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Improvement of verbal behavior after pharmacological treatment of developmental stuttering: a case study

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Abstract

Developmental stuttering is a disruption in normal speech fluency and rhythm. Developmental stuttering usually manifests between 6 and 9 years of age and may persist in adulthood. At present, the exact etiology of developmental stuttering is not fully clear. Besides, the dopaminergic neurological component is likely to have a causal role in the manifestation of stuttering behaviors. Actually, some studies seem to confirm the efficacy of antidopaminergic drugs (haloperidol, risperidone and olanzapine, among others) in controlling stuttering behaviors. We present a case of persistent developmental stuttering in a 24-year-old adult male who was able to control his symptoms to a significant extent after administration of risperidone, an antidopaminergic drug. Our findings show that the pharmacological intervention helped the patient improve on a set of fluency tasks but especially when the tasks involved the uttering of content words. Our results are discussed against the current theories on the cognitive and neurological basis of developmental stuttering.

1. Introduction

The World Health Organization classifies stuttering as a disruption of the normal speech rhythm, whereby the subject knows exactly what he or she wants to say but is unable to utter the intended words and sentences fluently [17]. This definition remains valid. However, in the last 10 years some experimental studies have suggested that the neurological component may play a more important role in this disorder.

Developmental stuttering is characterized by behaviorally evident sound and syllable repetition at the beginning of words, phrases, and sentences, sound prolongation, interruptions, silences or sound blocks, facial spasms and muscular tensions in the oro-facial district during speech.

Developmental stuttering usually begins in childhood between the age of 6 and 9 years and affects around 5% of the child and adolescent population to different degrees of severity. In most cases, spontaneous remission of the symptoms does occur but one percent of developmental stutterers are still affected by this problem in adulthood. Finally, male subjects are more affected than females [1].

A genetic component of the disturbance is suggested by Ambrose, Cox & Yairi [1] and Yairi, Ambrose & Cox [19].

Wu, Maguire, Riley, Lee, Keator, Tang, Fallon & Najafi [18] found a strong activation of the dopaminergic neurons in left caudate and in left amygdala in a group of stutterers vs. a control group. Such stronger activation was evident in the left insula and in the hearing cortex, too. A greater than normal dopaminergic activity in the left basal ganglia might induce a lower activity of the speech circuits in the dominant hemisphere and could partly explain the physiological mechanisms of stuttering. Actually, a finding common to many studies on the physiology of stuttering is the lower and different pattern of cerebral activation of the left hemisphere language cortex, combined with a stronger activation of the homologue areas of the right hemisphere [3]. These results allowed to hypothesize [9] that developmental stuttering might be considered as a mainly neurological dysfunction, and more specifically a dopaminergic dysfunction of the basal ganglia. This hypothesis is indirectly confirmed by the efficacy of antidopaminergic drugs such as haloperidol [11], risperidone [8] and olanzapine [6] which proved useful in controlling the stuttering symptoms. These drugs are principally presynaptic antagonists of dopaminergic receptor D2, which is largely present in human basal ganglia.

The exact functioning of antidopaminergic drugs is not completely clear [13], and it is impossible to generalize the results of these studies to suggest the efficacy of the pharmacological treatment of stuttering.

An indirect confirmation of the dopaminergic theory of developmental stuttering comes from Parkinson's Disease (PD). PD is characterized by a degeneration of dopaminergic neurons in the basal ganglia, and sometimes its first symptoms include speech difficulties [7].

Many researchers suggest that the neurotransmitters' balance as a whole (and not just a quantitative variation of a single neurotransmitter) is fundamental for the smooth execution of speech [15]. Actually, some studies report on the utility of SSRI (paroxetine) in the treatment of stuttering [4].

Finally, developmental stuttering closely resembles Tourette's Syndrome, a tic-disorder with an important obsessive-compulsive component. The resemblance between the two syndromes is evident when we analyze the secondary behaviors like repetitive or prolonged eye-blinks, jaw blocks and jaw tremors, or abnormal head and arms movements associated to dysfluencies in stuttering and typical of Tourette's Syndrome, as shown by Mulligan, Anderson, Jones, Williams & Donaldson [10].

On the other hand, but not necessarily in opposition with dopaminergic theory, Vasic and Wijnen [16] proposed a psycholinguistic theory about etiology of stuttering. The authors suggest that stuttering depend on an excessive attentive threshold level for speech.

2. Method

We present a case of developmental stuttering persisting into adulthood in a 24-year-old Italian male who, after administration of the antidopaminergic drug risperidone, could successfully reduce the symptoms of stuttering.

Risperidone is an "atypical" antipsychotic drug: its prolonged assumption causes a lower incidence of extrapiramidal adverse reactions and a lower incidence of Tardive Dyskinesia because risperidone is a D2 and 5HT-2a

antagonist. Therefore, the serotoninergic antagonism can help controlling the adverse reaction of the dopaminergic block.

2.1 Behavioral assessment

Initially, developmental stuttering was confirmed by means of the Stuttering Severity Instrument [14]. After obtaining the patient's informed consent to the treatment, including the possible risks, we obtained all the most important treatment-related biological health parameters like heart and liver functioning. Then, we have decided to operate as follows:

1. we established the patient's baseline level of stuttering on all experimental behavioral tasks;
2. 0.5 mg/d risperidone was administered for a six-week period as suggested by the literature [8];
3. At the end of the first treatment period, all experimental behavioral tasks were re-administered to investigate the efficacy of treatment;
4. The first treatment period was followed by a six-week washout period, while repetition of all experimental behavioral tasks came immediately after;
5. A second six-week drug intake period followed (same dose), and the administration of all experimental behavioral tasks was successively repeated;
6. Finally, during a washout period of 12 weeks the long-terms effects of the drug were explored and successively all experimental behavioral tasks were repeated.

2.2. Behavioral investigation measures

The following behavioral measures were administered:

- Stuttering Severity Instrument [14]: at baseline and at the end of all treatment and washout periods, a conversation sample of 150 words and a reading sample were recorded. The percentage of stuttering, the mean of the longest three blocks and the subjective evaluation of the secondary behaviors related to stuttering were computed. The degree of stuttering according to a graded scale was defined.
- Measures of verbal fluency at baseline and at the end of all treatment and washout periods: content word production, content word repetition and nonword repetition on the basis of the Italian versions of the FAS [2] and BAT [12] tests, originally designed for the assessment of aphasia deficits. We individually analyzed each test by calculating the percentage of stuttered syllables against the total number of syllables.
- Finally, secondary behaviors associated to stuttering were explored based on Mulligan & colleagues [10], at baseline and at the end of all treatment and washout periods: a subject's phone conversation of ca. 400 words with a familiar person was recorded. The video-analysis was later run at zero volume to avoid the influence of stuttered speech. The "yes or no" head movements were not counted. However, later it was decided to count all movements that were highly frequent at baseline like repetitive eye-blink, sustained left eyes, jaw jerking and sustained low head movements.

2.3. Experimental hypotheses

With reference to the available theories about the dopaminergic etiology of stuttering, we expected an improvement on language performance after the first pharmacological treatment period and a worsening of these results after the first washout period, and finally a further improvement after the second pharmacological treatment period.

3. Results

3.1. The Stuttering Severity Instrument

On the Stuttering Severity Instrument (SSI), after the baseline measures, the subject scored 34, with a percent value of 90-96, corresponding to a severe degree of stuttering.

After the first treatment period, the degree of stuttering decreased to moderate with a scale score of 22 and a percentile value between 24-40. This shows a strong improvement which was maintained during the first washout period. Stuttering was moderate with a mild worsening up to a scale score of 25 and percentile measures of 56-66. During the second drug period, the patient obtained the best results with a scale score of 18, a percent value of 5-11 and mild stuttering. Finally, after the 12-week period moderate stuttering was observed, with a scale score of 23 and 24-40 percent values (Fig. 1).

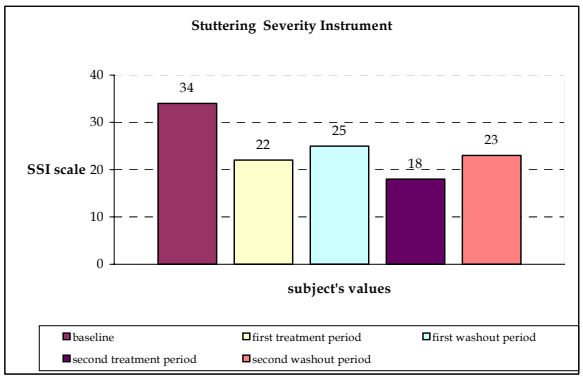


Figure 1: Scale scores on the Stuttering Severity Instrument.

The subject indicated a feeling of unprecedented easy speaking, with no muscular and jaw tensions and a new ability to manage the blocks among the major subjective sensations.

3.2. Verbal Fluency Analysis

Each of the three tasks were entered into the Logistic Regression Test. Then, all task results were compared by a simple Variance Analysis.

3.2.1. Content word production

At baseline, a Stuttered Syllable (SS) versus Total Syllable (TS) ratio of 35.13% was obtained. During the two treatment periods the SS/TS ratio was 8.24% and 6.77%, while during the washout periods it amounted to 7.39% and 7.02% (Fig. 2).

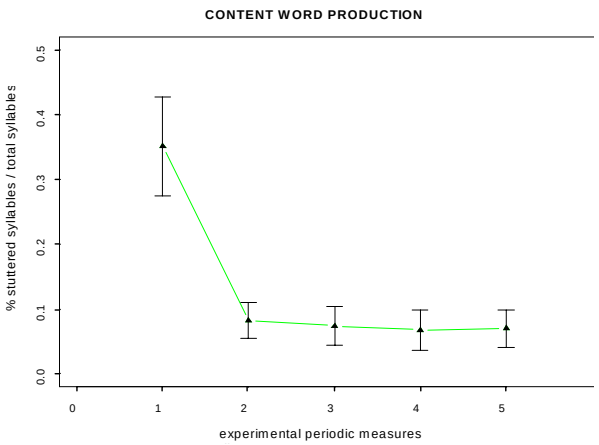


Figure 2: Content word production. 1-baseline; 2-first treatment period; 3-first washout; 4-second treatment period; 5-second washout.

The Logistic Regression Test results support all these findings in all comparisons between baseline and treatment and washout periods (z=-7.06, z=-6.73, z=-6.60, z=-6.93; p<0.001).

3.2.2. Content word repetition

On this task the patient scored 17.86% at baseline. The two six-week treatment periods demonstrate a significant improvement as shown by Logistic Regression (z=-2.86, z=-2.86; p<0.01), with 1.19% in both cases.

Significance is confirmed by the statistical analysis after the washout periods too (z=-2.85, z=-2.71; p<0.01), with SS/TS ratios of 2.38% and 3.57% (Fig. 3).

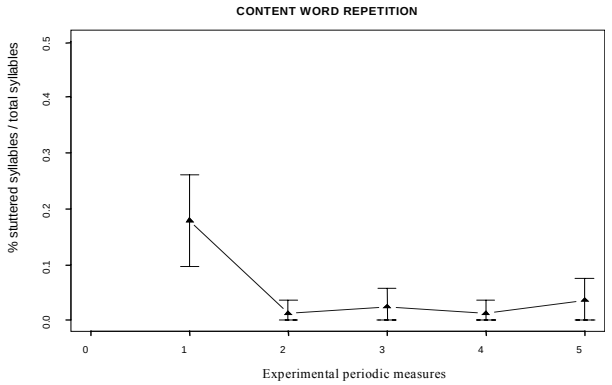


Figure 3: Content word repetition. 1-baseline; 2-first treatment period; 3-first washout; 4-second treatment period; 5-second washout.

3.2.3. Nonword repetition

On this task which is useful to determine verbal fluency, the SS/TS ratio was 9% at baseline. Statistical analysis reveals that the only two significant comparisons concern the treatment periods (z=-2.14, z=-2.14; p<0.05) with 1% of SS/TS. No significant variations were found between washout periods and baseline scores, with percentages of 6% and 3% (Fig. 4).

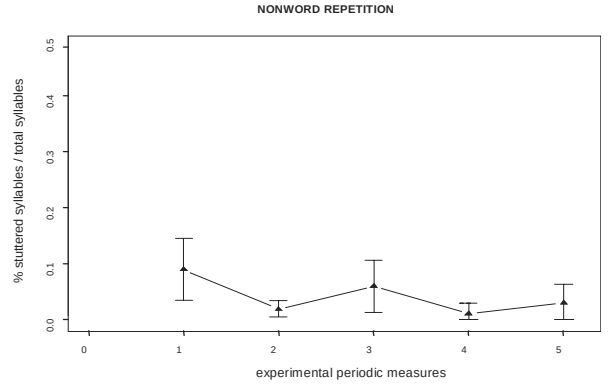
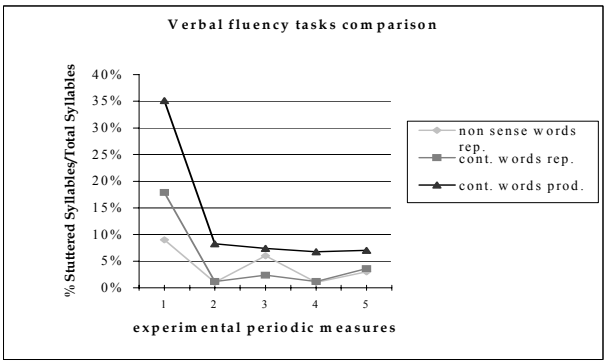


Figure 4: Nonword repetition. 1-baseline; 2-first treatment period; 3-first washout; 4-second treatment period; 5-second washout.

3.2.4. Comparison between all verbal fluency tasks

An Analysis of Variance was performed to compare all three verbal fluency tasks with the five treatment periods and verify whether the drug could have different effects on these tasks.

The statistical results are significant (F= 4.69; p<0.05) and indicate a probable different effect of risperidone on the



various verbal fluency samples (Fig. 5), where probably content word tasks obtained the most important fluency gain.

Figure 5: Verbal Fluency tasks comparison. 1-baseline; 2-first treatment period; 3-first washout; 4-second treatment period; 5-second washout.

3.3. Secondary behaviors associated to speech

A final statistical analysis was run on involuntary movements associated to normal and stuttered speech.

As previously said, only the movements that were more present at baseline were explored: repetitive eye-blink, sustained left eyes, jaw jerking and sustained low head movements (Tab. 1).

Table 1: Most relevant involuntary movements associated to speech classification.

musc. districts	Base-line	ther. per. 1	Wash-out 1	ther per. 2	Wash-out 2
Rep. eye-blinks	56	19	11	15	24
Sust. left eyes	12	4	12	2	7
Jaw jerk.	33	8	12	3	20
Sust. head mov.	16	9	15	10	14

A Two Proportions Test with single comparisons between consecutive treatment and washout periods was applied, with baseline as referent. Data interpretation is not simple, but significant results were found for repetitive eye-blinks between the second treatment period and the second washout period (χ^2=3.19; p<0.05). This suggests that the increase in involuntary movements during the second washout period is caused by the lack of risperidone. Sustained left eyes movements were significant for all comparisons (χ^2=12, p<0.0005; χ^2=17.14, p<0.00005; χ^2=4.44, p<0.05). Thus, it seems that risperidone is effective when it is taken, but not during washouts. Jaw-jerking results suggest the effectiveness of the drug only during the second treatment period, since these data are significantly different from the two washout periods (χ^2=6.99, p<0.005; χ^2=19.29, p<0.000001). Finally, sustained low head movements show significant differences between the two treatment periods and the first washout (χ^2=6, p<0.01; χ^2=4.57, p<0.05). This indicates the effectiveness of risperidone during therapy and the duration of the effects (only for this specific type of involuntary movement) after the second washout (Fig.6).

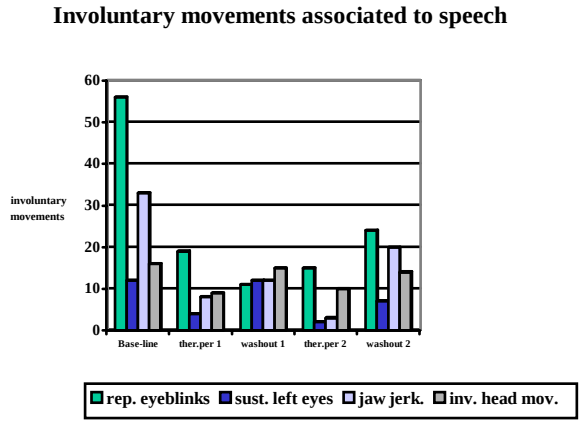


Figure 6: involuntary movements associated to speech

4. Discussion

Our findings support the hypothesis that pharmacological treatment with risperidone can help manage the typical dysfluencies of developmental stuttering. In fact, the Stuttering Severity Instrument scores demonstrate that under therapy stuttering changed from severe to moderate during the first treatment period and to mild after the second treatment period. The worsening after the washout periods is minimal because stuttering does not increase beyond the moderate level.

Verbal fluency measures suggest that both content word production and repetition and nonword repetition improved under therapy. These findings seem to confirm the findings that adult stutters produce more dysfluencies on content words [5]

Regarding the analysis of the secondary behavioral components of stuttering, the total amount of involuntary movements tends to diminish from baseline to the second treatment period but it increases sensibly after the second washout period. Thus, in this instance treatment with risperidone was effective only during its assumption, while it had a more enduring effect on verbal fluency measures and SSI.

These results allow us to confirm Mulligan and colleagues' [10] theory, which defines stuttering as a tic disorder. We could consider an excess of typical motor activity in the child, as a positive predictive factor for the development of stuttering behaviors. This, in turn, confirms the relevance of the neurological and motor component of stuttering. We may assume that risperidone can influence a cognitive component and/or a motor component of verbal fluency.

In conclusion, during treatment periods risperidone seems to be effective in controlling stuttering symptoms like dysfluencies and involuntary movements associated to stuttered speech. Most importantly, risperidone was well tolerated by the subject.

Therefore, our results confirm a possible dopaminergic etiology of stuttering that can be considered a psychomotor disorder with an important neurological component determining its manifestation.

5. References

[1] Ambrose N.G., N. J. Cox & E. Yairi 1997. The genetic basis of persistence and recovery stuttering. *Journal of Speech, Language and Hearing Research*, Vol. 40, No. 3, pp. 567-580.

[2] Benton A.L. & K. Hamsher 1989. Multilingual aphasia examination. Iowa City, IA: AJA Associates.

[3] Braun A.R., M. Varga, S. Stager, G. Schulz, S. Selbie, J.M. Maisog, R.E. Carson & C.L. Ludlow 1997. Altered patterns of cerebral activity during speech and language production in developmental stuttering. An H2(15)O positron emission tomography study. Brain, Vol. 120, Pt. 5, pp. 761-784.

[4] Costa D. & R. Kroll 2000. Stuttering: an update for phisicians. Canadian Medical Association Journal, Vol. 162, No. 13, pp. 1849-1855.

[5] Howell P., J. Au-Yeng & S. Sackin 1999. Exchange of stuttering from function words to content words with age. Journal of Speech, Language and Hearing Research, Vol. 42, pp. 345-354.

[6] N. Lavid, D.L. Franklin & G.A. Maguire 1999. Management of child and adolescent stuttering with olanzapine: three case reports. Annals of Clinical Psychiatry, Vol. 11, No. 4, pp. 233-236.

[7] Leder S.B. 1996. Adult onset of stuttering as a presenting sign in a parkinsonyan-like syndrome: a case report. Journal of Communication Disorders, Vol. 29, pp. 471-478.

[8] Maguire G.A., G.D. Riley, D.L. Franklin & L.A. Gottschalk 2000. Risperidone for the treatment of stuttering. Journal of Clinical Psychopharmacology, Vol. 20, No. 4, pp. 479-482.

[9] Maguire G.A., G. D. Riley & B.P. Yu 2002. A neurological basis of stuttering?. Neurology, Vol. 1, pag. 407.

[10] Mulligan H.F., T.J. Anderson, R.D. Jones, M.J. Williams & I.M. Donaldson 2003. Tics and developmental stuttering. Parkinsonism and Related Disorders, Vol. 9, pp. 281-289.

[11] Murray T.J., P. Kelly, L. Campbell & K. Stefanik 1977. Haloperidol in the tratment of stuttering. British Journal of Psichiatry, Vol. 130, pp. 370-373.

[12] Paradis M. 1999. Test per le afasie in un bilingue. EMF Edizioni Bologna.

[13] Rang H.P., M.M. Dale & J.M. Ritter 2001. Farmacologia. Casa Editrice Ambrosiana.

[14] Riley G. 1980. Suttering Seveirity Instrument for children and adults. Cs. Publications.

[15] Schreiber S. & C.G. Pick 1997. Paroxetine for secondary stuttering: further interaction of serotonine and dopamine. Journal of Nervous and Mental Disease, Vol. 185, No. 7 pp. 465-467.

[16] Vasic N, F. Wijnen 2001. Stuttering and speech monitoring. In DISS' 01, pp. 13-16.

[17] World Health Organization. 1977. Manual of the international statistical classification of disease, injuries and causes of death. Geneva, WHO, Vol. 1, International Classification of Disease.

[18] Wu J.C., G. Maguire, G. Riley , A. Lee, D. Keator, C. Tang, J. Fallon & A. Najafi 1997. Increased dopamine activity associate with stuttering. Neuroreport, vol. 8, No. 3, pp. 767-770.

[19] Yairi E., N. Ambrose & N. Cox 1996. Genetics of stuttering: a critical review. Journal of Speech and Hearing Research, Vol. 39, No. 4, pp. 771-784.