

NOTE

PURE DYSGRAPHIA WITH RELATIVE SPARING OF LOWER-CASE WRITING

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INTRODUCTION

Since the early 1980s writing disorders have been studied within information processing frameworks. Cognitive analyses of dysgraphia have often been based upon models of writing that propose some "central" linguistic processes and some "peripheral" processes related to the actual motor output and specific for written output (Ellis, 1982; Margolin, 1984).

It has been proposed that at the end of the "central" spelling routes a short-term retention buffer is aimed at storing orthographic representations of words and nonwords alike, before the actual realization of output. This so-called Graphemic Output (orthographic) Buffer (GOB) would also be implied in oral spelling. Beyond the graphemic buffer, "peripheral" processes are devoted to transforming the orthographic string into graphic motor programs specifying the sequences of movements necessary for handwriting. A specific impairment of the GOB would imply quantitatively and qualitatively similar graphemic errors in all writing tasks (delayed copy, naming, writing to dictation) and in oral spelling. In fact, such a pattern has been reported in the literature (Caramazza, Miceli, Villa et al., 1987; Posteraro, Zinelli and Mazzucchi, 1988; Hillis and Caramazza, 1989; Caramazza and Miceli, 1990).

Diagnostic criteria for selective impairments of the GOB can be summarized as follows (Hillis and Caramazza, 1989): (1) similar patterns of spelling errors for words and nonwords; (2) number of errors increasing as a function of item length; (3) no effect of lexical factors; (4) no semantic or morphological errors; (5) spelling errors consisting of deletions, insertions, substitutions and transposition of letters which may lead to the production of graphemically implausible nonwords.

Recently, some detailed observations have also been reported upon deficits of "peripheral" processes in handwriting (e.g., Baxter and Warrington, 1986; Papagno, 1992). Some of these patients, whose oral spelling was always correct, showed double dissociations between lower- and upper-case handwriting style (De Bastiani and Barry, 1986; Patterson and Wing, 1989; Kartsounis, 1992). In these cases, it was postulated that the selective impairment occurred in "peripheral" writing processes, where lower- and upper-cases style can be represented by independent graphic motor codes.

In the present paper we describe a patient affected by acquired pure dysgraphia with a relative sparing of lower-case writing. In this case the dissociation between upper- and lower-case writing does not seem related to defects in "peripheral" writing processes.

CASE REPORT

D.V.M. was a 72-year-old right-handed housewife with 7 years of education. Her native language was Italian. In June 1991, she sustained a left ischemic cerebral lesion whose clinical signs were a right homonymous hemianopia and slight weakness of right arm. CT scan showed a left parietal-occipital ischemic lesion, with enlargement of left occipital ventricular horn (Figure 1). Doppler fluximetry showed marked stenosis of left common carotid artery. D.V.M.



Fig. 1 - CT scan.

complained of mild amnesic and moderate writing disturbances. She was evaluated from October to December 1991. She was well-oriented and cooperative. The patient was given some neuropsychological tests from a standardized Italian battery (Spinnler and Tognoni, 1987). Immediate verbal memory (Bysyllabic word span=4), memory for complex verbal material (Story test=10.3/16), constructional, oral and limb praxis were in the normal range for age and education. She obtained a poor score at Corsi's visuospatial immediate memory task (3). Her Verbal I.Q. was in normal range (95).

Speech-language Evaluation

D.V.M. did not show overt aphasic disturbances. She performed flawlessly on tasks for spontaneous speech and naming, oral and written comprehension, repetition and reading aloud, comprised in an Italian standardized battery (Basso, Capitani and Vignolo, 1979). She could

TABLE I
Written Tasks of Standard Battery (scores expressed in percentages)

	Lower-case	Upper-case
Written naming	85	40
Writing to dictation		
Letters (10)	100	100
Syllables (10)	100	100
Words (10)	60	40
Nonwords (5)	80	40
Sentences (5)	40	0
Word copy (10)	80	60
Letter transposition		
Upper- to Lower-case (21)	100	—
Lower- to Upper-case* (21)	—	100

* This task is not comprised in the battery.

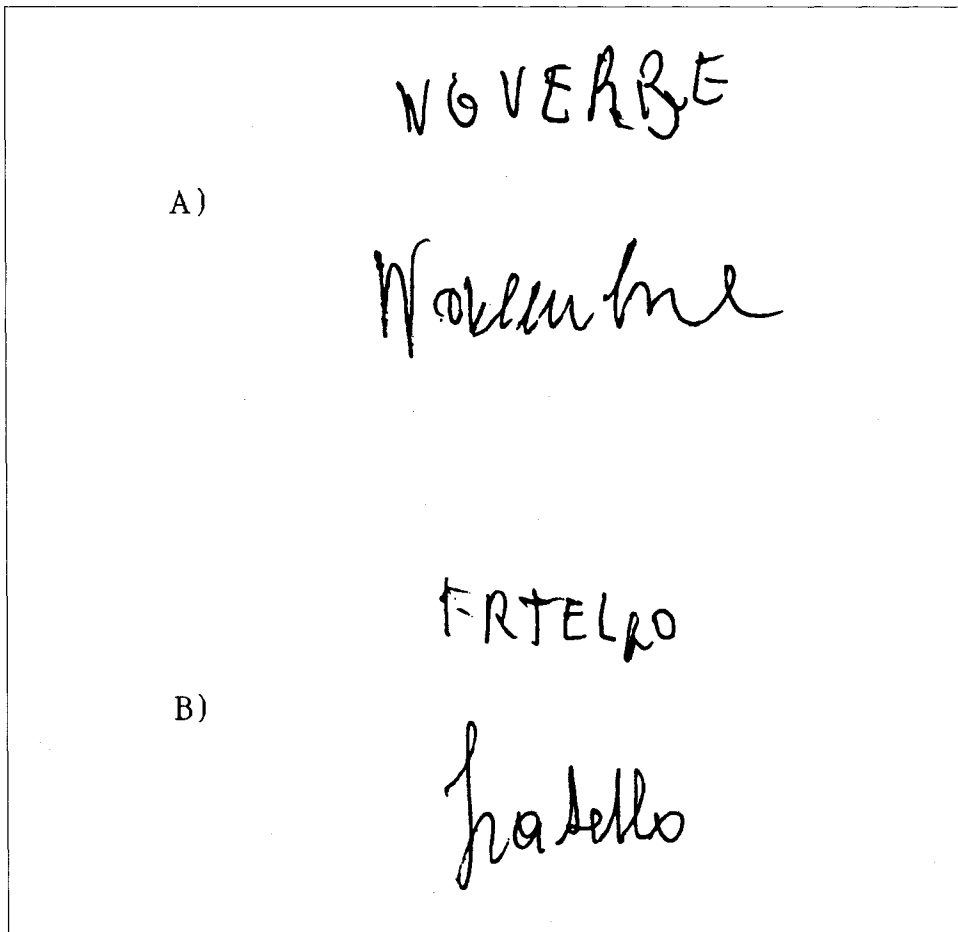


Fig. 2 - Examples of D.V.M.'s upper- and lower-case writing: A) delayed copy ("novembre" - november - written as "noverbe" in upper-case and correctly in lower-case); B) writing to dictation ("fratello" - brother - written as "frtello" in upper-case and correctly in lower-case).

sign her name but made many errors in writing tasks, and she reported more difficulties when writing upper-case letters. For example, when asked to write a letter to her husband, she managed to write two sentences in lower-case script, but refused to write in upper-case. For this reason, we asked her to perform standard writing tasks both in lower- and upper-case format. Results are shown in Table I.

D.V.M. wrote single letters and CV syllables without errors, and correctly transposed single letters from upper-case to lower-case and viceversa. By contrast, she made many errors in copying and in writing words and nonwords spontaneously and to dictation, with a better performance with lower-case format (Figure 2).

This picture suggested that D.V.M.'s impairment was neither in the selection nor in the actual motor output of single letters, but in the production of letter strings, especially in upper-case.

TABLE II
Correct Responses at Writing to Dictation and Spelling

	Lower-case	Upper-case	Spelling
Words (30)	27	17	10
Nouns (10)	10	7	5
Verbs (10)	8	5	2
Funcctors (10)	9	5	3
Bisyllabic (15)	14	9	9
Trisyllabic (15)	13	8	1
Nonwords (25)	21	18	19
Monosyllabic (15)	15	14	15
Bisyllabic (5)	3	2	3
Trisyllabic (5)	3	2	1

Special Writing Evaluation

In order to investigate D.V.M.'s difficulties in detail, we compared lower- and upper-case writing in several tasks (writing to dictation, copying and written naming) on different lists of words and nonwords. In particular, we tried to confirm discrepancies between lower- and upper-case writing and to identify the source of her spelling disorder. Part of the material used in this study was taken from an Italian battery for the cognitive analysis of language disorders (Miceli, Laudanna and Burani, 1990).

Test 1: Word and Nonword Writing to Dictation and Oral Spelling

The patient was presented with a list of 30 words, balanced for length and grammatical class, and with a list of 25 pronounceable nonwords, 2- to 8-letter long. During the same session, the patient first wrote the list in lower-case format, then in upper-case and finally she was asked to spell item orally. Results are given in Table II.

D.V.M. spelled words in lower-case significantly better than in the other modalities tested ($\chi^2 = 20$, d.f. = 2, $p = 0.001$). Upper-case writing and oral spelling did not differ from each other ($\chi^2 = 3.3$, d.f. = 1, $p = 0.7$), although the patient made more errors during spelling. Grammatical class did not affect either upper-case writing or spelling, while spelling showed a clear effect of stimulus-length (D.V.M. spelled correctly only one long word).

The few lower-case errors were single graphemic errors (1 insertion and 1 deletion) and one double error (1 deletion + 1 substitution), all resulting in nonwords. In upper-case writing and oral spelling, errors consisted of single or multiple letter deletions, substitutions, insertions and transpositions. Responses resulted in pronounceable and nonpronounceable nonwords. One deletion in oral spelling resulted in a real word ("puniti" – punished – was spelled as "punti" – points).

In nonwords writing, D.V.M. showed a similar accuracy in all three spelling tasks ($\chi^2 = 1$, d.f. = 2, $p = 0.5$). She made only 1 error with 2-letter items (in upper-case writing) but produced many errors with longer stimuli: length had a significant effect in all tasks ($\chi^2 = 7.2$, 8.4 and 14 for lower-, upper-case and spelling, d.f. = 2, $p < 0.02$, 0.01, and 0.001 respectively).

Also with nonwords, D.V.M.'s errors in all spelling tasks consisted of single and (rarely) multiple graphemic errors (mainly substitutions and deletions) resulting in pronounceable and nonpronounceable nonwords. A quantitative error analysis will be reported below.

Test 2: Word and Nonword Writing to Dictation and Copying

The patient was asked to write to dictation in lower- and upper-case, and to copy from the model in upper-case letters a list of 30 words, balanced for frequency and length, and 30 nonwords, balanced for length. D.V.M. read all stimuli flawlessly. Results are given in

TABLE III
Correct Responses at Word and Nonword Writing to Dictation and Copy

	Lower-case	Upper-case	Copy
Words (30)	29	17	21
High-Frequency (15)	15	7	10
Low-Frequency (15)	14	10	11
4-letter (6)	6	6	6
5-letter (6)	6	4	6
6-letter (6)	5	3	4
7-letter (6)	6	3	4
8-letter (6)	6	1	1
Nonwords (30)	22	16	23
4-letter (6)	5	5	6
5-letter (6)	5	4	6
6-letter (6)	5	4	5
7-letter (6)	5	2	3
8-letter (6)	2	1	3

Table III. The first relevant finding is that D.V.M.'s accuracy in upper-case copying was similar to that observed in upper-case writing to dictation. Moreover, D.V.M. made only one error in lower-case word writing to dictation. Performance on this task was significantly better than that on upper-case writing ($\chi^2 = 13.4$, d.f. = 1, $p = 0.0002$). Very good performance on lower-case spelling prevented the study of frequency and length. Upper-case writing ($\chi^2 = 10.5$, d.f. = 4, $p = 0.03$) and copy ($\chi^2 = 14.5$, d.f. = 4, $p < 0.005$) were significantly affected by stimulus length, but not by word frequency (upper-case writing: $\chi^2 = 1.2$, d.f. = 1, $p = 0.2$).

In nonword writing, accuracy did not differ significantly ($\chi^2 = 2.5$, d.f. = 1, $p = 0.1$) between lower- and upper-case format. Again, D.V.M.'s performance was poorer on longer items, but not significantly so for the three modalities tested.

Lower-case word writing to dictation was significantly better than nonword writing ($\chi^2 = 6.4$, d.f. = 1, $p < 0.01$), while neither upper-case writing nor copying differed for words and nonwords.

In lower-case word writing D.V.M. made only one literal substitution that gave rise to a nonpronounceable word ("membri" – members – written as "membni"). With nonwords, lower-case errors were predominantly letter substitutions resulting in some words, and in legal and illegal nonwords. Errors in upper-case writing and copying were similar for words and nonwords (single or multiple letter deletions, substitutions, insertions and transpositions). Responses in upper-case nonword writing and copying consisted mainly in (pronounceable or illegal) nonwords; only one response was a real word ("came" written as "cane" – dog).

Test 3: Delayed Copying

We asked D.V.M. to copy in lower-case stimuli written in upper-case and viceversa. We used 10 nonwords and 10 words, of different length, to be copied after an unfilled 10-sec interval. D.V.M. made no errors when copying in lower-case format. In upper-case copying, she achieved similar scores for words (6/10) and nonwords (5/10). Performance was affected by length (words: 85% correct for short stimuli versus 25% correct for long stimuli; nonwords: 75% versus 33%). Errors were similar to those observed in the other tasks.

Test 4: Written Naming

The patient was presented with two sets of line drawings: the first comprised 22 objects whose names were balanced for length and frequency, the second comprised 22 actions balanced for frequency. She was asked to write each list in lower-case first and then in upper-case.

TABLE IV
Error in Lower- and Upper-case Handwriting and in Oral Spelling

	Lower-case		Upper-case		Oral spelling	
	Words	Nonwords	Words	Nonwords	Words	Nonwords
Total	12/114	12/55	65/144	33/85	22/30	6/25
Single graphemic errors	8 (67%)	10 (83%)	47 (72%)	23 (70%)	14 (66%)	4 (66%)
Insertions	2	1	8	2	0	0
Deletions	3	2	15	7	7	2
Transpositions	0	0	6	3	5	1
Substitutions	3	7	18	11	2	1
Multiple graphemic errors	4 (33%)	2 (17%)	18 (28%)	10 (30%)	7 (33%)	2 (33%)
Responses consisting in:						
Words	0	5 (42%)	0	1 (3%)	1 (4%)	0
Nonwords	9 (75%)	6 (50%)	49 (75%)	26 (79%)	11 (52%)	4 (66%)
Illegal	3 (25%)	1 (8%)	16 (25%)	6 (18%)	9 (42%)	2 (33%)
Refusals	0	0	0	0	1 (4%)	0

Accuracy was similar for objects and actions. Once more, lower-case spelling was significantly better than upper-case writing in both object (86% versus 40%; $\chi^2=9.9$, d.f. = 1, $p=0.001$) and action naming (77% versus 40%; $\chi^2=7.5$, d.f. = 1, $p=0.006$). In lower-case writing, the effect of frequency was significant when object and action naming were considered together (95% correct for high-frequency items versus 40%; $\chi^2=5.8$, d.f. = 1, $p=0.01$), while length had no effect ($\chi^2=1.3$, d.f. = 1, $p=0.2$). By contrast, upper-case writing was significantly affected by stimulus length (57% correct for short words versus 12%; $\chi^2=4.1$, d.f. = 1, $p=0.04$), but not by word frequency.

Errors were graphemic in nature in both lower- and upper-case writing (mainly substitutions and deletions, but also insertions and transpositions) and responses resulted in pronounceable and nonpronounceable nonwords.

Error Analysis

D.V.M. showed a peculiar writing pattern. She showed similar numbers of errors in upper-case writing to dictation, copying and naming, and in oral spelling, with similar performance on words and nonwords. On the other hand, lower-case writing was more accurate for familiar stimuli (words versus nonwords; high-frequency versus low-frequency words), while item length did not affect *word* writing. These observations could suggest that lower- and upper-case writing relied upon different mechanisms. However, D.V.M. produced similar graphemic errors in lower- and upper-case handwriting and in oral spelling, as shown by the cumulative analysis of errors in all the tasks (Table IV).

The few lower-case errors on words demonstrated the same distribution as upper-case errors: they were represented mainly by single graphemic deletions or substitutions, resulting in legal or illegal nonwords. In lower-case writing of nonwords, D.V.M. produced a large number of single graphemic substitutions, often resulting in real words (5/12 errors; e.g. "sterzo" became "sterzo" — steering wheel). In oral spelling the error pattern was similar, although insertions were absent and deletions were very frequent. The distribution of spelling errors as a function of letter position also showed relevant findings. Cumulating words and nonwords processing tasks, which shared the same features, errors were most frequent in the relative medial position in all modalities (Figure 3).

These findings demonstrate that errors in lower- and upper-case handwriting and in oral spelling not only were similar but also respected the same spatial distribution. Thus, the source of errors was very likely to be the same.

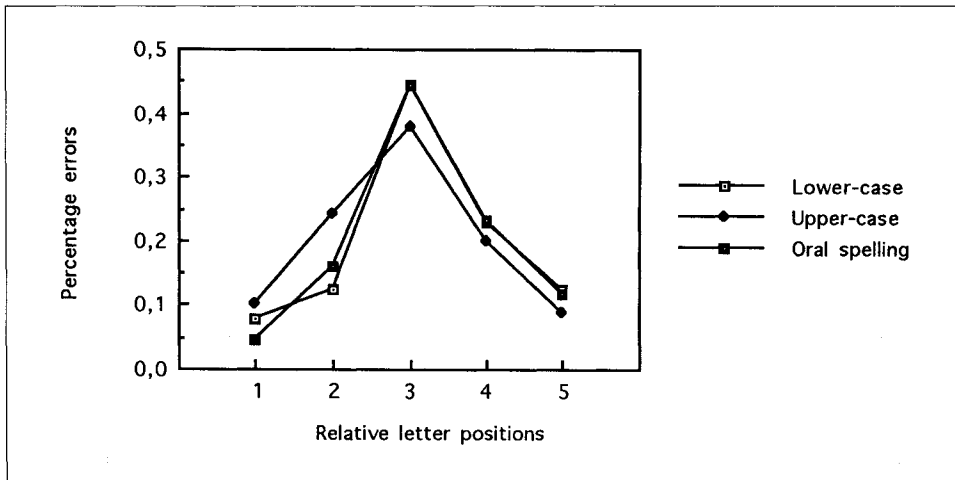


Fig. 3 – Incidence of errors as a function of relative letter position (computed according to Wing and Baddeley, 1980).

DISCUSSION

A selective impairment of lower- or upper-case handwriting, due to a selective damage of peripheral writing processes, has been reported in a few patients with difficulties in letter formation and unimpaired oral spelling (Patterson and Wing, 1990; Kartsounis, 1992).

Our patient did not present difficulty in retrieving the correct motor programs when writing single letters or longer strings: letters were clearly legible and easily identifiable on all occasions. We did not perform a systematic temporal analysis of written output (as suggested by Patterson and Wing, 1990). However, our patient appeared to spend comparable times in tracing lower- and upper-case letters, without relevant hesitation. D.V.M. repeatedly declared that she preferred lower-case writing because she was more familiar with it and especially because she was aware of the greater number of errors in upper-case writing. She often became aware of her errors either while she was writing her responses or after completing them. Then, she repeated the stimulus accurately, but corrected written responses only occasionally.

The lack of alterations of letter formation and the impairment of oral spelling ruled out a “peripheral” account for this patient’s defect. Instead, D.V.M.’s writing pattern suggested an interaction of central and peripheral writing processes: lower-case writing was relatively preserved, although a single cognitive impairment likely induced equivalent errors in the three modalities (lower-, upper-case and oral spelling).

D.V.M.’s upper-case writing showed qualitatively and quantitatively similar errors in writing to dictation, copying and confrontation naming. Lexical status of items did not affect performance; word and nonword copying were impaired as well; errors were always graphemic in nature and were a function of item length with a higher incidence in central positions. Oral spelling showed a qualitatively and quantitatively similar error pattern.

Thus, D.V.M.’s functional impairment appeared to be restricted to a component devoted to processing graphemic representations, independent of input modality, common to oral and written spelling, and shared by lexical and phonological mechanisms. These findings met the criteria outlined in the introduction for a GOB defect.

In the present case the GOB impairment would not result in a limited retention of information (Katz, 1991), nor would it be affected by attentional gradients (Hillis and Caramazza, 1989). D.V.M., as well as normal subjects (Wing and Baddeley, 1980), produced most errors in the medial position of letter strings, a pattern typical of patients with GOB impairments (Caramazza et al., 1987; Posteraro et al., 1988).

A GOB defect should not imply differences between lower-case and upper-case

handwriting because graphemic representation held in this post-lexical buffer are then treated in the same way. This assumption has not been tested specifically in other patients: some authors compared handwriting and typing, without relevant differences (Miceli, Silveri and Caramazza, 1985), while others studied only one response modality (e.g., Posteraro et al., 1988). The fact that D.V.M. was more accurate in writing familiar words in lower-case would be inconsistent with a GOB impairment. However, our analysis showed that the error pattern in lower-case script was similar to that observed in the two other modalities, and matched that expected in GOB defects on qualitative grounds. So, available data suggested that the GOB was the source of graphemic errors in all tasks, but that it was involved to a different extent in different output modalities. To account for the lexical facilitation in lower-case, one could assume that D.V.M. relied less on the GOB when spelling in lower-case because this is a simple and more automatized task.

In conclusion, D.V.M. showed a previously unreported writing pattern. Even though lower-case writing of familiar stimuli was relatively spared, errors showed a similar distribution across tasks. We suggest that a GOB defect could account for such findings, because lower-case word writing posed a minimum load on the buffer. Although admittedly ad-hoc, such an interpretation raises the issue of the equivalence of different handwriting styles, suggesting possible interactions of central and peripheral writing processes. Systematic investigations of performance on different spelling tasks in dysgraphic patients are necessary for addressing these theoretical aspects of writing models.

ABSTRACT

We describe the cognitive analysis of a patient with acquired pure dysgraphia. She presented a peculiar dissociation between lower- and upper-case handwriting: lower-case writing was relatively spared and showed a significant superiority of words versus nonwords. Upper-case writing and oral spelling did not show lexical effects, but were affected by item length. In all modalities errors consisted mainly of single graphemic substitutions, deletions, insertions and transpositions, resulting in legal or illegal nonwords, and showed a similar distribution across letter positions. These findings were suggestive of an impairment of the graphemic output buffer, which however revealed itself to different degrees in the two handwriting styles.

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