Cluttering in Down Syndrome

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Key Words

Cluttering · Predictive cluttering inventory · Down syndrome · Stuttering · Stammering

Abstract

Objective: To study the occurrence of cluttering in individuals with Down syndrome (DS) taking into account the wide range of symptoms that may be found in cluttering. **Patients and Methods:** Twenty-six speech language therapists, who had clients with DS among their caseload, administered the Predictive Cluttering Inventory in 76 individuals with DS aged between 3.8 and 57.3 years. **Results:** Sixty of the 76 participants (78.9%) obtained a score that classified them as a clutterer, and 13 other participants (17.1%) qualified as a clutterer-stutterer. In almost 65% of the participants, not all items had been scored; thus, compromising strict administration of the Predictive Cluttering Inventory. **Conclusion:** The results clearly illustrate the need for a consensus on the symptoms that are essential for a diagnosis of cluttering, and also indicate that an instrument like the Predictive Cluttering Inventory may not be suitable for every clinical population. Future research is necessary to determine if the disfluent speech exhibited by certain clinical subgroups (such as individuals with DS) represents a distinct pattern that is different from developmental stuttering, and does not form part of a syndrome of cluttering either. Copyright © 2008 S. Karger AG, Basel

Introduction

Cluttering is a disorder that, until some years ago, was largely neglected in the speech language pathology literature [2]. It is also a disorder that appears to be hard to define. There is no consensus on which features are essential to the disorder and which are incidental [3]. Moreover, some authors consider cluttering to be primarily a speech motor disorder, while others believe that a language impairment is at the core of the disorder [4]. An additional complication is that cluttering may coexist and perhaps even overlap with other disorders, especially stuttering [5–8].

One group of patients in whom the occurrence of cluttering has received some attention in the past are those with Down syndrome (DS). The differential diagnosis with stuttering in particular has been given some attention. According to Cabanas [9] and Weiss [6], the disfluencies demonstrated by individuals with DS represent cluttering rather than stuttering, because of the absence of secondary behaviors. Also Otto and Yairi [10], who compared the spontaneous speech samples of 19 institutionalized nonstuttering individuals with DS and 19 individuals of normal intelligence, agreed that ‘cluttering’ more correctly characterized speech in DS; their conclusion was based on the high speaking rate observed in many of their participants. Farmer and Brayton [11] con-

cluded that the disfluent speech of 6 of their 13 subjects with DS represented cluttering rather than stuttering because of poor ratings of intelligibility. Preus [12], on the other hand, who studied 47 individuals with DS in 2 Norwegian institutes for the mentally retarded, concluded that the disfluencies found in patients with DS may be classified as genuine stuttering. In one third of his participants, he observed secondary symptoms in the form of body movements, devices of masking (e.g. pretending to cough or changing word order when blocking on a sound) and other postponement and avoidance behaviors, indicating proof of a 'conscious experience of stuttering'. Preus [12] added, however, that the presence of stuttering in the speech of individuals with DS does not rule out the possibility that cluttering may also be found. Actually, only 27.7% of the participants were diagnosed as pure stutterers, whereas 12.7% were diagnosed as pure clutterers and 19.2% as combined cases. Of the total sample, 40.4% displayed neither stuttering nor cluttering.

Clearly, the diagnosis of cluttering in each of the above studies was highly dependent on a limited number of features (the absence of secondary features, a high speech rate, poor intelligibility) assumed to be characteristic of the disorder by the investigators. There is some consensus nowadays that cluttering, being a multifaceted disorder, requires a comprehensive assessment, and that a wide range of potential cluttering symptoms should be considered [3, 4, 13, 14]. One instrument that may be helpful in this respect is the Predictive Cluttering Inventory by Daly and Cantrell [1], recently presented at the Fifth World Congress on Fluency Disorders in Dublin, Ireland. This inventory, a further elaboration of the former Checklist for Possible Cluttering [15], was developed by having 60 recognized fluency specialists, educated and trained in different programs around the world, rate and rank-order 50 signs and symptoms believed to be characteristic of cluttering. The Predictive Cluttering Inventory contains 33 items covering 4 domains (pragmatics, speech-motor, language-cognition, motor coordination-writing problems; see Appendix). Each item is scored on a 5-point rating scale indicating how often the behavior is observed (1 = almost never, 2 = infrequently, 3 = sometimes, 4 = frequently, 5 = almost always), yielding a total possible score of 165. A score of 80 or more classifies a client as a clutterer, while scores from 40 to 79 are believed to indicate a diagnosis of 'clutterer-stutterer'.

In individuals with DS cluttering has, to the best of our knowledge, never been studied while taking into account the wide range of symptoms that may be found in cluttering. The primary purpose of this paper is to report the

results of a study that investigated the prevalence of cluttering in individuals with DS using the Predictive Cluttering Inventory of Daly and Cantrell [1].

Method

Twenty-six speech language therapists, who had clients with DS among their caseload, contributed to the data collection of this study. They were 25 therapists with Bachelor's degrees and 1 therapist with a Master's degree, recruited at a local symposium on DS and among members of the Vlaamse Vereniging voor Logopedisten (Flemish Organization of Speech Language Therapists). In the former case, potential participants had been addressed directly, while the other therapists were invited to participate via E-mail and/or by telephone. The invitation informed potential participants that they would have to complete a questionnaire for each of their clients with DS, but they were not told that the questionnaire was an instrument to diagnose possible cluttering, nor that the purpose of the study was to investigate the occurrence of cluttering in individuals with DS. The age of the participating therapists ranged from 22.4 to 56.2 years (mean 37.6 years). Experience with DS ranged from less than 1 year to 35 years (mean 13.2 years). The participants administered the Predictive Cluttering Inventory translated into Dutch. A total of 76 surveys of individuals with DS could be obtained, of which 51 were male and 24 were female (for 1 individual, gender was not disclosed). The age of these individuals ranged from 3.8 to 57.3 years (mean 22.8 years), with no significant age difference between the 2 gender groups (males: 3.6–57.0 years, mean 24.5 years; females: 4.6–57.3 years, mean 18.11 years; Mann-Whitney U test $Z = -1.533$; $p = 0.125$). The age distribution is shown in table 1.

Analysis and Results

An initial inspection of the 76 inventory results (table 2) revealed that for only 27 individuals (i.e. 35.5%) all the items had been scored. The overall scores of these 27 individuals ranged from 66 to 155 (mean 107.4, SD 23.7; median 103). Twenty-four of them obtained a score of 80 or higher, and thus were classified as clutterers. The remaining 3 individuals obtained a score between 40 and 79, and were classified as stutterer-clutterers. Forty-nine of the 76 returned inventories (64.5%) were incomplete, with the number of unscored items ranging from 1 to 27 (mean 6.8, median 3). In spite of the fact that their inventories were incomplete, 36 individuals still obtained a high enough score to be classified as a clutterer (i.e. >80) and 10 individuals to be classified as a stutterer-clutterer. Only 3 individuals obtained a score of less than 40; they were the ones with 27, 26 and 23 unscored items. The proportion of individuals classified as a clutterer (either with a complete or an incomplete inventory) was slightly high-

Table 1. Age distribution of the individuals with DS

Age, years	Number of individuals
<10	15
10–20	25
21–30	9
31–40	16
41–50	7
>50	3

Table 2. Number of respondents classified as a clutterer, clutterer-stutterer or other

Complete inventories ^a			Incomplete inventories ^b		
clutterer	clutterer-stutterer	other	clutterer	clutterer-stutterer	other
24	3	0	36	10	3

^a n = 27, 35.5%. ^b n = 49, 64.5%.

er in the female participants than in the male participants [21/24 (87.5%) vs. 38/51 (74.5%), respectively], but not at a statistically significant degree ($\chi^2 = 1.641$; $p = 0.200$).

An analysis of all 76 inventories showed that item scores ranged from 1 to 5 for all items but 1 (for item 11, scores ranged from 2 to 5). Means ranged from 2.3 to 4.1 (table 3).

An analysis of the responses for the individuals who were classified as clutterers showed that across subjects the highest scores (mean >4) had been assigned to item numbers 22 (poor language formulation, poor story-telling; sequencing problems), 11 (articulation errors), 27 (improper linguistic structure; poor grammar; syntax errors), 2 (lack of awareness of own communication errors or problems) and 7 (does not repair or correct communication breakdowns), in order of high to low. The 5 lowest scores (mean <2.4) were observed for item numbers 30 (writing includes omission or transposition of letters, syllables or words), 17 (initial loud voice trailing off to unintelligible murmur), 19 (repetition of multi-syllabic words and phrases), 16 (variable prosody, irregular melody or stress) and 15 (speech rate progressively increases, i.e. festinating).

An analysis of the incomplete inventories showed that the section 'motor coordination-writing problems' was most often incomplete, both in terms of individuals with

Table 3. Mean scores for each of the 33 items and the number of individuals each score was based on

Item	Mean	Number of individuals
1	3.8	76
2	4.1	76
3	3.2	68
4	3.8	70
5	3.6	73
6	2.5	72
7	3.9	76
8	3.4	70
9	3.7	72
10	2.6	70
11	4.1	71
12	3.4	71
13	3.6	72
14	2.9	69
15	2.3	66
16	2.5	59
17	2.4	62
18	2.7	61
19	2.3	63
20	2.6	64
21	3.2	71
22	4.0	71
23	3.9	68
24	3.0	64
25	3.0	63
26	2.9	61
27	4.0	69
28	3.7	72
29	4.0	50
30	3.9	35
31	3.8	50
32	2.6	61
33	3.0	59

missing scores and in terms of the absolute number of unscored items across subjects. The most completed section appeared to be 'pragmatics' (table 4). The items that across subjects remained most often unscored were number 29 [poor motor control for writing (messy); unscored 26 times], 30 (writing includes omission or transposition of letters, syllables, or words; unscored 41 times) and 31 (oral diadochokinetic coordination below expected normative levels; unscored 26 times).

A comparison of the completeness of the inventories of the participants according to age, distinguishing between 6 different age groups (table 5), yielded a significant difference (Kruskal-Wallis test, $\chi^2 = 23.531$, d.f. = 6; $p < 0.001$). As table 5 shows, it was especially in the youn-

Table 4. Completeness of the inventories: number of respondents with incomplete inventories and total number of unscored items (across participants) for each section

Section	Respondents with incomplete inventories	Total number of unscored items
Pragmatics	15	37
Speech-motor	20	102
Language-cognition	20	72
Motor coordination-writing problems	47	123

gest group (<10 years) that inventories remained incomplete. Further comparison of the results of the participants under age the of 10 years with those aged over 10 years indeed showed that in the former group on average significantly more items had remained unscored (11 vs. 2.75, respectively; Mann-Whitney U test, $Z = -3.771$; $p < 0.001$). Apparently, gender played a role too. The proportion of male participants with an incomplete inventory was significantly higher than the proportion of female participants [37/51 (72.5%) vs. 11/24 (45.8%), respectively; $\chi^2 = 5.056$, $p = 0.025$]. On the other hand, when looking at which items were most often not scored, the 2 gender groups showed a moderate correlation (Spearman rho = 0.595; $p < 0.001$).

Discussion

The findings of the present study are quite ambiguous. On the one hand, the results would seem to be in agreement with previous reports [6, 9, 10], and confirm the view that cluttering symptoms are common among those with DS. On the other hand, results of this study raise 2 major concerns: (1) 'cluttering' as a clinical entity, and (2) the validity of the Predictive Cluttering Inventory as an instrument to aid in the diagnosis of cluttering. The proportion of participants with a score indicative of cluttering was extremely high. Sixty of the 76 participants (i.e. 78.9%) obtained a score that classifies them as a clutterer and 13 other participants (i.e. 17.1%) qualified as a clutterer-stutterer. For comparison, the study of Preus [12] with 47 individuals with DS found only 12.7% were pure clutterers and 19.2% combined (cluttering/stuttering) cases. Our figures are all the more remarkable as the individuals in our study were unselected for the presence of disfluencies in their speech.

Table 5. Completeness of the inventories according to age: number of unscored items for each of the 6 age groups

Age, years	Number of unscored items			
	mean	minimum	maximum	SD
<10	11.0	0	27	10.1
10–20	1.0	0	8	2.0
21–30	3.4	0	14	5.2
31–40	5.5	0	17	6.1
41–50	2.0	1	3	0.8
>50	2.3	0	6	3.2

It is possible that these elevated figures represent a sampling error, and that the majority of participants in our study were indeed clutterers. A more likely explanation, however, is that these figures reflect the definitional problem that currently still surrounds cluttering. In the Predictive Cluttering Inventory, certain deficits in the domain of pragmatics and language-cognition are included as symptoms of cluttering. An analysis of the responses for the individuals who were classified as a clutterer showed that 4 of the 5 items with the highest average scores (namely item numbers 2, 7, 22 and 27) belong to the domains of pragmatics and language-cognition. Furthermore, pragmatics was also the domain with the lowest number of unscored items, followed by the language-cognition domain. It is presently unclear to what extent language and pragmatic problems are really implicated in the diagnosis of cluttering. St. Louis et al. [3], for instance, did not include language difficulties and pragmatic problems in their working definition of cluttering. Interestingly, Preus [12] pointed out that his findings did not apply to individuals with DS who have a less-developed language proficiency than those in his study.

The high number of clutterers found in our sample with the high number of incomplete inventories is a somewhat vexing finding. In almost 65% of the participants not all items had been scored; thus, compromising strict interpretation of the Predictive Cluttering Inventory. The section on 'motor coordination-writing problems' contained the 2 most frequently unscored items overall [number 29: poor motor control for writing (messy); number 30: writing includes omission or transposition of letters, syllables, or words]. The latter results are of course not totally unexpected. Although many individuals with DS can attain useful levels of reading and writing skills, it is a fact that the development of handwriting and correct spelling is often problematic [16]. It is

difficult to explain why there were proportionally more males than females with incomplete inventories.

In the current study, gender differences in the occurrence of cluttering were not observed. The proportion of individuals classified as a clutterer was slightly higher in the female participants than in the male participants, but not at a statistically significant level. This is at odds with what has been reported in previous studies, and also contradicts the professional opinion about the sex ratio in cluttering. Arnold [17], for instance, reported a male to female ratio in cluttering of 4 to 1, just like in stuttering. Surveys in the USA by St. Louis and Hinzman [18] and in the UK by St. Louis and Rustin [19], involving 156 and 130 speech language pathologists, respectively, have shown that professionals agree that there is a male to female preponderance in cluttering, without agreement, though, about the exact ratio (6.1 to 1 in the former study versus 3.0 to 1 in the latter). On the other hand, the absence of a gender difference is in line with the findings of an investigation of public awareness of cluttering and stuttering by St. Louis and McCaffrey [20]. The 57 lay persons involved in the latter study reportedly knew far more males than females who stuttered, but as many females as males who clutter. Figures about the sex ratio are important as indicators of a possible genetic origin of cluttering, but evidently more research is necessary about this aspect.

The results of the present study clearly illustrate the need for a consensus on the symptoms that are essential for a diagnosis of cluttering. In its present form, the Predictive Cluttering Inventory covers a wide range of potential cluttering symptoms. On the one hand, this is commendable, but, on the other hand, this approach ignores the fact that certain characteristics have a higher diagnostic value and are more agreed upon by experts to be indicative of cluttering than others. A possible solution would be to develop weighted scores. The ranking of items found by Daly and Cantrell [1] could perhaps serve as a starting point.

Results also indicate that an instrument like the Predictive Cluttering Inventory in its present form may not be suitable for every population. Perhaps the development of a questionnaire specifically designed for administration in individuals with DS should be considered. Most critical in the development of such a specific instrument would be, in our opinion, the selection of items that are relevant for this population. The results of the present study might be used as a guide in this item selection. It is clear, for instance, that items pertaining to writing (items 29 and 30) are not really meaningful for most individuals

with Down syndrome. Alternatively, one could try to develop separate norms for each of the 4 domains (pragmatics, speech-motor, language cognition, motor coordination-writing problems), so that partial scoring and interpretation becomes possible in accordance with individual capabilities. Furthermore, it would seem necessary to develop different versions of the Predictive Cluttering Inventory for different age groups. The number of unscored items, for instance, was significantly higher in the participants under the age of 10 years than in those over the age of 10 years. This need to adapt for different age groups holds, most likely, not only for the population of DS.

Future research may also want to reckon with the possibility that the disfluent speech exhibited by certain clinical subgroups, such as individuals with DS, represents a distinct pattern that is different from developmental stuttering, and does not form part of a syndrome of cluttering either, but nonetheless includes some cluttering characteristics. In this respect, the notion of 'cluttering spectrum behavior' as proposed by Ward [4] may be interesting. The author proposes to use this term as a descriptor for speakers who display some cluttering characteristics, but for whom a diagnosis as a 'clutterer' is less certain. One group of speakers mentioned in particular are those who show elements of other speech language disorders that cannot be ascribed with certainty to cluttering. Indeed, individuals with Down syndrome would seem to fit well into this category.

Finally, the Predictive Cluttering Inventory in its present form applies a definite cut-off score of 80, above which cluttering is diagnosed and below which a stuttering-cluttering combination is diagnosed. Given the fact that there is still much controversy about the scope of behaviors that diagnose cluttering, one may wonder if such a definite cut-off score is tenable. Also, we know of no experimental studies to confirm the interpretation that individuals with scores between 40 and 79 always present with an additional stuttering component. In summary, it currently seems more indicated to conceive of the Predictive Cluttering Inventory as an adjunct in the process of diagnosis of cluttering, rather than as a genuine diagnostic tool.

Appendix: The Predictive Cluttering Inventory by Daly and Cantrell (2006)

Pragmatics

1. Lack of effective self-monitoring skills
2. Lack of awareness of communication errors or problems
3. Compulsive talker: verbose; tangential; word-finding problems
4. Poor planning skills; misjudges effective use of time
5. Poor social communication skills; inappropriate turn-taking; interruptions
6. Does not recognize or respond to listener's visual or verbal feedback
7. Does not repair or correct communication breakdowns
8. Little or no excessive effort observed during disfluencies
9. Little or no anxiety regarding speaking; unconcerned
10. Speech better under pressure (improves short-term with concentration)

Speech-motor

11. Articulation errors
12. Irregular speech rate; speaks in spurts or bursts
13. Telescopes or condenses words
14. Rapid rate (tachylalia)
15. Speech rate progressively increases (festinating)
16. Variable prosody; irregular melody or stress pattern
17. Initial loud voice trailing off to unintelligible murmur
18. Lack of pauses between words and phrases
19. Repetition of multisyllabic words and phrases
20. Coexistence of excessive disfluencies and stuttering

Language-cognition

21. Language is disorganized; confused wording; word-finding problems
22. Poor language formulation; poor story-telling; sequencing problems
23. Disorganized language increases as topic becomes more complex
24. Many revisions; interjections; filler words
25. Seems to verbalize before adequate thought formulation
26. Inappropriate topic introduction, maintenance or termination
27. Improper linguistic structure; poor grammar; syntax errors
28. Distractible; poor concentration; attention span problems

Motor coordination-writing problems

29. Poor motor control for writing (messy)
30. Writing includes omission or transposition of letters, syllables or words
31. Oral diadochokinetic coordination below expected normed levels
32. Respiratory dysrhythmia; jerky breathing pattern
33. Clumsy and uncoordinated; motor activities accelerated or impulsive

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