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Could the frequency of Stuttering-Like-Disfluencies predict persistent stuttering in children who have just started to stutter?

Stuttering onset occurs for 95% of people who begin to stutter before the age of 4 years, tipically in the third year of life. *Spontaneous recovery* during childhood is common, with recovery rates estimated at 68-96%, usually no later than the fourth year post-onset. If symptoms persist beyond this time, the efficacy of treatment might result more problematic. As a result it is important to refer the subjects that tend to persist to early treatment. The CNR project "Phonetic indexes predictive of chronic stuttering in preschool children" started in 2008, and aimed to identify, among different behavioural indexes, the ones which are able to predict stuttering persistence at early ages, in order to assure to more at-risk subjects the best therapeutic interventions. The aim of the current study is to evaluate the clinical efficacy of the *Disfluency Profile* (i.e. the percentage of Stuttering-Like Disfluencies over 100 spoken syllables) in identifying children at greater risk of persistence. Results of the study suggest that 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 over the next six months, albeit not to the standard minimum of 80%. However, the use of the Disfluency Profile is preferable to the *Stuttering Severity Instrument* (third edition), which has been proposed as a predictive tool by some researchers.

Key words: disfluencies, children, stuttering persistence, stuttering recovery, developmental stuttering.

Introduction

It is well known that stuttering is characterized by involuntary part-word and monosyllabic word repetitions, as well as disrhythmic phonations which consist of prolongations of sounds and/or arrests of speech. Yairi, Ambrose (2005) referred to these as 'Stuttering-Like Disfluencies' (SLD) to distinguish them from 'Other Disfluencies' (OD). OD typify the speech of people who do not stutter, and include multisyllable/phrase repetitions, interjections, revisions and incomplete utterances. According to Yairi, Ambrose (2005), for 95% of people who begin to stutter, the onset occurs before 4 years of age (see also Reilly, Onslow, Packman, Cini, Ukoumune, Bavin, Prior, Eadie, Block & Wake, 2013). Among preschool children who experience stuttering, approximately 90% will recover spontaneously, usually within 4 years post-onset (Yairi, Ambrose, 2013). If symptoms persist beyond this time, then natural recovery rarely occurs without any intervention, and treatment itself risks not to be successful. Furthermore, it is well known that stuttering is characterized by a strong hereditary component (Kraft, Yairi, 2011) and is often associated with negative attitudes towards communicative situations (Clark,

Conture, Frankel & Walden, 2012). Because of these evidences, the clinical importance of early treatment for children at high risk of persistent stuttering becomes apparent.

The purpose of the research project "Phonetic indexes predictive of persistent stuttering in preschool children" (CNR-RSTL n. 995, granted in 2007 to the first author) is to identify some behavioural indexes that might serve as predictors of persistence at early ages.

Specifically, several clinical factors in previous researches have been proved quite able to differentiate children at high risk of chronic stuttering from those who are likely to recover (Yairi, Ambrose, 2005: 348). Apart from biological factors, like to be male or to have one or both the parents who still stutter, we choose to verify the following statements in the second year after the onset there is a risk of persistence if: 1) the SLD percentage (%) remains relatively high (Yairi, Ambrose, 2005); 2) there is a high degree of Consonant-Vowel coarticulation in perceptually fluent speech (Subramanian, Yairi & Amir, 2003); 3) there is a mal-attitude toward speech (negative attitude towards communication is high). This contribution only deals with the first assumption, and it is based on the SLD's developmental paths of the persistent and recovered stutterers as described by Yairi, Ambrose in the *Illinois Longitudinal Study* (2005, Chapt. 5, see table 1). Our choice of this measure was driven by current best research evidence and by the criterion of clinical suitability, and for the last reason we did not consider other not frankly clinical measures (such as the acoustic ones).

Table 1 - *Mean (standard deviation) of SLD and OD for Persistent, Recovered and Control Groups of Children (modified from Yairi, Ambrose, 2005: 173)*

Subjects	0-6 months	7-12 m.	13-18 m.	19-24 m.	25-36 m.	37-48 m.	49-60 m.
Persistent							
SLD	11.31 (6.12)	9.76 (6.32)	7.82 (5.31)	7.34 (6.75)	7.93 (6.40)	5.85 (8.37)	3.61 (4.42)
OD	11.03 (6.74)	5.41 (2.09)	5.42 (2.48)	5.75 (3.57)	7.49 (4.20)	5.54 (1.67)	6.38 (3.01)
Recovered							
SLD	11.03 (6.74)	5.38 (4.37)	3.01 (2.65)	1.99 (1.51)	1.62 (1.56)	1.10 (0.81)	0.91 (0.64)
OD	5.85 (3.00)	5.21 (2.34)	5.13 (2.92)	4.80 (2.25)	4.93 (2.89)	5.07 (2.06)	5.75 (2.51)
Controls							
SLD	1.42 (1.01)		1.11 (0.79)		1.08 (0.97)	0.93 (0.89)	
OD	4.42 (2.27)		4.39 (1.60)		4.67 (2.17)	5.42 (2.02)	

As shown in table 1, the SLD percentage of the recovered stutterers was diminished by nearly half between the first (11.03%) and the second semester post-onset

(5.38%), and it was systematically reduced in the following semesters. Conversely, the SLDs rate reduction (if any) of the persistent stutterers was very low over the same period (11.31% in the first and 9.76% in the second semester).

This kind of results could be discovered only by research projects that are longitudinal in design. In fact, the emerging view on stuttering considers it as a neurodevelopmental disorder involving multiple variables, including motor, language and emotional factors (Smith, 2016). Only projects designed to follow children who stutter from the onset to the final remission or persistence of the disorder (at least 5 years long), like the Illinois Longitudinal Project (Yairi, Ambrose, 2005) or the Purdue Stuttering Project (Smith, 2016), not to say of the prospective "Victoria Study" (see Reilly et al., 2013) are equipped to track these factors.

We started from the results of our recent report on 10 Italian children who stutter (Zmarich, 2015), whose persistence was better predicted by the "16-22 months" scores of the *Disfluency Profile* than the "9-15 months" scores. However, the accuracy of prediction of disfluency profile at 16-22 months was not much better than the prediction based on the severity scores (SSI-3, Riley, 1994). As a consequence, the application of these instruments seemed result in similar outcomes.

Here we present the data regarding 13 children (they constitute the whole sample at the last session of the project), ten of which were already participants of the previous report (Zmarich, 2015). The present update is justified by two main reasons: (1) the increase of the sample by three subjects, whose analyses have been completed after Zmarich (2015); (2) the increased post-onset interval (almost two years more), which adds reliability to the assessment of the final clinical outcome. The experimental purpose is to evaluate which of the following scores could best predict persistence at different stages and with different tools: the *Disfluency Profile* at 9-15 months post-onset, the *Disfluency Profile* at 16-22 months post-onset, or the SSI-3 scores at 16-22 months?

1. *The CNR Project*

The CNR project started in 2008, and aimed to identify some behavioural indexes to predict persistence or recovery from stuttering.

Forty families were enrolled in the study when the following criteria/conditions were satisfied: at least one member who stuttered at that time (or had already stuttered) and a child aged 12-23 months in the same family. In this way we could be sure to maximize the probability to collect stuttering cases (see Kloth, Kraaimaat, Janssen & Brutten, 1999, for a similar design).

All children were first audio and video recorded when they were 24-months-old, and as soon as a child showed the first symptoms of stuttering, he/she was addressed to the "Centro Medico di Foniatria" (CMF) in Padua in order to receive a formal diagnosis, and to be evaluated for speech and language abilities in the attempt to rule out other main diseases (Zmarich, 2015). 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 the phonetic development (TFPI, Zmarich, Fava, Del Monego & Bonifacio, 2012; "routines" for verbal play, Stoel-Gammon, 1989), lexical development (MacArthur-Bates CDI, Caselli, Pasqualetti & Stefanini, 2007), severity of stuttering (SSI-3, Riley, 1994), and communication attitude (KiddyCAT, Vanryckeghem, Brutten, 2007). If, at the end of that period, the child was still stuttering and the parents requested it, a treatment was initiated at the CMF and the experimental observation consequently ceased.

2. *Materials and Methods*

2.1 Subjects

The subjects of the current study are 13 children of the 14 who began to stutter after the recording at 24 months of age (table 2).

Table 2 - *Subjects (*treated subjects), gender, age at the stuttering onset, months elapsed from stuttering onset to first recording, months elapsed from stuttering onset to final clinical outcome*

subjects	*gender*	*age onset*	*months post onset at the first recording*	*months post onset at the final evaluation*
BM	F	51	2	42
CA	F	30	0	68
CG	M	30	1	38
FM	M	29	0	46
FMd	F	36	3	37
GS	F	30	6	31
MG	M	40	0	67
*ML	M	30	6	39
*RF	M	36	3	41
SC	M	31	2	24
*SL	M	21	5	33
*TA	M	31	3	40
VL	F	33	4	33
Mean		32,27(m,d)	2,21(m,d)	41,15 (m,d)

According to parents' reports, children started to stutter between 21 and 51 months of age (mean: 33 months) and they were first recorded from 0 to 6 months after the onset (mean: 2,21; months, days). Four of the subjects at the end of the observation period were treated.

In order to determine the final clinical outcome of each child, a structured telephone interview to parents was made, after an average of 41 months and 15 days from stuttering onset (range: 24-68 months). Based on these interviews, 3 children

had become persistent stutterers (S) and 10 had recovered spontaneously (NS). S children were among the children who attended a therapy.

2.2 Instruments

Disfluency Profile: it is an index drawn from an original method for counting disfluencies put forward by E. Yairi and N. Ambrose in several studies, collected in Yairi, Ambrose (2005). It is based on the calculation of the percentage of SLD out of 400 target syllables;

Stuttering Severity Instrument – Third Edition (SSI–3; Riley, 1994): it is a reliable and valid norm-referenced stuttering assessment tool that can be used for clinical and research purposes. It has been developed to evaluate stuttering severity both in adults and children. Three features have to be obtained to calculate an SSI-3 score: (1) Frequency of stuttering, (2) Duration of the three longest stuttering moments, and (3) Physical Concomitants. Frequency is expressed in percent syllables stuttered and it is converted to scale scores of 2-18. Duration is timed to the nearest one tenth of a second and it is converted to scale scores of 2-18. The four types of Physical Concomitants are summed and converted to scale scores of 0-20;

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 stutterer), and they also had to place the level of stuttering severity along a 7-point Likert scale, where 1 represents the absence of stuttering and 7 the maximum of severity.

2.3 Procedure

As regards the experimental session, the child was audio and video recorded in a play activity where she/he was manipulating objects belonging to three different structured daily activities (from Stoel-Gammon, 1989), and while she/he was describing a picture from SSI-3. The recordings lasted around an hour, and allowed the collection of a sample of at least 500 syllables. Once transcribed, the same syllables were selected for calculating the *Disfluency Profile* as well as the SSI-3 score.

The experimental design consists in predicting the persistence of the disorder from the *Disfluency Profile* scores, calculated at 9-15 months or at 16-22 months post-onset, and from the SSI-3 scores (at 16-22 months post-onset).

To this end, on the basis of the *Illinois Longitudinal Study*, as described by Yairi, Ambrose (2005, Chapter 5th), we tried to formulate some operating criteria for adapting the SLD's developmental paths of the persistent and recovered stutterers to prognostic purposes. Based on these criteria, the child candidate to recover spontaneously would be characterized by the following conditions, to be applied in sequence:

– at *n* semester from the stuttering onset, the reduction of the SLD % should be stronger (or equal) than the reduction of SLD % of the recovered subjects

reported in tab. 5.5 by Yairi, Ambrose (2005: 173). This reduction could be quantified by the ratio of the SLD percentage of the equivalent *n* semester divided by the SLD percentage at the onset;
– if the reduction rate does not decrease, the SLD percentage should be closer to the average SLD % of the recovered subjects, as reported in tab. 5.5 by Yairi, Ambrose (2005: 173) at the equivalent *n* semester from the onset, than to the average SLD % of persistent stutterers in the same table.

The use of SSI-3 as predictor of persistent stuttering, however, is more problematic, because the scores for the preschool children do not present any cut-off value able to distinguish the non stuttering children from the children with *Very Mild* stuttering (Riley, 1994). Hence, we decided to refer to the proposal of Howell, Davis (2011) which assume that, in order to be considered recovered, a child who stutters should 1) score less than 24 at the SSI-3 and 2) present a reduction of at least 2 points compared to the evaluation made at stuttering onset. To evaluate the effectiveness of the prediction, we referred to clinical standards (Meisels, 1988), according to which the instrument under evaluation must simultaneously exhibit a level of sensitivity (measurement accuracy in detecting S) and specificity (measurement accuracy in detecting NS) of at least 80%, although we are aware that such a small sample size would not formally allow this kind of analysis (Jones, Gebski, Onslow & Packman, 2002). We calculated an a priori simulation about the size of the sample needed to reach statistical power for a sensitivity analysis (Jones, Carley & Harrison, 2003), obtaining that, with a likely recovery rate of 88% (Yairi, Ambrose, 2013), a sample of size 40 participants was needed. Regrettably, this size was very far from the possibilities of our low-funded project, but nevertheless we wanted to use this analysis in order to find a clue that we were on the right way.

3. *Results*

Table 3 shows the scores of the *Disfluency Profile* (SLD) and the SSI-3 for the subjects in three different sessions (0-6 months, 9-15 months, 16-22 months post-onset). The predictions of final clinical outcome, S (stutters) or NS (non stutterers), are based on the scores of the last two sessions and were checked against the results of the structured telephone interview.

The values of the *Disfluency Profile* at the 9-15 months post-onset session are correctly predicting the outcomes for 2 S and 6 NS, while producing 4 false positives and 1 false negative (sensitivity: 66.3%, specificity: 60.0%).

Table 3 - *Individual percentages (%) of SLD at the Disfluency Profile and SSI-3 scores, at three evaluation sessions, and prediction (Pre) of persistence (S) or recovery (NS). The rightmost column shows the final clinical outcome (*treated subjects)*

Subjects	*0-6 months*		*9-15 months*		*16-22 months*				*Clinical outcome*
	SLD	SSI-3	SLD	Pre	SLD	Pre	SSI-3	Pre	
BM	4,7	23,0	4,1	S	2,3	NS	18,0	NS	NS
CA	12,0	20,0	8,8	S	6,0	S	20,0	S	NS
CG	5,5	13,0	1,3	NS	0,7	NS	10,0	NS	NS
FM	3,0	14,0	9,0	S	4,3	NS	17,0	S	NS
FMd	15,1	21,3	7,7	NS	7,9	S	20,0	S	NS
GS	4,4	12,3	2,0	NS	2,4	NS	8,0	NS	NS
MG	5,3	19,0	4,0	NS	3,5	NS	18,0	S	NS
*ML	7,0	21,0	2,8	NS	5,3	S	22,0	S	S
*RF	3,3	32,0	5,3	S	6,0	S	29,0	S	NS
SC	4,0	8,0	3,0	NS	2,0	NS	8,0	S	NS
*SL	1,9	5,2	2,2	S	14,0	S	25,4	S	S
*TA	20,0	26,0	24,0	S	10,8	S	28,0	S	S
VL	2,6	10,0	1,6	NS	0,5	NS	0,8	NS	NS
Mean	6,8	17,3	5,8		5,0		17,2		

Half a year later, the predictive power increases: the values of the Disfluency Profile correctly predict the final outcomes for 3 S and 7 NS, with 3 false positives and 0 false negatives (sensitivity: 100%, specificity: 70.0%). For this session (16-22 months after the onset) there are also the SSI-3 scores: by applying the cut-off criterion (Howell, Davis, 2011), 3 S and 4 NS are correctly predicted, while there are 6 false positives and 0 false negatives (sensitivity: 100%, specificity: 40.0%). Results are summarized in table 4.

Table 4 - *Results for the sensitivity & specificity analysis after an average interval of 41;15 (months;days) from the stuttering onset (final evaluation: September 2015). S=stutterers; NS=non stutterers*

		Actual clinical output by september 2015	
		S (3)	**NS** (10)
Disfluency profile prediction by 9-15 m. post-onset	S (6)	2 (66.6%)	4 false positives
	NS (7)	1 false negative	6 (60%)
Disfluency profile prediction by 16-22 m. post-onset	S (6)	3 (100%)	3 false positives
	NS (7)	0 false negative	7 (70%)
SSI-3 prediction by 16-22 m. post-onset	S (9)	3 (100%)	6 false positives
	NS (4)	0 false negative	4 (40%)

4. *Discussion*

The aim of the present study was to determine whether the *Disfluency Profile* or SSI-3 scores, measured at different age levels, might have potential as a clinical marker of stuttering persistence in Italian children.

The findings of the present study suggest that the predictive power of the *Disfluency Profile* at the session of 9-15 months post-onset is low, according to clinical standards, but it increases in the next six months, albeit not to the standard minimum of 80%. However this index performs better than SSI-3, which has been proposed as a predictive tool by Howell, Davis (2011). To date, there is no validated instruments for clinicians to predict the persistence of stuttering; therefore, the *Disfluency Profile* exhibits, compared to the SSI-3, the advantage of being free, less demanding (easy-to-use), less arbitrary (for instance, no need to evaluate tension on monosyllabic-word repetitions) and faster to administer and process (for instance, no need of video recording).

In conclusion, we are aware of the low statistical power of our results, and of the not "ideal" current process of verification of clinical outcome. Furthermore, for almost all the subjects currently at least 5 years have passed since the onset of stuttering, and this interval is deemed sufficient to exclude possible variations of the clinical status in the future (Yairi, Ambrose, 2005). Indeed, we have already planned to check again the clinical outcomes of the subjects by collecting updated and more reliable information directly at the subjects' home. Even so, larger studies will be essential to verify the extent to which these effects generalize across individuals and the degree to which they are driven by other factors such as the process of verification of clinical outcome and/or the attitude and capability of clinicians to work with these instruments.

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