</td>
<td>-0.563 (0.578)</td>
<td>-2.567(0.016)*</td>
<td>-2.871 (0.008)**</td>
</tr>
</tbody>
</table>

Significancy in level 0.01 shown with two stars (**) and in level 0.05 shown with one star (*)
T ratio and P value in rows show Independent T- test between case and control before or after intervention. T ratio and P value in column show Pair T- test between initial and final evaluation in case or control group. Underlined T ratio and P value is Independent T- test related to difference of final and initial evaluation in case and control group.

**Conclusion:**
Findings show that executive function in stuttering could be trained with cognitive rehabilitation. Recent models of stuttering postulate that executive attention and other self-regulatory processes may play an important role in fluent speaking (Conture et al., 2006; Hubbard Seery, Watkins, Mangelsdorf, & Shigeto, 2007). Attention has a close relation to language so that patients with specific language disorder have impairment in their ability to inhibit prepotent responses (Bishop & Norbury, 2005) and sustain attention (Finneran et al., 2009).

Present study shows that stuttering reduced with attention training. We can discuss this finding in different levels. Firstly, in motor control level, stuttering defined as a disruption of the motor sequence of the word, as a result of disturbed timing (Alm, 2005). In another word, stuttering is impairment in speech as a motor sequence (Ludlow and Loucks, 2003). Each motor sequence needs attention in learning and execution (Nejati et al., 2008). In motor level, attention caused higher performance in motor sequence of speech. Selective attention has been related to temporal processing (Simon, 2010) and training it could be enhance timing of motor sequence and reduced dysfluency of speech.

Sommer et al. (2002) show cortical disconnection below the laryngeal and tongue in sensorimotor cortex. They conclude that persistent developmental stuttering results from disturbed timing of activation in speech relevant brain areas.

In a cognitive approach, fluent speech requires starting (initiation), sustaining, shifting and stopping (inhibition) part of attention. Fluent speech need proper initiation different part of speech in a defined timing so that insufficient initiation of speech segments may be a core mechanism of stuttering (Alm, 2004). Such premature initiation of speech segments may be a core mechanism of cluttering, resulting in shortened or omitted segments, incomplete articulation, increased speech rate or spurts, and disturbed prosody. Present study confirms this finding in an experimental study so that training inhibitory control reduced stutter severity.

Attention is has a central role for plenty of cognitive function such as working memory. Researchers tend to name this cognitive function as executive function.
Executive functions contain attention as a fundamental function and planning, working memory and inhibitory control in higher level (Gothelf et al. 2007; Kates et al. 2007; McVay & Kane, 2009; McCabe et al, 2010).

Attention (Guion & Pederson, 2007) and working memory (Baddeley, 2003) play an important role in language processing.

Some researcher stated that clinical attention deficits and developmental language impairments are both a result of similar underlying neurodevelopmental deficit, whereas others noted that deficits in one area may contribute to deficits in the other (see Redmond, 2005). Working memory is impaired in child with stuttering (Anderson and Wagovich, 2010)

Training attention in present study as a basic executive function could be trained other executive function such as working memory and reduced stuttering with this manner.

Another component of attention that requires for fluent speaking is inhibitory control so that impairment in inhibitory control caused prolonged pausing on speech segment and halting speech. One cognitive component that trained with NEJATI is inhibitory control. This claim could be confirmed with higher performance of Stroop test after intervention. Wolf and Bell (2004) noted that language is an exact predictor of working memory and inhibitory control task. Eggers et al (2011) with Go No go task as a powerful neuropsychologic indicator of inhibitory control indicated that inhibitory control impaired in stuttering. Our result shows that training inhibitory control in stuttering could be reduced stuttering severity.

Another reason for reducing severity of stuttering with enhancing inhibitory control is empowering of trained children in emotion regulation. Karrass et al (2006) indicate that child with stuttering experiences greater emotional reactivity than child who did not stutter. They noted that child who stutter show inability to inhibitory control of attention and regulate the emotions they experience and this
impairments may contribute to the difficulties these children have establishing reasonably fluent speech and language. 

There are several strategies for regulation of emotional reactivity. One regulatory strategy is the allocation of attention resources to face with a problematic situation. Attention regulation helps to individual to shift their attention resource from emotionally arousing stimuli to a safe stimulus (Izard et al, 2008). Finally, speech production in person who stutter requires greater sustained attentional processing than person who do not stutter. This hypothesis confirmed with stuttering severity in dual task paradigm. (Bosshardt, 2006) Present study show that attention training expands attentional resource span and reduced stuttering specially in dual task conditions.

**Acknowledgment**

The Authors wish to thanks all participants for assistance on the provided data. This work was supported by Shahid Beheshti University.

**References:**


Riley, G. D. (1994). Stuttering severity instrument for children and adults (3rd ed.). Austin, TX: Pro-Ed. attention, along with executive function, are primary factors affecting auditory processing ability.

Sato1,2*, Y., Mori1, K., Koizumi1,3, T., Minagawa-Kawai1,4, Y., Tanaka1,5, A., Ozawa6, E., & et al. (2011). Functional lateralization of speech processing in adults and children who stutter. *Frontier in psychology*, 70(2), 1-10.


