

Proceedings of the 2nd International Conference on Stuttering

Edited by Donatella Tomaiuoli

Erickson

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Conference on Stuttering

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Donatella Tomaiuoli

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Etiology and features of stuttering

USING GENETICS TO UNDERSTAND THE CAUSES OF STUTTERING

Dennis Drayna, Carlos E.F. Domingues and M. Hashim Raza¹

Abstract

Although the causes of stuttering remain poorly understood, numerous twin studies have demonstrated that genetic factors make a significant contribution to this disorder. The identification of the genes that make up this contribution could provide an understanding of the molecules and cells involved and lead to improved diagnosis and treatment. Genetic linkage studies in our laboratory have identified locations on chromosomes 2, 3, 10, 12, 14, 15, and 16 that contain genes associated with persistent developmental stuttering in families. Studies on chromosome 12 have identified mutations in the *GNPTAB* gene and subsequent studies identified mutations in the related *GNPTG* and *NAGPA* genes that cause stuttering in populations worldwide. These three genes encode enzymes that attach a mannose 6-phosphate residue to a diverse group of ~50 enzymes, which acts as a targeting signal that causes the cell to route these enzymes to their proper

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location in the lysosome compartment within the cell. Severe mutations in *GNPTAB* and *GNPTG* have previously been associated with Mucopolipidosis Types II and III, rare fatal disorders characterized by multiple pathologies. The mutations in these genes present in stuttering have a less severe effect and occur at positions different from those found in Mucopolipidosis, which is consistent with the lack of mucopolipidosis symptoms present in stuttering subjects with mutations in these genes. Mutations in the genes identified to date may account for 8-16% of persistent developmental stuttering cases worldwide. While more genes remain to be identified, the genes found to date indicate that deficits in intracellular trafficking are important in stuttering.

Introduction and results

While numerous alterations of speech, neuroanatomy, and neural activity have been associated with stuttering, the disorder remains poorly understood at the level of cells and molecules. A better understanding of the molecular and cellular alterations that underlie stuttering would be a substantial aid to the development of improved diagnosis and therapy for the disorder. A longstanding question is the relative contribution of innate and learned factors to the disorder. For example, it has long been known that stuttering clusters in families, leading to suggestions that shared genetic factors, shared environmental factors, or both contribute to stuttering. Efforts to disentangle genetic and non-genetic contributions to stuttering have been greatly aided by twin studies. There have been nine independent twin studies of stuttering published to date, and these are summarized in Table 1. These studies differed from each other in many ways, including the number of subjects enrolled, the types of stuttering (persistent vs. recovered), the language spoken by the subjects, and the methods used for ascertainment, diagnosis, and statistical analysis. As a result most of these studies are not directly comparable. However all of them found a higher rate of co-occurrence (known as twin concordance) of stuttering in identical (monozygotic) twins compared to fraternal (dizygotic) twins. The heritability estimates for stuttering, defined as the fraction of the trait due to shared genes, were variable across these studies, and ranged from .42 to .85. It is

noteworthy that as twin studies have become larger and more sophisticated, the heritability estimates have risen, and the more recent estimates exceeding 80% represent very high heritability.

TABLE 1
Summary of twin studies of stuttering

Reference	Population	Concordance	Estimated genetic component	
		MZ	DZ	
Godai et al., 1976	Italy	.83	.10	nr
Howie, 1981	Australia	.77	.32	nr
Andrews et al., 1991		.20	na	.71
Felsenfeld et al., 2000		.62	.26	.70
Ooki, 2005	Japan	.52	.12	.80 to .85
Dworzynski et al., 2007*	United Kingdom	.19	.00	.60
van Beijsterveldt et al., 2010**	Netherlands	.53 to .61	.34 to .36	.42
Fagnani et al., 2011	Denmark	.47 to .53	.08 to .10	.81 to .84
Rautakoski et al., 2012	Finland	nr	nr	.82

* Persistent stuttering group; ** phenotype used was «probable stuttering»; (M) male; (F) female; (MZ) monozygotic; (DZ) dizygotic; (nr) not reported; (na) not available.

The involvement of genetic factors in stuttering is now well established, but while twin studies can estimate the overall contribution of genes to a trait, they provide little information about the underlying genetic architecture of that trait. For example, although familial clustering of stuttering in families is well documented (Kidd et al., 1981), efforts to assign a mode of inheritance (dominant, recessive, X-linked) have produced inconsistent results, and it is likely that stuttering can display different modes of inheritance in different families. Nevertheless the substantial heritability estimates

for stuttering have motivated many genetic linkage studies that sought to identify chromosomal locations where causative genes reside. More than 20 chromosomal loci have thus been identified, and these studies are summarized in Table 2. While early studies identified loci with only suggestive statistical support (Shugart et al., 2004), or only under conditional linkage models (Suresh et al., 2006; Wittke-Thompson et al., 2007), later studies produced stronger results.

TABLE 2
Genetic linkage studies of stuttering

Reference	Methodology analysis approach	Chr. region	Linkage Score	P values
Shugart et al., 2004	Nonparametric linkage	18p	4.72	
		18q	5.14	
Riaz et al., 2005	Linkage	12q	4.61	
Suresh et al., 2006	Nonparametric linkage Parametric model HLOD* Linkage	2q	3.16	
		7q	2.99	
		9p	2.3	
		12q	2.66	
		13q	2.8	
		15q	1.95	
		20p	2.18	
		21q	4.5	
Wittke-Thompson et al., 2007	Linkage Nonparametric linkage	3q		0.013
		13q		0.012
		15q		0.02
	Association with stuttering and linkage	9q		0.0067
		3q		0.0047
		13q		0.0055

Reference	Methodology analysis approach	Chr. region	Linkage Score	P values
	Linkage	2q		0.013
		5q		0.015 to 0.0051
Raza et al., 2010	Parametric linkage	3q	4.23	
Raza et al., 2012		16q	4.42	
Raza et al., 2013		2p	3.86	
		3p	3.18	
		3q	3.47	
		14q	3.45	
		15q	4.69	
Domingues et al., 2014		10q	4.28	

* HLOD: Heterogeneity LOD score analysis.

The finding of linkage to stuttering at a particular chromosomal location means that there is strong evidence for a causative gene at that location. Identification of these genes has been a long and difficult process. An early statistically strong linkage for stuttering was identified by Riaz et al. (2005) on chromosome 12 in Pakistani families that carried a high degree of consanguinity. Within this region on chromosome 12 more than 70 genes were evaluated in affected and unaffected family members. A variant that substitutes the amino acid lysine for the normal glutamic acid at position 1200 in the *GNPTAB* gene was found to co-segregate with stuttering in one large Pakistani family, and the same mutation was subsequently found in the affected members of three other Pakistani stuttering families (Kang et al., 2010). Many other different mutations in this gene were subsequently found in other individuals who stutter from North America, South America, Asia, Europe, and Africa (Raza et al., 2015). These data provided strong evidence that mutations in the *GNPTAB* gene underlie the linkage signal at this region on chromosome 12. The product of the *GNPTAB* gene forms part of a well-studied enzyme called Glc-NAc-phosphotransferase. Another gene designated *GNPTG* encodes another component of this enzyme. Because of this functional

connection to *GNPTAB*, we screened the *GNPTG* gene in individuals who stutter. This effort identified a number of different mutations in this gene in these individuals. The Glc-NAc-phosphotransferase enzyme acts in concert with a second enzyme, encoded by the product of the *NAGPA* gene. A survey of this gene also identified many different mutations in stuttering subjects (Kang et al., 2010). A survey of >1000 individuals who stutter from worldwide populations produced an estimate that mutations in these three genes together account for 8-16% of stuttering cases worldwide (Raza et al., 2015).

The products of the *GNPTAB*, *GNPTG*, and *NAGPA* genes act to produce a signal that controls the movement of components to their proper location within the cell. This signal is comprised of a mannose 6-phosphate, which is attached to ~ 50 different enzymes that degrade and recycle cellular components, and reside within a sub-cellular structure known as the lysosome. Our results in stuttering subjects were surprising because mutations in the *GNPTAB* and *GNPTG* genes have long been known to cause mucopolipidosis (ML) Types II and III, which are rare fatal medical genetic disorders. Detailed clinical examinations failed to identify any symptoms of ML II or ML III in our stuttering subjects carrying mutations in these genes. Subsequent studies showed that the mutations in these genes that cause mucopolipidosis are more severe and occur at different places in these genes than those found in stuttering (Raza et al., 2015).

Lysosomes exist in all cells in the body, and how mutations in these genes result in a deficit that affects only speech is unknown. We have introduced human stuttering mutations into the analogous positions in the mouse genes, and these result in mice that are viable, fertile, and have normal growth with no obvious abnormalities. More detailed studies of these mice, including their ultrasonic vocalizations, are underway.

Additional studies in our lab have focused on a large family from Cameroon, West Africa that has more than 100 individuals, more than 40 of whom display persistent developmental stuttering. Linkage analyses have shown that these cases are associated with genes residing at four different locations, on chromosomes 2, 3, 14, and 15, with different genes acting in different branches of the family (Raza et al., 2013). Studies of the region on chromosome 15 have tentatively identified a new gene in which mu-

tations are found in individuals who stutter from Africa, North America, South America, and Asia. Mutations in this gene may account for 2-4% of stuttering cases worldwide.

Conclusion

While the genetics of stuttering has significant complexity, it is possible to identify chromosomal loci at which causative genes reside. It is clear that there are multiple such genes and it is likely that more, perhaps many more, remain to be identified. Three genes have been identified to date, and mutations in these genes account for 8-16% of persistent developmental stuttering cases worldwide. Although much of the genetic contribution to stuttering remains to be identified, the genes identified to date indicate that deficits in intracellular trafficking are important in stuttering. Such deficits are being recognized as a common feature of many neurological disorders, and our results suggest stuttering may best be considered as a neurological disorder.

Acknowledgements

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ON BRAIN PROCESSES AND STUTTERING

Martin Sommer²

Individuals who stutter experience stuttering events as a loss of control of the speech muscles — they know what they want to say, but cannot articulate it in a fluent manner. This loss of control suggests a dysfunction of the speech motor system. In at least some stuttering events, the dysfluency is preceded by a precognition or perception of the impending dysfluency. Such precognitions are reminiscent of premonitory urges reported by people with Tourette's syndrome, wherein many affected individuals perceive an urge to produce a tic before the tic actually happens. We recently adapted a questionnaire for premonitory urges, used in Tourette's syndrome, and applied it to stuttering. Two groups were tested – one group of adults with stuttering and one age matched control group. The two groups could be clearly differentiated by these premonitory feelings preceding speech dysfluencies. The ability to anticipate movement lapses is a typical feature of basal ganglia disorders and presumably related to feedforward control of speech execution.

From the perspective of movement disorders, the repetitions seen in stuttering can be clinically analogous to the freezing of gait in Parkinson's disease patients, who «stutter with their feet». They are stuck in repetitions of the initial movement segment, because they are unable to shift from the initial set into the subsequent movement segments of gait. This clinical analogy made us wonder whether the motor cortex of adults who stutter prepares them for the transition between subsequent articulatory sets, in a fashion equivalent to Parkinson's disease.

We investigated the excitability of the tongue motor cortex during the transition between the initial and the subsequent segments of an utterance. While a group of fluent speaking control subjects showed an increase in excitability, in preparation for the second segment of the utterance, this dynamic modulation of motor cortex excitability was lacking in a group of

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adults who stutter. Not only was this difference significant between groups, but so was the extent to which the lack of motor cortex excitability correlated with general stuttering level. Since patients were fluent in most speaking situations during this study, it appears to be a trait marker of stuttering, rather than a state marker.

In summary, recent data uncover a disease core of sequential movement execution with both basal ganglia involvement and a linguistic component. Having clarified the pivotal involvement of the left hemisphere motor cortex, it remains to be seen whether modulation of excitability of this area or the afferent brain areas may serve to improve the outcome of speech fluency treatment.

TONGUE MOTOR REPRESENTATION IN ADULTS WITH PERSISTENT DEVELOPMENTAL STUTTERING

Pierpaolo Busan³, Giovanni Del Ben⁴, Fabrizio Monti⁵, Simona Bernardini⁶, Giulia Natarelli⁷, Piero Paolo Battaglini⁸

Abstract

The neurophysiology of developmental stuttering is not completely clear, but increasing evidence suggests the existence of differences in the structure and the functioning of brain motor systems in adults with persistent developmental stuttering (PDS).

A group of 11 right-handed males with PDS was evaluated by using transcranial magnetic stimulation (TMS) on their tongue representation in primary motor cortex and data were compared with those obtained in 15 matched normal speakers. Motor representations in both hemispheres were evaluated, recording motor evoked potentials. Variables such as motor thresholds, silent period thresholds and silent period durations were evaluated. Non-motor variables were also taken into account, such as the psychological profile (CBA 2.0) and speech-associated attitudes (BigCAT).

Adults with PDS showed higher motor thresholds in the left hemisphere with respect to the right hemisphere, especially when considering silent period thresholds. Silent period duration resulted significantly increased in PDS when stimulating the left hemisphere. This suggests the presence of a particular intracortical balance in the tongue motor cortex of adults with PDS. More specifically, while the higher silent period threshold in adults

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with stuttering could also speculatively suggest a tendency toward a lower intracortical inhibition, the presence of longer silent period duration, when stimulating the tongue representation of the left hemisphere, points toward a greater inhibition in the left motor cortex of stutterers. From a behavioral and cognitive point of view, principal findings indicated that adults with PDS confirmed a negative attitude toward speech situations in comparison to fluent speakers. This evidence, in line with previous ones in stuttering research, supports the existence of dysfunctional motor circuits in the left hemisphere of adults with PDS. Present suggestions could be useful for a better focusing of rehabilitative purposes.

Introduction

Developmental stuttering (DS) is a disruption of the rhythm of speech: subjects know what they want to say but they could be unable to do it in a fluent manner. DS is a complex and multi-factorial disorder: the stuttering brain shows a series of differences with respect to fluent speakers (Craig-McQuaide et al., 2014). It is not clear if these abnormalities are a prerequisite for the appearance of stuttering, or if they could be the result of a long-term stuttering in adults. However, overt disfluency is not needed to differentiate the stuttering brain from fluent individuals (e.g., Busan et al., 2013) and thus neurophysiologic non-invasive tools such as Transcranial Magnetic Stimulation (TMS) could be suitable to shed further light on the neural characteristics of stuttering. Studies that used TMS in stuttering are quite limited (e.g., Sommer et al., 2003; Alm et al., 2013; Busan et al., 2013) and even less studies directly investigated the characteristics of speech effectors with TMS (e.g., Neef et al., 2011; Neef et al., 2015). Weaker inhibition in the right hemisphere and a bilaterally reduced facilitation have been described in the tongue motor representation of stutterers (Neef et al., 2011). Moreover, a lack of left-hemisphere facilitation during speech, in relation with stuttering frequency, has been also reported (Neef et al., 2015). As a consequence, we aimed to further investigate abnormalities in TMS indexes of motor tongue excitability (speech specific effector) in DS persistent in adulthood (PDS). We investigated if PDS might show abnor-

malities in measures such as silent period, an index of motor excitability and intracortical inhibition. Differences in psychological profiles between PDS and controls were also evaluated using different self-report measures.

Materials and methods

Participants

A group of 11 adults with developmental stuttering (age range 24-47 years, mean 32.8 years) and a group of 15 matched normal, fluent speakers (age range 22-42 years, mean 29.5 years) were evaluated by using single pulse TMS. Participants signed an informed consent before the evaluation and the experimental protocol was approved by the local ethical committee.

TMS settings

TMS was used to obtain motor evoked potentials (MEPs) from the tongue, by stimulating its primary motor cortex representation. TMS was delivered on the left and right hemisphere. Muscular activity was recorded by means of classic surface electrodes. They were placed on the surface of the tongue. Two electrodes were placed about 0.5 cm from the midline of the tip of the tongue, on the right and left side. The remaining electrodes were placed at a distance of about 1.5 cm, toward the posterior part of the tongue. A series of indexes were obtained such as the bilateral motor threshold (MT; lower muscular activation), the bilateral active threshold (AMT; higher muscular activation) and silent period threshold (SPT; higher muscular activation). Finally, also the duration of the silent period (SP; higher muscular activation, stimulating at 130% of SPT) was bilaterally measured. Primary motor cortex tongue representation regards both sides of the tongue, thus measures obtained from the contralateral and ipsilateral side of it were considered. MT and AMT were considered as the stimulation intensity able to evoke a MEP in half of about ten consecutive stimulations. SPT was defined as the stimulation intensity that was able to evoke a silent period in half of about ten consecutive stimulations, while the SP duration

was measured from the appearance of the MEP until the first reappearance of muscular activity.

Behavioral and cognitive measures

Both groups were evaluated also from a cognitive and behavioral point of view by administering the BigCAT (Vanryckeghem and Brutten, 2012), a self-report measure of speech-associated attitude, and the Cognitive Behavioral Assessment 2.0 scale (Sanavio et al., 1997), a wide-ranging battery created for offering a general psychological assessment useful and adequate for the clinical practice.

Statistical analyses

Statistical analyses were performed by using mixed model analyses. MT, AMT and SPT were expressed as percentages of the maximal stimulation intensity of TMS. Silent period duration was expressed in msec. Factors were groups, stimulated hemispheres, and, in case of the silent period duration, also the side of the tongue. When considering post-hoc analyses and analyses related to behavioral and cognitive measurements (BigCAT and CBA 2.0), differences in non-normally distributed data were assessed by non-parametrical tests, while in normally distributed data differences were assessed by Student's t-test. A $p < 0.05$ was considered significant.

Results

Motor thresholds

Findings showed a tendency toward a significant difference for MT (interaction between experimental groups and stimulated hemisphere, $p = 0.079$), indicating the presence of a difference between MT in normal speakers (left MT lower than right MT, $p = 0.03$) that is not evident in adults with PDS. A similar significant difference was evident in AMT

(interaction between experimental groups and stimulated hemisphere, $p = 0.081$). In this case, the PDS group showed a difference between left and right hemisphere AMT ($p = 0.062$), pointing toward a lower excitability in the left hemisphere, perhaps compensated by a higher excitability in the right one. A similar but most convincing finding was evident in SPT (interaction between experimental groups and stimulated hemisphere, $p = 0.027$). The difference was between left and right hemisphere in PDS (higher thresholds in the left hemisphere vs. lower thresholds in the right one, $p = 0.032$).

Silent period durations

A significant effect was evident when measuring the interaction between groups and the stimulated hemisphere ($p = 0.04$). More specifically, PDS showed longer silent period durations when stimulating the representation of the tongue in the left hemisphere with respect to normal speakers ($p = 0.006$).

Main findings are summarized in Table 1.

TABLE 1
Summary of TMS differences between adults with PDS and normal speakers

Groups/TMS indexes	Adults with PDS	Normal speakers
LH Motor Threshold	58.3 ± 11.3	50.8 ± 11.5
RH Motor Threshold	57.8 ± 11.4	55.9 ± 8.1
LH Active Threshold	49.1 ± 10.8	43.6 ± 13.4
RH Active Threshold	44.7 ± 11.9	45.6 ± 7.1
LH Silent Period Threshold	50.2 ± 9.9	44.1 ± 12.9
RH Silent Period Threshold	45.4 ± 11.7	47 ± 7.7
LH Silent Period Duration (both sides of the tongue)	51.3 ± 6.1	43.4 ± 7.9
RH Silent Period Duration (both sides of the tongue)	48.5 ± 9.3	47.0 ± 11.2

Significant differences are indicated in bold, tendency toward significance is indicated in italic. LH = left hemisphere; RH = right hemisphere. Thresholds are expressed in % of maximal stimulator output; silent period durations are expressed in msec.

Behavioral and cognitive measurements

The BigCAT evidenced that stuttering group present a more negative attitude toward speech situations in comparison to fluent speakers ($p < 0.001$). Results on CBA 2.0 suggested that the stuttering group reported significantly higher mean scores than their fluent counterparts on the following scales: Eysenck Personality Questionnaire (EPQ/R-N, $p = 0.03$); Psychophysiological Questionnaire (QPF-R, $p = 0.003$), and Fear Survey Schedule (I.P.-PH, $p = 0.001$). Even if PDS showed significantly higher mean scores than controls, the average scores of stuttering participants on CBA 2.0 scales fell within the normal range of functioning (< 95 percentile).

Discussion

The present findings suggest the existence of a particular cortical/intracortical pattern of excitability in the neural projections of the left primary motor cortex of the tongue in adults with PDS. Specifically, while AMT tends to be higher in the left hemisphere of stutterers with respect to the right one, longer silent periods were also evident when stimulating the left hemisphere of PDS. Finally, also thresholds of left hemisphere silent period resulted higher in PDS, with respect to those of the right hemisphere. Thus, it is evident that left hemisphere motor structures of speech effectors could be less excitable in adults with PDS, also during tasks that do not directly involve speech. Silent period is an index of intracortical inhibition, and the presence of a higher silent period threshold in the left hemisphere of PDS could be also speculatively viewed as a counterpart of the higher inhibition in left hemisphere motor structures of PDS, as suggested by findings related to AMT and silent period durations. In this context, the right hemisphere could speculatively try to compensate a possible deficit in the left hemisphere of stutterers as showed by the absence of MT asymmetry in PDS. From a behavioral and cognitive point of view, the present work confirms that stuttering could be related to the presence of negative speech attitudes.

Theories suggesting that DS could be the result of an incomplete left language lateralization in the brain, resulting in a conflict for the execution of speech motor tasks are very old (e.g., Travis, 1978), but they are supported by imaging studies (e.g., Braun et al., 1997). Previous TMS studies on stuttering showed that motor excitability in the left hemisphere was reduced also in not directly speech-related effectors, suggesting the presence of a widespread motor inhibition in PDS (e.g., Sommer et al., 2003; Busan et al., 2013), as well as an aberrant balance between hemispheres when considering motor excitability (e.g., Alm et al. 2013). With respect to speech-specific effectors, a weaker inhibition in the right hemisphere, and a bilaterally reduced facilitation was evident in the tongue motor representation of DS (Neef et al., 2011). Also a lack of left-hemisphere facilitation (and thus of a motor planning-related asymmetry controlling speech motor plans) during speech, related with stuttering frequency, has been recently reported (Neef et al., 2015).

Conclusion

The present work contributes to the comprehension of DS, supporting the view that disfluency could be only the symptom of more subtle motor problems. Present data will be useful to define the neural targets for helping to disentangle new treatment options, as for example new neuromodulation or neurofeedback protocols.

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List of abbreviations

- Active threshold (AMT)
- Adults with persistent developmental stuttering (PDS)
- Developmental stuttering (DS)
- Transcranial Magnetic Stimulation (TMS)
- Motor evoked potentials (MEPs)
- Motor threshold (MT)
- Silent period threshold (SPT)
- Silent period duration (SP)

Session 2

Comorbidity

A MULTIFACTORIAL NEURODEVELOPMENTAL APPROACH TO STUTTERING: (1) LANGUAGE AND MOTOR FACTORS AND (2) PATHWAYS TO PERSISTENCE AND RECOVERY

Anne Smith⁹

Abstract

Stuttering is a neurodevelopmental disorder which emerges in early childhood when the multiple neural networks involved in language processing and speech production are being formed. We present a theoretical account of stuttering as a neurodevelopmental disorder involving multiple factors, including motor, language, and emotional factors. This account is motivated by recent findings from our laboratory showing that the well-known characteristics of stuttering present in adults, including speech motor instabilities and atypical language processing functions, are also present near stuttering onset in 4 and 5-year-old children.

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Introduction

The major goal of this presentation is to outline a neurodevelopmental, multifactorial theoretical account of developmental stuttering and to describe the many lines of evidence that support this account. Our theoretical account builds on our earlier proposal (Smith, 1990; Smith, 1999) that stuttering is a nonlinear, emergent disorder arising from the complex interactions of multiple underlying factors, including motor, language, and emotional factors. Our current account arises from the proliferation of many new experimental results related to neurodevelopmental disorders in general, and to stuttering in particular. It is now clear that stuttering should be considered within the class of neurodevelopmental disorders, that includes, for example, specific language impairment and dyslexia. A neurodevelopmental disorder is one that arises during childhood due to atypical development of the central nervous system. Stuttering, unlike many other disorders in this class, has received relatively little experimental attention. Yet, many lines of evidence suggest that genetic and epigenetic factors play an important role in the emergence of stuttering in childhood, and many neuroimaging studies indicate that there are widespread, atypical structural and functional aspects of the central nervous system associated with stuttering.

A central tenet of our earlier account (Smith, 1999) remains the same: developmental stuttering is a speech motor disorder, but its emergence depends critically upon factors both within and outside the neural systems mediating speech motor control. Particularly significant for the development of stuttering are linguistic and emotional factors. In this presentation, we make the case for a multifactorial account of stuttering that encourages a clear delineation of the factors that contribute to its onset and persistence. Another key component of our account embraces the essential heterogeneity of this disorder by positing that the underlying factors have dramatically different weights in different individuals who stutter as suggested many years ago by Van Riper (1982).

The Purdue Stuttering Project

The evidence I will present to support the ideas outlined above comes from many different laboratories in many different countries. I will, however, highlight recent work emerging from our laboratory, which houses the Purdue Stuttering Project. This project has been funded by the National Institutes of Health since 1988 and has produced a large range of behavioral and physiological findings related to stuttering in both children and adults. Our initial work focusing on motor, language, and autonomic factors related to stuttering was conducted on adult participants. Over the years we modified all of our experimental techniques, including recording movements of the articulators, electromyography to record orofacial muscle activity, electroencephalography to measure event-related potentials (ERPs) of the brain, and skin conductance and blood pulse volume signals as indices of autonomic nervous system function. We can record and analyze data from all of these signals in children as young as four years.

In the most recent phases of our project, we have introduced a new, longitudinal approach to investigate early stuttering. We recruit children who are stuttering at age 4 to 5 years and we follow them for five years with visits to the laboratories every year. Using this approach, investigators can provide critical insights into the development of chronic stuttering and recovery from stuttering in the school-age years. Tracking the progress of the children from age 4-5 years until age 9-10 years allows us to determine whether the child recovered or persisted, and to relate this later persisting/recovered status to both behavioral and physiological parameters we observed in these children in the preschool years. We have been able to complete a wide variety of both cross-sectional studies of children at various ages and longitudinal studies to track development of specific factors. In addition, we have completed studies in which data from four and five year olds is analyzed in relation to their ultimate stuttering status: recovered, persisting, or never stuttered. To date we have recruited approximately 120 children who stutter (CWS) and 70 typically developing matched controls (CWNS)

into the project. Though we include emotional factors as a significant factor in developmental stuttering, we are in the process of analyzing those data and will not include results related to emotion, temperament, or autonomic processes in this report.

A neurodevelopmental perspective

The structure of the brain changes dramatically with development, and at any given point in time, brain structure is a product of interactions among genetic, environmental, and epigenetic factors (Lenroot and Giedd, 2006). Most of us know what genes are: segments of DNA that are fixed at birth; but the term «epigenesis» is one that has more recently entered our vocabularies. Epigenesis encompasses the timing and intensity of gene expression throughout life, and as such, epigenesis is the process by which genes and the environment interact. The pattern of growth of the CNS, then, is not fixed at birth; rather it arises over development through the interaction of genes, epigenetic processes, and experience. In recent accounts of neurodevelopmental disorders, epigenetic factors have been given greater emphasis, because it is through these factors that experience can help to shape the ultimate structure of the brain, and the ultimate phenotypic outcome — stuttering versus not stuttering.

From this neurodevelopmental perspective, then, we understand that stuttering outcome is not fixed at birth; rather for each infant there is a probability at birth that the child will experience stuttering and ultimately recover or persist. Stuttering emerges through development of the brain in early childhood when the neural circuits that mediate speech motor, language, and emotional processes are being established and modified. The critical role of experientially mediated epigenetic factors in stuttering is clearly demonstrated by the fact that twins with identical genes at birth (monozygotic twins) are not always concordant for stuttering. Studies of monozygotic and dizygotic twin pairs with a stuttering proband produce estimates that concordance for stuttering is approximately 18% for DZ twins and 55% for MZ twins (Felsenfeld et al., 2000; Howie, 1981). This is lower than the concordance rate for specific language impairment in MZ

twins, suggesting that genetic factors fixed at birth play a smaller role in emergence of stuttering compared to SLI. A central question for researchers and clinicians is: what are the environmental factors that play a critical role in determining whether, once stuttering has started in the preschool years, the child will recover or persist? In this presentation, we will consider (1) what are the critical factors for stuttering emergence in the preschool years, and (2) which factors appear to be related to the probability of persistence or recovery.

Motor and language factors

Clinicians and researchers broadly agree upon the fact that stuttering is expressed as a problem in speech motor planning and execution resulting in perceptible disfluencies of varying severity and duration. Disfluencies arise when the CNS fails to generate the appropriate neural command signals to control the timing and amplitude of muscle activity necessary for fluent speech production. From studies of adults who stutter, we know that not only are their speech motor systems behaving atypically during perceptibly disfluent speech, but that instabilities in speech motor dynamics are present during their fluent speech. For example, when adults who stutter learn novel non-words, they show an immature pattern of speech motor learning, suggesting that their speech motor systems never develop stable, reliable central motor programs for speech (Smith et al., 2010).

In experiments from our laboratory, we have asked when these atypical speech motor dynamics develop. Is stuttering, even in the early stages, characterized by immature speech motor developmental patterns, such that children who stutter lag their normally fluent peers? We have completed large-scale cross-sectional investigations of activity in orofacial muscles (Walsh and Smith, 2013) and articulatory kinematics during speech production (Walsh, Mettel and Smith, 2015) in preschool children who are stuttering and their fluent counterparts. In EMG recordings of lip muscle activity from the four perioral quadrants, we observed no atypical characteristics of muscle activation in CWS compared to their fluent peers. Muscle activity was not different for the CWS in its amplitude, nor in terms of

bilateral synchrony of activity. We also observed that muscle activity during disfluencies in CWS was smaller in amplitude compared to the amplitude of the EMG associated with the surrounding fluent speech.

In our investigation of speech articulatory kinematics in large groups of preschool children who stutter and matched controls (Walsh, Mettel and Smith, in press), we did find significant differences in motor dynamics between the children who are stuttering and those who were not. These differences were observed in the perceptibly *fluent* speech of the CWS. We used a measure of the consistency of inter-articulatory coordination and found that only the male CWS showed clear evidence of a lag in speech motor development. Their indices of speech motor coordination showed higher variability in repeated productions. These results provide evidence of sex-linked developmental differences in boys and girls who are stuttering in the preschool years.

Many investigations of adults who stutter have explored motor processes underlying nonspeech oral and limb motor tasks (e.g., Smits-Bandstra, De Nil and Saint-Cyr, 2006), in order to ascertain whether stuttering is a speech specific motor disorder. Speech and other motor behaviors share common features (e.g., Franz, Zelaznik and Smith, 1992), and some areas of the brain are activated for both speech and nonspeech oral and limb motor tasks (e.g., Chang et al., 2009), thus the general hypothesis of these studies has been that motor aspects of stuttering should be observable in other motor behaviors. To explore this hypothesis in preschool children, we used a simple bimanual clapping task and asked children to clap to a target beat. After a pacing phase, the beat went off, and children continued to clap trying to match the target beat. We found no differences between children who stutter and those who do not in their motor timing performance on this clapping task (Hilger et al., in preparation).

It is also widely acknowledged that language factors play an important role in the development and maintenance of stuttering. In adults who stutter, disfluencies are more likely to occur in longer, linguistically complex utterances, and speech motor stability is decreased during the production of such sentences (e.g., Kleinow and Smith, 2000; Ratner and Sih, 1987). The onset of stuttering typically occurs when the child's linguistic abilities are developing very rapidly (e.g., rapid growth of MLU and phonological

abilities). Thus the onset of stuttering occurs in a period in which both linguistic and motoric demands on the speech production system are dramatically increasing. Some preschool children who stutter show lags in measures of language and articulation performance compared to their typically developing peers (e.g., Ntourou, Conture and Lipsey, 2011), while others do not (Watkins, Yairi and Ambrose, 1999). It is well-established, then, that the «language factor» in developmental stuttering is heterogeneous among different children. Stuttering occurs in children with robust language systems, those with normal to sub-clinical low language skills, and those with frank language disability.

In our laboratory we have taken a cognitive neuroscience approach to investigate the role of language processing in stuttering. Event-related brain potentials (ERPs) are sensitive indices of the neural processes mediating different aspects of language, including semantic, syntactic, and phonological processing. We have employed classic paradigms that elicit well-known language-related ERP components (e.g., N400, P600) to ascertain whether language processing differs in normally fluent versus stuttering children and adults. In adults who stutter, even when no speaking is required of them, we have observed atypical ERPs for language stimuli presented in both auditory and visual modes (Cuadrado and Weber-Fox, 2003; Weber-Fox, 2001; Weber-Fox et al., 2008). Such results suggest that in addition to the neural circuits mediating speech motor processes, stuttering is associated with atypical development of language processing networks.

We have also completed identical ERP experiments with school-age CWS and found that their results are qualitatively different from those we observed in earlier studies of adults (Weber-Fox et al., 2003). For example, we have observed that an ERP component elicited in a rhyming task was reduced in school-age CWS compared to peers, while this response was identical for adults who stutter and their controls (Weber-Fox et al., 2008). Such differences in finding for school-age children and adults who stutter highlight the need for studies of the factors underlying stuttering across the developmental continuum.

Additionally, we have examined language processing in preschool CWS and matched controls (Weber-Fox et al., 2013). In this study we used child-directed cartoon videos to investigate the brain's processing of semantic and syntactic violations in spoken language. No overt responses or judgments

were required of the children. CWS had slightly longer latency N400s for processing semantic information and a more right lateralized P600 for processing violations in syntactic phrase structure. Thus, these initial findings suggest that even in the preschool years, children who are stuttering are experiencing atypical growth patterns in neural circuits mediating language processing.

Pathways to stuttering persistence/recovery

We have stated that stuttering is an *emergent* disorder in order to emphasize that the child who begins to stutter goes through his/her own developmental trajectory, which may end with recovery or persistence of a lifelong stuttering problem. Perhaps the most important question facing those of us working in the area of stuttering is how to predict the likelihood that a preschool child who is stuttering will persist. A major goal of the Purdue Stuttering Project is to aid in the development of a test battery that can be administered to preschool children to determine the probability of persistence/recovery from stuttering. Those children likely to persist can then receive early, targeted therapy.

By following preschool children who are stuttering over five years, we can retrospectively classify preschoolers as those who will eventually persist and those who will eventually recover. Using this approach we have examined a range of measures from the first year of recruitment into the project, including standardized test scores, speech motor measures, ERP indices, as well as nonspeech motor timing scores to search for useful predictors of persistence. Our results to date have uncovered variables that appear to have no predictive value for persistence/recovery, and others (e.g., speech motor coordination scores) that preliminary results suggest will have predictive value.

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STUTTERING AND READING: PRELIMINARY STUDY FOR SPEED READING DETECTION

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Francesca Del Gado¹⁴ and Donatella Tomaiuoli¹⁵

To assess a reading disorder in presence of stuttering is a difficult task. Although previous studies have explored reading skills in adults and children who stutter (for a review see Nippold and Schwarz, 1990), there are no studies to the present that have investigated the quality of reading in cases of comorbidity.

In our clinical experience, in children who stutter (CWS), reading speed is often affected by the presence of disfluencies, making the data clinically unreliable for a diagnosis of dyslexia.

Consequently, stuttering could create problems in identifying the conditions in which there is a problem in the decoding process. The majority of studies have investigated the relationship between stuttering and language in pre-school age (e.g. phonological encoding, phonetic segmentation, judgments of rhymes, repetition of non-words), but there are only few ones which have investigated phonologic and linguistic skills of school-age CWS.

Weber-Fox and colleagues (2008) have examined behavioral responses and event-related potentials (ERPs) in a sample of 10 school-age CWS during a visual rhyming task and a non-word repetition task. However, results didn't provide univocal evidence to support a clear relation between phonological working memory and stuttering.

Sasisekaran et al. (2013) in a recent study compared receptive and expressive vocabulary, articulation and short-term memory of 9 CWS aged between 10 and 14 years and 9 fluent peers. The results pointed out that CWS showed more difficulties in a test of phonemic awareness

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and in the articulation task, slower phoneme monitoring and delayed encoding of phonemes (with no significant differences in monitoring percentage errors).

Finally, Sasisekaran and Byrd (2013) found a directly proportional relationship between phonological complexity (e.g. consonant cluster) and task difficulty in a small sample of stutterers (9 children aged from 7 to 13 years). However, the authors didn't provide univocal results about this relationship.

The same skills that come together in phonological processing define the prerequisites for reading (Snowling and Hulme, 2011); for this reason maybe it could be more appropriate to determine whether the reading disorder/difficulty in CWS is due to difficulties in speech motor control or altered auditory feedback, rather than in language issues.

As shown in the picture, the production in oral reading tasks can be affected by phonetic, phonological and orthographic skills and by lexical processes (Fig. 1).

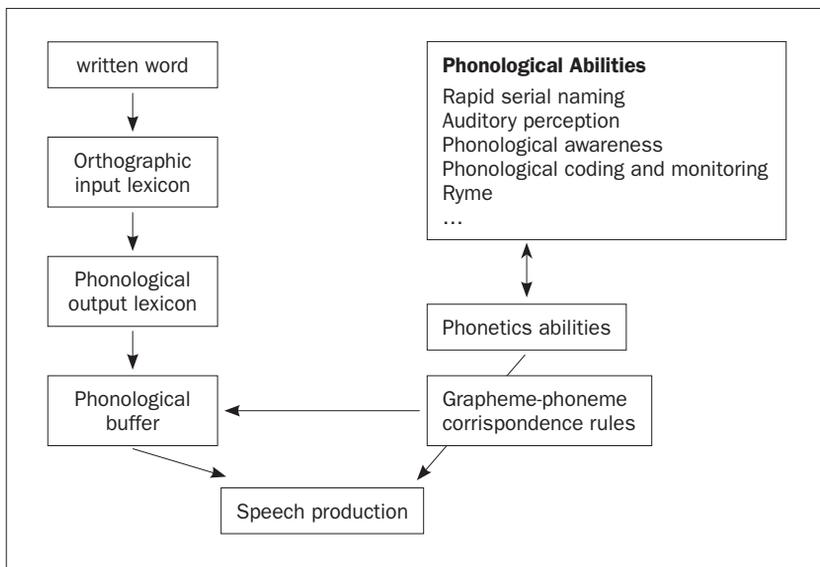


Fig. 1 Speech patterns in reading tasks.

Concerning reading evaluation in stutterers, at present, there aren't enough information about the assessment instruments that could help to define the actual reading speed in individuals who stutter and to identify any risky conditions for a reading disorder.

However, there are documented conditions in literature, defined as facilitating conditions, which can improve verbal fluency.

The mechanisms by which these conditions cause a reduction of disfluencies are not known yet.

In particular, we can assume three possible ways to investigate the reading characteristics in CWS:

1. the masking noise effect (MAF, Masking Auditory Feedback; Antipova et al., 2008), which can be used as a way to assess the actual reading quality in CWS, taking advantage of its characteristic to «wipe» the speech by any possible disfluency. In particular, it was reported that under the effect of masking noise it can be expected a reduction in the percentage of disfluencies produced in adults who stutter. It has been suggested that the mechanism behind this improvement can be attributed to: 1) anxiety reduction related to hear one's own stuttering (Adams and Moore, 1972); 2) «coverage» of primarily altered auditory feedback, on which stuttering would depend (Foundas et al., 2004); 3) increase in the voice's tone, which would be the real facilitating factor (Adams and Hutchinson, 1974). However, it should be also noted that nowadays there are no studies on the masking effect's influence on reading of CWS.
2. whispered reading, another condition which facilitates verbal fluency (Bloodstein, 1950); in this case facilitation is given by the modification of the voice's tone, without interference on the auditory feedback.
3. Finally, another possibility of investigation is represented by silent reading. It was shown that during silent reading tasks, an increase in reading speed can be observed in silent mode compared to reading aloud in both normal adult readers and dyslexic ones. Normal readers' performance improved by about 50% in speed while reading silently, while the increase in people with dyslexia didn't exceed 25% (Ciuffa et al. 2014).

Aims

The purposes of this paper are different: a) to study the influence of different reading modes in dyslexics and stutterers compared to normally fluent peers and normal readers, b) to investigate the qualitative and quantitative modifications of the reading characteristics (speed and accuracy) and disfluencies' characteristics during different tasks and to identify some specific clinical indexes of dyslexia in school-age CWS.

Data and methods

Participants

A sample of children was selected (n. = 32, M age 9.8; Min: 9.2 years; Max: 11.3 years; sex: 25% F, 75% M), among those referred to the «CRC Balbuze» of Rome and to the public Neuropsychiatric Clinic «San Donà al Piave» for a rehabilitation treatment for Stuttering or Dyslexia between 2014 and 2015. A further criteria for the inclusion was the attendance from 4th to 6th grades of primary school, to minimize the influence of a possible insufficient or poor reading automatization.

Clinical samples' performances were compared to those in a control group attending the 4th grade of a public primary school in Rome (n. = 14; Min: 9.5; Max 10.3; sex: 57.14% M, 42.86% F) randomly selected. Inclusion criteria for the control group were the absence of any learning disorder, stuttering and/or other fluency disorders, $IQ \geq 85$, no neurological neither sensorial deficits.

All the children underwent a multidisciplinary assessment of overall development, neuropsychological and learning skills, and a multidimensional assessment of stuttering. It was therefore possible to identify 4 subgroups of children, based on the diagnosis:

- Only Stuttering (Group 1, n. = 17);
- Stuttering plus Dyslexia (Group 2, n. = 8);
- Only Dyslexia (Group 3, n. = 7);
- Controls (Group 4, n. = 14).

Procedures

Stuttering diagnosis and dyslexia diagnosis

Participants were classified as stutterers or dyslexics according to DSM-5 criteria (APA, 2013); in particular, all the participants underwent a multi-dimensional evaluation of fluency and academic skills that lead to a DSM-5-based diagnosis. In particular, each child was assessed with an Italian standardized comprehensive battery of oral reading accuracy and speed/fluency (MT tests, DDE-2), writing (DDE-2, BVN 5-11), reading comprehension (MT tests), arithmetical (AC-MT tests) and cognitive (WISC-III or WISC-IV) tests.

Speech disfluency and reliability measures

Specifically trained professionals conducted the multi-dimensional evaluation to identify the severity of overt aspects. This was done by qualitatively and quantitatively assessing of disfluencies. The speech sample comprised 200 words and was collected during conversation. For each test it has been defined the average score of stuttered syllables (%). Patients were video-recorded to collect speech samples and to capture the qualitative and quantitative stuttering aspects.

Reading procedures/tasks

Each participant also underwent an experimental reading evaluation (250 or more syllables) in four different modes using different reading tests:

- whispered reading (278 syllables);
- reading under MAF condition at 75dB (300 syllables);
- silent reading (248 syllables);
- oral reading (minimum 200 syllables).

An analysis of accuracy, speed and of the disfluencies produced by the four subgroups was carried out during each reading test and compared to the others.

Data analysis

A descriptive and inferential analysis of the data was made; in particular, the data collected were analyzed in SPSS version 20.

Results

ANOVA and post hoc analyses

Using ANOVA approach and through group post hoc comparisons, performances of the four different groups were investigated in relation to the three principal selected variables:

– Reading speed:

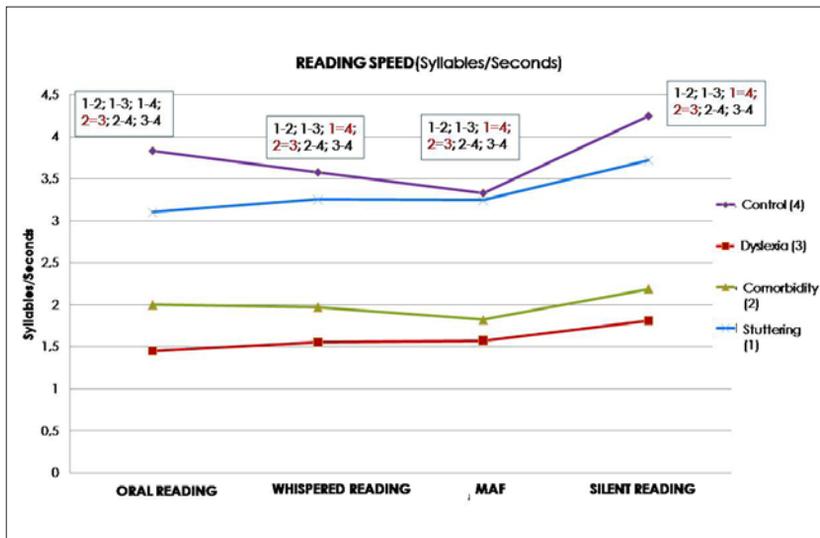


Fig. 3 Reading speed through different reading modes; post-hoc group comparisons were computed following Tukey’s method; not significant post-hoc group comparisons are reported in red.

Results showed that:

- Speed reading rate in CWS was slightly slow on average when compared with controls ($p = .04$), falling within normality parameters; however, reading speed could be improved by experimental reading modes (whispered reading, silent reading, MAF). In fact, during experimental reading tasks, no significant differences were detected between Stuttering group (Group 1) and controls (Group 4).
- No significant differences emerged between Comorbidity Group and Dyslexia Group, independently from reading modes. Comorbidity group showed slower reading performance in every reading mode, thus confirming a reading disorder. According to this preliminary data, in presence of comorbidity between stuttering and dyslexia, reading speed could be considered a reliable parameter to identify slow readers, beyond disfluencies exhibited during the tasks. In conclusion, speed reading appears to be affected significantly and mostly by decoding skills, rather than stuttering.
- Reading accuracy:

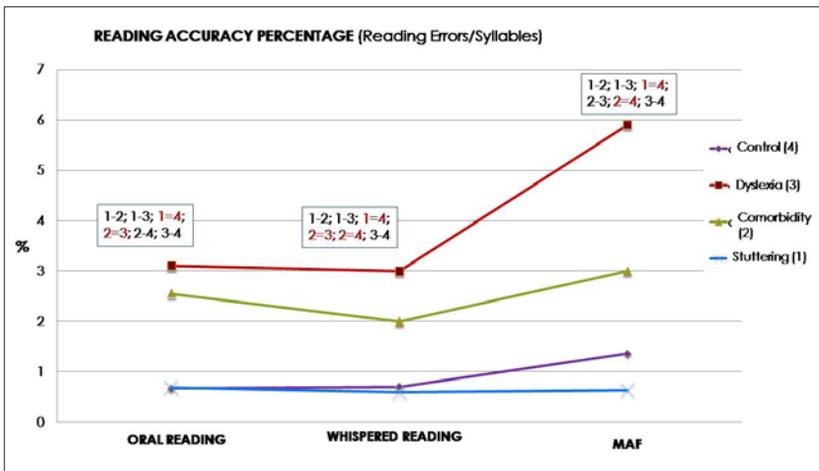


Fig. 4 Reading accuracy through different reading modes; post-hoc group comparisons were computed following Tukey's method; not significant post-hoc group comparisons are reported in red.

Group comparison failed to detect significant differences between Group 1 and Group 4 on accuracy reading in all reading modes. In addition, there was no significant difference on reading accuracy between Group 2 and Group 3 in oral and whispered reading; however, the two groups were significantly different in MAF ($p = < .01$): in Group 2 reading mistakes decreased significantly. This latter result may highlight a new causal mechanism which explains the mistakes detected in oral reading mode in Group 2, which could be caused by stuttering disorder rather than by dyslexia. In other terms, some disfluencies were erroneously confused with reading mistakes.

In conclusion, the simple detection of reading speed and accuracy parameters is not sufficiently alone, especially in case of suspect comorbidity between stuttering and dyslexia. A careful qualitative analysis of mistakes and disfluencies produced during the reading tasks it's mandatory (see Table 1 for clarifications).

TABLE 1
List of possible false reading mistakes, produced in reality by stuttering

Stuttering versus Dyslexia	
Dysfluencies	Reading Errors
Repetitions	Hesitations
Silent blocks	Pauses (> 5")
Interjections	Insertion of syllables/words
Broken words/phrases	Omissions (syllables, words or whole phrases)
False starts/Corrections	Rereading of the same word/line

– Disfluencies' percentage:

A qualitative analysis showed that Group 2, when compared with Group 1, exhibited a worse stuttering symptomatology in every task, thus confirming an higher verbal fluency impairment (see Fig. 5).

An analysis of profiles distribution by gender was not carried out, due to the presence of a small number of females for each group.

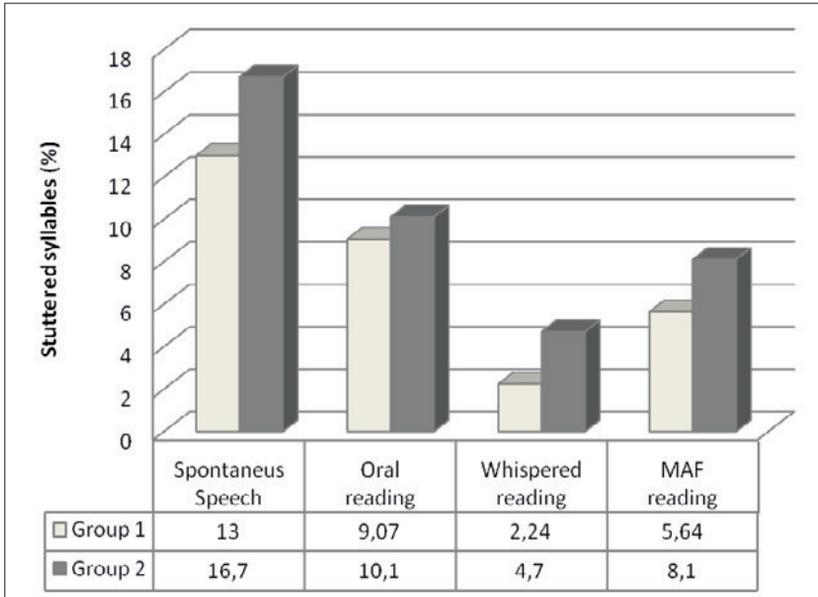


Fig. 5 Modification of dysfluencies (% Stuttered Syllables) during different reading task.

Conclusions

The current study represents a preliminary stage of a larger work that is being conducted, in which the sample has been enlarged.

Notwithstanding this initial research stage, we were able to formulate the following remarks:

- to evaluate reading characteristics (speed and accuracy) in CWS is complex but absolutely possible, even in cases of comorbidity with dyslexia. Facilitating conditions may represent valid and low cost clinical support factors to analyse reading characteristics in CWS; children with comorbidities have more complex clinical situations, both to identify and to treat.

Future directions:

- to increase sample size;
- to define sample inclusion criteria which take into account the heterogeneity of stuttering and the age of the CWS (severity of disfluency symptomatology, relations with overt aspects, especially in case of comorbidity);
- to consider the physiological modifications in both speech and reading skills with respect to the age;
- to distinguish pre- and post- therapy's effects of fluency inducing conditions on spontaneous speech;
- to consider stuttering covert aspects, especially in the comorbidity group;
- to investigate the facilitating conditions effect also in spontaneous speech;
- to define the quantity and quality of disfluencies.

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*Session 3***Assessment****THE CROATIA PROTOCOL FOR CLINICAL TRIALS OF STUTTERING**Mark Onslow¹⁶

There are many treatments for stuttering with many different intended benefits. At present, the different benefits of all those treatments cannot be captured with a single, overarching primary outcome that reflects a common goal for all of them. This makes it currently impossible to directly compare effect sizes of different treatments with different goals in a single, comparative randomised trial. The Croatia Protocol for Clinical Trials of Stuttering proposes a solution to that problem with an overarching measure intended to reflect the benefit of any treatment for stuttering, and which is valid and inexpensive. The present proposal overviews the Croatia Protocol and presents research that establishes its validity and describes its relationship to other primary outcomes currently used in clinical trials.

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EVALUATING STUTTERING FROM THE SPEAKER'S PERSPECTIVEJ. Scott Yaruss¹⁷**Abstract**

Stuttering is a complex disorder that has primarily been defined from the perspective of the listener. Common descriptions enumerate observer-defined moments of «stuttering» behavior, along with associated measures of the frequency, duration, and severity of those observed behaviors. It is clear, however, that speakers and listeners do not always view these moments in the same way. Key aspects of the speaker's experience of stuttering that may be overlooked by a purely observer-based approach include times when the speaker feels that he or she might stutter but does not, due to avoidance or the use of treatment strategies; negative reactions associated with speaking, even when speaking does not occur; missed opportunities for social interaction; and an overall adverse impact on the speaker's quality of life. This paper seeks to refocus the attention of researchers and clinicians on the experiences of those who stutter, both for the purpose of clinical assessment and for the evaluation of treatment outcomes.

Stuttering is typically defined as a speech disorder characterized by certain types of disruptions, or speech disfluencies, in the forward flow of speech (Bloodstein and Bernstein Ratner, 2008). These disfluencies have been categorized in different ways by different authors, and a considerable amount of research has sought to determine what types of speech disfluencies typify stuttered speech as opposed to fluent speech. Often, the delineation of «stuttered» or «stutter-like» disfluencies (Ambrose and Yairi, 1999) includes a list of specific types of speech or nonspeech behaviors, such as parts of words, prolongations, and blocks, as well as other disruptions in speech that are judged by listeners to be in some way atypical.

Such descriptors are certainly important, for it is necessary for clinicians and researchers to be able to describe and document the phenomenon they

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wish to study. Still, there are several other aspects of the stuttering disorder that are also of importance to the speaker, even though they may be harder for listeners to perceive directly (e.g., Yaruss and Quesal, 2004). Examples include the negative emotional reactions that speakers might have their speaking difficulties (e.g., feelings of embarrassment, anxiety, or shame), the thoughts that speakers might have about their communication abilities (e.g., low self-confidence, poor self-esteem), the «tricks» that speakers might use to hide moments of stuttering from others (e.g., changing words, circumlocution; avoiding sounds, words, situations, or people that might evoke stuttering), difficulties in social interaction and participation in life activities due to the reactions of other people (e.g., subtle or direct discrimination by others, negative judgments about the speaker's communication abilities), the overall impact of stuttering on the speaker's quality of life (e.g., reduced satisfaction with education, employment, family, or civic life), and more.

One of the most common ways of describing these «below-the-surface» aspects of the disorder is the «iceberg analogy» (Sheehan, 1970). Many authors have called upon this image and others to highlight the fact that «stuttering is more than just stuttering» (Reeves and Yaruss, 2013) — at least, from the perspective of the speaker. When researchers consider the effects of stuttering on the lives of those who experience the disorder, it becomes increasingly clear that what an observer sees on the surface as involving certain types of disfluencies does not adequately reflect what a speaker experiences under the surface or in life as a whole. Simply counting disfluencies (or utterances or time periods containing disfluencies) cannot tell clinicians or researchers how people who stutter feel about speaking, whether they view themselves as good communicators, whether they participate in social activities, or whether have felt held back in their ability to achieve their goals in life. Of course, such information can be obtained through other means, such as a client interview — a standard part of a comprehensive assessment — or other assessment tools (e.g., Yaruss and Quesal, 2006), but these broader features of the speaker's experience have only partially been incorporated into definitions of stuttering and protocols for assessment.

This discrepancy between the listener's viewpoint and the speaker's viewpoint can be seen in every aspect of the condition, not just those related

to life experience as described above. Disagreements about what stuttering is even can be found in regards to the moment of stuttering itself. For example, Perkins (1983; 1990) suggested that the behavior that is perceived by listeners as a stutter (including repetitions, prolongations, blocks, or other behaviors) may actually be perceived by speakers as resulting from an underlying sensation of «loss of control» of their speech mechanism — something that happens under the surface that is not observable by the listener. Certainly, speakers may also be aware of the speech behaviors, as well as associated (or secondary) characteristics, including eye blinks or physical tension. From perspective of those who stutter, however, the salient feature of the moment of stuttering appears to be the sensation that speakers know what they want to say but, for some reason that they cannot necessarily explain, are temporarily unable to say it. Perkins suggested that this feeling of loss of control is the defining feature of stuttering, though his argument was met with skepticism by several other authors (Bloodstein, 1990; Ingham, 1990; Martin and Haroldson, 1986; Smith, 1990).

Regardless of whether the moment of stuttering actually involves a sensation of loss of control, the fact remains that people who stutter and listeners perceive different events and characteristics during the moment that is often referred to as «stuttering». Sometimes, people who stutter may exhibit stutter-like disfluencies even when they do not experience any underlying sensation of being stuck. This may occur because the speaker is actually experiencing a typical disfluency just like any other speaker, for not all repetitions, prolongations, or blocks constitute stuttering. Or, the speaker might be exhibiting voluntary stuttering or pseudostuttering (Moore and Perkins, 1990), that is, imitating the surface behavior of stuttering — which may be, to a listener, indistinguishable from the surface behavior exhibited during «real» moments of stuttering. Other times, people who stutter may feel a sensation of being stuck (or a «loss of control») even when they do not exhibit any observable behavior for listeners to identify. This is acutely seen in the case of covert stuttering (Murphy, Quesal and Gulker, 2007), in which speakers may go to great lengths to hide their surface stuttering behaviors from others, even though they are feeling that they are, or will be, unable to produce words fluently. Listeners may also be unable to perceive moments when speakers successfully manage stuttering using various strat-

egies taught in therapy, such as preparatory set (Van Riper, 1973), and they may be unable to detect increases in physical tension in certain areas of the body even when speakers are stuttering overtly (Tichenor, Leslie, Shaimain and Yaruss, submitted). Thus, defining the moment of stuttering based solely on what a listener can see or hear leads to an incomplete definition of the phenomenon, one that fails to describe what speakers actually experience and does not sufficiently consider the difficulties faced by those who stutter in producing sounds and words or in communicating with others. IN other words, it reflects a lack of empathy on the part of the judging listeners, if they do not recognize the broader consequences of the moment and the experience of stuttering (Quesal, 2005).

The solution to the problems raised above is not straightforward. Clearly, it is necessary to at least consider what listeners can perceive in the speech signal and in other related behaviors when engaging with individuals who may or may not stutter. Observation and documentation of such behaviors form at least part of the process of evaluating treatment outcomes. And, certainly, many people who stutter would like to exhibit fewer disruptions in their speech that might interfere with their communication with others. Much of speech therapy is based on changing the surface characteristics of speech production, so measuring the success of that therapy necessarily involves measuring observable changes in that behavior.

Defining stuttering based *solely* on the listener's perception is problematic; however, the notion that a speaker's experience must be entirely dependent upon what a listener perceives (Martin and Haroldson, 1981) reflects exactly the lack of empathy that Quesal lamented. Moreover, it opens the door for clinicians and researchers to underestimate the impact that stuttering can have on a speaker's life simply because they fail to see or hear a particular behavior that they, as listeners, have deemed to be problematic. Avoidance, circumlocution, covert stuttering, and other methods of minimizing the outward appears of speech disruptions are all undeniable aspects of the experience of stuttering for some speakers, even though they are difficult and at times impossible to observe. Also undeniable is the fact that speakers can often anticipating a moment of stuttering (Jackson, Yaruss, Quesal, Terranova and Whalen, 2015), and this, too, is not visible to listeners. Denying these experiences based on the apparent absence of

the selected behaviors in the speaker's behavior does little to enhance our understanding of the disorder. On the other hand, defining stuttering *solely* based on the speaker's experience is also problematic — at least until the field has a better understanding of what that sensation (for now, called by some the loss of control) might actually be.

This goal — understanding the underlying disruption when the speaker perceives a loss of control — would therefore appear to be an important step in furthering our understanding of stuttering. The field has made great strides in differentiating which observable behaviors are more likely to occur in some people than in other people, as well as in describing the genetic and neurological differences between people who exhibit those behaviors and people who do not. Increased consideration has been given to the thoughts and emotions of people who stutter, as well as the consequences that people may experience in their lives. The increased visibility of self-help and support groups has also helped to highlight the experience of stuttering, and greater partnerships between clinicians, researchers, and people who stutter have resulted in improved mutual understanding of how stuttering can affect people's lives. Still, there is more work to do, for our science has not progressed as far in determining what speakers are experiencing when they produce those observable behaviors — and what may happen at other times, when speakers report a disruption that remains entirely hidden to a listener.

A better definition of the moment of stuttering, based on an improved understanding of the neurological underpinnings of what has so far been referred to as a loss of control, would help to close this gap. Combining such investigations with ongoing work examining the impact of stuttering (again, from the perspective of people who stutter), would help researchers and clinicians development more holistic assessments and more effective interventions that can help people who stutter overcome the burden of their disorder.

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Session 4

The client and the treatment

EVIDENCE FROM NARRATIVES OF PWS: WHAT ARE THE IMPLICATIONS FOR INTERVENTIONS

Harsha Kathard¹⁸

Narratives as evidence: why clinicians must hear the voices of People Who Stutter (PWS)

Stuttering interventions are intended to make a difference in the lives of People Who Stutter. Therefore, it seems sensible that the voices of PWS must be heard and understood by speech-language therapists as a basis for developing personally-relevant interventions. In this paper, the importance of narrative evidence is explained by contrasting the difference between a case history which is typically used in clinical practice and a life history as a research method. The data generated from life history narrative study (Kathard, 2003) is used to explain core issues of self-identity and stuttering — who am I? The key constructs of self-identity as DisOther, Able and «un-disabled» are discussed illuminating the deeply human complexity of self-identity formations evident in the stories of adults who stutter. Given

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such complexity, the paper considers the implications of these narratives and emerging constructs for clinicians as they enter into the therapeutic relationship. It urges clinicians to approach their interventions with full awareness of how the personal, social and political forces are at play both for the client and clinician. It highlights the tensions and conflicts that emerge when approach to intervention is swayed by the underpinnings of the medical model while the lifeworld of the client calls for a deeper understanding of the stuttering experience through alternative lenses of the social model or capabilities model. The implications for clinical practice which value the depth of stories and subjective experience are explained.

SAFETY BEHAVIOURS AND STUTTERING

Robyn Lowe, Fjola Helgadóttir, Ross Menzies, Susan O'Brian, Ann Packman and Mark Onslow¹⁹

Background

For many, stuttering is associated with debilitating mental health problems. Social anxiety disorder is the most prevalent condition associated with adults who stutter.

Safety behaviours are a core component of social anxiety disorder. Those who are socially anxious use safety behaviours to avoid negative outcomes or events such as social humiliation during social encounters. The person attributes the non-occurrence of a negative event to the safety behaviour. Safety behaviours prevent the unlearning of fear and maintain anxiety.

In a recent report it was found that during routine clinical treatment of stuttering, speech pathologists commonly recommend that patients engage in certain tasks that might, under certain circumstances, be considered safety behaviours.

Aims

The aims of this study were (1) to identify safety behaviours used by adults who stutter and (2) determine whether a relationship exists between safety behaviour use and scores on psychological measures including fear of negative evaluation, avoidance and cognitions.

Methods

Participants were 134 stuttering patients who completed a cognitive behaviour therapy program for anxiety. The participants completed a range

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of psychological measures and identified safety behaviours, from a list of potential behaviours, they typically used in everyday speaking situations.

Results

Results were obtained that were statistically and clinically significant.

Conclusion

Future research is required to investigate safety behaviours, anxiety and stuttering treatment.

CLIENTS' DEMANDS AND CLINICIANS' CAPACITIES: CAN WE MEET?

Suzana Jelcic-Jakšić²⁰

Clinical wisdom, infused into the profession decades ago by Don Baer, is that the complaints of clients fundamentally drive clinical practice. Also of interest to clinicians is empirical information about how stuttering affects people and drives them to clinicians with such complaints. When clients present to clinicians, the cycle of evidence based practice begins with knowledge from clinical trials about effect sizes that can realistically be expected from treatments. This presentation synthesises available knowledge about what those affected with stuttering complain about, what is known about how stuttering affects them, and what is known about effect sizes that can be expected from treatment when they come to clinics. Conclusions are suggested that might inform clinical practice and research.

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Session 5

The social and structural frame

ATTITUDES OF NONSTUTTERING CHILDREN AND YOUTH TOWARD STUTTERING: WHAT WE KNOW AND WHAT WE NEED TO LEARN

Kenneth O. St. Louis and Mary E. Weidner²¹

Abstract

The adult nonstuttering public holds negative and biased attitudes toward stuttering and people who stutter. These negative beliefs and self reactions have been observed in nearly all populations around the world in which they have been studied. The research unambiguously documents that people who stutter experience stigma and discrimination as a result of their stuttering.

The *Public Opinion Survey of Human Attributes-Stuttering (POSHA-S)*, has been used extensively in research among adult populations worldwide. Even though differences in populations exist, overall results show that the similarities among adults in different study populations are greater than differences. Two questions logically arise: «Where do the negative attitudes so widely shared around the world come from?» and «How do the attitudes change from children to adults?».

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Emerging research is suggestive of disparate attitudes among different age groups across the lifespan (i.e., young children, children, adolescents, and adults). Attitudes of nonstuttering children are important because a host of anecdotal accounts and numerous studies have documented that children who stutter are subjected to teasing and bullying by their peers (Langevin, 2015).

Recent research on children's attitudes is summarized for various ages, from preschool through high school. Preliminary results from a new measure, the *POSHA-S/Child* developed by Weidner and St. Louis (2014), are highlighted to augment previous research with preschool and kindergarten samples. Three studies on changeability of stuttering attitudes in middle and high school are also described.

A possible explanation for observed changes in attitudes from young children to young adults is advanced. Some of these changes have been quite surprising and others relatively predictable. A call for additional research with nonstuttering children and youth is advanced, identifying especially promising or important areas of inquiry.

Public attitudes toward stuttering

A very large and growing literature has documented that people who stutter live in social environments that, despite the best intentions of many in society, hold incorrect beliefs about stuttering and behave in ways that are often not supportive or sensitive (St. Louis, 2015b). These include (a) the well-publicized «stuttering stereotype» (Woods and Williams, 1976) or belief that stutters are nervous, shy, introverted, weak, and so on and (b) listeners' behaviors, such as filling in stuttered words, or feelings of concern if stuttering were to occur in people with whom they associate. These negative beliefs and reactions have also been observed in children as documented in a review by Langevin (2015).

IPATHA Initiative

For 16 years, the International Project on Attitudes Toward Human Attributes (IPATHA) has sought to document similarities and differences

in public attitudes toward stuttering. To date, adult respondents from 39 countries have responded to a standard instrument, the *Public Opinion Survey of Human Attributes-Stuttering (POSHA-S)* in 25 languages (St. Louis, 2011). Similarities in attitudes have been found to outweigh differences, although important, geographic, national, cultural, vocational, economic, and other variables do affect attitudes (St. Louis, 2015a). *POSHA-S* components (means of clusters of similar items) contributing to three subscores are listed Table 1. Circa September, 2015, the median sample means for *POSHA-S* Beliefs and Self Reactions subscores were 34 and 1, respectively, based on 10,474 public respondents in 133 different samples. The vast majority of these studies have surveyed adults; yet, a few studies have explored children’s stuttering attitudes. Following is a summary of the results of those investigations.

TABLE 1
Mean ratings for POSHA-S items, components, subscores, and Overall Stuttering Scores (- 100 to + 100) for the mean ratings of samples of children and youth compared to the median sample mean from the POSHA-S database

Variable	Database Median ^a	High School Students (Poland)	High School Students (USA)	Middle School Students (USA)	Elementary Students 1 (Turkey)	Elementary Students 2 (Turkey)
Number	133 samples	111	83	55	89	101
Age (yr)	37.2	17.6	16.3	12.8	12.1	11.8
OVERALL STUTTERING SCORE	17	11	18	18	2	4
Beliefs About Persons Who Stutter	32	25	37	36	7	14
Traits/ Personality	17	26	13	38	-22	-15

Variable	Database Median^a	High School Students (Poland)	High School Students (USA)	Middle School Students (USA)	Elementary Students 1 (Turkey)	Elementary Students 2 (Turkey)
Stuttering Should Be Helped by...	15	3	25	16	0	5
Stuttering is Caused by...	32	19	34	30	-5	0
Potential	64	52	77	59	54	64
Self Reactions to People Who Stutter	2	-4	1	1	-3	-5
Accommodating/ Helping	40	25	64	44	34	25
Social Distance/ Sympathy	6	-7	17	25	-20	-13
Knowledge/ Experience	-34	-28	-53	-27	-35	-42
Knowledge Source	-9	-5	-30	-39	11	9
Obesity/ Mental Illness Subscore	-35	-33	-38	-23	-25	-50
Overall Impression	-15	-8	-14	21	-16	-35
Want/Have	-84	-87	-95	-74	-48	-79
Amount Known about	-5	-4	-6	-16	-12	-35

Variable	POSHA-S/ Child Variable	Kindergarten Students (USA)	Preschool Student (USA)	Preschool Student (Turkey)
Number		26	28	31
Age (yr)		6.2	4.5	4.3
OVERALL STUTTERING SCORE		6	-7	-7
Beliefs About Per- sons Who Stutter		15	8	15
Traits/ Personality		-18	-34	-23
Stuttering Should Be Helped by...		23	25	27
Stuttering is Caused by...		-1	-19	-4
Potential		57	59	58
Self Reactions to People Who Stutter		-4	-22	-28
Accommodating/ Helping		32	3	-1
Social Distance/ Sympathy		8	9	9
Knowledge/ Experience	Experience	-51	-80	-94
Knowledge Source	—	—	—	—
Obesity/Mental Illness Subscore	Obesity/ Wheelchair Subscore	-32	-41	-57
Overall Impression	—	—	—	—
Want/Have	Preference	-22	-18	-26
Amount Known about	Experience	-43	-64	-89

^a Median of 133 separate sample means from the POSHA-S database containing 10,474 respondents (circa September, 2015).

Children's and adolescents' attitudes toward stuttering

High School Students

Flynn and St. Louis (2011) reported that high school students at a high school in an American college town in West Virginia held attitudes that were comparable to those of the adult population. Table 1 shows that their components, subscores, and OSSs were similar to those of the database median values. These results suggest that adult public attitudes have assimilated to adolescents in high school by the age of 16 years. This was not the case in a recent study in Poland (Przepiórka, Węsierska and Błachnio, personal communication 5 September, 2015) wherein high school students' attitudes were less positive than those of university students, as well as university students and the general public in previous studies (e.g., Przepiórka, Błachnio, St. Louis and Wozniak, 2013). The previous samples held attitudes that were generally typical of the *POSHA-S* database.

Middle school students

In a recent unpublished study, Kuhn and St. Louis (2015), sampled middle school students in a school located in a small town in rural West Virginia, characterized by lower-than-average education and income levels. The students, whose mean age was nearly 13 held stuttering attitudes that were similar to those of the database median and of high schoolers, although their obesity and mental illness scores were more positive.

Elementary school students

Samples of 6th grade students in Turkey generated worse than average attitudes compared to the *POSHA-S* database (Özdemir, St. Louis and Topbaş, 2011b). Nevertheless, children's attitudes were remarkably similar to attitudes of their parents, grandparents or other adult relatives, and neighbors. The study involved two probability samples, each from a different

district of the city of 600,000 people, wherein differences in attitudes were extremely small. A companion study comparing the adult neighbors' attitudes to those of a convenience sample with considerably higher education levels (Özdemir, St. Louis and Topbaş, 2011a) showed that the differences observed in the probability and convenience samples were likely due to differences in socio-economic variables.

Kindergarten Students

The *POSHA-S* was designed to sample attitudes of 6th graders and older, literate respondents. It may well provide valid estimates of younger children's attitudes, perhaps as low as 4th grade, but no data from this age group is available. Weidner and St. Louis (2014) developed a young child version of the *POSHA-S* and named it the *POSHA-S/Child*. It followed the items and structure of the older child and adult version as closely as possible, but had some important modifications (Weidner, St. Louis, Burgess and LeMasters, 2015). These include: a short cartoon-based video depicting stuttering, oral, face-to-face administration instead of self-administration through reading, yes-no scoring with «I don't know» being inferred by the examiner, comparison of stuttering with anchors of obesity and wheelchair use because young children would likely not know anything about mental illness, and experience with stuttering and the other attributes being determined by combined ratings of parents and children.

An ongoing but unpublished study compares attitude ratings of adults who responded online to the *POSHA-S* and to the *POSHA-S/Child*. A large order-counterbalanced sample of adults generated belief subscores of 49 and 41, respectively; self reaction subscores of 16 and 20, respectively; and OSSs of 31 and 30, respectively. The *POSHA-S* Obesity/Mental Illness subscore was -27, and the *POSHA-S/Child* Obesity/Wheelchair subscore was -26. These results suggest strongly that the adult and child versions of the *POSHA-S* are numerically comparable.

Using the *POSHA-S/Child*, Weidner, St. Louis et al. (2015) sampled 5-7 year-old kindergarten children (mean = 6 years) from a rural small town in West Virginia, again, with lower-than-average socio-economic indicators.

The children's stuttering attitudes were less positive than average, especially comparing them to the middle school students sampled from another rural area of the same state. For example, the OSS for the kindergarteners was 6 compared to 18 for the middle schoolers (Table 1).

Preschool students

Weidner, St. Louis et al. (2015) also sampled 3-5 year-old preschool students (mean = 4.5 yr). These children were from an above-average socio-economic university town (the same one in the Flynn and St. Louis study). Surprisingly, the preschool students had worse attitudes than the kindergarteners, even considering a potential negative influence of socio-economic factors. Their subscores and OSS were 8-18 points lower than those of the kindergarteners who were about 1.5 years older on average.

The preschool age study was replicated in Turkey, with very similar stuttering attitude results (Weidner, Nakişçi, St. Louis and Özdemir, 2015). By contrast, the Turks' attitudes toward obesity and wheelchair use were less positive than those of the American preschoolers.

Changing children's negative attitudes toward stuttering

The studies by Flynn and St. Louis (2011), Przepiorka et al. (2015), and Kuhn and St. Louis (2015) are the only *POSHA-S* studies that have attempted to change attitudes of nonstuttering youth. High school students subjected to a humorous but frank presentation about stuttering by Flynn, a moderate-to-severe stuturer, showed a large positive change in measured attitudes. Additionally, a professionally produced film about three stutters, including Flynn, also was with substantial improvements in adolescents' stuttering attitudes, but not as large as the oral presentation. When the film was then followed up by a shortened version of the oral presentation, the resultant attitudes were similar to those following the longer oral presentation. By contrast, when high school students in Poland viewed a film about stuttering, their attitudes did not change.

Other high schoolers in the Polish study did show a very small positive change after a workshop on stuttering.

A current study that is underway is exploring the permanence of attitude change effected by Flynn's oral presentation (or in some cases the video as well) in the same individuals who were high school students seven to nine years earlier. Results are not complete, but preliminary analyses suggest that the positive attitude changes on the *POSHA-S* following the high school intervention were maintained through young adulthood.

The recent studies by Kuhn and St. Louis (2015) and by Przepiorka et al. (2015) did not corroborate the earlier finding that youngsters' attitudes are always amenable to change. These American middle school students, who held relatively typical adult attitudes, and Polish high school students, whose attitudes were somewhat more negative than normal, effectively did not change their beliefs and self reactions to stuttering after watching video about stuttering. Moreover, the middle schoolers' attitudes did not change in a follow-up condition one month later. In the case of a subset of 10 pilot study respondents whose follow-up was one year later, they actually held worse attitudes after watching the video (also observed in some Polish high schoolers), but these attitudes returned to their pre-video level one year later.

Weidner is currently conducting a pilot study designed to ameliorate uninformed or negative attitudes of preschool nonstuttering children. The study will involve a series of puppet-based video episodes designed to educate children about «differences», i.e., that «everyone is different and the same», and how to react to children who stutter or have other «differences». Data are as yet unavailable on the effectiveness of this intervention.

Implications

Given the ubiquity of teasing and bullying that occurs with child stutterers, the time has come to accelerate research efforts directed at measuring and improving public attitudes toward stuttering among children. The research reviewed has taken the approach that a standard measure is critical in documenting differences across populations as well as document-

ing changes effected by various educational program or interventions. The *POSHA-S* and *POSHA-S/Child* provide such standard measures.

At this point, there is much to learn about the acquisition, internalization, and manifestations of public attitudes toward stuttering. The limited research suggests that, contrary to expectations, negative attitudes can be acquired very quickly by young 3-5 year-old children with only brief exposure to stuttering. These appear to be directed more toward the stuttering behavior than the person who may stutter, e.g., preschool children believe that a person who stutters would be able to make good choices and make friends but simultaneously be worried if a friend stuttered. This is akin to Derman-Sparks' (1989) concept of «pre-prejudice» and supports related child psychology literature that suggests by the time children reach the age of 6-7 years, some of their negative attitudes toward others with differences have improved (Aboud, 1988). A large gap in the available data occurs between the 1st and 5th grades, but by the 6th grade, it appears that parents' attitudes have essentially determined children's attitudes. This situation appears to be maintained through middle school and high school. Interestingly, the limited data available suggests that middle school and high school students may not be as amenable to efforts to improve their attitudes as university students or adults (Abdulla, 2015; Kuhn and St. Louis, 2015; Przepiorka et al., 2015). While not yet understood, the actual interventions applied very likely have affected the differences. For this and many other puzzling questions, only further research can answer them. Accordingly, we call for additional studies of children and youth, especially preschool to 5th grade children, to explore the development and changeability of their stuttering attitudes.

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INQUIRY ON STUTTERING IN ITALY: CHARACTERISTICS OF THE PUBLIC SERVICE (REGARDING UNIVERSITY EDUCATION AND HEALTH CARE) AND CHARACTERISTICS OF LOGOPEDIC PRACTICE

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Abstract

In Italy, the diagnosis and treatment of stuttering is not included in the «essential levels of care» (Italian: «LEA») guaranteed everywhere by National Health System, although regions are able to devote their own resources to the issue. Possible inadequacies of the Italian welfare, academic, and research institutions could be both the cause and the effect of a very confused legal framework for the professions involved. Consequently, we set out to investigate these inadequacies through the use of three questionnaires. Two questionnaires were submitted online to almost all relevant Italian public services and universities with courses potentially involved addressing stuttering. The third questionnaire focusing on speech therapists' practices and materials was sent to more than 190 logopedists. Results confirm a problematic Italian situation for both the training of future speech therapists on disfluencies and access to public health services for the person who stutters (PWS). In addition, there is a shortage of etiological and clinical research and of evidence-based (EB) treatment programs and clinical materials.

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Introduction

Stuttering is a difficult disorder to understand because of its multidimensional and variable character (not only evident alterations in the verbal flow, but resulting also in secondary underlying symptoms that range in severity). This difficulty creates a fragmented and sometimes contradictory repertoire of knowledge, extending from scientific literature to clinical practice (Florio and Bernardini, 2014).

Some transnational studies have found an increased aversion from students and speech therapists to work on stuttering as opposed to other communication disorders (Yaruss and Quesal, 2002) and a reduction of specific training regarding stuttering (Yaruss, 1999; Yaruss and Quesal, 2002). This trend has led to a growing research interest on the academic and clinical education of speech language therapists (SLT).

A study commissioned by International Association of Logopedics and Phoniatics found that in Eastern Europe, SLT receive basic training in evaluation and treatment of stuttering, usually at the university level. Post-graduate training, however, is uneven in different states and there are generally few therapists who formally specialize in such disorders.

The literature shows that if the State does not provide free treatment, PWS may find it difficult to support the direct costs of treatment and/or to obtain reimbursement from a private insurance company (cfr. Fibiger et al., 2008; Blumgart, Tran and Craig, 2010).

In Italy stuttering is not included within the «essential levels of care» (LEA) guaranteed to all citizens by the National Health Service, but regions have the opportunity to use their own resources to provide additional services, resulting in very diverse regional frames.²⁸ In addition, the normative framework for health professions appointed to take charge of PWS is also very confusing.

The hypothesis that moved the creation of this study is that there are different realities in our country which moreover could not meet the need of people who stutter and speech therapists.

²⁸ *I Lea: livelli essenziali di assistenza*. Ultima consultazione: 30 marzo 2015 in http://www.salute.gov.it/portale/temi/p2_5.jsp?area=programmazioneSanitariaLea&menu=lea.

With either a complete lack of research or limited and outdated Evidence Based Medicine (EBM) studies, this work investigated the following issues in different Italian regions:

- the organization of theoretical and practical education on fluency disorders in university courses of Logopedics (UCL) (three years long);
- the presence and organization of services for diagnosis and treatment of stuttering in public health facilities and
- the tools and methodological approaches used by SLT.

Methods

To address the first two investigation points, two online questionnaires were used (Q1 and Q2, respectively), administered by *CAWI* method and using the *SurveyMonkey* platform. The third questionnaire (Q3) was disseminated via different modalities: manual delivery of a hard copy, e-mail delivery of an editable e-sheet, posting of the e-sheet on the website of *Società scientifica logopedisti italiani*, and through social networks of professional groups.

Q1 was addressed to the Directors of the 36 UCL active in 17 regions during the academic year of 2014/2015.

Q2 was sent to the Directors or Coordinators of 299 public health facilities, both hospital and territorial, in 20 Italian regions who are addressing children and/or adult (Child Psychiatry, Phoniatrics, and Rehabilitation). The inquiry was supported by *Federazione logopedisti italiani* (FLI), which assisted in sensitizing members of the most problematic regions.

Finally, Q3 was sent only to SLT.

Results

The responses to Q1 and Q2 were collected from 24 January 2015 to 14 March 2015 and responses to Q3 from February 2014 to September 2014. Overall, there were 21 respondents to Q1 (58.3%), 75 to Q2 (26%; 18 of 20 regions), and 90 to Q3 (47.4% of the contacts approached directly,

with almost all from northern Italy (92.2%), specifically Veneto, Emilia Romagna, Lombardia and Piemonte).

Q1: *Educational offerings on stuttering by university courses of Logopedics.* Analysis showed the existence of very different realities. In fact, 52.38% UCL offer theoretical education on fluency disorders with a course that includes other disorders (i.e. voice, articulation, swallowing disorders) and in other UCL it is an educational option. These educational courses, in most cases restricted to graduates in Logopedics (76.2%), last an average of 15.45 hours (range 2-60 hours; 12.96 SD). An internship (with an average duration of 24 hours, 5.47 SD) is possible only in a third of the courses, likely due to the lack of health facilities that treat the disorder.

Regarding the Core Curriculum of Logopedics (FLI, 2010), we can see that the levels of knowledge, skills, and abilities required for the four Elementary Teaching Units on the topic of fluency disorders are not achieved by most UCL.

Q2: *Characteristics of the public health services for the diagnosis and treatment of stuttering.*

The diagnosis of stuttering is performed in 63.4% of public health facilities, but treatment in only 42.3% overall, with just 23.1% in Southern Italy.

Almost all of these facilities (about 90%) cover the preschool and school ages, but only 39.4% cover adulthood.

Thirty-eight percent (38%) of respondents reported that people are expected to pay a ticket for both the diagnosis and treatment in preschoolers, but by adulthood, the percentage rises to 58% for the diagnosis and 62% for treatment.

The assessment of stuttering is performed mainly by logopedists (81,1%), child psychiatrists (52.8%), phoniatriests (43.4%), and psychologists (34%). Most of the time treatment is performed by logopedists (81,1%) and by psychologists (26.4%).

Only 48.1% of the services delivering diagnosis and/or treatment for fluency disorders are diagnostic-therapeutic coded and only 38.5% had, in the last five years, a specific training/update on fluency disorders. However, 93.6% of respondents feel the need for the enactment of National Guidelines. Finally, a high percentage of responses were unable to indicate if stutterers' associations were present in their region (53.2%).

Q3: Tools and methodological approaches characterizing the work of Italian logopedists.

51.1% of the respondents deal with disorders of fluency, and they were primarily aimed at preschool (76.1%) and school subjects (71.7%), with only 58.7% and 26.1%, respectively, focusing on adolescence and adulthood.

70% had training in fluency disorders, although 79.4% considered both the academic education/training and the available updating/refreshing courses in Italy insufficient, and as much as 95.7% noted it would be useful to issue National Guidelines.

All respondents would like to extend multidisciplinary training to better address the multidimensionality of the disorder.

It was found that SLT use quali-quantitative measurements of dysfluency (70.7%), audio-video recordings (65.9%), self-assessment questionnaires (56.1%), protocols (53.7%), specific tests (36.6%), and others assessment tools (14.6%).

With regard to treatment, there is a tendency to use an eclectic combination of different methodologies, rather than a single EB program. Among the EB therapeutic strategies, Stuttering Modification techniques are the most common (17.5%), followed by Fluency Shaping techniques (15%) and the Lidcombe program (15%).

41.3% of respondents indicated the following critical points in the assessment and therapy of stuttering in Italy: lack of standardized instruments validated on the Italian population (47.4%); practical difficulties, such as counting syllables stuttered online (26,3%); the need to integrate multiple methods (15.8%); and difficulties in involving the people most important to the patient into the diagnosis/treatment process (15.8%).

Discussion and conclusions

Although the data collected with Q1 and Q2 showed, as hypothesized, very different situation from region to region, in general the amount of theoretical and practical education/training and updating/refreshing is insufficient for all professionals involved in the diagnosis and treatment of stuttering.

This could create a dangerous circle that could eventually lead to the reduction of SLT experts in this area, and thus putting existing and future services at risk (Yaruss and Quesal, 2002). On the contrary, given the complexity of stuttering and the discomfort reported by health professionals to treat it (Yaruss and Quesal, 2002), a greater amount of education, training, and clinical experience is needed, both during and after the UCL.

As for Q2, however, the better public service for assessment/diagnosis rather than treatment and the greater difficulties and higher costs found in adulthood (Fibiger, 2008) may lead PWS to turn increasingly towards the private sector, where «miraculous» programs, lacking any evidence base or foundation of good practice, are proposed by individuals often lacking any professional qualifications. In addition, the rarity with which there is professional collaboration during the assessment and treatment of fluency disorders goes against the most recent evidence that to decrease the risk of relapses and facilitate the generalization of the modifications learned in therapy, it is essential to consider not only the overt symptoms, but also underlying symptoms through a multidisciplinary approach to the problem.

The under-representation, especially in the Centre and South Italy, of prevention programs, information campaigns, and self-help associations of stutterers, and especially the high percentage of respondents who do not know this information even when it does exist, stresses the need to increase the exchange of information between all those who for various reasons are concerned with this disorder.

Finally, we need to invest more on research in order to standardize the assessment tools and test the effectiveness of international programs on the Italian population. It is, in fact misleading and limiting to generalize the results obtained on patients with different culture and language (Andretta, Chiari and Stella, 2015).

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Session 6

Onset, recovery, persistence and assessment

VERBAL FLUENCY, LEXICAL PRODUCTION AND SOCIO-ECONOMIC CHARACTERISTICS IN A GROUP OF 24-MONTHS-OLD CHILDREN WITH A FAMILY HISTORY OF STUTTERING, IN RELATION TO POSSIBLE STUTTERING ONSET AND FINAL RECOVERY OR PERSISTENCE

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Abstract

The research project «Phonetic indexes predictive of persistent stuttering in preschool children» (CNR, RSTL n. 995), examines 40 subjects, divided in 26 children who have never started to stutter, and 14 who developed stuttering. As to the latter, 4 have become persistent and 10 recovered spontaneously.

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At 24 months of age, the Disfluency Profile (Yairi and Ambrose, 2005) was calculated in a representative sample of connected speech. The occurrences of Stuttering-Like Disfluencies and the occurrences of Other Disfluencies (equally common among stutterers and not stutterers) were counted. Lexical age (MacArthur-Bates, CDI) and socioeconomic score of parents (SES) were also obtained. The main result shows that future stutterers produce more SLD (3.01% vs 0.75%) and have smaller vocabulary size than future non-stutterers.

Introduction

In recent years, it has been given importance to the role played in stuttering by socioeconomic factors and family education (SES). SES is related to the sum of the sociological and economic indexes (of a person or a family), based on level of education, financial resources and type of occupation. Morgenstern (1956) studied the prevalence of stuttering in 7,000 children at the age of 11 years, suggesting that this prevalence was higher where the parents of stutterers, to ensure their children the best chance to improve their social situation, exerted a lot of pressure to increase their language skills. A second study (McKinnon, McLeod and Reilly, 2007), which examined a sample of patients with three different types of speech disorders (included stuttering), also found no significant relationship between the prevalence of disease and the family SES. Reilly et al. (2009; 2013), who investigated the onset of stuttering in 1,619 Australian children at the age of 2- and 3-years respectively, found the children who were stuttering to have mothers who were graduate or had followed postgraduate courses in greater proportion than the population in general (see also Howell, 2010). In contrast, Boyle, Boulet, Schieve, Cohen, Blumberg and Yeargin-Allsopp (2011) found a significant connection between SES and the prevalence of stuttering (and for speech-language pathology in general) for the families of children and adolescents (aged from 3 to 17 years) characterized by lower SES scores. A recent study led by Richels, Johnson, Walden and Conture (2013), suggests that the level of education of both the parents, the SES and the size of the child's vocabulary may be strong predictors of the skills development in language and vocabulary, and maintain significant relations with stuttering.

However, these studies have not evaluated the percentage of Stuttering-Like Disfluencies (SLD) and Other Disfluencies (OD) in 24-month-old children who subsequently begin to stutter and in those who will not. Our study aims at analysing the role of some social factors, such as the socio-economic status of the family (SES) and the level of parental education, which may influence the incidence and prevalence of stuttering. In our study we have considered it's important to calculate the percentage of disfluencies already at 24 months, taking into account a number of factors: stuttering begins between 2 and 4 years of age after a first period of fluent speech; several researchers have linked the frequency of lapsus, disfluencies and self-corrections with the rising of syntactic ability for utterance production (for a review, see Zmarich, 2012). Furthermore, it is worth of noting that the Mean Length of Utterance of the Italian children at 24 months already exceeds the 4 words (Caselli, Pasqualetti and Stefanini, 2007).

Method

Forty children (18 females and 22 males) participated in this study. All the participants were audio and video recorded at 24 months. Only those who started to stutter were assessed directly, with a series of audio and video recordings, for at least 16-22 months (every 3 months), and indirectly using phone interviews, up to 48-54 months post-onset. For each stage, the stuttering severity score, using the SSI-3 (Riley, 1994) was also calculated. At 24 months of age, the Disfluency Profile (Yairi and Ambrose, 2005) was calculated in a representative sample of connected speech, counting the occurrences of SLD such as repetitions of part of words or monosyllabic words as well as dysrhythmic phonations, and the occurrences of OD (equally common among stutters and not stutters). Children's lexical age (MacArthur-Bates CDI, Caselli et al., 2007) and educational and socioeconomic score of parents (similar to SES, Hollingshead, 1975) were also obtained through anamnestic and socioeconomic questionnaires. To determine the final clinical outcome of each child, a structured phone interview was conducted with the parents after an average of 40 months from the onset of stuttering (range: 20-68 months). It was found that 4 children had become persistent stutters (S) and 10 had recovered spontaneously (NS). S are the same children who had undergone therapy.

TABLE 1
Frequency, average and standard deviation of familiarity with stuttering in relation to the Parents (P), Siblings (Sb), or both (PSb); education of Mother (M) and Father (F) and the SES based on the final outcome (NS and S)

	FAMILY HISTORY			EDUCATION		SES		FAMILY SES
	PSb	PSb	PSb	M	F	M	F	
NS	5	13	4	4,73 (1,40)	4,23 (1,72)	46,28 (11,76)	38,27 (16,48)	40,01 (14,48)
S	4	4	4	4,07 (1,44)	4,00 (1,56)	38,27 (13,66)	38,43 (19,57)	38,286 (14,75)

Results

Overall, both future S ($M = -3.42$) and future NS ($M = -1.90$) presented a delay in the lexical age considering the age-referenced norms. To verify if there was a significant difference between the two groups based on the age difference among lexical and chronological age, a t-test on the scores difference of future S and NS was performed. The difference resulted not significant [$t(31,74) = 1.72, p = 0.09$], but the trend of future S to have a smaller vocabulary approaches the significance. As to comparison between future S and future NS for disfluencies, the Mann-Whitney U test revealed that future S produced a significantly higher SLD percentage, but not higher OD percentage, than future NS ($U = 75,00, p = 0,002$).

TABLE 2
Frequency, Average and Standard Deviation of chronological age, lexical age; difference between these two measures and percentage of SLD and OD according with the clinical outcome (S and NS)

	GENDER		CHRONOLOGICAL AGE	LEXICAL AGE	DIFF. LEX-CHRON	% SLD	% OD
	M	F					
NS	13	13	24,59 (1,09)	22,74 (4,002)	-1,90 (4,40)	0,75 (0,69)	3,32 (2,54)
S	9	5	25,29 (3,31)	21,50 (4,68)	-3,42 (2,98)	3,01 (2,73)	4,20 (3,16)

Next, we wanted to assess if there is a difference between the clinical outcome (S/NS) and the single SES of each parent, and/or the family SES, and/or the mother and the father education. In order to verify the contribution of these factors on the overall variance (R^2) of the two groups, after adding the «lexical age less the chronological age», we performed a logistic regression using, as dependent variable, the outcome classification. Results showed that these factors can explain a significant proportion of the variance ($R^2 = 0.478$). The 85% of the observations are correctly classified by the model ($p = 0.016$) but only the «lexical age less the chronological age» score was significant ($p = 0.034$).

We performed then a regression analysis within each group, considering the data from the individual coefficients taken from SES and as the dependent variable the «lexical age less chronological age»: only the mother's occupation, for the group of NS, approaches the significance ($\beta = 0.909$, $t = 2.173$, $p = 0.062$).

As for the variables «lexical age less chronological age» and «SLD %», we verified if it is possible to link these differences to different levels of SES. For this reason, a t-test on the difference in the scores of «lexical age less chronological age» of future S and NS within each SES quartiles, was carried out, considering separately the lowest ($N=10$), the highest ($N=10$) and the union of the two intermediate SES quartiles ($N=20$). A significant result was found for the mid SES: the lexical abilities of future S result delayed ($M = -4.25$, $SD = 1.83$) when compared to NS ($M = -1.09$, $SD = 3.44$), $t(15.85) = -2.57$, $p = 0.020$.

To evaluate the significance of the difference in the percentage of SLD within the quartiles, a non-parametric Mann-Whitney analysis was performed. It showed that in the subjects in the highest quartile ($> 75^\circ Q$), the future NS produce a significantly higher proportion of SLD compared to future S ($U = 19.000$, $p = 0.045$). Conversely, for those belonging to the intermediate quartiles, future S produce a significantly higher proportion of SLD with respect to NS ($U = 75.500$, $p = 0.031$).

Discussion

The present results show that, as early as 24 months of age, the future S differ from NS in the number of SLD produced (3.01% vs 0.75%). The

data obtained are particularly relevant for the early detection of the disorder in children who are at risk (familiarity), in order to identify and prevent the full manifestation of stuttering. From literature we know that S have less ability in expressive vocabulary (Ntorou et al., 2011). Our results highlight the trend of future S to have a more limited vocabulary than future NS. Associating the level of familiarity with stuttering and the family SES to the difference between the lexical age and chronological age, our analyses show a significant correlation between the mother's occupation and future NS. A higher SES index seems to increase chances to properly develop communication skills. We also found a significant difference in the lexical development and fluency of future S and NS which is associated to the middle SES category: future S produce fewer words and more SLD with respect to future NS, confirming Morgenstern (1956) results.

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AWARENESS AND ANXIETY AMONG PRESCHOOL CHILDREN WHO STUTTER (CWS)Ruth Ezrati-Vinacour³⁵, Keren Mamistvalove-Amir³⁶, Daniela Lobel³⁷, Nitza Katz-Bernstein³⁸

In the majority of cases, the disorder of stuttering begins before the age of four. For many years the question of whether young children who stutter are aware of their stuttering has been central in both theoretical and clinical thinking. Recent studies show that from the age of three CWS show signs of awareness towards dysfluency (Ambrose and Yairi, 1994; Yairi, 1983) and that CWNS are aware between the ages of four to five (Ezrati-Vinacour et al. 2001). Starting at the age of four we notice negative attitudes towards stuttering speech (Ezrati-Vinacour et al., 2001; Vanryckeghem et al., 2005). CWS experience undefined stress and anticipation of being insulted that may lead to feelings of anxiety. Most research studies conducted among PWS support a positive relationship between stuttering and state anxiety, there is lower agreement between trait anxiety and stuttering. However, the relationship between stuttering and anxiety among preschool children remains vague. One study has been published regarding the question about the relationship between the level of anxiety and stuttering in CWS and that study found no difference in anxiety levels between CWS and CWNS (Craig and Hancock, 1996).

The purpose of this current study is to examine the existence of anxiety (trait and state), awareness and attitudes towards speech among preschool CWS in comparison with two groups CWNS: 1. Preschool children with articulation disorders 2. Preschool children with no speech disorders. The

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speech of 50 children was recorded, 15 of them were CWS, 15 children with articulation disorders and 20 children with no speech disorders. Speech samples were analyzed for severity of stuttering and speech intelligibility via instrumental and subjective measurements. Three self-report measures were used: the Hebrew version of the FSSC (Fear Survey Schedule for Children, Ollendick, 1983), a questionnaire which evaluates trait anxiety level; RCSS (Reaction of Children to Speech Situations), a questionnaire that was developed in this study to evaluate state anxiety level; and the Hebrew version of the KiddyCAT (Communication Attitude Test for Preschool and Kindergarten Children Who Stutter, Vanryckeghem and Brutten, 2007) in order to evaluate levels of awareness and attitude towards speech. Results show a significant higher level of trait and state anxiety in the group of CWS as compared with both groups of CWNS. In addition, the awareness and attitude towards speech among both the CWS and the children with articulation disorders was significantly higher than that of children with no speech disorders. These results emphasize the importance of early intervention in stuttering in direct or indirect therapy by technical and/or communicational-emotional therapy. This may reduce anxiety levels and prevent the side effects of anxiety in young CWS.

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DIFFERENTIAL DIAGNOSIS OF EARLY CHILDHOOD STUTTERING AND OTHER UNUSUAL DISFLUENCY PATTERNS

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Abstract

The differential diagnosis of childhood stuttering bears on issues of definition, measurement and interpretation of observed characteristics, and alternative diagnostic distinctions to be made. There are many factors to consider in assessment. These factors are reviewed: speech sample procedures and contexts, sample size, word and syllable counts, stuttering and disfluency types and frequency measures, locations and nature of the observed characteristics, secondary (concomitant) behaviors, speech rate and intelligibility, demographics (age, gender, family background) and social communication behaviors.

Although diagnosis of childhood stuttering may be based primarily on an established definition and measures of observed speech characteristics, additional considerations are noteworthy, such as a child's awareness of speech disruptions. The evaluation of a child's developmental abilities (i.e., cognitive, linguistic, motor, articulatory, sensory-perceptual) close to and after stuttering onset also contributes importantly to understanding the fluency disruptions and associated communication behaviors. An analysis of risk factors will aid interpretation, as well.

The majority of diagnostic decisions about stuttering can be made confidently by the application of research-based diagnostic criteria, but sometimes a child presents with speech characteristics that are different from the classic profile. The pattern of disfluent speech may suggest an alternative clinical interpretation. A child's speech characteristics sometimes are not «stuttering-like» but are not «normally» disfluent, either. Consideration is given to other patterns and forms of fluency disruptions, including the types and patterns characteristic of language impairments. Other unusual cases are encountered when a child inserts pauses at inappropriate locations,

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for example, in the middle of words, but displays no other stuttering-like disfluencies. Yet another diagnostic challenge may be faced if disruptions are stuttering-like, but occur predominantly in word-final position. Does an atypical pattern signify a different disorder subtype? When and how may the term «stuttering» still apply?

The nature of diagnosis, definitions and symptom characteristics of stuttering and other fluency disorders will be reviewed. A framework of classifications will be examined. Perspectives on the application of diagnostic and prognostic criteria will be considered, and clinical decision trees will be shared.

Introduction

A *diagnosis* is «the identification of a specific condition usually not apparent at the beginning» (Yairi and Seery, 2015, p. 199), but a *differential diagnosis* identifies that a condition (i.e., disorder) is different from other possible clinical conditions. When disfluency patterns are evaluated in young children, there are three main questions of differential diagnosis for speech-language clinicians: 1) Is this stuttering or normal disfluency?; 2) If it is stuttering, is this the type that will persist or recover?; and 3) If it is not stuttering nor normal disfluency, what other type disfluency disorder is it?

Consideration of differential diagnosis criteria

An ideal set of criteria yields 100% accuracy in the discrimination of cases. We want differential diagnosis criteria with sensitivity (correct identification of true positives) and specificity (correct identification of true negatives). This ideal outcome is impeded by challenges in several areas: How do we define possible fluency disorders? How do we measure observed characteristics? How do we best distinguish and interpret observed characteristics?

To decide when to diagnose a fluency disorder, we can refer back to Van Riper's perspective that speech is defective when it: «deviates so far from

other people that it calls attention to itself, interferes with communication, or causes its possessor to be maladjusted» (Van Riper, 1963, p. 16). Van Riper defined stuttering as occurring when the «forward flow of speech is interrupted by a motorically disrupted sound, syllable, or word or the speaker's reaction thereto». (Van Riper, 1971, p. 15).

Near the time of onset, children who stutter are often unaware of their speech disruptions (Ambrose and Yairi, 1994; Bishop, 2013). The reduced awareness of speech in very young children was recently underscored by research comparing responses to altered auditory feedback among non-stuttering adults, young children, and toddlers. Although 4-year-olds and adults adjusted their vocal productions responsively to altered auditory feedback, 2-year-olds did not (MacDonald et al., 2012). Yairi and Ambrose (2005) reported a mean age of stuttering onset of 33 months (2 years, 8 months). This early age makes it appear unlikely that the emergence of stuttering-like disfluencies stems from a child's awareness of a speech problem. The level of awareness of a disorder may be considered based on a pyramid model (based on Barco et al., 1991) with three components: intellectual awareness, emergent awareness, anticipatory awareness. Even if awareness is not an essential element in the diagnostic criteria, if a child expresses a complaint or shows signs of frustration and awareness, the diagnosis of stuttering is easier to make.

Because the disorder of stuttering may exist even if a child is unaware of speech disruptions, differential diagnosis when stuttering is first suspected relies more heavily on the observations of surface speech characteristics. Yairi and Ambrose (2005) have provided criteria based on multiple disfluency indicators to aid diagnosis of stuttering vs. normal disfluency. These criteria were established conservatively to minimize instances of false positives.

Several important considerations in disfluency measurements have been highlighted by Yairi and colleagues, including: disfluency classifications, syllable (vs. word) counts, sampling in multiple contexts (e.g., at home vs. the clinic), samples of sufficient length (Sawyer and Yairi, 2006), and parent and clinician severity ratings. It is especially important to classify and quantify the observed types of disfluencies, differentiating the stuttering-like disfluencies (SLD) from less disruptive (Other) types. Despite care taken with measurements, the high levels of variability in disfluencies

and stuttering-like events can still cast shadows of uncertainty about the diagnosis in the minds of both parents and clinicians.

Does a certain level of struggle or tension need to be present to be confident of a «stuttering» diagnosis? Is the faster rate of a disfluent event a potential indicator of tension or ‘loss of control’? The dimension of tension is difficult to measure, although it may be reflected in measures known to differ between children who stutter and age-matched controls, such as the average numbers of sound or syllable repetition units, or the time intervals between repetition units (Yairi and Ambrose, 2005).

Uncommon disfluency patterns

Knowing that we will encounter considerable variability across stuttering cases, how generally present across situations do SLD types need to be before we are confident of the diagnosis? What do we decide about the diagnosis of a child who only stutters noticeably in isolated conditions (e.g., only in extreme excitement), but rarely if ever displays stuttering in the routine course of each day? Are we sometimes challenged to justify a decision to intervene based on parent report that the child stutters, because both parents and clinicians are failing to catch events on the recordings made at home and in the clinic?

Still another challenge to differential diagnosis arises when the SLD criteria are met for stuttering, but the locations of the SLD are atypical. Sometimes word-final sound or syllable repetitions are observed in children (McAllister and Kingston, 2005; Rudmin, 1984). Should these cases be considered stuttering? Cases are especially challenging when there is a mix of *both* word-initial and word-final disfluencies. When the disfluencies are almost exclusively in word-final locations, greater doubts arise about whether the pattern should be called «stuttering». In one published case study, the pattern was described as «articulatory oscillations». In another case, it was proposed that the pattern may be a phonological process (Camarata, 1989); it occurred specifically on voiceless consonants. Can such patterns be similar to the phonological processes of reduplication (e.g., «baba» for bottle) or doubling («car-car» for car)?

Evaluation of a child's developmental abilities (i.e., cognitive, linguistic, motor, articulatory, sensory-perceptual) close to and after stuttering onset also contributes importantly to understanding the fluency disruptions and associated communication behaviors. Interpretation is also affected by demographics (age, gender, family background) and social communication behaviors.

Stuttering can co-exist with language disorders, or a language disorder may be the main reason for excessively high levels of disfluent speech. A language-based fluency disorder can be differentiated based on assessment of language skills as well as the observation of predominant disfluencies in the «Other» types including revisions, interjections, multisyllable and phrase repetitions (Befi-Lopes et al., 2014). In these cases, the disfluencies stem from difficulties with word-finding and formulating sentence structures. In cases of severe language disorders however, such as autism, children may be observed to engage in fewer interjections (ums and uhs) or revisions, and produce more silent pauses and repetitions instead (Lake, Humphreys and Cardy, 2011). These characteristics have been explained as they relate to speaker-oriented vs. listener-oriented strategies by the speaker.

The predominance of non-SLD disfluency types in older children can still mask a stuttering disorder, however. This means another challenge for disfluency classification arises when unusual clustering of disfluencies is observed. For example, what classification is appropriate if a child's sound repetitions occur predominantly on interjection types ('um or uh')? Will the measurement criteria for differential diagnosis of stuttering still be appropriate to apply?

Finally, clinicians may face a diagnostic challenge if children display atypical locations of hesitation pauses. What diagnosis is appropriate if the clinician observes a highly unusual pattern of inserting pauses to breathe mid-word rather than at clause or sentence junctures? Inexperienced clinicians may be tempted to classify such events as broken words (i.e., disrhythmic phonations), but this identification of the disfluent characteristics may be inappropriate. The existence of an unusual behavior in isolation (i.e., relatively few disfluencies otherwise), and absence of struggle or tension during mid-word breaks may be important to making an appropriate interpretation when the pattern is not «stuttering».

Conclusion

In summary, differential diagnosis of childhood fluency disorders includes the consideration of definition, diagnostic criteria, measurement processes, overall disfluency levels, and classification of disfluency types. Knowledge of the typical patterns of observed characteristics in stuttering and normal disfluency aids the recognition of uncommon disfluency patterns that may be isolated to specific types or unusual locations. Word-final repetitions and mid-word pauses for breath, are examples of uncommon disfluency patterns that may not be «stuttering». A child's overall profile of assessment results, the nature and level of speech awareness, and the signs of whether there is tension or struggle, can aid the interpretation and differentiation of uncommon disfluency patterns.

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ASSESSMENT OF BILINGUAL CHILDREN WHO STUTTER: WHAT CAN WE LEARN FROM NON-STUTTERING BILINGUAL CHILDREN

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Abstract

With about 60% of the world's children being raised as dual language learners and approximately 6,000 languages spoken throughout the world, globalization has re-emphasized the benefits of being bilingual. This comes with the need for better understanding of the impact of bilingualism on all areas of child development. With the trend toward more children being raised bilingual, it is inevitable that, as speech pathologists, we must update our techniques to properly treat these children. In order to treat stuttering in bilingual children, we must first understand where speech patterns of monolingual and bilingual children differ, so that our diagnostic tools can be updated to reflect these differences.

Introduction

Globalization has re-emphasized the need for better understanding of the impact of bilingualism on all areas of child development. At least 60% of the world's children are dual language learners. In the European Union, 10% of school age children speak a language other than the majority language of the country that they live in.

With the number of children from diverse linguistic background increasing, most Speech Language Pathologists are not adequately prepared to assess bilingual children who may be at risk for stuttering and have little normative data with which to make comparisons.

In order to understand the impact of bilingualism on children who stutter, speech pathologists need culturally appropriate protocols that first differentiate types of bilingualism. These assessment protocols will aid clini-

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cians in accurately identifying true stuttering from normal speech disfluency in bilingual children. However, since normal speech patterns of bilingual children may differ from those of monolinguals; we must therefore first establish norms by evaluating the language and fluency development of bilingual children who have never stuttered (BCWNS) in order to identify deviations from normal speech in bilingual children who stutter (BCWS). This will lead to better identification and, ultimately, better treatment of those children at risk for stuttering. This presentation will discuss the language and fluency development of bilingual children who have never stuttered in order to develop a protocol for assessment of bilingual children who are at risk for stuttering.

Findings for language and disfluencies in the speech of young bilingual children who never stuttered, will be used to inform the assessment of bilingual children who are at risk for stuttering.

Bilingualism

With limited normative data on bilingualism and little exposure to case studies, speech pathologists may see bilingualism as unfamiliar and complex, perhaps even abnormal.

The reality is that bilingual is another form of normal. Contributing to the perceived complexity are the wide variations in patterns of bilingualism, including the age at which children become bilingual, the settings in which they use each language, the extent that they must rely on each language and for what purpose(s), and their motivation to learn both languages. Bilingualism can represent a cultural heritage, a scholastic goal, or a local necessity. And of course, bilingual children have unique individual characteristics, personalities, abilities, strengths, and weaknesses. These unique individual characteristics are relevant to assessment and treatment.

There are two basic categories of bilingualism: Bilingual First Language Acquisition (BFLA), in which two (or more) languages are acquired around the same time, and Sequential or Early Second Language Acquisition (ESLA), in which children acquire one language first and a second language later, often upon school entry. In young children stuttering may manifest differently in

these two patterns of bilingual language development. In the pre-school-age population, the majority of studies focus on BFLA. This presentation will therefore focus primarily on BFLA with some compensation for ESLA.

Bilingualism is a continuum rather than a categorical measurement, and performance on language tasks fluctuates depending upon language dominance and proficiency (Grosjean, 2004). Level of proficiency tells us about individual capabilities in understanding, speaking, reading and writing in each language spoken, whereas dominance refers to many dimensions of language use and experience including proficiency, ease of processing and frequency of use. These dimensions are not independent and have considerable overlap: however, even though «levels of proficiency and degrees of dominance tend to correlate» (Birdsong, 2006; p. 47), bilinguals can have almost native-like proficiency in both languages but still consider one language to be better than the other. Alternatively, they may be dominant in one language (L1 or L2) but not necessarily highly proficient in that language. With no standard measure, detailed descriptions of exposure and usage are critical factors to determining levels of bilingualism.

Bilingualism language development

Evidence shows that bilingualism does not impair language and should not be discouraged, even for children with Language Impairment (LI) (Thordardottir, 2006). Research in Canada shows that pre-school children with impairment in their first language, enrolled in bilingual immersion programs, performed as well as peers enrolled in monolingual programs (Bruck, 1982). Even children with more severe developmental disorders such as Down Syndrome, in bilingual education, evidenced language characteristics similar to those documented for their monolingual peers (Kay-Raining Bird et al., 2007).

Few studies have compared young BFLA children's lexical development to that of comparable monolinguals (see De Houwer et al., 2015 for a review). In BFLA, where multiple languages are acquired at the same time, lexical development may be situation-specific and linguistic proficiency may not be the same in both languages (Grosjean, 1989). BFLA children are usually more proficient in one language and there is a strong relationship between

language exposure and bilingual development (i.e., a higher level of exposure leads to stronger linguistic performance) (De Houwer, 2009; Pearson, Fernandez and Oller, 1997; Thordardottir, 2011). For example, a child immersed in a second language in school may not know specific school words in his/her home language. As such, while both expressive and receptive vocabulary may be smaller in each language when compared to monolingual language development, the total vocabulary of both languages combined is generally the same as that of monolinguals. De Houwer and colleagues (2015) found that their sample of bilingual Dutch infants understood more words than the monolinguals. Hence there is no consistent difference between the vocabulary sizes of young bilinguals and monolinguals, and no evidence that exposure to two languages from birth slows down lexical development and in fact bilingual infants may benefit from the diversified language input.

Current research suggests that being bilingual is as natural a process as monolingual language development. The benefits of bilingualism are equally clear, and include access to two language communities and a richer linguistic environment that can positively affect vocabulary and cognitive development. Bilingual children should be assessed in relation to a bilingual rather than monolingual standard, with both languages taken into account to determine proficiency. Assessment must consider both the amount of input in each language as well as the structural and pragmatic differences between languages (Thordardottir, Rothenberg, Rivard and Naves, 2006).

Bilingualism and stuttering

It is not clear whether bilingual development increases the risk for stuttering. Stuttering typically has its onset in the preschool years, during a period of dynamic linguistic development, and the imbalance of language proficiency during this time may impact fluency. During this critical period of dual language development, uncertainty with two languages might lead to increased disfluencies, and the distinctive motoric differences in the production of two languages could uniquely compromise a bilingual child's ability to establish and maintain fluency. This additional burden could result in bilingual children being more susceptible to fluency breakdown and po-

tentially lead to stuttering. Some feel that the exposure to a second language prior to entering school increases the likelihood of stuttering (Howell, Davis and Williams, 2009; Packman et al., 2009) and would therefore support the elimination of one language during this critical period. However, a certain amount of normal speech disfluency is expected in monolingual children, and perhaps the aforementioned difficulties represent the normal speech disfluencies of pre-school-age bilinguals.

Research that has examined the speech disfluencies of normally fluent children has historically focused on descriptions of monolingual English speaking children in order to identify stuttering and predict the risk of persistence (Yairi and Ambrose, 2005; Thornberg and Yairi, 2001). Studies of other than English speaking children are limited (Robinson, Davis and Crowe, 2000, Bernstein-Ratner and Benitez, 1985; Carlo, 1996). Although patterns of disfluency may be similar across languages in monolingual children, there may be differences in bilingual speakers. Therefore, the use of monolingual guidelines to evaluate disfluency in bilingual children can be misleading.

In monolingual children who have never stuttered, disfluencies are increased in more complex tasks. Bilingual children who never stuttered (BCWNS) seem to produce a greater frequency of disruptions in the forward flow of speech than their monolingual peers (Bedore, Fiestas, Pena and Nagy, 2006; Lennon, 1990; Lofranco, Pena and Bedore, 2006; Rieger, 2003; and Wiese, 1984) and may have a higher frequency of disfluencies in complex language tasks. Bryd and colleagues (2015) evaluated narrative samples of bilingual children who have never stuttered (BSWNS) and found that the frequency of stutter-like disfluencies ranged from 3-22%, exceeding the monolingual English standard of 3 stuttered-like disfluencies per 100 words spoken. It was also found that there was no significant frequency difference in stutter-like or typical disfluencies produced relative to language dominance. There were, however, significant differences depending on the language that the child was speaking, e.g. more stutter-like disfluencies in Spanish regardless of language dominance. In an ongoing study in a population of English/French bilinguals (Shenker and MacLeod, in preparation), preliminary results are similar, with more stutter-like disfluencies found in the French samples.

Summarizing, the assessment of disfluencies in the narrative samples of BCWNS has suggested that the frequency of disfluencies exceeds monolin-

gual guidelines. One reason for this could be attributed to increased levels of linguistic uncertainty that leads to increased disfluencies. However, types of disfluencies are consistent with the findings for monolingual children, with monosyllabic whole word repetitions being manifested by more than 3%, particularly at the onset of phrases or utterances. Additionally, the number of iterations per moment may be higher than what has been reported for monolingual children. Bryd and colleagues found no instances of audible/inaudible prolongations in their samples. These factors, plus irregularities in timing and tension may be predictors of increased risk of stuttering in bilingual children.

While bilingual acquisition is as natural as monolingual, and feature the same developmental milestones, there are individual differences that must be accounted for in assessing the risk for stuttering. ESLA children who speak a minority language as L1 may face unique problems in regard to the risk of stuttering. In these cases, socio-economic factors and the relationship between amount of exposure to the majority language of the community and the performance of the child in that language may provide additional challenges that result in a weaker performance and higher rate of disfluency.

It is concluded that linguistic uncertainty during early language development may temporarily lead to increased typical disfluencies in young bilingual children. The findings described support the recommendation that monolingual disfluency frequency guidelines may be too low for what might be indicative of stuttering in bilingual children. This has the potential to increase the risk of false positive identification of stuttering in a bilingual child. What is needed is reconsideration of distinctive findings from bilingual children who have never stuttered in order to construct unique standards for assessing the risk of persistent stuttering in a bilingual population.

Clinical implications

Despite the paucity of normative data, clinicians are increasingly asked to assess bilingual children. The information that we do have does suggest guidelines for clinicians. In assessment of bilingual children who may be at risk for stuttering, clinicians should follow these guidelines:

- Always evaluate in both languages regardless of dominance;
- If a bilingual child seems slow in lexical or language development, try to understand the nature of the delay rather than attribute it to bilingualism;
- Assess input experiences of the child in each language spoken;
- Rather than compare to monolingual standards, examine variations in exposure to understand the variability in early lexical development;
- Include a narrative task for assessment of disfluency in complex language when a child is older than 4 years;
- Compare typical disfluencies to stuttered-like disfluencies by type and loci;
- Since increased disfluencies may be related to linguistic proficiency it is important to remember that unequal proficiency development may be reflected by different disfluency patterns in each language spoken;
- Factors that may predict increased risk for stuttering in bilingual children include clusters of disfluencies, the presence of audible/inaudible prolongations and an uneven rhythm or rate of iterations per moment of disfluency.

Children who are introduced to a second language when they start school will benefit from the support of the first language (L1) at home. These children often start a second language (L2) while the L1 is neglected or not properly stimulated at home. This may apply in cases where the language of the home is a minority language in the community. Additional factors to keep in mind when assessing these children include:

- Parent educational level and language proficiency in the majority language of the country;
- Length of time that the child has lived in the country;
- Length of time that the child has been exposed to the majority language in general, at home, at school;
- Languages used by key adults in the child's environment.

Conclusion

In conclusion, there is a need for normative assessment standards for bilingual children, and using monolingual guidelines to determine prevalence and risk of stuttering in young children is cautioned against. Further research

is needed to create norms for bilingual speakers and, in particular, norms for comparison of similar/dissimilar languages. Better protocols for assessment will provide a clearer picture of the risk for stuttering and increase awareness of complexities of assessing ESLA children, highlighting the issues related to cultural diversity that may affect fluency. The establishment of normative standards for the evaluation of dual language learners at risk for stuttering will impact positively on treatment recommendations and outcome.

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Session 7

Treating children who stutter

**RELATION BETWEEN STUTTERING AND ANXIETY DISORDERS IN
ITALIAN CHILDREN AND ADOLESCENTS WHO STUTTER**

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Abstract

Purpose

The relationship between anxiety and stuttering has been widely studied. Indeed, over the past 50 years this has resulted in compelling and controversial theories and hypotheses that tried to shed light on the nature of this relationship. Previous researches have shown an increase of negative emotional reactions, as anxiety, related to particular speech

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situations in people who stutter. Specifically, it was found that levels of trait anxiety of children and adolescents who stutter do not differ from that one of non stutter peers.

However, children and adolescents who stutter display a significant increase of the state anxiety in speech situations, due to the fear of receiving a negative evaluation.

Current study aimed at assessing

1. possible difference in negative emotional reaction to speech situations and general anxiety between children and adolescents who stutter and non-stuttering peers;
2. possible associations between emotional reactions in speech situations and general anxiety in both groups.

Methods

In order to evaluate the general level of anxiety and its component the Italian version of the Revised Children's Manifest Anxiety Scale, Second Edition, (RCMAS-2) and the Speech Situation Checklist–Emotional Reaction (SSC-ER) were administered to 65 children and adolescents who stutter and 65 non stuttering peers aged from 9 to 17.

Results

A series of univariate ANOVAs, with Status, Age and Gender as independent variables and SSC-ER scores and RCMAS-2 total and subscales scores as dependent variables, were performed. The stuttering group was significantly higher than non stuttering group on the subscales Defensiveness and Social Anxiety scores of RCMAS-2, in addition, the interaction between Status and Age resulted significant. A series of separate correlations between SSC-ER scores and RCMAS-2 showed that in both groups levels of anxiety were associated to levels of emotional reactions to speech domain.

Conclusion

Children and adolescent who stutter did not result more anxious than fluent peers, however, presented higher levels of emotional reaction in social communication that might increase over time. They also referred higher levels of anxiety in social situations than non-stuttering peers although these levels are not clinically significant.

Introduction

Stuttering is a communication disorder that may be associated over years with negative consequences, such as negative mood states, distress and reduced feelings of self-efficacy, impairments in social interactions and lower quality of life (Craig et al., 2009). In this regard, anxiety is one of the most widely observed and extensively studied psychological concomitants of stuttering (Iverach and Rape, 2014). Despite the evidence of social anxiety disorder among stuttering adults, we still know very little about the onset of anxiety disorders in children and adolescents who stutter, although this identification would lead to a clinical implications for the management of both disorders across the lifespan. Smith et al. (2014) tried to fill this gap by publishing an important review of the research evidence relating to anxiety for children and adolescents who stutter. In sum, they found that anxiety tends to increase over time until it exceeds normal limits in adolescence and adulthood, and from this moment the risk of chronic anxiety become more likely. At the same time the authors concluded that findings from the studies focused on anxiety and stress in school-age children and adolescents who stutter were inconclusive. Below are described main findings emerged in previous studies. Blood, et al (2007) administered to 18 stuttering young people and 18 non-stuttering controls (aged from 12 to 18 years) the RCMAS scale. The stuttering group showed levels of anxiety within normal limits but higher than those of age-matched, non-stuttering controls. One year later Mulcahy et al. (2008) investigated social anxiety in 19 stuttering adolescents and 18 age-matched, non-stuttering controls (aged from 11 to 18 years), using the STAIC. Results showed that stuttering group evidenced significantly higher

levels of state, trait and social anxiety, when compared with non-stuttering controls; however, it is to note that anxiety levels were in the average range. Subsequently, Gunn et al. (2013) investigated anxiety in 37 stuttering adolescents (aged from 12 to 17 years) using a battery of assessments including the RCMAS-2. The 38% of stuttering adolescents received at least one diagnosis of a mental disorder, with the majority of these diagnoses involving anxiety. More recently, Messenger et al. (2015) administered the RCMAS to 18 school-age boys and 5 girls and to 41 adolescent boys and 9 girls who were seeking treatment for stuttering; age of participants ranged from 6 to 18 years. The authors found that all mean scores of RCMAS subscales of both groups of boys were within normal limits and only the scores on the Lie Scale were significantly higher than scores on the other three subscales. The purpose of this study is two fold: 1) verifying whether the amount of negative emotional reaction to particular speech situations and the amount of anxiety reported by children and adolescents who stutter is significantly higher compared to non-stuttering peers 2) investigating the hypothesized associations between state anxiety (in speech situations) and general anxiety.

Methods

In order to evaluate the general level of anxiety and its components, the Italian versions of the RCMAS-2 (Scozzari et al., 2011), and the SSC-ER, (Brutten and Vanryckeghem, 2007) were administered to 65 children and adolescents who stutter (20 females and 45 males) between 9 and 17 years old, matched by age and gender to 65 children and adolescents who do not stutter. The severity of stuttering was established at the time of this investigation using the SSI-4 (Riley, 2009).

Statistical analysis

The original groups were recoded into two subgroups: the younger (children), from 9 to 12 years ($N = 72$), and the older (adolescent), from 13 to 17 years ($N = 58$). In order to test the statistical significance of the differ-

ence in scores between children and adolescents who stutter and children and adolescents who do not stutter, and the influence of age and gender, a series of 3-way ANOVA analysis were performed, in which the SSC-ER summed scores and RCMAS-2 scores (total scale and four subscales) were the dependent measures and Group membership (stuttering/nonstuttering), Gender (male/female) and Age (younger/older) were the between factors. Finally, in order to examine the relationship between the SSC-ER summed scores and the RCMAS-2 scores a series of Pearson correlation coefficients were computed for each group separately.

Results

Difference between groups in emotional reactions to speech situations and anxiety levels

As far as concern the SSC-ER questionnaire the 3-way ANOVA evidenced a significant interaction between group membership and age [$F(1,122) = 8.206, p = .005 \eta_p^2 = .063$]. *Post hoc* analysis using Bonferroni's test revealed that the younger subjects who stutter ($M = 105,83; DS = 31.140$) reported significantly lower mean scores than the older ones ($M = 121.79; DS = 30,92$) ($p = .006$), whereas the subjects who do not stutter did not show any significant differences between the younger and older groups ($p = .217$).

In regard to the RCMAS-2, the analysis yielded a significant main effect for group membership in the subscales Defensiveness (DEF) [$F(1,122) = 7.257, p = .008 \eta_p^2 = .056$] and Social Anxiety (SOC) [$F(1,122) = 4.317, p = .040 \eta_p^2 = .034$] with the stuttering group obtaining significantly higher mean scores than nonstuttering group on both DEF ($M = 51.7, DS = 8,80$ vs. $M = 47.3 DS = 10$) and SOC ($M = 51.5, DS = 8.9$ vs. $M = 48,8 DS = 9.6$) although within the normal range of functioning. A significant main effect was also found for gender on the Total Anxiety Scale (TOT) [$F(1,122) = 5.148 p = .025 \eta_p^2 = .040$] and SOC [$F(1,122) = 5.623, p = .019 \eta_p^2 = .044$], with females scoring significantly higher than males on both TOT ($M = 51.4, DS = 6.8$ vs $M = 47.2, DS = 10,3$) and SOC ($M = 53, DS = 7,9$ vs. $M = 48.9, DS = 9.6$), however within the normal range. No other significant main effects or interactions emerged.

Associations between emotional reactions to speech situations and general anxiety

For both groups, significant and positive correlations emerged between the SSC-ER and the subscales of the RCMAS-2. In particular, among children and adolescents who do not stutter moderate and strong significant correlations were found between SSC-ER score and four RCMAS-2 scales such as TOT ($r = .53$), Physiological Anxiety (PHY; $r = .55$) Worry (WOR; $r = .64$) and Social Anxiety (SOC; $r = .53$). Conversely, children and adolescents who stutter showed no significant correlations between SSC-ER scores and RCMAS-2 PHY but only significant moderate correlations between SSC-ER with the subscale TOT ($r = .37$), WOR ($r = .30$), SOC ($r = .41$) of the RCMAS-2, respectively.

Discussion

Present findings highlight, as has been demonstrated in several cross-cultural investigations, that children and adolescents who stutter show higher levels of emotional reaction in communication experiences than fluent peers. Furthermore, unlike fluent speakers, children and adolescents who stutter increase the levels of anxiety in speech situations as they grow up. During the period of adolescence they experience more frequently environmental risk factors like teasing, bullying and negative peer reactions, that may increase the development of emotional reaction (as worry and anxiety) in communication situations. However, in the group who stutter, anxiety seems to be not only a contingent phenomenon restricted to speech situations but it is rather diffused to general social situations. Indeed, in this study children and adolescents who stutter tend to report greater levels of social anxiety than the non stuttering group, although these levels are not clinically significant. Furthermore, the evidence that children and adolescents who stutter present a higher defensive responses than fluent peers, may suggest a tendency for the first group to desire to be approved and accepted or may also reflect feelings of social isolation or rejection which are themes of particular interest for children and adolescents who stutter. The absence of significant differences between the two groups on RCMAS-2

total anxiety scale could indicate that adolescents who stutter are not more anxious than fluent peers. Finally, to date, the nature of correlation between anxiety responses and emotional reaction to speech situations of adolescents who stutter and non stuttering peers was consistent with the finding that these groups exhibited more differences than similarities.

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**EFFECTIVENESS OF THE CLASSROOM COMMUNICATION RESOURCE
IN IMPROVING PRIMARY SCHOOL LEARNERS' ATTITUDES TOWARDS
CHILDREN WHO STUTTER: LESSONS LEARNT FROM A RANDOMISED
CONTROL TRIAL FEASIBILITY STUDY**

Freda Walters, Rizwana Badroodien, Vivienne Norman, Harsha Kathard⁴⁷

Abstract

Children who stutter are bullied and teased by their peers, especially in the primary schooling years. The Classroom Communication Resource (CCR) was developed as a teacher-administered classroom-based education programme aiming to improve peers' attitudes towards CWS. The focus of this feasibility study was to determine the initial treatment effect of the CCR to improve peers' attitudes towards CWS and the procedural aspects for a larger scale cluster randomised trial (CRT) in future. Peer attitudes were determined via a Likert scale questionnaire, the Stuttering Resource Outcomes Measure (SROM), completed by primary school learners. A quantitative, CRT design with a control group was utilized to describe the direction and magnitude of changes in Grade 7 peers' attitudes towards CWS following the administration of the CCR at one-month and 6-month intervals. The evidence confirmed that the SROM was a valid and reliable outcomes measure. Attitude changes after the administration of the CCR were analysed through inferential statistics. A significant positive change in SROM scores were found in the experimental group ($p = 0.005$) when compared to the control group ($p = 0.41$) at one month as well as at 6 months ($p = 0.002$). Further improvement in SROM scores was found in the intervention group at 6-month follow up. The study indicated initial positive treatment effect of the CCR and validates further investigation. Participation rates decreased at the 1-month interval (91.5%) and even further at the 6-month interval (65.2%). This was mainly due to poor questionnaire completion and absenteeism. The implications of these findings for future full scale CRTs are discussed.

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ABC BALBUZIE®: AN EFFECTIVE INTEGRATED FLEXIBLE APPROACH TO STUTTERING FOCUSING ON SCHOOL AGE CHILDREN AND THEIR FAMILIES

Luisella Cocco, Monica Anisio, Simona Bernardini⁴⁸

Abstract

The ABC BALBUZIE® project, based in Turin, is a set of activities aimed at: increase knowledge and raise awareness of issues related to stuttering in Pediatric and School field, replace common stereotypes with more accurate evidence-based information; provide comprehensive information to the parents of children who stuttering (CWS) about risk factors, chronic stuttering and facilitating speech fluency; help the CWS to realize how they can become active and effective communicators, modify their dysfluency and improving their communication skills. The methodological approach run by specialized professionals, speech therapists (ST) and psychotherapist (PSY) integrates numerous aspects in compliance with the International Intervention Guidelines, (ASHA Guidelines, 1995) and the CALMS Model (Healey and al., 2004).

One of the goals of our structured Preliminary Therapy Course (PTC) for CWS and their families is to develop in the participants positive attitudes toward stuttering.

The Rational Emotive Behavior Therapy (REBT) is incorporated to the treatment to change the irrational beliefs about self-worth who generates emotional upset, reduced feelings of self-efficacy and lead the CWS to avoid the speech situation in social contexts perceived as demanding.

It had become very useful to involve parents, as a strong point of the program, to change their speech associated negative attitude, which tends to be corrective and leads as a result, to believe that stutter is a wrong way of talking. At the same time, the children will carry out activities in which they

⁴⁸ ABC BALBUZIE®, Turin.

will learn to identify their stuttering characteristics, the maladaptive emotions that they could feel and will work on improving fluency, changing the negative speech-associated attitude and learning problem solving strategy to deal with teasing or bullying. We have had positive feedback in this structured approach from CWS, parents and teachers. At the moment we are collecting result data, based on assessment, post treatment and follow-up through structured interview, stuttering severity rate, speech-associated attitude.

Introduction to aims of the project

A global approach to stuttering had already been put in place in year 2000 and structured in a project presented to the Sixth Oxford Dysfluency Conference (Cocco, 2002) which was carried out over the following years to address the critical issues that emerged during that period such as: incorrect common places about stuttering, for example «stuttering is caused by anxiety or emotional trauma»; almost total absence of prevention and tendency to send to the specialist from six years of age onwards; long term treatment focused mainly on individual treatment with the aim to reach fluency with very little involvement of parents and teachers; poor attention on the negative impact that living with stuttering over the years could have on health domains; no mention of negative emotional reactions or negative speech attitude associated.

In 2012 we decided to join forces combining our different professional skills in a more structured way improving, integrating and modifying the activities and goals of the therapeutic program which has continued to evolve over the years, according to the analysis of critical issues emerging time by time. Over the years, we have seen that some elements, which we focused and worked on, have changed and improved such as: paediatricians and parents referring children to specialist earlier than years ago (from age 3 and 4); teachers more concentrated about stuttering in classrooms; increased focus on the improvement of positive communicative attitude as well as improving fluency; a better understanding of the importance of a stuttering assessment before treatment; parents more involved and concerned about the etiology of stuttering; increased demand for evaluation also from territories outside Turin or request for a second expert opinion.

On the other hand, we know that further work is needed to modify the persistence of some critical issues: still lack of correct information about stuttering risk factors and characteristics of stuttering itself, because still view as a result of emotional causes; parents too centred on the symptom «stuttering», reaching for fluency recovery from stuttering, without considering the influence of other predisposing, triggers and maintenance factors; persistence of common places and incorrect beliefs about stuttering.

Therapeutic approach

The ABC BALBUZIE* program for school age is structured as follow:

- assessment: varies according to age and includes a structured interview with the parents and with the child (Florio and Bernardini, 2014); the severity of stuttering established using the SSI-4 (Riley, 2009); the speech associated attitudes evaluated by the Italian versions of the CAT (Brutten and Vanryckeghem, 2007); the concerns and emotions about stuttering assessed by the Italian version of the pencil test «Framing my speech» (Florio and Bernardini, 2014); observation of communicative parent-child interaction (Rustin, Botterill and Kelman, 1996);
- preliminary Treatment Course in small group (2 to 3 children and families) organized by age groups;
- follow up after six months re-evaluates the communicative attitude, the severity of stuttering, the expectations of CWS and their parents about the recovery from stuttering, the management of stuttering at home and at school;
- further treatment sessions, directed to improve fluency shaping or/and speech associated attitude, and working to transfer in different life situation.

Specifically in this presentation we will focus on children aged 7 to 11, who attended the PTC in 2014.

We carry out activities in small groups of two children and in parallel, in the same hour of therapy, which are also working with parents, in a nearby room. In this way, we optimize the intervention time, to meet the family's needs who are often very busy.

The PTC is divided into two parts: «Learning and discovering together» and «Working with REBT».

The first part consist of:

- six therapy sessions during which parents and children work in two different rooms with speech therapists on: managing the stutter rather than «cure it» — motivation to treatment — discovering stuttering symptoms and characteristics and how it manifests itself differently in each of us — communication skills — communicative interaction parent-child — learning techniques to control and manage the stuttering to be able to improve his/her communication — working on stuttering modification and fluency shaping — modelling slower speaking rates and pausing which allows the child to synchronize the components of spoken language — creating positive attitudes toward speech and stuttering;
- one parent session with the psychotherapist (one family at a time) where each family will have the chance to talk about concerns regarding their child's stuttering, to overcome the feelings of shame and guilty;
- final group session for parents held by ST and PSY. All the parents of children (aged from 4 to 16), who attended the courses can meet and share their worries, identify useful elements that they have started to change to support their children and themselves in the process of management of stuttering. In this session we highlight the importance of motivation as the basis for the efficacy of treatment and we teach parents some useful strategies for working with praise and reinforcement in according to Behavior Therapy principals. These strategies are important for transferring what they have learned in therapy session in everyday life;
- one briefing at schools of the children who attended the courses to give advice for managing a child with stuttering.

The second part consists of working with the Rational Emotive Behavior Therapy (REBT) on change maladaptive emotions that the CWS feels related to stutter events. The REBT is a cognitive behavioral approach to psychotherapy developed by Albert Ellis (1962; 1994). We incorporated a three-term REBT's model (A-B-C) for the analysis of behavior and emotion. According to Albert Ellis emotional upset occurs when people attempt to fulfill their goals and encounter an activating event (A) that blocks the goal. In turn,

people have beliefs (B) about this activating event which influences how they feel and act (C). Behavior and emotion are consequences of an interpretation of (B) about some prior activating event (A). Thus, the (A) does not create the feeling (C), but beliefs about the event (B) contribute to the emotional disturbance. If the behavior is irrational, it must be due to faulty thinking. Irrational beliefs (Ib) represent demanding and unrealistic perceptions of how things should be, statements of blame directed at self and others, «awfulizing», «poor self-worth» statements that reflect an exaggeration of the event and the inability to tolerate frustration. A therapist challenges, questions, and disputes (D) the Ib. Leads the CWS to recognize irrational thought and to self-dispute. At the same time Problem solving strategy are taught to deal with teasing and bullying. Self-acceptance («I am ok although I stutter») and learn to feel health emotions are the goals of the REBT therapy session.

Pre treatment data

For this qualitative study we collected data from a group of 31 children who arrived at the Studio Multicodex where the ABC BALBUZIE® program is held, aged 7 to 11 (22 males and 9 females), that attended the PTC with their parents. The parameters considered to evaluate the effectiveness of the therapeutic approach for a qualitative comparison pre-treatment and a 6 month later follow-up are: the severity of stuttering, speech associated attitude, information about stuttering and its causes and management of stuttering (children and parents), management in school, parents expectations on recovering and treatment, parents worries.

Stuttering severity: 11 were mild, 20 moderate; 22 children with negative attitude; 24 children did not know why they stutter, 7 thought it was due to anxiety or psychological factors; almost all of them on the arrival of dysfluencies use strategy such as «stop and try again», «push the sound out», «take a deep breath and keep calm», «avoiding behavior», 19 children have experienced teasing, 15 children state that their parents and teachers do not like the way they talk because they give advice to not stutter.

Most of the parents thought the stutter was due to emotional factors although there was a family history of stuttering, they are concerned about

teasing and that stuttering may have an impact on their children's social and school life; 3 families state emotional causes and «so many things to say very quickly». All parents were convinced that working only on stuttering modification techniques «remove stuttering» and giving their children advice such as «slow down, stay calm, take a deep breath» or correcting them while stutter could have been helpful. Many of them had never talked about stuttering with their children in a straightforward manner.

Conclusions

The evaluation during the follow-up gave us these data: parents acquired more and specific information about the etiology of stuttering, dispelling wrong beliefs, became more aware and active toward realistic recovering expectations, motivating and facilitating verbal fluency in their children. They also found the chance very useful to share their feelings about stuttering with specialists (ST and PSY) and other parents during group sessions, were also becoming less focused on the symptom stuttering, recognizing the strength of their children in managing stuttering. We found out that the speech-associated attitude in the 22 children moved from negative to positive and it remained unchanged in the other 9. The severity of stuttering has not improved in all children but 12 of them went from moderate to mild. All the children had a better understanding of why you can stutter and could explain it to other children. Some made a speech in class to explain to classmates and teachers as be helped while stutter. They feel more secure to respond to teasing. Most of them have realized that they cannot have total control over the stuttering and tolerate more its variability. Our opinion is that the PTC gives a different opportunity to parents and children to deal together stuttering, guides them to a more functional and active way of managing stutter in everyday life, school included, and reducing stuttering symptoms and family stress. It had become very useful to involve as well parents and children together as a strong point of the program, to change their expectations and communicative behaviors.

We are aware of the limitations of this qualitative study and aware that this preliminary study will need further analysis.

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ISSUES TO CONSIDER WHEN WORKING WITH SCHOOL-AGED CHILDREN WHO STUTTER: FOR CLINICIANS

Susan L. Block⁴⁹

Introduction

Treating children who stutter should be a quick and easy process. However, it can often be long and complex, much to the frustration of the children, their parents and their clinicians. It is vital that we improve this situation as school-aged children become increasingly vulnerable to the devastating effects of developing chronic stuttering and all the implications that follow. Treatment in preschool children has ample and robust evidence demonstrating positive outcomes in relatively short timeframes. Similarly, treatment in adults has much evidence supporting positive fluency outcomes. Despite this, adults who stutter can also present with accompanying high levels of speech-related anxiety, which can impact the outcome of treatment and the maintenance of fluency. At what stage do children experience such negative impacts?

However, there are some very important considerations with school-aged children. They are increasingly aware of the stuttering, increasing adept at self-monitoring and self-evaluation and usually very motivated to stop stuttering. They experience increasing communication demands as they progress through school. Often they also have parents who are increasingly concerned about their child's communication problems and their consequent well-being. Parents also have to contend with the impact of the stuttering on the family — in a multitude of ways.

With scant, varying and conflicting information about the treatment of stuttering in school-aged children, clinicians can be confounded in choosing appropriate, evidence-based strategies. This can result in them being reticent to work with this age group. The complexity of co-ordinating children, parents, families and teachers can be overwhelming for many clinicians.

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It can also be problematic in clinical situations which can be bound by administrative regulations or restrictions that do not allow best practice or sufficient treatment.

One of the best things researchers can do for clinicians working with school aged children who stutter is to translate the research findings into clinical implications and strategies. We have been hampered at times by adherence to the golden model of RCTS, without acknowledging other levels of evidence. This other evidence may give clinicians some strategies and considerations to enhance fluency and treatment outcome in the absence of high level research evidence for this school-aged group.

Clinicians can also be hampered by their lack of awareness of the social context in which their school-aged clients now function. What are the social determinants of their lives? Do we address these in treatment? How can we meet the challenge of finding quicker, easier and more positive treatment outcomes?

Background

Making the decision to bring a child to treatment maybe one of the most difficult decisions some parents of a young child will make. Their precious and perfect child needs to be subjected to the influence of a clinician who is unknown to the parents, who is not known to the child and potentially who might make the stuttering worse — a parental nightmare. This puts significant responsibility on anyone working with children and we need to take this fact seriously. Parents need to feel confident in our ability to understand and like their child, they need to feel confident that their child will like us and enjoy their time with us and they need to trust our ability to improve the stuttered speech their child exhibits. At the very least, parents must be assured that we will take the time to get to know their child and that the treatment will be enjoyable and directed to the individual needs of their child. This involves gathering information about the family context and the social environment in which the child must function. However, most significantly it involves us working directly on the stuttering rather than being distracted by other issues that may result

from the child's experiences with stuttering. It is in fact for this reason that the most parents present their children for treatment in the first place. We need to honour this fact.

The issues

Parents

Preschool treatment routinely necessitates the involvement of parents at every stage of the treatment process. However, once the child starts school, treatment often occurs at school and parents are not always included in the process. As a result, transfer or generalisation are difficult potentially little carryover to home and situations outside the school. This incurs the risk that parents may think treatment has little affect. While school-based treatment is often provided as a support for teachers to enhance the education process, children are ultimately the responsibility of their parents. We need to let parents assume that responsibility in the treatment process. Denying them access to the treatment sessions is counterproductive. It is also not fair to the child who is trying to manage the process of treatment alone. Further, stuttering has been demonstrated to have negative impacts on the parents and families of children who stutter (Erickson et al, 2013). Engaging parents in the treatment process with older children can be empowering for parents as well as a relief as they assume part of the responsibility for the progress of their child in the treatment process.

Evidence

In his tutorial in 2008, Onslow reported that the clinical trials that met his definition of a true clinical trial reported some results for children aged 7-12 years using speech restructuring, regulated breathing, EMG Biofeedback and Verbal Response Contingent Stimulation, but varied effects were reported. Similarly, reports of the same procedures were reported for children in the 13-17-year-age group. However, poorer results were reported

for EMG biofeedback. Using speech restructuring and EMG feedback with children has the potential to affect speech naturalness. They are also complex treatments which require careful and consistent focus by children and significant support from their parents.

There is now ample evidence for the success of the Lidcombe Program with pre-schoolers and young primary school-aged children. There is also emerging evidence for group delivered Lidcombe (Arnott et al., 2014) and skype-delivery (Bridgman et al., 2014). However this evidence does not extend to older children. Similarly, there is also emerging evidence for the use of Syllable-timed speech with preschool children (Trajovski et al., 2009), but this evidence too does not extend yet to older children. There is ample evidence for speech restructuring, both intensively and using the Camperdown Program with adolescents and adults (e.g. Block, 2012; Block et al., 2005; Carey et al., 2009; 2014). However, again, this evidence generally does not extend to primary school-aged children.

Dual tasking has been demonstrated to be an issue with adults who stutter (Metten et al., 2010). As a result, it is being incorporated into some speech restructuring programs with adults in an attempt to facilitate generalisation of fluency in particular. Utilising dual tasking with school-aged children may also be beneficial to ensuring stability of fluency in older children in particular.

The use of video self-modelling has been reported by Cream et al. (2010). While it has not been trialled with young children, it is possible that children would benefit from viewing themselves performing the target behaviours, which in this case would be stutter-free speech.

Mental Health is a significant issue for many adults who stutter (Iverach, Jones et al., 2009; Iverach, O'Brian et al., 2009) with evidence indicating anxiety and mood disorders can result from growing up stuttering. Similarly, Gunn et al. (2014) have reported concerning results for adolescents who stutter. Alternatively, Kefalianos et al. (2012; 2014) provide data indicating that stuttering children younger than 4 years of age do not present with mental health attributes any different from normally fluent children. Similar results are evident in children up to 7 years old. Thus, it is incumbent upon clinicians to rectify stuttered speech before children reach the stage of developing negative mental health effects. These effects can also impact the parents and families of children who stutter (Erickson et al., 2013).

Goals of treatment

Fluency

In presenting their precious children for therapy, parents of children who stutter expect a focus on fluency (Erickson et al., 2013). They may have to manage the reactions of their child to teasing and bullying as a result of stuttering. They may have to manage school refusal that can be the result of bullying. They may have to manage avoidance of school and other activities, such as participation in oral activities in class, team membership, assuming leadership roles, withdrawal of social interactions etc. They may have to manage numerous restrictions on participation and activity. All of these factors must be managed at the same time as there are increasing communication demands in the classroom. While their fluent peers are increasing in their communication prowess, the child who is stuttering is often decreasing in both their communication effectiveness and their enthusiasm to communicate. While it is important to support children who are stuttering and to help them maintain a positive outlook, it is vital to increase their fluency because of the very concerning implications if stuttering continues.

Well-being

With increasing age comes increased susceptibility to the development of anxiety. Gunn et al (2013) reported that 38% of their teenage sample achieved at least one diagnosis of a mental health disorder, usually involving anxiety. Although many of the scores on psychological measures fell within the normal range, older adolescents (15-17 years) reported significantly higher anxiety, depression, reactions to stuttering, and emotional/behavioral problems, than younger adolescents (12-14 years). To address this situation, researchers at the Australian Stuttering Research Centre have developed a self-administered Cognitive Behaviour Therapy web-based program, initially designed for adults and now for adolescents (Helgadottir et al., 2014). This is available for adolescents who need cognitive restructuring whilst they are

also working on their fluency. Indeed, reducing anxiety has been shown to decrease factors such as the fear of negative evaluation and enhance well-being, each of which may influence the use of fluency strategies the child has learned.

General communication and socialisation

Much has been written about the language abilities of children who stutter. Most recently, Watts et al. (2015) demonstrated that children aged up to 7 years who stutter do not have language abilities that are inferior to those of their fluent peers. Thus, again, the evidence suggests that it is more appropriate to focus on fluency strategies rather than enhancing language ability. Nevertheless, if a child has spent years avoiding talking, it may be the case that they are not well practised at general social skills such as conversation starters, topic maintenance, responsible turn-taking etc. as well as other pragmatic aspects of interaction.

An advantage of working with school-aged children who stutter is that they usually have a social environment with adults who are able to support them in the treatment process. Parents and family members know the communication environment in which they need to function; teachers know the communication environment in which they are learning and socialising; other adults such as sports coaches or specialist teachers know the environment and language they need to use as they participate in sport, music, arts activities etc. These adults also can be vital supports for both fluency and well-being during the child's journey to fluency.

Conclusion

Despite a lack of high level evidence for treatment efficacy with school-aged children, we can make sensible clinical decisions based on research findings that certainly relate to school-aged children. We could reasonably decide to incorporate aspects of the Lidcombe Program, or at the very least, some aspects of Verbal Response Contingent Stimulation.

The use of Syllable-Timed Speech may be an appropriate decision for some children. Using aspects of Prolonged Speech would be appropriate for some children. Dual tasking or video self-modelling may be sensible additions to enhance generalisation of fluency. The addition of strategies for managing anxiety or other well-being impacts would be sensible, not only to improve the child's quality of life, but also to maximise the likelihood of long-term fluency maintenance.

Working with children is rewarding and important. It is important because of the ramifications of continuing through adolescence stuttering. It is a huge responsibility because of the expectations of parents and the responsibility they hand to us. We need to honour that responsibility by utilising current evidence and altering the communication future of the children with whom we work. We can change their lives and we need to do so by focussing on fluency as a priority. We have no more important task.

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A COMMON FACTORS APPROACH TO TREATING CHILDHOOD STUTTERING

Ellen M. Kelly PhD, CCC-SLP ⁵⁰

Abstract

Early intervention for stuttering is intended to help avert the potentially adverse, life-long impact of persistent stuttering on communication, social, academic and vocational achievements and to help parents understand how best to help their young children who are stuttering. Divergent parent-based treatment approaches appear to be similarly successful in reducing stuttering in young children, doing so more rapidly than without formal intervention in many cases, yet mirroring natural recovery rates. What, then, do these findings tell us about with whom it is best to intervene, when, why, and how? In this presentation, we will build a framework for addressing these questions by: (1) examining what we presently know about (a) children whose stuttering persists versus those who recover and (b) treatment for early childhood stuttering, (2) contrasting medical and contextual models of intervention, (3) identifying common elements across treatment approaches, in general, and for early stuttering, in particular, and, (4) using the common elements to propose an integration of medical and contextual approaches to inform future research and intervention in early childhood stuttering.

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Session 8

Psychology and wellness of the person who stutters

PSYCHOSOCIAL ASPECTS OF STUTTERING

Jan McAllister⁵¹

Abstract

Much recent research in the field of stuttering has focused on its psychosocial aspect, that is, the way that perceptual, cognitive, emotional and biological factors in the individual interact with social and physical factors in the environment. Although much of this research has highlighted social anxiety disorder in people who stutter, stuttering is also associated with other forms of anxiety, as well diminished psychological wellbeing across various other domains. This paper outlines current research into the psychosocial factors associated with stuttering from the preschool years to adulthood.

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Introduction

The *Oxford English Dictionary* defines «psychosocial» as «pertaining to the influence of social factors on an individual's mind or behaviour, and to the interrelation of behavioural and social factors». «Psychosocial» is an umbrella term that refers to an interaction between perceptual, cognitive, emotional and biological factors in the individual with social and physical factors in the environment. Psychosocial factors are therefore pertinent to a diverse range of phenomena, from mental health disorders like depression and anxiety, through quality of life, self-identity, self-stigma, self-esteem and self-efficacy, to engagement with others in interpersonal relationships and activities such as education and employment.

The psychosocial dimension of stuttering has long been recognised, and clinical guidelines for speech and language therapists (e.g. Royal College of Speech and Language Therapists, 2005) acknowledge the importance of this aspect of the condition.

Psychosocial factors and stuttering

The association between stuttering and social anxiety disorder in adults has been well documented (for a recent review, see Iverach and Rapee, 2014). The *Diagnostic and statistical manual of mental disorders 5th ed. (DSM-5)* (APA, 2013) defines social anxiety disorder as a persistent fear (i.e. of at least 6 months' duration) of one or more social or performance situations in which the person is exposed to unfamiliar people or to possible scrutiny by others, where the person fears that their behaviour will be embarrassing or humiliating. Fear of negative evaluation by others and avoidance of the situations that cause anxiety are characteristic of social anxiety disorder. *DSM-5* specifies that the anxiety, fear or avoidance must be unrelated or out of proportion to any other medical condition that is present (including stuttering, which is one of the examples cited).

A well-known cognitive model of social anxiety disorder (Clark, 2001) focuses on the way that information is processed in social situations. According to the model, people with high levels of social anxiety develop

three assumptions based on prior experience: excessively high standards for their own social performance, maladaptive beliefs about what will happen if they perform in a certain way, and unconditional negative beliefs about themselves. For example, a socially anxious person who stutters might believe that only fluent speech can be tolerated, that others will think them stupid if they stutter, and that the fact that they stutter means that no-one could like them. Because of these assumptions, social situations are seen as a source of threat. The anxious person then enters a vicious cycle in which they adopt safety behaviours such as avoiding speaking, avoiding looking at the other people in the situation, or indeed withdrawing completely from the situation. Paradoxically, adopting such behaviours prevents the person from gathering information that would disconfirm their maladaptive assumptions: for example, by suppressing speech they do not give others the opportunity to react neutrally to their dysfluency, and by avoiding looking at their interlocutor they fail to notice when they are being given non-negative social signals. Lowe, Guastella, Chen, Menzies, Packman, O'Brian and Onslow (2012) used eye movement technology to investigate the way that adults who stutter appraise a social situation in which they are involved (in this case, via a TV screen). Compared with controls, participants who stuttered looked less at the TV display overall and, in particular, spent less time looking at the positive facial expressions than other expressions or the background of the TV display. Attentional biases of this kind are characteristic of social anxiety disorder. During the early stages of processing, socially anxious individuals are hypervigilant for sources of threat (e.g., disapproving facial expressions), but once identified, they direct attention away from them (Pishyar, Harris and Menzies, 2004). McAllister Kelman and Millard (2015) found that 8- to 18-year-olds who stuttered and were socially anxious were biased towards schematic faces displaying negative expressions, unlike their non-anxious peers who stuttered.

Messenger, Packman, Onslow, Menzies and O'Brian (2015) noted that the literature regarding the emergence of social anxiety among children and adolescents who stutter has delivered inconsistent results. In their own study, they used a measure that can pinpoint the onset of social anxiety. Contrary to predictions, they found that scores for the 6-18-year-olds who stuttered fell within normal limits. They suggested that a possible reason for

this null finding might be that the boys in the sample were concealing their true levels of anxiety in order to present themselves in a more positive light. This explanation was consistent with their data. However, when McAllister et al (2015) gathered data from both parents and 8- to 18-year-olds who stuttered on a similar measure of the latter's anxiety, they found a significant correlation ($p < .001$) between mothers' and clients' responses on the social anxiety scale, suggesting that the clients' responses were accurate. Clearly this issue requires further investigation.

Social anxiety is not the only form of anxiety that has been documented in people who stutter. Craig and Tran (2014) carried out a meta-analysis which indicated that adults with chronic stuttering have elevated levels of both trait and social anxiety. However, Craig and Hancock (1996) found that 9-14-year-olds who stuttered did not differ from controls on measures of state or trait anxiety. Children and adolescents who stutter may experience elevated levels of other kinds of anxiety. McAllister et al (2015) screened for generalized anxiety disorder, separation anxiety, panic, social anxiety disorder and school avoidance in 8- to 18-year-olds attending pre-treatment sessions for stuttering. Clinically significant levels of anxiety were 3-4 times more common in their participants than in the general population. Overall, anxiety increased with age. Generalized anxiety disorder, panic, social anxiety disorder and school avoidance were all more common in older than younger participants; as expected, only separation anxiety was more common in younger than in older participants.

Anxiety is just one of several areas in which adults who stutter experience decreased psychological wellbeing. Craig, Blumgart and Tran (2015) used a measure of mood states that covered 9 domains including obsessive-compulsiveness, depressive mood, paranoid ideation and psychoticism as well as anxiety. Comparing the results with community norms, they found that adults with chronic stuttering fared significantly worse on all nine mood scales. They also found that self-efficacy, a person's expectation of being able to function effectively in the present and in the future, could protect against the impact of negative mood states such as anxiety and depression.

It was suggested above that psychosocial factors can have an important impact on individuals' participation in activities such as education and employment. Some people who stutter feel that their speech has had a

negative effect on their own life chances in such areas (e.g. Crichton Smith, 2002; Klompas and Ross, 2004), but this is not necessarily the case for people who stutter as a group (McAllister, Collier and Shepstone, 2012; Tran, Blumgart and Craig, 2012). It is clinically and theoretically important to determine the processes that lead to restricted participation. Corrigan, Larson and Rusch (2009) confer a key role in this process to the concept of self-stigma and the so-called «why try» effect. According to their model, self-stigma consists of three stages. First, the individual becomes aware of a stereotype (for example, a person who stutters becomes aware that some members of the public hold a negative view of people who stutter); next, they agree with it; finally, they apply it to themselves. Doing so leads to a reduction of self-esteem and self-efficacy, resulting in a sense of futility («why try») which deters them from pursuing activities that could enhance achievement and therefore help them to challenge their adopted stereotype. Boyle (2015) applied this model to stuttering. He found that 84% of his sample of 354 adults who stutter reported moderate to high levels of awareness of negative societal attitudes to stuttering and nearly a third applied the stigma to themselves. Self-stigma was positively associated with anxiety and depression and negatively associated with hope, empowerment, quality of life and social support.

The origins of psychosocial difficulties

Although the onset of social anxiety disorder is usually reported as typically occurring in late childhood or early adolescence, around half of the sample in a large epidemiological study (Schneier, Johnson, Hornig, Liebowitz and Weissman, 1992) reported experiencing the condition from a far earlier age. Hudson and Rapee (2000) explored the origins of social anxiety disorder and a range of related conditions including social anxiety, embarrassment, shyness, self-consciousness, social isolation, social withdrawal, audience sensitivity, and peer neglect. The studies that they reviewed were consistent with an early onset of some of these conditions: for example, children as young as 2 years old could be sensitive to negative evaluation by others and could experience embarrassment, and most 3-year-olds were

capable of doing so. Their review implicated various factors associated with the emergence of social anxiety disorder and related conditions: genetics, possibly in the form of an inherited predisposition towards anxiousness, or of social anxiety disorder specifically; an overly controlling parenting style; childhood illness; social isolation; being a firstborn or only child; and traumatic social experiences such as being bullied or teased by peers. The authors emphasised the importance of looking at children's early behaviour and temperament as antecedents of these later psychosocial conditions.

Temperament refers to the individual's genetically-endowed tendencies to react to and interact with the environment in particular ways (Buss and Plomin, 1984). Frameworks for describing temperament vary in terms of the number and nature of constructs that they employ, but of particular relevance here is behavioural inhibition, that is, avoidance, withdrawal, wariness and shyness with strangers and in novel situations (Hudson and Rapee, 2000). Children whose behavioural inhibition is maintained over a period of three years are more at risk than their peers of developing anxiety disorders, including avoidant disorder (Biederman et al., 1993, cited by Hudson and Rapee, 2000). The relationship between stuttering and temperament has been the subject of several reviews (Alm, 2014; Conture, Kelly and Walden, 2013; Kefalianos, Onslow, Block, Menzies and Reilly, 2012) but these have reported that the findings of studies are inconclusive.

In a systematic review of risk factors for anxiety in children and adolescents who stutter, Smith, Iverach, O'Brian, Kefalianos and Reilly (2014) reported that there was currently insufficient evidence to allow conclusive identification of the age of onset of such difficulties in this group. If this timing could be established, it might be possible to plan for the provision of preventative interventions. Hudson and Rapee (2000) have argued that it is just as important to pinpoint the emergence of behavioural problems, as these may be antecedents of anxiety disorders and other mental health difficulties. Given the association between negative peer reactions and behavioural difficulties (Hudson and Rapee, 2000), findings of negative peer attitudes to stuttering even among pre-schoolers (e.g Langevin, Pachman and Onslow, 2009; Weidner, St Louis, Burgess and LeMasters, 2015) might predict that behavioural difficulties could begin to emerge as early as the pre-school years. Contrary to this prediction, in a prospective community-based study of 4-year-

olds, Reilly, Onslow, Packman, Cini, Ukoumune et al. (2013) reported that children who stuttered did not differ significantly from their typically-developing peers on any aspect of the Strengths and Difficulties Questionnaire (SDQ), a measure of behavioural, emotional and social development. The findings were controversial and resulted in commentaries by many researchers and clinicians in the field (e.g. Bernstein Ratner, 2014; Donaher and Kelly, 2013; Howell, 2009; Yairi, 2014). However, a larger community-based study which also used the SDQ (McAllister, submitted) found that 3-year-olds who stuttered had higher levels of difficulty than their typically-developing peers, especially on scales relating to behavioural difficulties; among 11-year-olds, children who stuttered performed more poorly across all SDQ difficulty scales (behaviour, hyperactivity, emotional and peer difficulties).

Addressing psychosocial aspects of stuttering

A number of studies cited here have demonstrated an association between stuttering and mental health problems, including depression and various forms of anxiety. Cognitive behavioural therapy (CBT) (Beck, 2011) has been effective in addressing such problems in the wider population, and this approach has been shown to alleviate social anxiety in adults who stutter, both in its traditional face-to-face mode of delivery (Menzies, O'Brian, Onslow, Packman, St Clare and Block, 2008), and in an on-line form (Helgadóttir, Menzies, Onslow, Packman and O'Brian, 2014).

Although CBT is an effective treatment for social anxiety disorder in stuttering, both Menzies et al. (2008) and Helgadóttir et al. (2014) reported that it did not improve stuttering frequency. An alternative to CBT is Cognitive Bias Modification (CBM), which implicitly re-trains the maladaptive attentional biases that underpin social anxiety disorder (e.g. Schmidt, Richey, Buckner and Timpano, 2009) and is also an effective intervention for the disorder. It is possible that the unconscious mechanism used in CBM may permit improvements to fluency as well as psychological health. A possible advantage to the CBM approach is that it may be able to be used as a preventative intervention for psychological disorders (Woud, Postma, Holmes and Mackintosh, 2013).

Other recently-published studies suggest that alternative approaches may be effective. Craig et al. (2015) study indicates that self-efficacy is a protective factors when adults who stutter encounter negative mood, and clinicians need to be aware of this finding and deliver appropriate interventions for clients with poor levels of self-efficacy. Boyle's (2015) finding of issues around self-stigma in adults who stutter also suggests features that could be addressed in therapy.

Behavioural problems may be precursors of serious mental health difficulties (Hudson and Rapee, 2000). Pre-schoolers who stutter may be more likely to develop such behavioural problems (McAllister, submitted). Clinicians who work with pre-schoolers who stutter need to be aware of the possible developmental trajectory associated with such behavioural difficulties, and provide appropriate interventions and advice to parents, including addressing issues such as parenting style (Hudson and Rapee, 2000).

Conclusion

Psychosocial factors are theoretically and clinically relevant to stuttering in children, adolescents and adults. People who stutter are more susceptible than others to various kinds of psychosocial problem including low mood. The precursors of serious mental health issues may be apparent in children who stutter at an earlier age than their typically-developing peers. Factors that mediate poor psychosocial outcomes for people who stutter, such as self-stigma, and those that offer protection against such outcomes, such as self-efficacy, could be used to develop useful interventions for people who stutter.

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Session 9

Treating young and adults who stutter

PROLONGED SPEECH INTENSIVE TREATMENT MODEL FOR ADULTS WHO STUTTER: A STUDENTS' TRAINING DELIVERY

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Abstract

Background

It has been suggested that to increase the clinicians' (master degree students) competences in Bulgaria is necessary to update the Speech Language Pathology (Logopedics) educational curriculum in terms of clinical training with clients who stutter, and to provide relevant clinical training during the academic preparation (Georgieva, 2012).

This study describes adults who stutter (AWS) intensive treatment (IT) that was conducted by students in the SLP master degree programme: «Logopedics Management in Fluency and Voice Disorders».

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Objective

To increase the quality of the academic Speech Language Pathology (SLP) training of master degree students through application of an intensive treatment programme for adults who stutter (AWS). A very important part of that clinical educational experience is to apply fluency shaping approach through prolonged speech technique introduction. The aim of the present research was to establish speech fluency outcomes for the IT and to prove that the master degree programme meets the good IALP and CPLOL standards for successful students' training. This study is the first and unique trial of such intensive treatment at university level in Bulgaria and follows the Australian leading experience of the La Trobe University speech clinic.

Methods and procedure

The IT was conducted by eleven SLP students, under supervision, during their master degree training. 12 adults participated and were treated in the Prolonged speech model. The treatment was a traditional intensive speech restructuring known as Prolonged (Smooth) speech. Speech fluency assessment was conducted 1 week and 1 day before the beginning of the IT programme. The post-treatment evaluation was performed immediately after the intensive programme application.

Results: Outcomes were reported for an IT programme delivered by master degree students in SLP. Primary outcomes were measured according to (i) per cent syllables stuttered (%SS) within and outside university stuttering research center, (ii) speech naturalness score evaluation from pre- and immediately after the intensive treatment stutter-free speech samples, and (iii) self-report inventory scores.

Conclusions

The IT offered to AWS by master degree students during their professional clinical preparation was effective. The master degree students felt

more confident to deliver such a logopedical care for AWS. They produced objective speech fluency data and statistically important differences before and immediately after the IT regarding %SS and speech naturalness.

Keywords: stuttering, adult, treatment, intensive, student.

Introduction

A total of 150 questionnaires was sent to 3 different universities and 107 returned. One of the conclusions was that most of the Bulgarian students in the SLP BA programme observed an average of 3 stutterers throughout their study. This number increased to 5 up to 10 clients during the specialized master degree course, which indicated that observation and especially direct clinical experience were seriously emphasized (Georgieva, 2012).

Future research should be oriented toward projects for program effectiveness and the evaluation and elaboration of cost-effective diagnostic tools (Georgieva et al., 2014).

Objective

The *specific purpose* of this study was to find if an improvement was achieved during the IT delivered by master degree students. An *additional purpose* was to obtain information about AWS' perceptions of their speech behavior after completing the IT using Participant Self-Report Inventory.

Treatment programme

The research design of the present study was tailored with the special support of Dr. Susan Block. La Trobe University intensive programme of prolonged speech was implemented (Block and Dacakis, 2003). The treatment applied was an adapted Bulgarian model of La Trobe Intensive Program: student delivered treatment for adults. There were three treatment stages: instatement stage, transfer stage and maintenance stage (see the Table 1).

The reason to select the prolonged speech model in intensive format was that speech restructuring treatments have proved to be very efficient in adults who stutter (Boberg and Kully, 1994; Onslow et al., 1996; O' Brian et al., 2003; Block, Onslow, Packman, et al., 2005; Cocomazzo, Block, Carey, et al. (2012).

TABLE 1
The Intensive Treatment (IT) prolonged speech stages and their description

	Instatement stage	Transfer stage	Maintenance stage
Total duration: 54 h., 8 weeks	3 days, 8 hours each day	4 th and 5 th days, 8 hours each day	7 Follow-up sessions, conducted for 7 weeks, 2 hours weekly
Stages description	Prolonged speech Focus on speech naturalness Gentle onsets Sliding in Elongating vowels Linking words together	Hierarchy of speaking situations constructed by client prior to treatment Situations are different for each PWS (from five to sixteen steps in these hierarchies)	The clients discuss setting tasks they would like to practice between each session. The focus is on using their strategies all the time. Self management strategies taught: Self monitoring and Self evaluation Parents and friends involved
Forms of the treatment	Intensive and spaced Individual and group treatment	Intensive and spaced Individual and group treatment	Individual (2h) and group opportunities (2h). Could be different and is tailored to the client's needs.

Method

The essential method of treatment was a clinical modeling pattern.

Participants

Participants in the present study were 12 AWS: 10 males and 2 females with an age range of 18-29 years (mean 22.5 years).

Students involvement

Eleven students were enrolled in the present trial: 9 master degree students were in the final year of their graduate course in SLP programme, 2 were in the last year of their undergraduate training in SLP. A PhD student was also involved in this IT format. Prior to participation in the IT programme, all students had completed (i) a 60 hour BA level fluency disorders course, and (ii) 60 hour MA degree course for stuttering management. Before the participation in the IT course, all students received a 40 page copy of the treatment materials in Bulgarian developed by the PhD student.

Outcome measures

The speech outcome measures were: per cent syllables stuttered (%SS), speech naturalness, and self-report inventory scores.

Per cent syllables stuttered (%SS)

Stuttering was assessed as a percentage of syllable stuttered using the following model:

$$\frac{\text{Total SS}^*}{\text{Total number of syllables}} \cdot 100 = \%SS$$

*SS = syllable stuttered

Speech naturalness

A 9 degree scale, developed by the Australian Stuttering Research Center, was used. The higher scores indicated more unnatural speech. Speech naturalness was judged from 15-second audio taped speech segments, extracted from the pre-treatment clinic assessment and immediately after IT clinic assessment.

Participant Self-Report Inventory (PSRI)

The 20 items self-report inventory developed by O' Brian, Onslow, Cream, et al. (2003) was filled out by the participants in the two cohort study.

Reliability

Interrater agreement for %SS was assessed by randomly selecting 12 speech samples from the two observers – three pre-treatment and three post-treatment samples for an observer.

Data analysis

A nonparametric Friedman test was chosen for %SS interpretation. For the NAT analysis a paired *t*-test was selected.

Results

TABLE 2
Mean and standard deviations (SD) for percentage of syllables stuttered (%SS) and naturalness (NAT) scores at each assessment

Measure	Situations	1week pre-treatment	1 day pre-treatment	Immediately after the treatment
Mean %SS (SD)	Monologue-clinic	11.9 (1.38)	12.4 (1.33)	2.6 (0.5)
	Conversation-clinic	10.8 (1.37)	11.4 (1.4)	2.5 (0.9)
	Telephone-home	9.8 (1.19)	10.3 (1.22)	1.4 (0.69)
	Conversation-home	7.3 (1.21)	8.1 (1.22)	0.9 (0.72)
Mean NAT	Monologue		6 (1.044)	1.58 (0.51)

Regarding the %SS the results revealed that the two cohorts of AWS experienced a sharp decrease in %SS from pre- to post-treatment period. Friedman analyses was significant ($p < 0.05$) comparing 1 week and 1 day pre-treatment and immediately after the IT data regarding:

- monologue within clinic ($\chi^2 = 24$, d.f. = 2, $p = 0.000$)
- conversation in clinic ($\chi^2 = 12$, d.f. = 1, $p = 0.001$)
- telephone situation home ($\chi^2 = 12$, d.f. = 1, $p = 0.001$)
- home conversation ($\chi^2 = 12$, d.f. = 1, $p = 0.001$).

The mean naturalness score was 6 at pre-treatment period and 1.58 immediately after the IT. The Friedman analyses ($\chi^2 = 12$, d.f. = 1, $p = 0.001$) as well as the Wilcoxon signed rank test analyses ($Z = -3.082$; $p = 0.002$) showed significant change ($p < 0.05$).

As another measure of treatment outcome all the participants in the study completed Self-Report Inventory (SRI) items immediately after the third phase of IT.

The first, second and third SRI questions were filled in one day before and immediately after the treatment. Before the IT all the subjects reported they have severe stuttering (Q1 average score 6), rated their speech on an average of 6 score (Q2) and were dissatisfied with their present level of fluency (Q3 score 4). Immediately after the IT the subjects reported remarkable changes in the registered scores of the same Q1, Q2 and Q3 – respectively 1.5, 5.7 and 1.1., i.e. decreased severity of stuttering, increased fluency pattern including established naturalness as one of the essential components of the prosody.

Discussion

This was the first study in Bulgaria to report a short term outcome for stuttering intensive treatment conducted by master degree students. It was also the first study in the country that measured results from an IT model using a prolonged speech technique with AWS. The results showed that the student delivery treatment can ensure *objective* speech outcomes.

It was encouraging and stimulating that the master degree students reported feeling well prepared, both theoretically and practically, to offer

an efficient treatment for AWS. The student involvement in the adapted Bulgarian model of La Trobe University programme of prolonged speech facilitated evidence-based learning in a university clinical setting. The successful combination of theoretical and practical learning was achieved.

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THE RATIONAL EMOTIVE EDUCATION IN STUTTERING SPEECH THERAPY

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Abstract

Objectives

Stuttering is a complex, multifactorial disorder, so speech therapists must often work not only on the fluency disorders but also on the attached negative emotions. Rational Emotive Education (REE) is recognized as an efficient psychoeducational method for promoting a harmonious affective growth of the individual in presence of difficulties. The aim of our study was to evaluate the effectiveness of a REE path in association with speech therapy on 15 stutterers of age 723.

Material and methods

The treatment consisted in 8 weekly sessions. The REE Questionnaire was used to measure patients' ability to cope with stress associated with stuttering before and after treatment.

Results and conclusions

85% of patients improved their REE score, with an overall significant increase ($p = 0.036$), witnessing the development of a more rational way of thinking and a greater selfacceptance.

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Introduction

Stuttering is a global, multidimensional disorder, in which alterations of the normal flow of speech may elicit a negative communicative attitude, with behavioral symptoms of avoidance, not only against words judged difficult to pronounce, but also against people and social situations (Bernardini S., Vanryckeghem M., Brutten G.J., Cocco L. and Zmarich C. 2009; Bleek B., Reuter M., Yaruss J.S., Cook S., Faber J. and Montag C. 2012; Blomgren, 2013). This scenario calls for a holistic approach, capable to integrate the speech therapy for the reduction of stuttering with training for the management of its emotional and behavioral aspects.

Rational Emotive Education (REE) (Knaus, 1974), stemmed from Rational Emotive Therapy (RET) (Ellis, 1957), has been recognized as a valid tool for helping people with different kinds of difficulties to cope with taunts or emotional distress. We hypothesized that such an approach could help also stuttering subjects to develop a positive communication attitude, making them capable to accept their limits and perform efficiently within them.

To the best of our knowledge, the literature does not report examples of REE applied to patients with speech disorders; we thus designed a trial to evaluate the outcome of a REE treatment complementary to speech therapy on subjects of different age, level of stuttering and communication attitude. This paper reports the protocol followed and the results obtained.

Rational Emotive Therapy (RET)

The basic premise of RET is that emotions and behaviors originate from cognitive processes, i.e. are the result of what people think or believe (about themselves, other people, the world in general), not of the situations they have actually to face.

In Ellis' «ABC» model (1962) «A» represents an event and the person's «inferences» as to what is happening; «B» represents the «evaluative» beliefs that follow from these inferences; «C» represents the emotions and behaviors that follow from those evaluative beliefs. The central proposition of the model is that the emotional and behavioral consequences (C) are

not caused directly by the activating event (A), but by the interpretation or belief (B) one has of that event.

Ellis defined two broad categories of thoughts: rational and irrational. The former involve the concrete assessment of reality, which allows resizing the emotional impact and facilitates the achievement of specific objectives. Irrational thoughts, on the contrary, are apt to leading to negative emotional reactions. There are four categories of irrational thoughts: i) «demandingness», which considers as absolute need what is instead only a preferable option, ii) «awfulizing», which evaluates an event as the worst possible, causing reactions of anxiety, iii) «frustration intolerance», which defines a situation as unbearable, magnifying the unpleasant aspects and generating anger or avoidance and iv) «global evaluation», which can lead to selfdowning (selfcontempt) or otherdowning (rejection) and/or lifedowning (general despair).

The educational principles and methods of RET are at the basis of the REE approach for teaching sane mental health concepts and the skills to use these concepts.

Materials and methods

Study population

The original test sample included 15 stuttering patients referred to the Speech Therapy Centre of the «AOU Città della Salute e della Scienza», in Turin, Italy, where they had already followed a treatment program of speech therapy to reduce stuttering. The program involved various techniques of verbal fluency control: Smooth Speech (Ingham, 1984), Easy Relaxed Approach–Smooth Treatment (ERASM) (Gregory, 1986), pullout, cancellation, in block and after block correction and preparatory set (Van Riper, 1973).

Trial protocol

Prior to the treatment, all patients were assessed for the entity of dysfluency by means of the SSI3 (Stuttering Severity Instrument) (Riley, 1994),

which is based on the percentage of repeated syllables: 02.5% corresponds to normal fluency, 35% to mild dysfluency, 610% to moderatetosevere dysfluency and $\geq 11\%$ to severe dysfluency.

Our trial protocol included eight weekly sessions in which the REE approach was to be applied for overcoming the disease connected with stuttering. Each session concentrated on training the patient to build critical thinking resources by identifying and challenging irrational thoughts, to discriminate between frustrating solutions aimed at changing the triggering event and more effective solutions concentrated at transforming the emotional reactions related to the event. The REE protocol ultimate goal was to automate this mode of rational thinking in all situations of everyday life, developing coping competencies. The training was mainly based on colloquia and reports of their everyday performance for adults, and pictures, drawings and games for children.

At the beginning and at the end of the treatment, patients underwent two evaluations. They were scored for their tendency to think and react rationally by the REE Questionnaire in the Italian version (Di Pietro, 1992) on a scale from 0 to 18. They were assessed for their communication attitude in an informal colloquium, where they were scored following the OASES (Overall Assessment of the Speaker's Experience of Stuttering) (Scott Yaruss and Quesal, 2010) classification as mild, mildtomoderate, moderate, moderatetosevere and severe.

At the end of the training sessions, patients were asked to express their level of satisfaction about the treatment filling in a Qualitative Questionnaire: the questionnaire was appositely drafted and included 7 questions with score from 0 to 5.

Data statistical analysis

The data analysis used the nonparametric Wilcoxon's test for the comparison of matched continuous prepost data. Correlation between distributions was studied by Pearson's linear correlation coefficient r . Dichotomic variables were studied with Fisher's test. Statistical significance was set at null hypothesis $p \leq 0.05$.

Results

During the trial two patients dropped out. Table 1 displays the baseline features of the 13 patients who completed the treatment: 23% females, 61.5% grammar school children, 23.1% teenagers, and 15.4% young adults; 46% diagnosed with a moderatetosevere dysfluency and 69% with a moderatetosevere communication attitude. The communication attitude seemed to be unrelated to the dysfluency level: 11/13 (85%) patients suffered from a moderatetosevere or severe attitude in spite of a mild or moderate dysfluency, whereas 2/13 (15%) patients showed a mildtomoderate attitude even with moderatetosevere dysfluency.

Figure 1 illustrates the pre and posttreatment scores obtained at the REE questionnaire: 11 out of 13 patients (85%) showed an improvement, with the overall REE score increasing significantly ($p = 0.036$) from $(10.1 \pm 3.1)/18$ to $(12.9 \pm 2.5)/18$. The percentage increase had a wide range of values: from 0% for two males, both with moderatetosevere dysfluency and communication attitude, to 250% of a male teenager with mild dysfluency but severe communication attitude. The patient's percentage increase seems

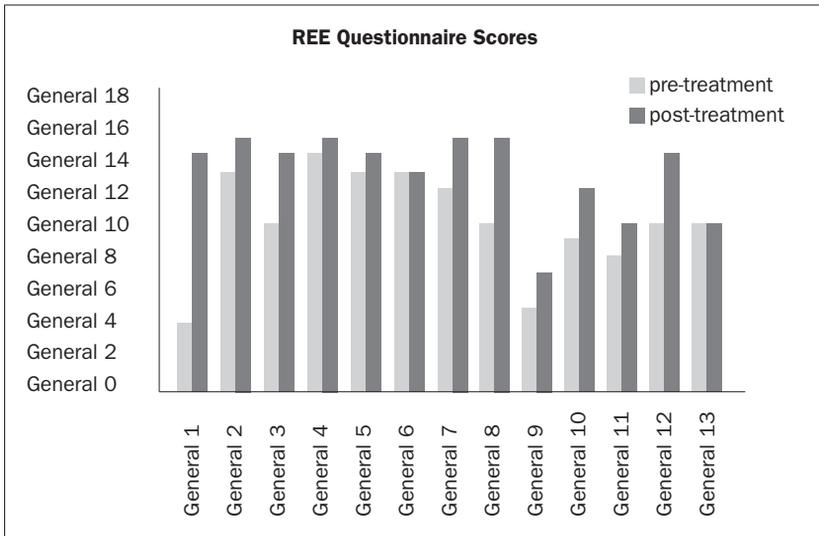


Fig. 1 Pre- and post-treatment scores at the REE questionnaire (maximum 18/18).

to be unaffected by gender, age, severity of dysfluency and communication attitude; however, the small sample sizes and the large variability among patients prevent any definite conclusion.

The post treatment communication attitude showed a definite improvement: the mild, mild to moderate and moderate subset increased from 4/13 to 12/13, while the moderate to severe and severe subset shrank from 9/13 to 1/13 ($p = 0.004$).

The answers to the Qualitative Questionnaire witnessed the high level of satisfaction of all participants to the treatment (31 ± 3)/35. The level of satisfaction was not correlated with their performance on the posttreatment REE questionnaire or their improvement.

Discussion

The efficacy of our REE treatment on stuttering subjects is proved by the significant improvement witnessed by both REE questionnaire score and communication attitude assessment after the treatment, a quantitative result that reflects our qualitative assessment on the behavioral change induced in the patients.

The mismatch between dysfluency level and communication difficulties observed before the treatment confirms that patients often have a perception of their disorder independent of the extent of the disorder itself. A negative perception (as observed in 85% of our patients) may actually lead to the creation of a psychological and emotional discomfort that speech therapy alone cannot counteract. At this purpose it is worth pointing out that the two patients who showed the greater improvement in their REE scores (50/50%) had originally a moderate or severe communication attitude in spite of their mild dysfluency, i.e., were patients with an emotive distress not commensurate to their physical disorder.

Conclusion

The program outcome in terms of patients' satisfaction, improved communication attitude and therapeutic success was remarkable. A trial

with a larger sample will be needed to confirm our results and search for relationship between the baseline features of patients and their proficiency.

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TABLE 1
Baseline features of patients who completed the trial.

<i>P</i> <i>t</i>	Gender	Age	Dysfluency (SSI3)	Communication attitude
1	m	teenager	mild	severe
2	m	teenager	moderatetosevere	mildtomoderate
3	m	teenager	mild	moderatetosevere
4	m	young adult	mild	moderatetosevere
5	m	child	mild	moderatetosevere
6	m	child	moderatetosevere	moderatetosevere
7	m	child	moderatetosevere	mildtomoderate
8	f	child	mild	moderatetosevere
9	m	child	moderatetosevere	moderate
10	f	child	mild	moderatetosevere
11	m	child	mild	moderate
12	f	child	moderatetosevere	severe
13	m	young adult	moderatetosevere	moderatetosevere

THE CAMPERDOWN PROGRAM AND STEPPED CARE FOR STUTTERING

Mark Onslow⁵⁵

Abstract

There are many who stutter who wish to obtain behavioural control of their stuttering. However, there are so many of such clients, and so few speech-language pathologists, that we have a pressing public health problem. This presentation overviews how the Camperdown Program has been adapted to a standalone Internet version, and how that Internet treatment can be a part of a stepped care approach to clients who stutter. It is suggested that such an approach can be part of the solution to the public health problem of stuttering.

Background

The current estimate is that the lifetime incidence of stuttering is 10 percent (Bloodstein & Bernstein Ratner, 2008). In other words, one in ten people will be affected at some time during their lives. How many of those who stutter and seek clinical help will benefit from being able to control its outward behavioural manifestations: the repetitions, fixed postures and extraneous behaviours that disrupt their attempts to speak?

Not all who stutter, it seems, but quite a few, can benefit from behavioural control of stuttering. You might think that assertion is self evident. I certainly do. But if you have any doubts, the claim is substantiated by research findings. In 2002, Scott Yaruss and colleagues reported a survey from the members of the National Stuttering Association in the United States (Yaruss, Quesal and Murphy, 2002). They found that 69% of participants reported that, and I quote, their “treatment involved learning techniques to help them speak as fluently as possible” (p. 120).

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We don't need to go into any elaborate health economics to make this point. In New York City with its population of 8.4 million people, there would be 170,000 adolescents and adults in the city affected by stuttering at any one time if we estimate the point prevalence there as 2% for that age range. According to the website of the American Speech Language Hearing Association, there are 21 speech language pathologists who treat stuttering in that city. That is one speech-language pathologist for every 8,000 people who stutter. That qualifies as a public health problem.

And since my challenge is to present something new here, there you have it. A public health problem plus the Internet, and there is something new for stuttering treatment: stepped care intervention for stuttering.

What is a stepped care health intervention? It is a simple idea that has been around for a while but to my knowledge has not been applied to health care for those who stutter, only mentioned once as a possibility by my colleague Rosemarie Hayhow (Hayhow, 2007). The stepped care model of healthcare delivery contains two fundamentals (Bower and Gilbody, 2005). It provides the simplest and most cost efficient method of health care that is efficacious. It is self-correcting so that clients progressively escalate to more resource intensive, and more costly, models of health care if they are shown to need it.

The final step in a stepped health care sequence for those who stutter and seek control of stuttering is reasonably self evident: it is standard care. We could argue a bit about whether that standard care should be a group treatment format or individual face-to-face treatment. To be sure, many of the clinical trials of stuttering control methods have been intensive, sometimes residential, one- or two-week treatment formats. However, I do not know of any health economic analysis that shows any benefit from that treatment format. And I certainly know of data to show that it is not necessary. But I am not here to talk about that issue.

What I am here to talk about is what the first step in a health care sequence might be for helping adolescents and adults control their stuttering, if that should be their clinical need. What about this, an Internet-driven treatment that requires no clinician and costs nothing? An Internet-driven treatment that can provide treatment to anyone provided that they have access to the Internet? A treatment that bypasses the need for regular face-

to-face access with a specialist stuttering clinician? Surely that could help us deal with this public health problem of too many who stutter and too few speech-language pathologists for them.

Can this be true? Yes, my colleague Shane Erickson has built such a website. It exists. There is clinical trial evidence to support the Camperdown Program (O'Brian, Carey, Lowe, Onslow, Packman and Cream, 2015), which includes a randomised controlled trial (Carey, O'Brian, Onslow, Block, Jones and Packman, 2010; in press). His website is based on the principles of the Camperdown Program. Shane has conducted and published nonrandomised clinical trials of this website method (Erickson, Block, Menzies, Onslow, O'Brian and Packman, 2012), and we are currently refining the website in preparation for further clinical trialing. This is an ideal first step in a stepped care approach to behavioural control of stuttering because it is better than economical; it costs nothing. The speech-language pathologist can simply direct the client to the website and treatment is self directed from there.

Based on the principles of the Camperdown Program, the standalone Internet treatment website has nine phases. Within the website, phases are only unlocked when participants have completed the goals for the previous phase. Participants are permitted to return to past phases if more practice at that level is required. Participants receive a reminder email after not logging in to the program for 1 week, and a subsequent email if they have not logged in for 2 weeks.

During Phase 1 clients learn to measure their stuttering severity with a 9-point scale. During Phases 2 to 5 they learn to imitate the Camperdown Program video training model, and evaluate how well they have done so. At this stage, their speech should not contain stuttering, but is unnatural sounding. Then, during Phase 6 they learn how to use a 9-point scale to measure speech naturalness. That scale is essential to the remainder of the treatment process as clients shape their speech toward being natural sounding and containing no stuttering. Then, during Phase 7 clients engage in the trademark Camperdown Program fluency cycles, where they practice the speech pattern, experiment with using it to control stuttering and sound natural, and then decide if the next step in the cycle is to do further practice imitating the model, or to do more experimenting with the fluency technique

to try to sound more natural while still controlling their stuttering. Then, during Phase 8, clients work through strategies to enable them to apply their fluency technique without stuttering and sounding natural during everyday speaking situations. Phase 9 is a standard maintenance phase.

For our recent nonrandomised trial (Erickson et al., in press) we recruited 20 adults. Two of our primary outcome measures were stuttering measured with percent syllables stuttered and quality of life measured with the Overall Assessment of the Speaker's Experience of Stuttering (OASES). The percent syllables stuttered measure was obtained by telephoning participants unexpectedly twice pre-treatment and twice six months later. We obtained the 6-month outcome data for 17 of our 20 participants. As is common with standalone Internet treatment sites, not all participants complied fully. Five of the 17 completed all nine treatment phases.

Using intention-to-treat analysis, including all participants, there was a modest Cohen's effect size of .24 from pre-treatment to follow-up, and a medium effect size of .53 for quality of life measured with the OASES. This change represents an improvement from "moderately severe" impact of stuttering to "moderate" impact.

However, in the context of stepped care, here is an important point. When we consider the five participants who completed all phases of the treatment, there was a large Cohen's effect size of .97 from pre-treatment to follow-up, and a large effect size of .96 for quality of life measured with the OASES. Two participants reduced their stuttering severity by more than 80%. What I particularly like about these results is that correlation analyses confirm the potential value of the website. For example, there was a moderate but significant correlation of .52 between the number of phases completed and OASES improvements.

So, what might be concluded so far from this research? Simply, that this nonrandomised trial gives a green light for randomised controlled trials of the procedure. We are currently refining the website and planning such trials. However, for now, it seems not going too far to argue that standalone Camperdown Program treatment may be a useful first step in a stepped care approach for some clients who need to control their stuttering to some extent during everyday life. Even taking account of the well-known limitations of nonrandomised trials as this one, it seems certain that many

clients will obtain significant clinical benefit from this website; perhaps even one quarter of them.

So, that would not be so bad, would it, if we could manage a quarter of clients who approached us needing to control their stuttering without having to spend any time or effort on them at all? If we could just send them to this website. And if it worked for them they would have it there at all times to help them maintain the benefits of their treatment; a treatment maintenance service that is completely cost free.

All well and good. But what about the others who do not obtain benefit from the standalone Camperdown Program website? That is where the stepped care approach escalates to a more costly treatment than our website.

Supposing you have a client who seeks to control stuttering and the website does not provide enough direction and support to achieve the desired outcomes? Obviously, that is going to happen. How often it happens remains to be seen after we have refined the website and done randomised controlled trials of it. But it will happen.

During our planned randomised controlled trials, things will complicate a little for our stepped care sequence. Let me return to that 2002 paper by Scott Yaruss and colleagues that I mentioned earlier (Yaruss et al., 2001). In addition, they reported that 49% participants said, I quote, their «treatment involved reducing the fear of stuttering or of speaking situations» (p. 120). Indeed, according to the best estimates available, somewhere between 40% and 60% of those who stutter will have so much fear of stuttering that they qualify for a diagnosis of social anxiety disorder.

I know of two cohorts that speak to the implications of this situation for control of stuttering. One of them was published by Lisa Iverach and colleagues in 2009 (Iverach, Jones, O'Brian, et al., 2009). Two thirds of her cohort of adults who sought treatment for stuttering had one of the mental health problems common among those who stutter, most of them related to anxiety. That group clearly was able to learn to control their stuttering, but showed clear evidence of losing that clinical gain six months later. In contrast, the one third of the cohort who had no mental health problems were easily able to maintain the benefits of their treatment and control their stuttering.

So obviously this issue complicates a stepped health care sequence for those who wish to control their stuttering, for many such clients will require anxiety management procedures as well as speech control procedures. We are currently dealing with that issue with development of our stepped care approach to stuttering, but let me take it out of the equation for now and just focus on what we are doing for that one-third of clients, or thereabouts, who require stuttering control and do not have any mental health issues to complicate things.

So back to the matter of when the first step in a health care sequence does not work for a client. No health care expenses have been directed at the client so far, so a next logical step would be to direct some minimal health care resources to the client. The next step could be that the standalone Internet treatment is supplemented by regular contact with a speech-language pathologist, say once per month. It ultimately will be an empirical question, but it does sound right to reason that there is evidence for some treatment effect from the standalone website, and if an individual client does not attain a treatment effect from it alone, then that client may attain a treatment effect if a clinician offers guidance and support on the journey through the site.

There is some empirical guidance here about how this might be done with nonrandomised and randomised clinical trials of the Camperdown Program with adolescents and adults who stutter using a telehealth approach. Our clinical trials have shown promise for high tech telehealth interventions using webcam (Carey, O'Brian, Onslow, Packman and Menzies, 2012), and low tech interventions using even a telephone (Carey, 2010). The results of the latter trial showed treatment effects to be the same, but the important issue is that the telehealth treatment version was a mean of 3.7 hours more cost efficient than the standard, in-clinic version of the treatment. With a mean treatment time per client of 11 hours, that represents a cost efficiency improvement of one-third, which is obviously worth having. There are additional economic cost benefits to society for a telehealth step in a health care sequence for stuttering. In my experience many who seek control of stuttering are young adults who do so for occupational reasons. Treatment by telehealth removes the economic cost of their travel time to and from the clinic. And of course,

in the case of adolescent clients, the burden of parents needing to take them to and from the clinic is removed.

So what then, if the Internet treatment, supplemented by webcam clinician contact once per month, does not do the trick? Well, the rest of my presentation is not rocket science. The next health care step for clients seeking stuttering control would be weekly webcam appointments, and for those clients who still fail to respond, there is the speech-language pathologist in person. A next step could be the webcam appointments supplemented by, say, one visit per month to the clinic to see the speech-language pathologist. And the final step, the most expensive one, for those who need it, weekly visits to the speech-language pathologist in the clinic. Or, if you see things differently to me, there is the intensive stuttering treatment option.

So where to here? Am I all talk? No, my colleagues and I have started doing this and we plan to finish, eventually, with randomised controlled trials of a stepped care approach to stuttering control compared to the standard methods that are tried and true in 23 clinical trials. This has been done for other health issues besides stuttering, such as smoking, alcohol abuse, obesity, and various mental health problems. We want it to be done also for stuttering.

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STUTTERING AND WEB-RADIO. A NEW ART-MEDIATED ACTIVITY

D. Tomaiuoli⁵⁶, M. De Meis⁵⁷, R. Siddi⁵⁸, E. Capparelli⁵⁹

Introduction

The effectiveness of a stuttering treatment can be primarily evaluated on the basis of the clients' ability to transfer in their everyday life what they learned during therapy (see among others Gregory, 1995). This means being able to maintain target fluency levels during verbalization using the tools and the learnt techniques; and this implies that they know, recognize and are able to manage their stuttering in both its overt and covert aspects (Conture and Curlee, 2007).

For long the international scientific literature has been showing (see among others Ingham and Onslow, 1987; Gregory, 2001) the difficulties people who stutter encounter to maintain therapy outcomes in the course of time. In fact, maintenance is often considered as the main drawback of the program with a client (Boberg, 1981). Even very brilliant results obtained by clients through therapy, often do not appear to be lasting in the long run, with a high relapse index (Craig and Hancock, 1995; Block, Onslow, Packman and Dacakis, 2006).

A high stuttering severity is often associated with the highest levels of stuttering regression (Craig, 1998; Gregory, 2001; Finn, 2003; Huinck et al., 2006), but most of all high covert stuttering aspects do limit or prevent the generalization and outcomes maintenance (Plexico et al., 2005).

In many clients who stutter, covert aspects of the disorder, by the effect of personal past experience, tend to get structured through time in a negative attitude towards stuttering and themselves as for emotions and opinions. The main difficulty in the maintenance stage for people who

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stutter is to oppose themselves to the tendency to go back to cognitions and behaviors settled in the years previous to treatment (Manning, 2009) and so previous to the obtained changes. So, it is possible to observe that outcomes maintenance appears to be notably favored in children (Hancock and Craig, 1998; Silverman, 1992; Gregory, 2001; Finn, 2003), rather than adolescents and young people.

Features of maintenance stage

Treatments for stuttering require a post-therapy maintenance stage, to keep in everyday life therapy outcomes in terms of cognitive, communication and behavioral changes (Conture and Curlee, 2007). At the end of the treatment clients define with their therapists the maintenance goals in the short and in the long run, the specifics to self-manage (Conture and Curlee, 2007) and the most functional situations as field of practice and check.

Clients are encouraged to make a daily communication practice, paying attention to their attitudes. Any kind of activity can be a goal oriented transfer one: first of all routine activities (e.g. family conversations), which are frequent and functional to generalization (Stokes e Baer, 1977), but also non-routine activities, as long as useful to practice some specific abilities. This stage is often made of follow-up sessions with the therapist and refresher clinics when necessary, through individual or small group sessions.

In the follow-up sessions the client meets again the therapist to share and verify the maintenance of therapy outcomes (Conture and Curlee, 2007) or eventual further progresses. This happens always less frequently. The presence of the therapist in this stage is less frequent than during the treatment (Manning, 2009) and this makes the maintenance stage less monitored. Therapists will know from the clients their self-perceived fluency quality. At most, they will have the possibility to vision and listen to some verbalization samples recorded in daily interactive situations by clients for their self-modeling and fluency self-regulation, and to check with them their progresses (Harasym et al., 2015).

Maintenance stage has to favor in people who stutter their responsibility and individual self-regulation (Finn, 2003) through self-monitoring and self management regarding target behaviors, fluency and changes of attitude. This is particularly true for adolescents and young adults, who, as said, encounter more difficulties to stabilize treatment outcomes.

Radio in the maintenance stage of the MIDA-SP program

The MIDA-SP (Tomaiuolo et al., 2012) is a multidimensional, integrated, differentiated program which uses art-mediated activities to make clients practice the acquired competences. This takes place in demanding situations, characterized by time pressure and the interaction with one or more other persons (not rarely an audience), in a creative and motivating way.

The treatment period is almost twelve months and is followed by a maintenance program helping clients to stabilize the generalization of the use of the learnt techniques and to promote their autonomy and self-regulation. As for the treatment, also the maintenance program structure depends upon the client's profile (Tomaiuolo et al., 2012). For clients with a low stuttering severity rate, the maintenance program is made of follow-up sessions with the therapist. These go on for almost a year. At the beginning of the maintenance stage, these sessions are two in a month; then their frequency gets lower, according to the evaluation of client's needs.

Besides the traditional maintenance program, we designed and are testing an advanced one. This specific and potentiated program is targeted on those subjects researchers recognized as more exposed to relapse, by both age and characteristics of their disorder, that is adolescents and young adults with a high severity of the disorder and in particular in its covert aspects. Even though these have gained good results through therapy, they still need to work on their communication to stabilize their outcomes. Covert aspects of their disorder can, in fact, make them feel inhibited in those situations requiring a communicative exposure, most of all in case of time pressure. As the traditional program, the advanced one starts one month after the end of the treatment and is made of follow-up sessions with the therapist, for a

length of 12 months. Besides, in the first 5 months of the program clients undergo advanced *refresher sessions* in small groups, consisting in an activity of radio speaking on a purpose-built web radio.

Radio speaking is a very interesting training ground for practicing and checking personal communication abilities and fluency. As a matter of common knowledge, radio speaking requires a particular ability to manage own verbal communication. As for the telephone, the absence of video prevents clients from using their non-verbal communication, doesn't allow them to see their interlocutors and get from them any feedback.

This activity in the advanced program at the end of MIDA-SP, follows its approach of using art-mediated activities and allows clients practice on those communication specifics they had been working during their treatment. So, clients generalize verbal facilitation techniques in a demanding and semi-structured communicative situation, characterized by personal exposure, high time pressure and problem solving. Clients can practice starting speaking and their verbal fluency, as they are called to build and offer their communication in a given time in terms of seconds; e.g. particularly useful is the exercise to introduce a song in the seconds of the instrumental intro.

Clients practice reading, spontaneous monologue and dialogue, expressing own ideas on specific topics. Clients can do these demanding tests only after treatment, as they have learnt the necessary techniques.

Clients who speak on the radio are in a non-routine situation, which integrates the client's communication practice in daily contexts. They work on covert aspects of stuttering, those which most inhibit the maintenance of outcomes, and on the management of verbalization anxiety. This takes place in a supervised, semi-structured context without the therapist.

Clients work in small groups in a recording studio, with the help of a professional radio speaker.

Being alone with peers and the speaker in a demanding situation requires clients a high command of their verbal communication, gives them a more pro-active role and makes them be and feel more responsible, promoting their self-regulation ability.

The client meets the therapist, as said, during the follow-up sessions, in order to verify goals and results, progresses and difficulties encountered

during the radio sessions, as well as during daily activities. The therapist verifies the use of the techniques through the interview and the analysis of some recorded verbalization samples.

After the first five months of the advanced maintenance program, the client goes on with the periodical follow-up sessions, up to the completion of the program. The frequency of these sessions can be reduced to the therapist discretion.

A preliminary study

Aim

The aim of this preliminary study is to explore the possibility for adolescents and adults who stutter and show a high severity rate of the disorder (especially as for covert aspects) to integrate the first part of their maintenance program through an art-mediated experience of radio speaking.

Method

Participants

The study has been conducted on a sample of ten people who stutter: three of them were females and seven males. They were from 15 to 19 years old, and had all been treated at the CRC Balbuze between May 2014 and April 2015 with the MIDA-SP.

The subjects in the sample took part to this first pilot study one month after they finished their treatment.

They all showed severe stuttering levels as for both overt and covert aspects (a D-profile, according to the MIDA-SP classification; see Tomaiuolo et al., 2012).

A control group (which had undergone the same treatment) has started a traditional maintenance program in parallel.

The advanced program

Radio speaking implies a specific «language», to which participants were trained since the first steps of the course, which have been going on for five months, for 20 weekly sessions, each of two hours.

It was trained by two professional radio speakers. The program was made of four stages:

- I stage: during the first stage (four sessions, for a total amount of eight hours), the radio speaker explained participants what a web-radio is and how it works. He described how to manage the radio clock, which is the daily programs schedule, and how to use the radio technical equipment (microphone, headphones and mixer). Participants worked on the potentiality of their voice and its use through some exercises of vocal warm-up, breathing, articulation, volume, rhythm, clear and correct pronunciation.
- II stage: during the second stage (six sessions, for a total amount of twelve hours), participants were called to a direct contact with the radio equipment, with a particular focus on the use of the microphone and self-listening. Participants were engaged in reading and analyzing different types of text (spot, news, radio commentary, documentary). They were called to break news: in the first time with no time limit, later in one minute limited time.
- III stage: the third stage (five sessions, for a total amount of ten hours) took place in a recording studio. Participants practiced breaking news and simulated a real radio program. At the same time, they were called to show the most relevant aspects of a topic through a more effective use of their voice. The most difficult aspect of this stage for the clients was to keep their communication in the limited time of the «intro», which puts them under a high time pressure, which can generate a high number of disfluencies (Lugo-Neris, 2005). Lastly, participants engaged in both an individual speaking and a cross-talk, through quick verbal exchanges.
- IV stage: in the last stage (five sessions, for a total amount of ten hours) participants built the radio scheduling and aired the web-radio programs.

Materials

The tests used in the study were: SDA test (Systematic Disfluency Analysis, Campbell and Hill, 1987) and ACES/OASES (according to the age), both administered at the beginning and at the end of each stage; SSS (Stuttering Severity Scale), administered before and after each session.

Results

Data gathered during four tests administrations: pre-therapy, intermediate, at the end of the treatment and six months after its end.

By the effect of this integrated work, average scores collected six months after the end of the treatment were better in the group undergone the advanced maintenance program than in the control group, for both overt and covert aspects of their disorder (see tables 1, 2a and 2b).

TABLE 1
Fluency evaluation: average scores (test: SDA)

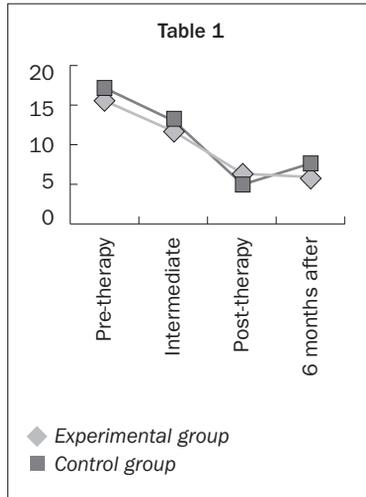
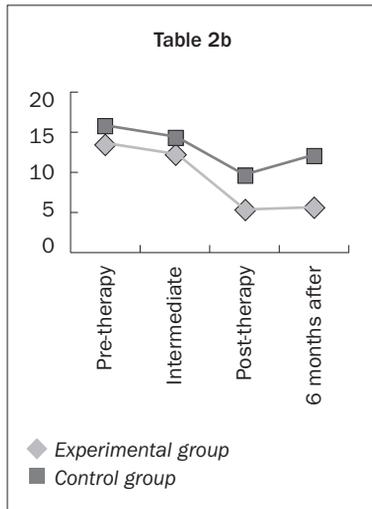
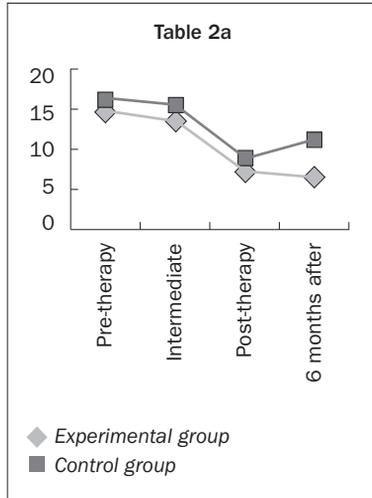
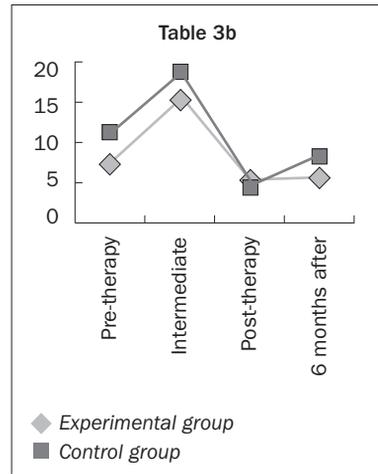
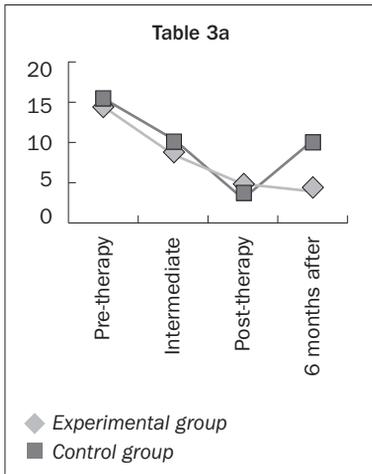


TABLE 2
Covert aspects evaluation (test: Aces/Oases):
whole test (Tab. 2a) and a focus on part III (Tab. 2b)



As observed from tables 3a and 3b, confronting average scores, subjects of the experimental group gave a better self-evaluation about their stuttering, for both fluency and naturalness.

TABLE 3
**Stuttering self-evaluation (test: SSS):
 fluency (Tab. 3a) and naturalness (Tab. 3b)**



Conclusions

The radio experience leads us to reflect on the need to identify specific maintenance programmes for each patient who has completed the rehabilitation programme based on the MIDA-SP profile. In particular, the data collected revealed the importance of ensuring advanced post-therapy maintenance sessions in patients with high stuttering severity levels. The advanced maintenance phase can be thought of as a stage of integration between traditional follow-up sessions and art-mediated activities. Specifically, Radio hosting, due to its characteristics, seemed to be an interesting testing ground.

Despite the interesting results achieved, it is necessary to set out on further reflection on the job. The data presented show first evidence considering the fact that this study is the first trial of the advanced maintenance programme. Moreover, it should be noted that it involved a small number of samples (experimental and control samples) distributed along a reduced timeline for the first results (6 months). Therefore, the research group sets itself the future goal to continue the data collection by expanding the samples, checking the maintenance of results over time (12 months) and envisaging the involvement of children in the advanced maintenance programme..

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Session 10

Final thoughts

WHAT'S THE REAL GOAL OF STUTTERING TREATMENT?

J. Scott Yaruss⁶⁰

Abstract

Different viewpoints about the goals of stuttering therapy lead clinicians to pursue different strategies in therapy. This is understandable, given the complexity of the disorder and the wide range of experiences reported by people who stutter. Unfortunately, however, current opinions about the goals of therapy do not appear to have sufficiently accounted for the views of those who stutter. This brief paper seeks to refocus the attention of speech-language pathology researchers and clinicians on the expressed desires of each specific client who stutters in an attempt to identify a more responsive and individualized approach to stuttering treatment.

Recent years have seen a renewed debate regarding the purpose of stuttering therapy. While it has long been assumed that the goal of stuttering therapy is to reduce or eliminate stuttering, questions have (again)

⁶⁰ Ph.D. University of Pittsburgh, PA; Stuttering Therapy Resources, Inc. — <http://www.StutteringTherapyResources.com>

arisen about exactly what this goal means — and about what clinicians and researchers should do if, for some reason, a speaker is unable to achieve the presumed goal. At times, the discussion has taken a controversial tone. Some writers have strongly dismissed the idea that therapy should focus on something other than (or in addition to) fluency as its «primary» goal. Other writers have argued just as strongly that the experience of stuttering involves more than just speech behaviors, so treatment for stuttering should also involve more than just speech behaviors.

Both sides have cited studies, personal reports, and anecdotal evidence in their favor — and, correctly so. Research does show that people who stutter can improve their fluency through various speech and stuttering modifications, though research also shows that many people who stutter have difficulty achieving or maintaining more fluent speech when using such strategies. Different research shows that many people seek other outcomes from their treatment in addition to improved fluency, such as reductions in negative reactions, and improvement in overall communication skills. At the same time, the volume of research addressing these aspects of therapy is limited, and questions about maintenance of therapy gains remain. Neither side seems likely to convince the other, though there is one common point of agreement: both sides concur that more research on the treatment of stuttering is sorely needed.

As the debate has raged on, it seems that one important set of voices has been underemphasized — that of people who actually live with stuttering on a daily basis and who are, ultimately, the subject of these various discussions about treatment goals.

Until relatively recently, most observations of stuttering were based on the perspectives of listeners (communication partners, researchers, or clinicians). Less credence has been given to the opinions of people who stutter. As self-help and support groups have become more prominent, and as some researchers have implemented the methods of qualitative or phenomenological analysis, this tendency to ignore the person who stutters has, fortunately, been reduced.

By listening to their clients who stutter, rather than assuming that they know what clients must want, practitioners can become better equipped to help people achieve their own goals in therapy.

That said, it is safe to assume that most people who stutter would probably rather not stutter, if given the chance. Stuttering has well-known negative consequences, and it is understandable that people would not want to have to experience those consequences if they did not have to. If a simple cure were available, people would likely flock to it. Unfortunately, despite generations of research, such a cure has not presented itself thus far. It is clear that presently available treatment can minimize stuttering and its consequences; it does not appear that presently available treatment can completely eliminate stuttering or its consequences for all speakers at all times. In this way, stuttering is like many other potentially chronic conditions. Analogies have often been made to the study and treatment of disorders such as asthma or diabetes: people with these problems can learn to manage their difficulties, though a cure is not presently available. If research were focused *only* on curing asthma or diabetes, then important advances in daily management of lifestyle and exposure to risks would not have been made. Likewise, if research were focused *only* on managing symptoms, then important advances in treating the underlying condition would not have been made. These fields have advanced because researchers and clinicians have approached the problems broadly — thinking about both the observable characteristics of the disorders as well as the life impact that may result. In doing so, practitioners have achieved improvements at multiple levels, thereby improving the lives of their patients.

The same has happened, though perhaps to a lesser degree, with stuttering. As noted above, a wide range of strategies have been developed in an attempt to help people who stutter enhance their speech fluency, improve their communication, and reduce the burden of stuttering on their lives. Even though some practitioners tout high success levels with these therapy approaches, however, not all people who stutter are able to benefit from them to the same extent. The reasons for this lack of uniform success are not entirely clear, though one reason may simply be the variability of stuttering. Different people who stutter have different experiences in their lives — they have different personalities, different needs, different histories, different inherent abilities, different coping mechanisms, and different skills. And, most certainly, they have different goals, both for their speech fluency and for their lives as a whole. Gaining a better understanding of these differences

in people's choices in (and out) of therapy would represent an important step in the process of improving treatment outcomes for people who stutter.

Another factor that likely contributes to differential success in therapy — regardless of the specific goal that is selected or approach that is followed — is the inherent difficulty associated with modifying speech production and attitudes toward stuttering. Many authors have acknowledged that changing communication and interaction patterns is hard. Nevertheless, there is often an implicit assumption that people who stutter simply need to «try harder» in order to be more successful in therapy: practice more, modify speech more, desensitize more, monitor more, etc. Certainly, people who put in more effort at any endeavor are more likely to achieve success, but the effort required in stuttering therapy is extreme, and it is not clear that researchers and clinicians have always been sensitive to the difficulties experienced by their clients. Monitoring speech at all times can result in a burden as great as or greater than the stuttering behavior itself. Many speakers engaging in fluency-focused therapy report concerns about unnatural speech patterns, lack of spontaneity, increased desire to hide stuttering, and fear of failure — even after they invest significant time and energy trying to modify their speech. At the same time, many speakers engaging in therapy focused on reducing negative attitudes report difficulties with overcoming their fears about speaking and accepting themselves as people who stutter, when what they would really like to do is just become people who do not stutter. Thus, simply selecting a different set of therapy goals is not the solution it would at first appear to be.

Given these inherent challenges regarding speech therapy for stuttering — which are present regardless of clinicians' or researchers' biases about whether fluency or something else is appropriate goal treatment — it would seem that one way to enhance the outcomes of therapy would be for clinicians and researchers to devote more of their energy to seeking out and listening to the goals and aspirations of the individuals they serve and less energy arguing about what those goals and aspirations ought to be. Specialists can advance the field by recognizing that different people who stutter are likely to have different goals in therapy — and by seeking to match treatments to the goals that people who stutter actually present, rather than the goals that match a particular paradigm or bias about the disorder.

To make this effort successful, however, it will be necessary to approach the problem of stuttering with an open mind about what people who stutter *might* want. If a practitioner focuses primarily on fluency, it is not surprising that he or she would have difficulty accepting that people who stutter might want to work on something other than just fluency. Likewise, if a practitioner focuses primarily on acceptance or stuttering modification, it is not surprising that he or she would have difficulty accepting that people who stutter might want to work on fluency. What is needed is a flexible framework that accommodates all aspects of different people's experiences of stuttering: management of fluency, reduction in negative emotional and cognitive reactions, changes to tension and struggle, increases in functional communication difficulties, minimized impact of stuttering on quality of life, and more. Fortunately, such frameworks are available, though of course more work is needed to match these frameworks to the specific needs of those who stutter.

Interestingly, the debate between different understandings of stuttering mirrors similar debates seen in other fields. Medicine, psychology, social work, and other aspects of the field of speech-language pathologist have struggled (or are still struggling) with finding a balance between different philosophies about various conditions. Researchers and clinicians in these other areas have increasingly come to realize that different approaches to intervention share commonalities, and that these commonalities can form the foundation for individualized yet comprehensive treatment — especially as it is increasingly informed and supported by input from clients. Some researchers and clinicians in stuttering have started to the same, though much work remains. Through partnership with individuals who stutter — and with one another — we can work toward a broader understanding of stuttering that embraces differences between philosophies as well as differences between speakers. Consensus about a single goal of therapy will not (and should not) be reached; consensus about a broad set of goals that may be appropriate to different degrees for different people is attainable.

In the end, then, the real goals of stuttering therapy will need to be multi-faceted, based specifically on the needs of those who stutter, and supported by research that considers the impact of the disorder on the entire person. Clinicians and researchers will need to listen to people who

stutter to understand what is truly their primary complaint and adapt their clinical methods and studies to address what really matters to those who live with the condition. Along the way, practitioners will need to collect data to support unbiased evaluation of treatment outcomes, influenced not by a particular philosophy or set of beliefs about what stuttering should be, but driven by an informed understanding of what stuttering actually is. Working toward a future in which such individualized yet comprehensive therapy is widely available is a worthwhile and appropriate response to the field's history of turmoil.

FUTURE TRENDS IN RESEARCH AND TREATMENT OF STUTTERINGEhud Yairi⁶¹

Considerations of stuttering treatment are closely tied to information concerning the epidemiology of the disorder: such as incidence, prevalence, and natural recovery. It has direct implications to (a) level of research funding, (b) the focus of research, (c) training of clinicians to work with age groups most affected, and (d) availability of clinical services where mostly needed.

Past studies have indicated that the overall incidence of stuttering is around 5% with 1% prevalence. Based on present world population, a 1% prevalence means that there are approximately 72 million people worldwide who currently exhibit stuttering. How can such a large number of people be evaluated, counseled, and treated? Furthermore, recent studies have reported an even larger incidence (data will be presented). Also, the number of speech clinicians in many countries is way too small for serving so many cases. Some countries lack any speech therapy experts.

Quite likely, in the near future, research will continue the current emphasis on three domains: (a) genetics, (b) brain morphology/physiology, and (c) motor aspects. Keeping in mind that the typical age of stuttering onset is early childhood, a large percent of people who stutter are preschool children. Hence, future research activities in the above domains, as well as clinical innovations should be directed toward issues involving young children and their parents. Success with this age group will substantially reduce costs. In this respect, research of the phenomenon of natural recovery is of central importance. Studies conducted in different countries during the past 10 years found natural recovery above 80% (more specific data will be presented), higher than reported in previous studies by the Illinois Stuttering Research Program (Yairi and colleagues).

One overall research objective will be to end up with a much more sophisticated, yet practical clinical tool, administered by speech/language clinicians, combining multiple data from the above three research domains

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as well from current information, fed into a computer program that yields highly accurate prognoses for persistency or recovery (more specifics will be presented). A second, related line of future research would be aimed at identifying factors associated with natural recovery. What happens? For example, what biological changes take place that lead to natural recovery? This line of research will also be highly relevant for the question of how long we wait before starting therapy.

As for clinical progress, keep in mind that the overall objectives of therapy are effective, efficient treatment that should be available to the population of people who stutter world-wide. Future treatment of stuttering will have to deal with the reality of: (a) the growing world-wide demands for help, especially in countries with large populations where economies have improved, and (b) a very limited number of clinical personnel (data will be presented). With so many millions of people who stutter, and so many more at the time of onset, current standard therapeutic methods with face-to-face individual or small group therapy sessions, will not do nearly enough to meet the demands. In the future, we will witness several significant developments:

1. Conventional speech therapy will utilize modern mass delivery (telepractice) modes to reach large numbers of clients in many different locals simultaneously. Outstanding clinicians will be available to the many.
2. Instrument-aided therapy will be readily available to many via modern communication technology.
3. Computer-based therapy for individual use will incorporate new parameters (e.g., biological) that can be modified/controlled to reduce stuttering impact.
4. New effective, inexpensive drugs will become available world-wide and capable to at least substantially improve fluency.
5. For very severe, therapy-resistant stuttering, various methods of brain stimulation will be applicable to individual clients.
6. Gene therapy will be developed for treating stuttering.
7. In general, therapy will be aimed to change not only overt speech but also brain anatomy and physiology, for example, repairing white brain matter. More specific data, examples, and illustrations will be presented.

OTHER CONTENTS

Workshops

AN INTRODUCTION TO COGNITIVE-BEHAVIOUR THERAPY AND ITS USE WITH PEOPLE WHO STUTTER

Jane Harley⁶²

Abstract

Cognitive behaviour therapy (CBT) is widely used as a treatment of choice for people experiencing a range of emotional disorders. It is concerned with the relationship between an individual's thoughts, feelings and behaviours and on working with unhelpful patterns and biases which have a role in maintaining problems. CBT integrates cognitive and behavioural strategies and uses a collaborative therapeutic style to promote self-understanding, empowerment, greater flexibility in thinking, self-compassion and resilience. It is used with children, young people and adults, and in individual and group settings. CBT for people who stutter has been particularly influenced by models of social anxiety (Clark and Wells, 1995; Rapee and Heimberg, 1997). These emphasise the role of fear of negative

⁶² The Michael Palin Center, London UK.

evaluation, attentional bias, pre- and post-event processing and the use of safety behaviours in maintaining social anxiety.

This workshop will give an introduction to the core theory, principles and techniques of CBT and an overview of its current use with people who stutter. Specific theoretical models of social anxiety will be discussed in relationship to work with people who stutter and key therapy steps will be presented. Participants will be shown techniques to help clients identify unhelpful patterns of thinking, attentional focus and ways of responding to events together with strategies for working with these in order to nurture more adaptive self-talk and behavioural choices.

Video excerpts from individual and group therapy sessions will be used to illustrate selected skills and to demonstrate how these core CBT skills may be applied with children, young people and adults.

OVERVIEW OF THE LIDCOMBE PROGRAM

Mark Onslow⁶³

Abstract

The Lidcombe Program is a treatment for preschool children younger than 6 years old who stutter. Researchers at the Australian Stuttering Research Centre at the University of Sydney led its development. The goal of the Lidcombe Program is no stuttering. Hundreds of clinicians around the world each year are trained in the use of the program, thousands of children are being treated with this program, and researchers from several countries are researching this treatment. But what is the Lidcombe Program, and how is it used? What do parents need to learn to do the Lidcombe program with their children? What are the components and stages of the program? What evidence is there for its benefits to children? What difficulties are commonly experienced by parents who do the Lidcombe Program? And why, 25 years after the first clinical trial was published, is the Lidcombe Program still being used? This workshop will provide an introduction to the Lidcombe Program, will allow participants to see some videos of children and parents doing the treatment, and hear what some clinicians and parents say about it. Participants attending this workshop will have the opportunity to ask questions about all aspects of the Lidcombe Program.

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STUTTERING: FOR BETTER OR FOR WORSE USE CREATIVITY «CHOOSE FOR BETTER»!

Joseph G. Agius⁶⁴

Abstract

The presentation highlights the importance of shifting attitudes in stuttering intervention. Changes in attitude of clinicians, adults who stutter and school-age children who stutter could determine the success of therapy. Research has also shown that children who stutter view speaking more negatively and experience more negative speech-related emotions than non-stuttering peers do. While practical strategies for helping children who stutter change their feelings and beliefs about stuttering are widely available, speech language pathologists feel uncomfortable targeting such goals. Our attitudes also have an influence in the therapeutic process. In this presentation, the results of a study exploring shifts in the attitudes and feelings of school-age children who stutter following a thinking skills program will be presented. The findings of this study led to a suggested model of intervention, the *Smart Intervention Strategy* (SIS), with school-age children who stutter. An overview of the SIS is highlighted and introduced.

Learning outcomes

Participants will:

1. become familiar with the results of a study exploring shifts in attitudes and feelings following a thinking skills prog;
2. appreciate the importance of positive attitude during stuttering intervention;
3. describe the structure and content of the *Smart Intervention Strategy*.

⁶⁴ University of Malta.

Posters

USING EVIDENCE TO INFORM STUTTERING INTERVENTION PLANNING: IMPLICATIONS FOR TEACHER INTERVENTION

Harsha Kathard, Kristen Abrahams, Michal Harty, Rizwana Badroodien, Freda Walters⁶⁵

Speech-language therapists are increasingly having to develop strategies for intervention at a population level particularly in contexts of minimal resource. In school contexts, teachers are a key resource in stuttering intervention as they are routinely available in the everyday contexts of children who stutter and their peers. Their roles are crucial in all contexts and particularly in contexts where there are few or no therapists available. While teachers do not replace the therapist, they play key role in creating positive environments to facilitate the inclusion and participation of CWS. However, teachers require training and support to become effective supports. It is not feasible or practical to provide individual training and support — on an individual basis particularly in contexts of under-resourcing. Therefore, it is likely that teachers — as a group — will require education and support. However, prior to planning such intervention, the opinions generally held by the group to stuttering should be established. In addition, the factors

⁶⁵ University of Cape Town – South Africa.

influencing their opinions and attitudes should be established as a basis for planning intervention. It is important to ascertain what demographic and school-related factors are associated with their attitudes. To illustrate the importance of using evidence to inform interventions, this paper uses the data generated from a large scale POSHA survey of 469 teachers in primary schools in Western Cape, South Africa. The results of the study indicated that teaching-related factors: quintile and years of teaching experience and for personal factors: gender, first language, familiarity and age were associated with their opinions. The implications of these findings for tailoring interventions with teachers, is discussed.

DEVELOPMENT OF THE LIDCOMBE PROGRAM IN FRANCE: FEEDBACK FROM CLINICIANS AND PARENTS

Rosalee C. Shenker⁶⁶; Veronique Aumont Boucand⁶⁷

Abstract

For several years treatment of stuttering in preschool age children in France utilized a counseling model based on psychodynamic understanding. This belief was based on the idea that reactions of the parents could create persistent stuttering, similar to the diagnosogenic philosophy of Wendell Johnson. Treatment was not usually initiated before the age of 6, and parents of children who stuttered were often advised in consultation with psychologists if stuttering persisted.

In 2000, an overview of the Lidcombe Program was first presented in France. The introduction of this program posed a challenge to the beliefs of many clinicians, who were initially sceptical and critical of these ideas. In 2010, the first workshop for the Lidcombe Program was introduced in Paris as part of Quebec/France collaboration. While efficacy for the Lidcombe Program has been demonstrated primarily in English speaking countries with one randomized controlled trial done in Germany, this was the first attempt to provide training in French. Subsequently there have been 8 basic workshops, and several articles written in French suggesting that the outcomes of the program provided in French are consistent with those found in English-speaking countries.

This presentation highlights parent/clinician feedback gathered through an internet questionnaire to clinicians who received training, and qualitative interviews of parents. Questionnaires were emailed to 167 French clinicians with a response rate of 42%. The attitudes of the French clinicians, as well as parents compares positively overall to those of their English-speaking counterparts. Results of the clinician's questionnaire will

⁶⁶ Montreal, Quebec, Canada.

⁶⁷ Paris, France.

discuss feedback on the components of the Lidcombe Program including measurement, parent verbal contingencies, treatment in practice versus natural conversations and satisfaction with the program. Parent interviews suggest that although parents initially may find that it is unnatural to give the verbal contingencies since it is not their habit to use the types of reinforcement used in English-speaking countries, they adjust after a few sessions. Initially comments for stutter-free speech might take the form of more neutral acknowledgement of fluency, but the parents quickly accommodate to the verbal contingencies and provide them warmly to their children. Parents soon begin to generalize positive comments to other situations and this has been seen as a positive change to the parental interaction with their child. Discussion of the impact of cultural differences between the South European and the English Culture, and preliminary outcomes of the treatment in France will also be discussed.

EVALUATING THE LIDCOMBE PROGRAM IN FRANCE: FEEDBACK FROM PARENTS AND CLINICIANS

Veronique Aurmont Boucard
Université Paris 6

Rosalie C. Shenker
Montreal Fluency Centre

1. Goal

Evaluate satisfaction and ease of use of the Lidcombe Program in France

Evaluate:

- Use of Lidcombe Program by clinicians
- Benefits/challenges identified
- Challenges attributed to cultural differences

2. Methodology

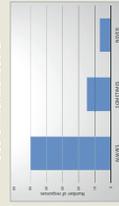
- Feedback from clinicians; interviews with 3 Parents
- Questionnaire
 - Sent to 167 French clinicians
 - Trained in the LP since 2011
 - 43% response rate

What is the Lidcombe Program?

- A verbal response contingent treatment
- Presented by parents
- For children younger than 6 years

3. Findings: Clinicians

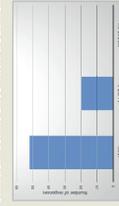
Use of manualized LP



Satisfaction compared to previous method of treatment



Clinician confidence with treatment



4. Findings: Parents

Challenges

1. Initial difficulty maintaining daily severity ratings
2. Moving from practice to natural conversations
 - Difficult at first
 - Contingencies unnatural
 - Parents not accustomed to terms

Benefits

- 1. Once accustomed to daily ratings routine, viewed as useful
- 2. Positive comments generalized to other behaviours (outside of speech)
 - Improved parent/child relationship
- 3. Parents became comfortable with process
 - Quickly accustomed to verbal contingencies
 - Learned to manage more severe situations
 - Children more confident

5. Conclusions:

1. French Clinicians have adapted well to the Lidcombe Program
 - Few ad-hoc deviations from treatment guide were used
 - Need time to adapt to terminology of treatment/measurement concepts
2. Parents need time to adapt to the treatment but adjust well and are satisfied
3. Preliminary results show that the Lidcombe Program can be adapted across cultures/languages

COARTICULATION AND VOWEL CENTRALIZATION IN FRENCH AND ITALIAN STUTTERING ADULTS AND CHILDREN

Marine Pendelieu⁶⁸; Anaïs Tissot-Dupont⁶⁹; Solange Rossato⁷⁰; Claudio Zmarich⁷¹

Abstract

Many studies on stuttering focused on degrees of coarticulation in fluent speech as a way to observe speech motor control (Van Riper, 1982; Wingate, 2002; Sussman et al., 2011). Findings based on $\Delta F2$ and locus equation have shown various behaviors, mainly depending on the place of articulation of the consonant, but also on prosody, word stresses, speech rate and language. In Verdurand et al. (2013), French and Italian stutters tend to coarticulate less than fluent speakers, with a significant role of the language spoken. Concerning the area of vocalic triangles (AVT), smaller values were found for stutters in some studies (Klich and May, 1982; Blomgren, Robb and Chen, 1998), whereas no significant difference in vowel centralization was found (Prosek et al., 1987). The AVT of stutters does not seem to decrease when speech rate increases, as opposed to what is seen in fluent population (Hirsch et al., 2009).

These phenomena could be considered as strategies to try to prevent stuttering events (Van Lieshout et al., 2004); therefore, children who stutter (CWS) and adults who stutter (AWS) might show different patterns of coarticulation as those strategies may not be installed yet.

This present work aims to compare coarticulation of stutters according to their age, between a population of school-age French and Italian children and adults. The corpus used was recorded with 40 French speakers (10 AWS and 10 fluent adults, and 8 CWS and 12 fluent children) and 39 Italian speakers (11 AWS and 10 fluent adults, and 8 CWS and 10 fluent children) (Pendelieu-Verdurand, 2014). Each subject had to repeat syllables

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(CV with C = {/b/, /d/, /g/} and V = {/a/, /i/, /u/}) and isolated vowels (/a/, /e/, /i/, /o/, /u/). All fluent syllables were kept and manually annotated. For each subject, formants were measured and used to calculate the AVT on isolated vowels and the k slope for each consonant (/b/, /d/, /g/) using the locus equation (Lindblom, 1963).

First results on Italian adult speakers have shown that there was no significant difference between the AVT of Italian stutterers and the one of Italian fluent speakers. This supports the results that have been found (Prosek et al., 1987). We are currently analyzing data of Italian children speakers, French adult and children speakers, in order to compare the results to those obtained for Italian adult speakers.

Concerning intra-syllabic coarticulation, k slopes of French and Italian child speakers are being estimated so that we can find out if the difference found between adults in (Verdurand et al., 2013) also exists between CWS and fluent children, and if the role of language is already significant at their age.

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EVALUATION OF THE «DISFLUENCY PROFILE» AS A PREDICTOR OF PERSISTENT STUTTERING IN CHILDREN WHO HAVE JUST STARTED TO STUTTER

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According to Yairi and Ambrose (2005), for 95% of people who begin to stutter the onset occurs before the age of 4 years, most of the beginnings occur in the third year of life, and 90% of children who start to stutter recover spontaneously, usually no later than the fourth year post-onset. If symptoms persist beyond this term, treatment becomes difficult, so it is important to submit the subjects that tend to persist to early treatment.

The project «Phonetic indexes predictive of chronic stuttering in preschool children» (Dr. Claudio Zmarich, CNR-RSTL n. 995) started in 2008, and aimed to identify some behavioral and clinical indexes that predict persistency. 40 families where at least one component stuttered at that time, for as many children, participated. All children were first audio and video recorded when they were 24-month-old, and as soon as a child showed the first symptoms of stuttering, he was addressed to the «Centro Medico di Foniatria» in Padua for receiving a diagnosis and for being evaluated for speech and language abilities and for ruling out other diseases. Subsequently, the child began to be recorded at home every 3 months up to 16-22 months, to collect data on phonetic development, speech-associated attitude and severity of stuttering.

Here we present data about 12 children which started to stutter. The experimental design is to predict the persistency based on the value of the «disfluency profile» at 9-15 months post-onset (counts of repetitions of part

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of words or monosyllabic words, as well as disrhythmic phonations, Yairi and Ambrose, 2005). At the end of the observation period this measure was compared with the SSI-3 score (Riley, 1994). To better determine the final clinical outcome of each child, a reference was also made to a telephone structured interview to parents after an average of 43.2 months from the onset of stuttering, which determined that 3 children had become persistent stutterers (S) and 9 had recovered spontaneously (NS).

In order to be considered a good clinical marker, the disfluency profile must simultaneously exhibit a level of sensitivity (accuracy in detecting S) and specificity (accuracy in detecting NS) of at least 80%. Comparing the stage of 9-15 months post-onset to the final outcome, you get 2S and 5NS, including 4 false positive and 1 false negative (sensitivity 66.3%, specificity 55.5%). A half year later, the predictive value increases: you get 3S and 6NS, with 3 false positives and 0 false negatives (sensitivity 100%, specificity 66.6%). For the stage of 16-22 months we also have the SSI-3 scores: applying the criterion of cut-off used by Howell and Davis (2011), and comparing the results to final outcome, you get 3S and 5NS, including 4 false positives and 0 false negatives (sensitivity 100%, specificity 55.5%). In conclusion, the predictive value of the disfluency profile at 9-15 months post-onset is lower than the clinical standards, but increases in the following semester, and it is a better and more «user-friendly» predictor than the SSI-3.

Evaluation of the "disfluency profile" as a predictor of persistent stuttering in children who have just started to stutter

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1. Abstract
Rationales: to identify some behavioral indexes able to predict persistency at early ages, in order to assure to more at-risk subjects the best therapeutic strategies
Aims: to evaluate the clinical efficacy of the Disfluency Profile in identifying children at greater risk of persistency
Results: the predictive power of the Disfluency Profile at the session of 9-15 months post-onset is low, according to clinical standards of sensitivity and specificity, but it increases in the next six months, albeit not to the requested minimum of 80%. However this is better than SSI-3, which has been proposed as a predictive tool by Howell & Davis (2011).

2. Introduction
 • Yairi and Ambrose (2005) have called the typical dysfluencies produced by stutters **stuttering-like dysfluencies (SLD)**, to distinguish them from **other dysfluencies (OD)**, which are typical of the stutters' as well as non stutters' speech.
 • According to Yairi & Ambrose (2005), for 95% of people who begin to stutter, the onset occurs before the age of 4 years.
 • Around 90% of children who start to stutter recovers spontaneously (Yairi & Ambrose, 2013), no later than the fourth year post-onset.
 • If symptoms persist beyond this term, treatment becomes difficult, so it is important to submit the subjects at high risk of persistence to early treatment
 • It is further well known that stuttering is characterized by a strong hereditary component (Kraft & Yairi, 2011)

3.1. The "Phonetic indexes predictive of persistent stuttering in preschool children" project (CNR-RSTL n. 995, 2007): Aims
Aims: to identify some behavioral indexes able to predict persistency at early ages.
Assumptions: high risk of persistency if in the 2nd year post onset:
 • 1) the SLD % remains relatively high (Yairi & Ambrose, 2005);
 • 2) the CV coarticulation is high (Subramanian, Yairi, Ofek, 2003);
 • 3) the negative attitude towards communication is high.
 This contribution deals only with the first hypothesis.

3.2 The "Phonetic indexes predictive of persistent stuttering in preschool children" project (CNR-RSTL n. 995, 2007):
 • 40 families, where at least one relative stuttered at that time, for as many children (aged between 12 and 23 months)
 • All children were first audio and video recorded when they were 24-months-old (see Quartarone et al., this conference)
 • As soon as a child showed the first symptoms of stuttering, if any, he/she was addressed to the "Centro Medico di Fonia-tri in Padova for receiving a formal diagnosis, for being evaluated for speech & language abilities and for ruling out other main diseases
 • At the same time, the child began to be audio and video recorded at home, every 3 months up to 16-22 months post-onset (for a total of 6 recordings), in order to collect data on:
 • severity of stuttering (SSI-3, Riley, 1994)
 • Disfluency profile (% SLD)
 • phonetic development (Zmarich e al. Articulation test, Acoustic analysis of coarticulation)
 • lexical development (MacArthur-Bates CDI)
 • communication attitude (KiddyCAT, Vannirkegem & Bruten 2007)

If at the end of this period the child was still stuttering and if parents demanded it, a treatment was initiated at the CMF and the experimental observation consequently stopped.

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GLOSSARY
STUTTERING-LIKE DYSFLUENCIES (SLD):
 • part-word repetitions
 • monosyllabic words repetitions
 • Disrhythmic phoneations
OTHER DYSFLUENCIES (OD):
 • Interjections;
 • Repetitions of polysyllabic words and phrases
 • Revisions and incomplete phrases.

4. Aims of the current study
 • to evaluate the clinical efficacy of the Disfluency Profile in identifying children at greater risk of persistency

5. Subjects
 • 13 children who began to stutter after the recordings at 24 months of age
 • Age of stuttering onset: between 21 and 51 months of age (mean: 33 months) according to parents,
 • Age of first recording: from 0 to 6 months after the onset (mean: 2.21; months;days).
 • Clinical Outcome: a telephone structured interview to parents was made, after an average of 41;15 from stuttering onset (range: 24-68 months): 3 children had become persistent stutters (S) and 10 had recovered spontaneously (NS). S and 10 are the same children who had undergone therapy

6. Instruments
 • Disfluency profile: the SLD% out of 400 target syllables (Yairi & Ambrose, 2005);
 • SSI-3 (Riley, 1994), calculated on 400 target syllables (the same as for SLD);
 • Structured telephone interview: parents had to respond to a series of 9 YES or NO questions (answering yes to at least three of them qualified the child as stuturer), and also to place the level of stuttering along a 7-point Likert scale, where 1 represents the absence of stuttering and 7 the maximum of severity.

7.Procedure
 • The child was audio and video recorded, for about an hour, and a sample of never less than 500 syllables was collected. We tried to predict the persistency of the disorder from the Disfluency profile scores at 9-15 months or 16-22 months post-onset, or from the SSI-3 scores (16-22 months post-onset).
 • We adapted to prognostic purposes the data on the SLDs developmental paths of the persistent and recovered stutters in the Illinois Longitudinal Study, as described by Yairi & Ambrose (2005, 5th chapter); the SLD percentages of the recovered stutters was halved in passing from the first to the second semester post-onset, and systematically reduced in the following semesters; conversely, the reduction rate (if any) of the persistent stutters was very low.
 • For using the SSI-3 as predictor, we referred to the proposal of Howell & Davis (2011), to be considered recovered, a stuturer must 1) score less than 24 at the SSI-3 and 2) present a reduction of at least 2 points compared to the onset.
 • To evaluate the effectiveness of the prediction, we made reference to clinical standards: the instrument have to simultaneously exhibit a level of sensitivity (measurement accuracy in detecting S) and specificity (accuracy of the identification of NS) of at least 80%, although we are aware that such a small sample size would not formally allow this kind of analysis.

8. Results (sensitivity & specificity analysis)
S=Stutters; NS=non-stutters

		Actual clinical output by september 2015		
		S (3)	NS (10)	
Disfluency pr. prediction by >=4.5 m. post -onset	S (6)	2 (66.6%)	4 false positives	
	NS (7)	1 false negative	6 (60%)	
Disfluency pr. prediction by 16-22 m. post -onset	S (6)	3 (100%)	3 false positives	
	NS (7)	0 false negative	7 (70%)	
SSI-3 -prevalence by 16-22 m. post -onset	S (9)	3 (100%)	6 false positives	
	NS (4)	0 false negative	4 (40%)	

9. Discussion & Conclusion
 • The predictive power of the Disfluency profile at the session of 16-22 months post-onset is better than at 9-15 months post onset, and it is better than the SSI-3 at 16-22 months.
 • Since no reliable clinical tool exists yet for predicting the persistency of stuttering, the Disfluency profile maintains, compared to the SSI-3, the advantage of being free, less demanding, less arbitrary (no need to evaluate lesion) and faster to administer and process.

Best Graduation Thesis Award

The Conference established a special award for the best B.A. graduation thesis in Speech Therapy whose topic is stuttering, presented in the last two Academic Years. This Award is a recognition of the fundamental role Universities play in preparing tomorrow's therapists and researchers on stuttering.

USING DELAYED AUDITORY FEEDBACK (DAF) AS A REHABILITATION INSTRUMENT IN DISFLUENCY

Thesis discussant: *Dr. Martina Andriollo*

Supervisor: *Dr. Gian Vincenzo Zuccotti*

Institution: University of Milan

Summary

Stuttering is a complex disorder, hard to define in an unambiguous way, generally considered a problem of the flow of the speech. In the last years various clinical trials have been contributing in strengthening the hypothesis which consider disfluency as a disorder of coordination and senso-motory feedback systems regarding the movements underlying speech production.

Stuttering rehabilitation is typically divided into two steps:

- Learning of facilitating techniques: stuttering modification techniques and fluency shaping exercises;

– Generalization.

Next to these approaches, there are also various methods like Delayed Auditory Feedback (DAF).

As of today there are scientific evidence suggesting the use of DAF as a functional instrument for disfluency treatment (Kalinowski et al., 1993, 1996; Stuart et al., 1997; Stuart and Kalinowski, 2004; Van Borsel et al., 2003); nevertheless, it's to be recognized that not all individuals achieve improvements in fluency using this device.

The work's aim is to investigate and to evaluate whether the use of Delayed Auditory Feedback (DAF) in a «classic» rehabilitation treatment for stuttering (made out of the exercises mentioned above) significantly reduces the episodes of stuttering in both teenager and adult subjects, thus improving verbal fluency and communication efficiency compared to the «classic» rehabilitation alone.

Methods

Once inclusion/exclusion criteria has been defined, it was decided to work in this way: the 18 chosen participants to the study (4 females, 14 males – mean age 18 years) have been divided in two groups with a single-blind randomization. *Group «A»* is the experimental group: all the subjects underwent a «classic» rehabilitation treatment plus DAF usage (medium delay: 86 ms). *Group «B»* instead is the control group.

Treatment duration was set to be 3 months, once a week.

An informed consent for audio/video recordings (crucial for an accurate analysis) was presented and signed by all the patients.

Each subject has been evaluated before and after the treatment; Stuttering Severity Instrument-3 (SSI-3, Riley) has been used for this purpose.

The initial assessment for speech sample gathering has taken place during two different communication tasks: conversation and reading aloud.

The treatment was the same for both groups, with various activities being proposed:

- exercises for breath control and pneumophonic coordination;

- praxis-phonemic discrimination;
- orofacial proprioception;
- deconstruction and desensitization;
- fluency shaping and stuttering modification techniques.

Group «A» used DAF during these activities. During a 45 minutes therapy, 20 minutes were dedicated to conversation (C) and newspapers reading (R) under DAF.

The final assessment, after 3 months of treatment, used the same reading tasks given out during initial assessment.

Results

With informal observation and data analysis of values regarding pre/post evaluation of both groups, a quantitative improvement in global fluency and other aspects connected to it (or the achievement of a «functional disfluency») has arisen for each subject. More specifically:

GROUP A (MEANS)				
	More typical disfluencies		Less typical disfluencies	
	C	R	C	R
PRE	9,6	8,3	12,3	13,8
POST	5,9	7,3	7,4	14,9
p-value (t-test)	0,04	0,5	0,02	0,7

GROUP B (MEANS)				
	More typical disfluencies		Less typical disfluencies	
	C	R	C	R
PRE	7,4	5,0	13,0	11,8
POST	4,2	2,6	9,3	7,7
p-value (t-test)	0,0007	0,002	0,0006	0,0015

GROUP A (MEANS)				
	Words per minute		Syllables per minute	
	C	R	C	R
PRE	108,9	119,8	201,9	227,4
POST	73,2	86,1	134,0	157,7
p-value (t-test)	0,05	0,04	0,04	0,05

GROUP B (MEANS)				
	Words per minute		Syllables per minute	
	C	R	C	R
PRE	105,9	93,9	190,2	194,8
POST	93,7	93,3	167,4	179,8
p-value (t-test)	0,3	0,9	0,2	0,4

	GROUP A (MEANS)			GROUP B (MEANS)		
		Duration of blocks (seconds)			Duration of blocks (seconds)	
		C	R		C	R
PRE		1,2	1,1		2,2	1,7
POST		0,9	0,8		1,6	1,2
p-value (t-test)		0,09	0,04		0,001	0,01

GROUP A (MEANS)			
	PRE	POST	p-value (t-test)
Distracting sounds	0,9	0,2	0,02
Facial grimaces	2,9	1,9	0,01
Head movements	1,1	0,4	0,08
Movements of the extremities	1,2	0,4	0,04

GROUP B (MEANS)			
	PRE	POST	p-value (t-test)
Distracting sounds	1,7	0,6	0,05
Facial grimaces	3,1	1,6	0,0001
Head movements	1,9	0,4	0,01
Movements of the extremities	2,7	1,1	0,0002

From a statistical perspective using DAF with traditional treatment does not quantitatively improve subject's final performance against the traditional treatment alone.

Conclusions

According to the results previously described, DAF has been proven to be a clinically valid added value for disfluency management with a traditional treatment; it does not represent an alternative, but a supporting feature for a more efficient objectives achievement.

From scientific literature, positive effects on several stutterers thanks to DAF usage cannot be denied; however, data available nowadays can't be used to predict whether a subject could benefit from it or not, considering the multifactorial and varying nature of the disorder. The statistical results obtained do not express strong evidences of a greater effectiveness of DAF treatment; it would be complicated to assert how much the improvements resulted from using DAF or from treatment itself.

Nevertheless, subjects who experimented DAF were widely satisfied and they are still using the device even after therapy period.

Three factors still need to be investigated about the device use:

- Residual effect
- Contained cost
- Aesthetics

To answer questions emerged from the study further studies are necessary, in which an initial skimming based on the susceptibility to DAF and a wider and less heterogeneous sample will be essential.

It also remains to investigate the real effectiveness of the Delayed Auditory Feedback in reading aloud tasks, since scientific literature provides limited data about it.

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