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Where do Perseverations Come From?

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Abstract

RB, a patient with probable Alzheimer's disease, makes continuous perseverations of single letters when writing (e.g. *fruit* → fr*uu*it), particularly on high frequency letters. An analysis of her errors reveals that her perseverations do not reflect letter substitutions or transpositions, nor do they suggest difficulty with geminates. No continuous perseverations were found in oral production, in graphic and simple motor tasks, and in oral spelling. RB's data do not support an attention deficit as the basis of her continuous perseverations. It is proposed that a deficit at the level of abstract letter representations is the source of RB's perseverations. The implications of this conclusion for accounts of perseveration and of spelling models are discussed.

Perseveration, the incorrect continuation or recurrence of a response, is frequently observed in patients who have suffered brain damage (Hudson, 1968). The term *perseveration* is used to describe a wide range of phenomena, from the recurrence of the same noun in naming a series of different pictures, to the failure to switch to a new task because of the continuation of responses from a preceding task, to errors observed in writing that consist of the repetition of letters (e.g. *lamp* → lam*pp*). Such diversity has led several investigators to conclude that perseveration is not a unitary phenomenon and has prompted various attempts to define the perseverative behaviors. A classification that currently finds wide application was proposed by Sandson and Albert (1984, 1987), who distinguished three patterns of perseverative behavior: (a) recurrent, the incorrect recurrence of a previous response within a task, (b) stuck-in-set, the incorrect maintenance of a given activity or type of response, and (c) continuous, the incorrect prolongation of a current behavior. In the present paper, we focus on the last type of perseveration and we describe the continuous perseverations produced in writing by a patient, RB, with probable Alzheimer's disease (AD). RB's continuous perseverations in word writing consisted of the addition of letters, as in the errors *lobster* → lo*bb*ster and *door* → d*ooo*r. The primary objective of our investigation is to determine the causes of RB's perseverative errors and, more generally, to elucidate the mechanisms that give rise to (at least some forms of) continuous perseverations.

Comprehensive accounts of perseveration have eluded investigators, which is not surprising if one considers the heterogeneity of perseverative behaviors and their manifestation in multiple pathologies – from dementias, to Parkinson's disease, to schizophrenia. The account proposed by Sandson and Albert (1984, 1987) is an exception and represents an attempt to define the functional impairments, along with their neuroanatomical correlates, responsible for the various forms of perseverative behaviors. Of interest here is the explanation of continuous perseverations advanced by Sandson and Albert (1987), according to which an attention deficit, probably related to right hemisphere damage, underlies perseverative errors of this sort (for a similar account, see also Goldstein, 1948; Wepman, 1972). Although the notion of attention is not articulated in the present account, the idea of an attention deficit seems to be consistent with the observation that in individual patients continuous perseverations appear in a variety of tasks. For example, patients who repeat letters when writing words have also been reported to incorrectly repeat elements in drawing or to continue to pronounce a given syllable or phoneme in word production tasks (e.g. Hudson, 1968; Sandson and Albert, 1984). By definition, attention is implicated in a variety of tasks and thus should be critically involved in tasks as different as writing, drawing, and word production. Consequently, an attention deficit that causes continuous perseverations is likely to cause the appearance of this type of error across a series of tasks, as indeed has been reported in a number of neuropsychological patients. However, results seemingly problematic with the attention account have been recently reported by Neils-Strunjas *et al.* (1998) in their patient (DT) with dementia due to AD. In addition to a severe writing impairment, DT produced a variety of types of errors, including omissions, additions, and substitutions of one or more letters, the execution of poorly formed letters, and a substantial number of errors

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characterized by incorrect letter repetition. We refer to the latter type of error as letter perseverations, and *under* → und*d*er or *faith* → fait*i*h are representative examples of errors of this sort. Remarkably, DT's oral spelling was also impaired, but letter perseverations were absent in this task. Such a stark discrepancy between the occurrence of letter perseverations in oral and written spelling is inconsistent with an attention account, which would predict continuous perseverations in multiple tasks, and particularly in oral and written spelling, tasks which arguably posit similar demands to the attention system.[1] Although motor processing was impaired in patient DT and was responsible for a deficit in motor tasks, damage at the level of motor processing is unlikely to be the cause of DT's continuous perseverations since no errors of this type were observed either in drawing or constructional tasks. Continuous perseverations limited to word writing such as those observed in patient DT are more in line with Goldberg's (1986) hypothesis that perseverative behavior is restricted to those cognitive domains that are impaired in conditions of brain damage. From this perspective, perseverative behavior would be expected to be confined to writing in conditions in which writing processing is selectively impaired. Given this latter account, the issue becomes one of defining which mechanisms implicated in writing (or in other cognitive domains) are impaired and give rise to continuous perseverations. This issue will be explored in the present paper.

Case report

RB is a 72-year-old, right-handed woman, with 12 years of education. Until approximately 7 years ago, she worked part-time as a salesperson, but was primarily a homemaker. RB began experiencing memory problems in 1993 and was self-referred to the Alzheimer's Disease Research Center at the Columbia University College of Physicians and Surgeons in 1996 where she underwent an extensive neurological examination, neuroimaging, and neuropsychological evaluation. An MRI of the brain without contrast from April 1996 revealed slight diffuse atrophy, ventricular dilatation, large sulci, and multiple small confluent periventricular white matter foci most compatible with infarcts. These infarcts are consistent with white matter disease and, according to NINCDS-ADRDA criteria, are not considered ''in and of themselves'' to account for her progressive cognitive deficits (McKhann *et al.*, 1984). These findings, coupled with her neurological and neuropsychological results presented in greater detail below led her to be diagnosed with probable Alzheimer's disease.

RB's diagnostic neuropsychological testing was conducted in May 1996 and revealed significant deficits in the areas of memory, abstract reasoning, and language (see Table 1). Performance on a selective reminding test (SRT; Buschke and Fuld, 1974) was severely impaired (total words recalled = 15/72), and long-term recall and recognition on this test fell below the 1st percentile. RB's score on the recognition subtest of the Benton Visual Retention Test (BVRT; Benton, 1955) revealed impaired non-verbal memory performance (6/10 correct; $z = -3$). Her language scores were more variable. For example, on a shortened (15-item) version of the Boston Naming Test (Kaplan *et al.*, 1983) she obtained a score of 14/15, placing her within normal limits for her age and education. Scores on the repetition and comprehension subtests from the Boston Diagnostic Aphasia Exam (BDAE; Goodglass and Kaplan, 1972) were comparable to those of age-matched controls. Performance was well within normal limits on a word production test requiring phonemic organization, the Controlled Word Association Test (CFL; 59th percentile), but was significantly below average on the Animal Naming Test, which requires semantically organized word production (6th percentile). The Rosen Drawing Test (Rosen, 1981), which assesses construction ability, revealed mild difficulty reproducing line-drawn figures: her score was 3/5 (z-score = -1). Like other AD patients, RB demonstrated a tendency to ''close in'' when performing this drawing test; that is, her drawings often overlapped with the stimulus figures that she was asked to copy. Visual attention, as assessed by a cancellation task (Stricks *et al.*, 1998), was poor (time to complete = 201 sec, $z = -2$). Overall, the findings of the 1996 evaluation conform to a diagnosis of probable AD.

Follow-up annual testing was administered for the next two years (later the patient refused it) and revealed an overall

Table 1. Neuropsychological Tests: RT's scores over three evaluations

Neuropsychological Tests	Range	Controls[a] Mean (SD)	RT's scores		
			5/96	5/97	5/98
Orientation					
Modified Mini Mental Status Exam	0–57	52 (3)	40	32	N/A[b]
Attention					
Cancellation Test (in seconds)	0–240	63 (27)	100	70	201
Construction					
Rosen Drawing Test	0–5	3 (1)	3	1	2
Benton Visual Retention Test: Matching	0–10	9 (1)	9	9	9
Memory					
Selective Reminding Test:					
(a) Total Recall	0–72	43 (9)	15	18	22
(b) Delayed Recall	0–12	6 (3)	0	0	0
(c) Delayed Recognition	0–12	11 (1)	4	8	0
Benton Visual Retention Test: Recognition	0–10	8 (2)	6	3	3
Language					
Boston Naming Test	0–15	14 (1)	14	15	14
Controlled Word Association (CFL)	0–100th ile	64 (31)	59	25	63
Category Fluency (Animals)	0–100th ile	36 (28)	6	13	5
Sentence Repetition (BDAE)	0–8	7 (1)	8	8	8
Comprehension (BDAE)	0–6	6 (1)	6	5	5
Abstract Reasoning					
Similarities (WAIS-R)	0–20	11 (3)	7	6	7
Identities & Oddities (Mattis DRS)	0–16	15 (1)	15	16	15

[a]Non-demented elderly (N = 155).
[b]NA = not administered.

decline in cognitive functioning (see Table 1). Functional performance declined as well, and in December 1997, it became apparent that RB could no longer care for herself and was admitted to an assisted living facility. Her functional ability has continued to decline, and in March 2001 she was transferred to a nursing home due to her need for more extensive custodial care. Her modified mini-mental state score (Mayeux *et al*., 1981) was 40/57 in May, 1996, and 27/57 in May, 2001. This most current score, combined with a rating of 2/5 on the Clinical Dementia Rating Scale (CDR) indicates that at present RB has moderate dementia. At the time of the present investigation, the cognitive manifestations of RB's dementia significantly affected her activities of daily living. For example, she did not remember the name of her deceased husband or even the fact that she was married, and required assistance in daily chores such as grooming and dressing.

During an evaluation of RB's writing in November, 2000, it was noted that responses were characterized by incorrect repetitions of letters. Additional testing was arranged to further examine this particular behavior and continued through May, 2001. Informed consent was obtained from the patient prior to the examination. A test for apraxia (Boston Diagnostic Aphasia Test (BDAE), Goodglass and Kaplan, 1983) administered in 2001 revealed that RB was able to execute sequences of complex movements described by verbal commands such as "show me how you would prepare a letter for mailing," thus ruling out ideational apraxia. Ten out of 12 actions were performed correctly in the ideomotor activities section of this test, which includes items such as "show how you would hammer a nail with your right hand." The two errors were both related to the action of hitchhiking; when asked to perform the action, both with the left and the right hand, RB commented that she had "never hitch-hiked before."

RB's perseverations in word writing

Our primary objectives in assessing RB's word writing were to describe the nature of RB's letter perseverations and to determine the variables affecting the occurrence of these errors. To this end, we asked RB to write to dictation a total of 349 words. RB was instructed to write the words in the format she preferred – she chose cursive. Of the total number of words written by RB, 128 were eliminated due to illegibility, leaving 221 responses for analysis. Legibility was scored separately by the first and second author. Inconsistencies in scoring were discussed so that a mutual decision could be reached. An inspection of RB's samples of pre-morbid handwriting revealed her deterioration: before the illness her words were well formed and intelligible, with no spelling errors.

Some of the words that we kept for additional analyses contained poorly formed letters, but no attempts were made to analyze these errors. The appearance of poorly formed letters is probably the result of the motor control impairment that was noted earlier in constructional tasks (see Case Report). As long as a word's letters were intelligible, the word was considered for analysis. We distinguished between single-type and mixed

Table 2. Examples of RT's spelling errors

Error type	Written spelling		Oral spelling	
	Target	Response	Target	Response
Single errors				
Additions	front	fron*e*t	robe	ro*u*be
	drive	dr*e*ive	car	car*e*
	car	car*u*	become	bec*h*ome
Deletions	bl*u*e	ble	we*i*rd	werd
	sum*m*er	sumer	apric*o*t	aprict
	aspe*c*t	aspet	ma*l*l	mal
Substitutions	cha*t*	cha*d*	fe*e*d	fe*a*d
	ear*t*h	ear*u*h	kit*t*en	kit*c*en
	lear*n*	lear*m*	blo*o*d	blo*u*d
Mixed	body	*lalary*	camera	*camruae*
	hotel	*lhalel*	squirrel	*squle*
	rabbit	*raket*	balloon	*blime*
Phonologically plausible errors	juice	*juise*	center	*senter*
	friend	*frend*	tongue	*tung*
	certain	*surten*	acre	*acquor*
Letter perseverations	touch	tou*u*ch	door	doo*o*r
	whale	whale*e*		
	street	strre*e*t		

errors (see examples in Table 2). Mixed errors consisted of those responses in which more than one type of error was observed. Single errors include *additions* (when an extraneous letter was produced), *deletions* (when a single letter or consecutive letters were omitted), and *substitutions* (when a letter was exchanged with another). We also scored for phonologically plausible errors (PPEs), that is, strings of letters that sound like the target word, as in *creep* → creap, and letter perseverations, errors in which a letter was incorrectly duplicated. Finally, we searched for responses in which RB wrote semantically-related or unrelated words, errors that would arise because of semantic and/or word comprehension deficits frequently encountered in patients with dementia related to Alzheimer's disease. The percentage of error types in RB's written spelling can be found in Table 3.

RB's writing was severely impaired; of 221 dictated words, she correctly wrote only 25 (11%). No semantically-related or unrelated words were noticed, a result that rules out a semantic or comprehension deficit as the basis of RB's poor performance in writing to dictation. We scored 15 PPEs (7% of the responses), and 76 words contained a single error (34% of the responses), while 105 words contained mixed errors (47% of the responses). It is probable that PPEs reflect the patient's lack of familiarity with the spelling of some words and reveal the relative intactness of mechanisms for converting word sounds into grapheme sequences. The analyses of the other errors will be presented along with the data from oral spelling, at which point it will be possible to advance hypotheses about the causes of these errors. Next we focus on letter perseverations.

Letter perseverations were frequent in RB's writing: we scored 111 of them in the corpus of the 221 erroneous words retained for analyses. Examples of these errors are shown in

Table 3. RT's responses in oral and written spelling

	N (%) responses/task		
	Written spelling[a]	Oral spelling[a]	Oral spelling (whole corpus)
Words			
Correct	25 (11%)	102 (46%)	150 (43%)
Phonologically plausible errors	15 (7%)	31 (14%)	58 (17%)
Single errors	76 (34%)	35 (16%)	59 (17%)
Mixed errors	105 (47%)	30 (13%)	59 (17%)
No response	0 (0%)	23 (10%)	23 (6%)
Total	221	221	349
Non-words			
Correct	0 (0%)	15 (68%)	15 (68%)
Total	22	22	22

[a]Sample of words retained for analysis of written spelling.

Table 4. Letter distribution

Letter	% in the corpus (Tot = 1560)	% in RT's perseverations (Tot = 113)
e	17	45
r	7	14
t	7	2
a	7	8
i	6	6
o	6	3
n	6	3
l	6	0
s	6	3
c	4	11
h	4	0
d	3	0
b	3	0
u	3	5
m	3	0
p	2	0
g	2	0
f	2	0
v	1	0
k	1	0
w	1	0
y	1	0
z	1	0
j	0.25	0
x	0.19	0
q	0.01	0

Fig. 1. As can be seen in Table 4, the incidence of perseverations varied across letters: it was exceedingly high with the letter *e* (55/111; 49%), relatively lower with the letters *c*, *r*, *a*, and *o*, and absent with other letters (e.g. *b*, *f*, *m*). With two exceptions, the perseverations consisted of duplications of a single letter, as in the error *lobster* → lo*bb*ster in which the letter *b* is repeated. The two errors in which a letter was duplicated multiple times were *stick* → sti*ccc*k and goes → go*eee*se (note the final *e* that was also added in this error). Twenty times, the duplication involved a geminate letter, as in the error *door* → d*ooo*r.

Before proceeding, it is important to determine the nature of RB's perseverative errors, as errors of this sort can be interpreted in a number of ways. One possibility is that RB's letter perseverations reflect a deficit with geminate letters similar to that observed in a few patients with acquired spelling deficits (Caramazza and Miceli, 1990; McCloskey *et al*., 1994; Taiturier and Caramazza, 1996). The errors of these patients suggest that information concerning the identity of a letter and whether the letter is doubled is distinctly represented at the level of the word orthographic representations accessed in spelling, as schematically shown in the example (1) below, in which we follow the notation of Caramazza and Miceli (1990).

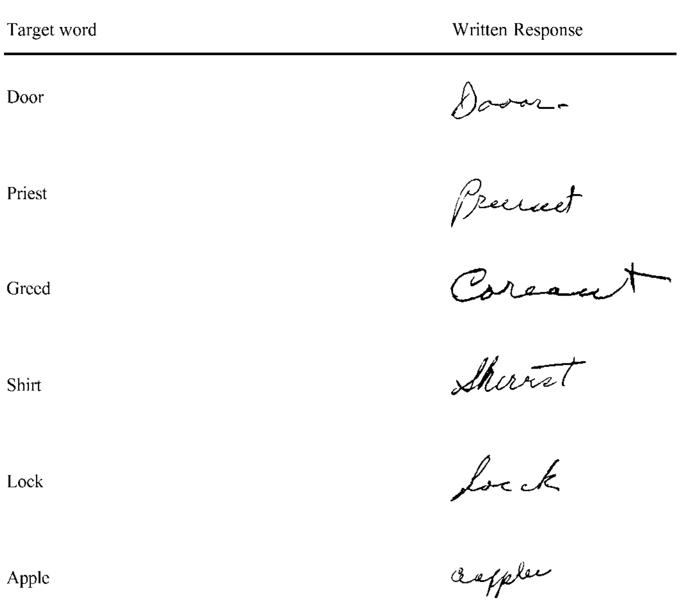

Fig. 1. Examples of RT's letter perseverations in the written spelling task.

G
|
DRES (1)

A geminate feature, represented in (1) as G, is linked to the letter *S* and indicates that this letter is duplicated in the word *dress*. Because the geminate feature is specified at another level of representation than that of letter identities, in conditions of brain damage, information about a letter's gemination (not its identity) can be selectively impaired. A deficit associated with the geminate feature that is of particular interest here is illustrated in (2).

G
|
DRES (2)

The geminate feature is incorrectly linked to the letter *E* and this gives rise to the error *dress* → dr*ee*s, in which the letter *S* is not duplicated but a letter perseveration error occurs with the letter *E*. Thus, letter perseverations can be caused by a transposition of the geminate feature, which migrates from its original position to an incorrect position. Can this be the source of RB's letter perseverations? This explanation seems untenable for two reasons. First, we should find that letter perseverations appear more frequently with words comprising geminate letters (e.g. *dress*) than with words without geminate letters (e.g. *shirt*). But this is not the case, since only 8/111 (7%) letter perseverations were encountered with words containing geminate letters. The second reason is related to the kind of errors anticipated by this explanation: not only should we observe letter perseverations, but also the deletion of a geminate letter. That is, expected errors should be like *pepper* → pe*perr*. But again, errors of this sort were very rare: only 7/111 (6%).

Another problem associated with geminate letters was observed by Venneri *et al*. (1994) in a brain-damaged patient. This problem is exemplified in (3) below:

```
 G G
 | |        (3)
DRES
```

Because the geminate feature was incorrectly duplicated and associated to a letter (*R* in our example), the error *dress* → d*rr*ess should appear, which contains a letter perseveration (the *R* is incorrectly duplicated). But this account seems also untenable for our patient. According to this account, letter perseverations would appear predominantly with words containing geminate letters, but our findings reveal otherwise, as we have described above.

Finally, we should explore the possibility that RB's letter perseverations represent letter substitutions. Errors in which a letter is substituted by another were encountered in the corpus of RB's errors, and one may suspect that perseverations are substitutions in which the exchanged letter happened to be identical to the letter immediately preceding it, as with the error *chair* → ch*aa*r, in which the letter *i* is substituted by the preceding letter *a*. Note that if RB's letter perseverations are letter substitutions, we should not expect errors like *chair* → ch*aa*ir – here the duplicated letter (*a*) does not replace another letter, we simply have a letter addition. However, this account is indefensible since RB produced, almost exclusively, errors like *chair* → ch*aa*ir (62/66, 94%; for this analysis we excluded written responses in which the perseveration involved the last letter of the word or occurred in fragments). In sum, it seems that RB's letter duplications should be interpreted as genuine perseverations, rather than errors associated with geminate letters or as substitutions.

Notably, in writing we found no instances in which RB repeated whole words. That is, she never wrote a word twice in response to two different stimulus words. For example, when asked to write *chair* and in a subsequent trial *dog*, she did not write *chair* in both trials. In terms of the taxonomy proposed by Sandson and Albert (1984, 1987), in writing RB committed only continuous perseverations, not recurrent perseverations. We also analyzed whether RB perseverated on letter strokes. In this analysis we considered only letters that were formed by more than one stroke and that appeared to have a consistent form. The sample of strokes that we examined (N = 100) includes the horizontal stroke of the letter *t*, the dot of the letter *i*, and the downward vertical stroke of the letter *p*. Out of 100 strokes examined, we did not observe any stroke duplication. In sum, RB's perseverations only occurred with *letters*, not with whole words or with letter strokes. This pattern suggests that mechanisms responsible for the processing of letters, rather than words, are implicated in the continuous perseverations observed in RB's writing.

Does RB perseverate on non-writing tasks?

Accounts proposing that continuous perseverations originate from an attention deficit are committed to the prediction that forms of perseverative behavior should be manifested in tasks that make comparable demands in terms of attention processing. Indeed, as attention is implicated in multiple tasks, continuous perseverations should also be evident in multiple tasks. The fact that only letters were involved in the perseverations found in RB's writing suggests that whatever the attention mechanisms responsible for these errors are, they affected the selection and ordering of the letters comprising written words. This fact further indicates that RB should perseverate on tasks that, like writing, involve the selection and ordering of individual components. Tasks demanding spoken words are ideal in these respects, in that they require the processing of phonemes and syllables. Notably, perseverations involving phonemes and syllables have been reported in neuropsychological patients (Buckingham *et al*., 1979; Papagno and Basso, 1996; Cohen and Dehaene, 1998). Various tasks requiring oral production were thus employed to determine whether RB also perseverates with phonemes or syllables. These tasks are described below. Since in these tasks the focus is on the perseveration of phonemes and syllables, we only report the occurrence of these errors without discussing the other types of errors we found.

(a) *Picture Naming* Throughout various testing sessions, RB named a sample of 15 pictures from the Boston Naming Test four times. RB correctly identified 58/60 pictures in total. In none of her responses were phonemes or syllables incorrectly repeated.

(b) *Verbal Repetition* RB was asked to repeat single words (N = 20), combinations of three letters not forming pronounceable syllables (e.g. *WGB* or *MBB*; N = 40), 1- to 4-digit numerals (N = 40), and short sentences (N = 15). The experimenter pronounced the items, which RB immediately repeated aloud. With the exception of two numerals, RB's responses were always correct. No phoneme or syllable perseverations were noticed in any of RB's responses.

(c) *Oral imitation* This task, from Sandson and Albert (1987), required RB to repeat a single letter and a letter pair ten times each. She performed both tasks correctly, with no perseverations.
(d) *Oral Reading* RB read aloud a set of words (N = 118) that had first been administered in the word writing task, in addition to ten pronounceable non-words (e.g. *chaip*) and ten short sentences composed of a total of 44 words. In the case of multiple responses, her last response was scored, although all responses were examined for phoneme perseverations. Correct response rates varied: 105/118 for words (89%), 6/10 for non-words (60%), and 9/10 for sentences (90%). None of RB's errors consisted of the incorrect repetition of a phoneme or a syllable.

Perseverations of the whole word were absent in tasks involving oral production. More importantly, in these tasks we did not observe any incorrect duplications of phonemes or syllables, a finding in sharp contrast to what we found in writing, where perseverations of individual letters were copious. In light of this discrepancy it is unlikely that mechanisms related to attention are responsible for the perseverations observed in writing. Such a conclusion, however, is open to criticism. It can be argued that attention is involved to a different extent in the tasks of written and oral word production, whereby attention demands are greater in writing, and hence perseverations are more likely to be visible in writing following an attention deficit. Written and oral spelling are similar in terms of the demands each task places on attention – if anything, it has been hypothesized that demands are greater for oral spelling (Aarsland *et al*., 1996; Croisile *et al*., 1996). The expectation based on the attention account is thus to find letter perseveration errors in RB's oral spelling.

Does RB perseverate on oral spelling?

There are three principal reasons for examining RB's oral spelling. First, as discussed above, oral spelling is particularly suited for testing the hypothesis that RB's perseverations arise as a consequence of an attention deficit. Second, the finding that letter perseverations do not appear in oral spelling would restrict our search for the causes of RB's perseverations to mechanisms specific for writing. Finally, a comparison between oral and written spelling also allows us to better characterize RB's spelling impairment.

For oral spelling, we administered the 349 words used for written spelling, in addition to 22 non-words. Half of the words were presented for the first time in the oral spelling task and the other half was initially presented in the written spelling task. Both tasks were administered within the same test period. Twenty-three oral responses were eliminated because RB lost track of the task at hand. Oral spelling was scored using the same criteria that were adopted for scoring her written spelling responses (see Table 3). RB correctly spelled 150/349 (43%) words overall and 102/221 (46%) words from the sample retained for written spelling analysis. As can be seen in Table 3, similar error distributions were found in the whole corpus and in the sample used for written spelling (for the sake of consistency, here we discuss the latter type of data). The number of PPEs, errors sounding like the word targets, is noticeable as they account for a third of the errors in the task (31/96; 32%). These errors suggest that because of a failure to access stored orthographic information, RB relied on phonology-orthography mapping. However, evidence that RB's oral spelling is impaired with non-words (15/22 correct) indicates that the phonology-orthography mapping procedure is not intact. A similar number of single errors (N = 35) and mixed errors (N = 30) were observed. As in written spelling, single letter errors involve the substitution, addition, or deletion of a letter (see examples in Table 2). These errors are likely to reflect impairments affecting both the lexical and sublexical processing of word orthography, a hypothesis that is further corroborated by the finding that these errors occur both in oral spelling and written spelling, tasks that share lexical and sublexical orthographic mechanisms (a more detailed discussion of the relation between oral and written spelling can be found below). Another component shared between oral and written spelling is the graphemic buffer – a structure within which information about the word spelling is temporarily held (Ellis, 1982; Caramazza *et al*., 1987). In contrast to patients with graphemic buffer impairment, RB's correct rate in oral spelling did not decline for longer words (see Table 5). This result rules out a graphemic buffer deficit.

Critical here are RB's oral spelling errors that are characterized by the repetition of a letter. She committed only three errors of this sort: *door* → d*ooo*r, *million* → mil*oo*un, and *debt* → d*ee*p. Note that the last two errors differ qualitatively from those she produced in written spelling, in which a letter was added. A striking dissociation was thus found between RB's written and oral spelling concerning letter perseverations: while abundant in the first task, letter perseverations rarely occurred in the latter task. This dissociation has straightforward implications for hypotheses concerning the damage causing the perseverations. On the one hand, it strengthens the conclusion that an attention deficit is *not* the source of the letter perseverations observed in written spelling. On the other hand, it confines explanations of the causes of RB's perseverations to mechanisms that when damaged have repercussions for written spelling, leaving oral spelling relatively intact.

Table 5. Number of errors (%) as a function of word length (letter number) in RT's oral spelling

Word length (N letters)	N (%) incorrect words
4	19/40 (47%)
5	14/35 (40%)
6	17/38 (45%)
7	10/21 (47%)
8	4/8 (50%)

Does RB perseverate because of neglect?

Perseverations similar to those encountered in RB's writing have been observed in patients with hemispatial neglect, an impairment in processing stimuli presented in a particular dimension of space, whether horizontal, vertical, or radial (Mennemeier *et al*., 1994). For example, Ellis *et al*. (1987) described a patient who neglected stimuli shown in the left space and who wrote words with duplicated letters as in *ladder*→laddd*e*r. These errors are explained by a failure to monitor those letters that have already been written. Once written, a letter falls in the left hemispace (at least in writing systems like English) and if the letter is prey to the neglect deficit, the patient may not recognize that the letter has already been written and may write it again. Importantly, at least some patients with neglect share another critical feature with patient RB: letter perseverations are not produced in oral spelling. In cases of neglect, perseverations have also been noted in other tasks (e.g. line cancellation tasks; Na *et al*., 1999). If we further consider that symptoms associated with neglect have been reported in AD patients (Ishiai *et al*., 1996; Bartolomeo *et al*., 1998), the hypothesis that neglect is at the basis of the perseverations observed in RB's writing acquires a certain plausibility. In contrast to patients with neglect, however, no signs of neglect were observed when RB performed a variety of tasks. For example, in oral reading, errors were not concentrated around one half of the word. Moreover, on a cancellation tasks (Stricks *et al*., 1998) she omitted or overlooked a comparable number of items in the left and the right half of the space (5 vs. 4 items). RB's performance on the cancellation task differs also in another respect from that of neglect patients: perseverations manifested as recurrent cancellations of the same items were not demonstrated by RB. In short, our findings do not support the hypothesis that neglect underlies her letter perseverations in writing.

Does RB perseverate on graphic and motor tasks?

Having ruled out an attention deficit and hemispatial neglect as plausible causes of RB's letter perseverations, we turn to the hypothesis that mechanisms involved in writing are the source of these errors. Current theories of spelling agree on a general model in which oral and written spelling share mechanisms responsible for the retrieval of stored representations of word orthography and for the conversion of a word's phonemes into their graphemes, in addition to the graphemic buffer (Ellis, 1982; Margolin, 1984; Goodman and Caramazza, 1986a, b; McCarthy and Warrington, 1990). A schematic illustration of this model is shown in Fig. 2. From the perspective of this model, the absence of letter perseverations in oral spelling indicates that the deficit responsible for this type of error cannot be at the level of the mechanisms shared by oral and written spelling. On the contrary, RB's data suggest that the locus is at the level of the mechanisms

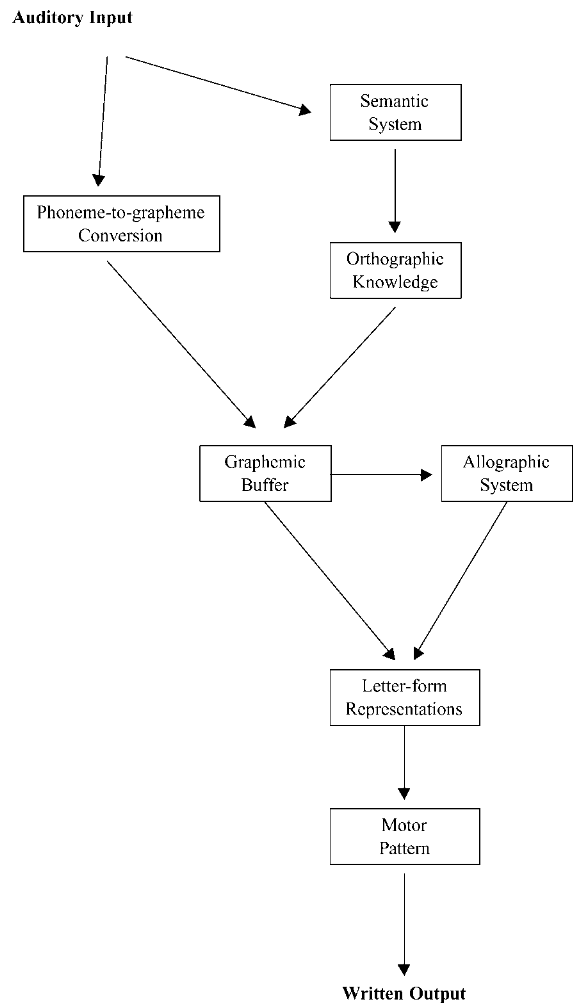

Fig. 2. Model of writing.

selective for writing. These mechanisms involve the selection of representations that specify the letter forms and motor plans for the execution of the hand movements.

Although cognitive scientists remain far from offering a detailed description of the processes involved in the planning and the execution of orthographic sequences, recent neuropsychological studies allow us to delineate at least some of the levels of processing, as illustrated in Fig. 2. Three types of selective deficits that share relevancy to the present topic have been described. The first type of deficit involves selecting the letter format – e.g. capital vs. cursive – and is evidenced by selective or more severe problems in writing words in one of these formats (De Bastiani and Barry, 1989; Patterson and Wing, 1989). The fact that these deficits were observed in patients who produced well-formed letters rules out a problem in accessing and executing letter plans; instead they indicate an impairment in retrieving representations that specify the letter format, or allographic letter representations. A second form of selective impairment relevant here relates to letter substitutions, errors in which a letter is substituted for another as in *b*ar → *d*ar. In some patients, these substitutions occurred predominantly between letters formed by common strokes

(Lambert *et al.*, 1994; Rapp and Caramazza, 1997; Zesiger *et al.*, 1997; Del Grosso Destreri *et al.*, 2000; Miozzo and De Bastiani, 2002). An example is given by the upper case letters *B* and *D*, which share a vertical stroke, typically executed starting from the top and proceeding downward. The fact that these patients produced well-formed letters and that their drawing abilities were preserved suggests that the functional impairment is localized at the level where letter motor plans, which encode the strokes comprising individual letters, are selected (for a discussion of the neuroanatomical correlates of this deficit, see Grossman *et al.*, 2001). These representations are referred to as abstract letter form representations, a term that emphasizes the notion that the parameters pertaining to the shape of letters (e.g. size) or the modality of their execution (e.g. speed) are not specified at this level. Thus, the same abstract letter representation is accessed to print a letter in different size or at different speed. The final writing deficit relevant here is more peripheral and is characterized by malformed letters and additional strokes (Margolin, 1984; Papagno, 1992), as well as impaired drawing, suggesting a deficit in the control and execution of fine motor plans.

The hypothesis that RB's perseverations originate from a deficit at the level of allographic representations can be ruled out because she was able to produce letters in both capital and cursive format. A deficit at the level where abstract letter forms are processed seems a more plausible hypothesis, particularly when we take into account the fact that RB's perseverations involve letters. However, we cannot exclude the possibility that RB's perseverations reflect a more peripheral deficit, a feasible hypothesis if we consider that drawing abilities are not intact in RB and the fact that continuous perseverations have been observed in patients with peripheral motor problems. Evidence that RB demonstrates continuous perseverations when performing graphic or motor tasks would lend support to this latter hypothesis. We tested this possibility by means of graphic and motor tasks that in earlier studies had elicited continuous perseverations. We used two tasks requiring graphic reproduction. In the first task (from Sandson and Albert, 1987), RB was presented with multiple loops or a series of *m*'s and *n*'s randomly alternating, which she was instructed to reproduce. We only scored those instances in which RB completed the test successfully – on a few occasions, RB appeared to fail to understand the task or she "closed in." For the second task, RB was presented with sets of two or three geometric shapes or lines and instructed to copy them. There were a total of 15 sets of items. On another occasion, RB was asked to draw five different geometric shapes orally presented by the experimenter (e.g. "draw a square"). Despite her poor drawing performance, RB did not incorrectly repeat lines or figures on any of these tasks.

RB was also asked to perform two motor tasks previously used in frontal assessments (Dubois *et al.*, 2000). Luria's "fist-edge-palm" hand motor series task requires subjects to reproduce a series of three actions multiple times – making a fist, extending the fingers outward, and rotating the hand palm up (Luria, 1966). Although RB refused to do this task with the left hand, inappropriate repetition of a given gesture was not observed when she performed it with her right hand. Instances of perseverative behaviors have been reported in tasks requiring the inhibition of a response (Dubois *et al.*, 2000), such as those related to the go-no go paradigm (Drewe, 1975). Patients must inhibit a response by providing an opposite response to the movement the examiner performs, such as tapping once when the examiner taps twice. On this task, RB required multiple trials to learn the task but demonstrated no difficulty inhibiting inappropriate responses, and made no perseverative errors in 10 trials.

The absence of perseverative behavior in graphic and motor tasks implies that a peripheral deficit at the level of the processes responsible for the execution of fine movements cannot be identified as the source of the letter perseverations observed in RB's writing. Converging evidence, although of a subtler type, emerges when we consider the similar movements that are involved in writing the cursive letters *e* and *l* and in drawing the loops in Sandson and Albert's (1987) task. If the cause of RB's perseverations was motor in nature, comparable results should have been obtained with these items; however, this is not what we found. We may rule out the hypothesis of a deficit at the level of allographic letter representations because, as we have argued above, RB is able to write letters in different formats. By the process of elimination, we are left with the hypothesis that RB's perseverations are caused by a deficit at the level of selecting abstract letter representations.

General discussion

We documented a patient, RB, with probable Alzheimer's disease and a severe spelling deficit. RB's spelling deficit is likely to reflect impairments at various levels of the spelling system, including mechanisms for converting phonology to orthography, the selection of letter identity in writing, and motor processing. Interestingly, RB exhibited perseverations that were specific in terms of both the elements that were incorrectly repeated – letters – and the tasks in which such perseverations occurred – only in writing. Within the classification proposed by Sandson and Albert (1984), RB's errors represent continuous perseverations. Errors determined by letter duplications may reflect a problem with geminate letters or can be letter substitutions in which an identical letter is accidentally repeated, as in *chair*→ch*aa*r. However, we have shown that these interpretations do not apply to RB's errors, which represent genuine letter perseverations. We administered other tasks to RB that resemble writing in terms of requiring the sequencing and integration of elements, such as word naming and graphic and motor tasks that involve the duplication of a given component (e.g. a loop, a line, or a simple action). Continuous perseverations were absent when she performed the latter type of tasks. The data collected from patient RB replicated and further extended those reported by Neils-Strunjas *et al.* (1998) with DT, another AD patient. Similarly to our patient, DT also produced frequent letter perseverations in writing, but not in oral spelling or in graphic and motor tasks.

The appearance of perseverations in a single task has immediate implications for accounts of perseverative behavior, particularly for continuous perseverations. The most accredited account of continuous perseverations was proposed by Sandson and Albert (1984, 1987), who attributed these errors to an attention deficit. If an account of this sort is consistent with the observation that continuous perseverations occur in different tasks, it is less in line with our finding that RB's continuous perseverations were isolated to a single task. It is improbable that there are attention mechanisms specific for writing. Within the framework of an attention account, it is more plausible to suppose that writing is a task that makes heavy demands of attention resources, so that perseverations are likely to appear in this task when attention mechanisms are impaired. However, this account provides no explanation as to why continuous perseverations were absent in RB's oral spelling, arguably a task comparable to, if not more demanding than writing in terms of attention resources required. A more feasible alternative is that an attention deficit is the cause of continuous perseverations only for some patients. For other patients, the cause is related to more focal deficits that affect performance only in certain tasks. Empirically, patients of the two groups can be distinguished based on whether or not continuous perseverations appear in multiple tasks. The issue, of course, becomes one of defining which cognitive domains are selectively associated with continuous perseverations. Both our study and that of Neils-Strunjas *et al*. (1998) reveal that continuous perseverations are circumscribed to writing, but the extent to which they selectively appear in other cognitive domains remains to be investigated. A second major issue is to determine which mechanisms are responsible for the appearance of continuous perseverations in a given task. The data of patient RB help us to address this issue in the context of writing.

The locus of the writing deficit responsible for RB's perseverations can be localized fairly precisely within the spelling model illustrated schematically in Fig. 2. The hypothesis that we find most plausible is that of a deficit at the level of abstract letter representations which are accessed for writing. A deficit at this level would result in errors that involve individual letters, and that would appear only in writing, as indeed was found with RB's perseverations. To explain why letter perseverations may occur at this level, we appeal to assumptions concerning selection that are widely adopted in other contexts (e.g. semantic, lexical, or phoneme retrieval) as well as in spelling (Rapp and Caramazza, 1997; Miozzo and De Bastiani, 2002). The probability that a letter representation is selected is a function of its activation level: the representation reaching the highest activation level at a critical point in time is selected. Thus, the letter *a* of the word *cat* is selected because its activation level is higher than the activation levels of the letters *c* and *t*. Later, if the activation level of the letter *a* decreases, either because of spontaneous decay or inhibition, while the activation level of the letter *t* increases, the letter *t* is selected because it reaches the highest activation level. However, if the activation level of the letter *a* does not decrease quickly, or the activation level of the letter *t* does not rise significantly, the letter *a* remains the letter with the highest activation level and the one that is selected, giving rise to the duplication of the letter *a* and to the appearance of a letter duplication error like those produced by patient RB. Thus, RB's incorrect letter duplications may arise because of problems related to the activation of abstract letter representations.

The account that we propose also offers us clues to understand why RB's perseverations are particularly numerous with some letters (*e*, *r*, *t*, *a*, and *i*), while absent with others (see Table 3). Various pieces of evidence indicate that the frequency with which letters are produced in writing affects the selection of abstract letter representations. Patients with putative impaired access to these representations produce significantly fewer errors with high frequency letters than low frequency letters (Black *et al*., 1989; Miozzo and De Bastiani, 2002), and neurologically intact subjects are faster at writing high frequency letters than low frequency letters (Van Gallen, 1980). Frequency is often assumed to affect activation level, so that the representations of high frequency letters reach higher activation levels than the representations of low frequency letters. If this is true, and if RB's letter perseverations reflect dysfunctions in letter activation levels, we would expect perseverative errors to be more common with high than low frequency letters. In fact, because high frequency letters reach relatively high activation levels, it takes longer for their activation level to decline to a critical level, increasing the probability of incorrectly re-selecting a high frequency letter. RB's data conform to this prediction. While the five most frequent letters of the English alphabet (*e*, *t*, *a*, *o*, and *i*; norms from Francis and Kucera, 1982) occur in our corpus 43% of the time, they account for 64% of the letter perseverations ($\chi^2 = 18.2$, $p < .001$). Furthermore, the largest number of duplications occurs with the letter *e*, the most frequent English letter.

If we are correct in attributing RB's perseverations to a problem in selecting abstract letter representations, analyses of these errors would shed light on how processing unfolds at this level of representation. One aspect of RB's repetition errors is of particular interest here and relates to geminate letters. All of RB's perseverative errors involving geminate letters consisted of the addition of a single letter, as in *cook*→coo*o*k. In this respect, the errors with geminate letters were identical to those recorded for non-geminate letters, which almost invariably (89/91 times; 98%) consisted of single letter additions, as in *sock* → soc*c*k. This striking similarity implies that geminate and non-geminate letters are similarly processed at the level of abstract letter representations. It seems then that we have to rule out the alternative account that (a) the abstract representation of a geminate letter is selected once and (b) there is information indicating that the execution of this letter has to be repeated. This account predicts perseverative errors consisting of the duplication of double letters, as *cook* → coo*oo*k. But errors of this sort were never committed by patient RB. On the contrary, it seems that to write the word *cook*, for example, two distinct retrievals of the letter *o* take place. It appears that information is encoded differently at prior levels of spelling processing. Here, the letter *o* in the word *cook* is specified once and a separate feature indicates that this letter is duplicated. Perseverations that involve an impairment at this level were observed by Venneri *et al*. (1994). In their patient (an Italian speaker), perseverations occurred predominantly with geminate letters and also consisted of duplication of geminate letters, as for example in the error *ginocchio* [knee]→gino*cccc*hio.

Before concluding, we should emphasize that we took a different approach to the examination of perseveration than has been taken in prior studies, which typically have attempted to provide a behavioral description of the phenomenon or to determine its neural correlates (notable exceptions are the studies of Cohen and Dehaene, 1998 and of Plaut and Shallice, 1993). Instead, we focused on the cognitive aspects and attempted to identify the causes of perseverative errors with reference to current cognitive models. Our study represents another example of the critical role that cognitive theories can play in the interpretation of neuropsychological impairments. Further, just as in the past where researchers have examined other types of errors made by brain-damaged patients, we examined RB's perseverations from the perspective of understanding the organization of the writing system. Our hope is that future analyses of perseverations will shed light on novel aspects of writing and other cognitive processes.

Acknowledgements

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Note

[1]It should be noted that Neils-Strunjas *et al*. (1998) interpreted their patient's perseverations in writing as due to an attention deficit. We question this interpretation and in our paper will present additional data that seem problematic for their view.

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Where do perseverations come from?

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Abstract

RB, a patient with probable Alzheimer's disease, makes continuous perseverations of single letters when writing (e.g. *fruit*→fr*uu*it), particularly on high frequency letters. An analysis of her errors reveals that her perseverations do not reflect letter substitutions or transpositions, nor do they suggest difficulty with geminates. No continuous perseverations were found in oral production, in graphic and simple motor tasks, and in oral spelling. RB's data do not support an attention deficit as the basis of her continuous perseverations. It is proposed that a deficit at the level of abstract letter representations is the source of RB's perseverations. The implications of this conclusion for accounts of perseveration and of spelling models are discussed.

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Primary diagnosis of interest

Alzheimer's disease

Author's designation of case

RB

Key theoretical issue

- Spelling, perseveration, attention

Key words: perseveration; Alzheimer's disease; writing, spelling; attention

Scan, EEG and related measures

MRI

Standardized assessment

Neuropsychological battery

Other assessment

WAIS-R, SRT, BVRT, BDAE, Rosen Drawing Test, CDR

Lesion location

- Diffuse atrophy

Lesion type

Atrophy

Language

English