# Peripheral Dysgraphia: Dissociations of Lowercase from Uppercase Letters and of Print from Cursive Writing

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Abstract: Clinical analyses of patients with acquired dysgraphia provide unique opportunities to understand the cognitive and neural organization of written language production. We report J.B., a 50-year-old woman with peripheral dysgraphia who had prominent dissociations in her ability to write in lowercase versus uppercase and print versus cursive. We gave J.B. a series of tasks that evaluated her skills at writing uppercase and lowercase print and cursive, spelling aloud and in writing, writing numbers and symbols, and visual letter recognition and imagery. She was impaired in printing letters, with lowercase more affected than uppercase, but her cursive writing was relatively intact. This pattern was consistent across letter, word, and nonword writing tasks. She was unimpaired on tasks assessing her visual recognition and imagery of lowercase and uppercase letters. Her writing of numbers was preserved. J.B.'s handwriting disorder was accompanied by a central phonological dysgraphia. Our findings indicate functional independence of graphomotor programs for print and cursive letter styles and for letters and numbers. We discuss the relationship between peripheral and central writing disorders.

Key Words: acquired dysgraphia, handwriting, neuropsychology, case report

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**Reader Benefit:** Peripheral dysgraphia may selectively impair handwriting of specific letter formats.

**JHDB** = Johns Hopkins Dysgraphia Battery. **PALPA** = Psycholinguistic Assessments of Language Processing in Aphasia.

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Writing involves a number of cognitive processes (Figure 1). Most models broadly distinguish between the central (linguistic) and peripheral (output) components (Ellis, 1982, 1988; Ellis and Young, 1996; Hillis and Rapp, 2004; Margolin, 1984; Purcell et al, 2011; Rapcsak and Beeson, 2002; Rapp and Caramazza, 1997). Central processes are involved in spelling regardless of whether the output modality is handwriting, oral spelling, or typing, while peripheral processes are outputspecific. Although in this report we describe these processes as if they were discrete, their mechanisms are highly interactive and likely implemented through distributed pathways.

Writing words that we have heard spoken aloud as "phonological input" (Figure 1) requires word-specific phonological and orthographic processing. Semantic processing typically occurs as well, although understanding word meaning may not be essential for accurate spelling (Patterson, 1986). This set of processes, known as the "lexical route" in dual-route models of reading and spelling, is engaged when we spell familiar words with either regular (eg, "middle," "chicken") or irregular (eg, "yacht," "sword") phoneme-to-grapheme correspondences. The lexical route cannot be used to spell unfamiliar words or nonwords (nonsense words that follow the rules of English spelling, eg, "dusp," "snite"), however, because these items do not have lexical representations.

For us to write nonwords heard aloud, a phonemeto-grapheme conversion process maps units below the level of words onto plausible spellings based on experience. This "sublexical route" can also be used to spell regular words, but not words with irregular phoneme-tographeme correspondences. The lexical and sublexical routes both converge in a graphemic buffer that maintains the abstract grapheme identities and order for translation into the selected peripheral output system.

The peripheral system for handwriting likely involves 3 main subprocesses (Ellis, 1982, 1988; Ellis and Young, 1996; Margolin, 1984; Rapp and Caramazza, 1997). The first subprocess converts graphemes to allographs, which are letter shape representations including variants in case (lowercase versus uppercase letters) and basic letter format (print versus cursive letters). The second subprocess translates allographs into graphomotor patterns that specify the relative size, position, direction, and sequence of the strokes to be made in writing letters. The third subprocess maps these graphomotor patterns

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FIGURE 1. A cognitive model of writing, showing central processes (above dotted line) and peripheral processes (below dotted line).

onto graphic codes, which provide specific neuromuscular instructions for the selected writing medium, such as pen on a sheet of paper or spray paint on a wall (Ellis, 1988).

Strong support for this general model comes from the many different ways that brain damage can disrupt these writing processes. Detailed case studies of dysgraphia (a disorder of writing resulting from a brain lesion; also called "agraphia") have shed light on mechanisms at each stage. Dysgraphias may be broadly divided into those that affect central or peripheral processes. In central dysgraphias, spelling is disturbed across all output modalities; in peripheral dysgraphias, a specific modality, such as handwriting, is affected.

# **Central Dysgraphia**

Impairments in specific central processes result in different forms of central dysgraphia. For example, surface dysgraphia results from damage to the lexical route. Patients with surface dysgraphia spell relying on a phoneme-to-grapheme conversion process, thus making errors mainly with irregular words (Beauvois and Dérouesné, 1981; Goodman and Caramazza, 1986a; Hatfield and Patterson, 1983). Their spelling errors tend to be phonologically plausible, and they tend to "regularize" irregular words, eg, spelling "yacht" as "yot."

Conversely, phonological dysgraphia is caused by damage to the sublexical route. Patients with phonological dysgraphia spell using a lexical-semantic strategy, which causes them particular difficulty with nonwords (Henry et al, 2007; Roeltgen et al, 1983; Shallice, 1981). To a lesser extent, they may have trouble spelling words that have weak lexical-semantic representations, such as function words (eg, "of," "when") and nouns low in imageability or frequency (eg, "analogy," "pact") (Ellis and Young, 1996; Hillis and Caramazza, 1994). When these patients make errors in words or nonwords, their spellings are usually implausible.

Many patients with phonological dysgraphia also have phonological dyslexia, the corresponding deficit in reading (Rapcsak et al, 2009). The co-occurrence of these deficits supports a model in which spelling and reading share central processing systems, that is, the same phonological system converts phonemes to graphemes and graphemes to phonemes (Rapcsak et al, 2009; Rapp and Lipka, 2011).

Damage to the graphemic buffer causes a central dysgraphia in which spelling mistakes increase with increasing word length (Caramazza et al, 1987;

Katz, 1991). The most errors tend to be in the middle letter positions (Caramazza et al, 1987). Interestingly, letter substitution errors tend to keep consonants as consonants and vowels as vowels (Buchwald and Rapp, 2006; Schiller et al, 2001). Some patients have specific deficits in their ability to write vowels (Cotelli et al, 2003; Cubelli, 1991) or consonants (Kay and Hanley, 1994; Miceli et al, 2004). These findings imply that the consonant-vowel status of a letter is set in the graphemic buffer and that vowels and consonants have distinct representations.

# Peripheral Dysgraphia

Damage to peripheral processes at the allographic level can impair handwriting in a number of specific ways. For example, disruption in selecting allographs can cause substitutions of similar-looking letters (Di Pietro et al, 2011; Ellis, 1982; Goodman and Caramazza, 1986b; Weekes, 1994). Disturbed control of letter case can cause "cASe-miXinG" within words (De Bastiani and Barry, 1989; Forbes and Venneri, 2003; Hanley and Peters, 1996; Menichelli et al, 2008; Weekes, 1994).

Peripheral dysgraphia can also affect uppercase versus lowercase and print versus cursive writing. The most common dissociations are between uppercase and lowercase printing (Graham et al, 1997; Hughes et al, 1997; Kartsounis, 1992; Patterson and Wing, 1989; Shuren et al, 1996) and between uppercase printing and lowercase cursive writing (Del Grosso Destreri et al, 2000; Menichelli et al, 2008; Silveri, 1996; Trojano and Chiacchio, 1994; Venneri et al, 2002; Weekes, 1994). We know of only 1 patient who was specifically impaired in printing lowercase letters but not in writing them in cursive (Hanley and Peters, 1996).

These selective writing deficits indicate that lowercase versus uppercase letters and print versus cursive letters are processed independently of one another functionally and potentially also neurally. It is not entirely clear which of the processing stages stores these representations. Most studies have attributed impairments in producing lowercase versus uppercase and/or print versus cursive to allographic damage (Hanley and Peters, 1996; Hughes et al, 1997; Menichelli et al, 2008; Patterson and Wing, 1989; Shuren et al, 1996; Venneri et al, 2002; Weekes, 1994). Many researchers reached this conclusion based on patients' poor performance on letter visual imagery tasks (eg, a patient is asked to imagine a specific letter and then say whether it has straight and/or curved lines) (Menichelli et al, 2008; Shuren et al, 1996; Venneri et al, 2002). A few authors, however, have argued that case- and print-versus-cursive-specific deficits develop from a disturbance at the graphomotor level (Del Grosso Destreri et al, 2000; Kartsounis, 1992; Silveri, 1996).

Damage to graphomotor patterns causes difficulties in forming letters, traditionally termed "apraxic agraphia" (Rapcsak, 1997; Roeltgen and Heilman, 1983; Valenstein and Heilman, 1979). Patients with graphomotor deficits write poorly formed letters with omitted, repeated, or misplaced strokes (Baxter and Warrington, 1986; Kartsounis, 1992; Zesiger, 1997; Zesiger et al, 1994). Patients may substitute letters that share graphomotor features with the target letters (Del Grosso Destreri et al, 2000; Lambert et al, 1994; Miozzo and De Bastiani, 2002). Few studies, however, have differentiated between the effects of graphomotor and visual similarity (but see Di Pietro et al, 2011, and Rapp and Caramazza, 1997, for careful analyses of these effects).

Letter frequency affects writing in some patients with graphomotor deficits (Black et al, 1989; Miozzo and De Bastiani, 2002), but not all such patients (Del Grosso Destreri et al, 2000; Lambert et al, 1994). For example, high-frequency letters like "A" may be easier to write than low-frequency letters like "Q." While patients with graphomotor deficits cannot copy letters completely fluently, many patients do better at copying than at spontaneous writing (Baxter and Warrington, 1986; Grana et al, 2001; Kapur and Lawton, 1983; Kartsounis, 1992; Roeltgen and Heilman, 1983).

Ever since Déjerine (1882), researchers have reported patients who had deficits in reading letters but intact reading of nonalphabetic symbols. Less is known about this dissociation in relation to dysgraphia, although some patients have been reported to have deficits in writing letters despite relatively preserved writing of digits (Anderson et al, 1990; Davies et al, 1997; Del Grosso Destreri et al, 2000; Delazer et al, 2002; Grana et al, 2001; Hanley and Peters, 1996; Kapur and Lawton, 1983; Macoir et al, 2011; Patterson and Wing, 1989). Most of these patients also had allographic or graphomotor disturbances, implying that letters and digits have independent representations at a peripheral level of function.

# **Introduction to the Reported Patient**

We report a patient with a peripheral handwriting dysgraphia caused by graphomotor impairment. She exhibited a unique writing profile: She was impaired in printing both lowercase and uppercase letters, but her deficit was worse with lowercase. Her cursive writing and her writing of numbers were relatively intact. Along with her handwriting disorder, she showed a central phonological dysgraphia. We examine the implications of her performance pattern for understanding the cognitive and neural organization of peripheral handwriting processes, as well as interactions between peripheral and central writing systems.

# CASE REPORT

J.B. was a 50-year-old right-handed woman, a native speaker of North American English who had a 12th-grade education. She was living with her family. She came to us because of difficulties "remembering how to form letters" when she wrote.

Her problem had started suddenly a year earlier. She first noticed it when her daughter was unable to read a grocery list that J.B. had printed because so many letters were missing; eg, J.B. reported having printed "ara rap" for "Saran Wrap." In spite of her writing problem, she had continued to work as the bookkeeper for her family business and she reported few difficulties because most of her tasks were computerized and she was able to recognize and type letters using the keyboard. When handwriting was required, she copied letters from a self-developed "cheat sheet" containing the uppercase and lowercase print letters of the alphabet. She reported no problems in writing numbers.

Her neurologic and general physical examinations were normal. She had undergone 3 operations to clip and coil brain aneurysms in her right internal carotid artery; these procedures had been done 21 years, 7 years, and 4 months before we evaluated her. She reported that after the first aneurysm clipping, she had developed problems with remote autobiographical memory; eg, she could no longer recall details of her childhood or her wedding. Although she still could not retrieve these remote memories, she denied any anterograde memory deficits or cognitive problems other than writing.

Computed tomograms taken 10 years, 7 years, and 9 months before our evaluation, as well as 7 months afterward, revealed a right inferior caudate infarct and moderate artifacts of her clip and coils. Thus, her infarct had long predated the onset of her dysgraphia.

Nine months after our evaluation, we gave J.B. a single photon emission computed tomogram, which showed mild-to-moderate hypoperfusion in the right caudate and the left frontal and parietal lobes. The transaxial slices shown in Supplemental Digital Content 1 (http://links.lww.com/CBN/A44) have arrows pointing to the left frontal and parietal hypoperfusion. (In the figure, R = right hemisphere and L = left hemisphere.) In keeping with her history of infarction and the sudden onset of her dysgraphia, we presumed that this lefthemisphere perfusion deficit had been caused by a frontoparietal stroke. This stroke may have been missed on the computed tomograms because of her clip and coil artifacts. Her neurosurgical implants precluded her from undergoing magnetic resonance imaging.

J.B. consented to our reviewing her business records from 4 years to 2 years before the onset of her dysgraphia, to evaluate her premorbid writing ability. These records were not an ideal writing sample as they consisted mainly of customer lists (names of people and companies, many of them abbreviated) and numbers (dates and dollar amounts), with only brief handwritten notes.

Nonetheless, this sample showed no evidence that she had had any difficulties forming letters, and provided examples of well-formed lowercase and uppercase print and cursive writing. She usually wrote in cursive, but occasionally mixed formats, eg, "PRozects," "mAil toNight." No specific letters appeared more often in the "wrong" format than any others. The handwriting sample had no lexical spelling errors. J.B. described herself as having been an "okay" speller before her problem began.

Four months after we first evaluated J.B., we gave her a neuropsychological assessment that included tests of handwriting. We followed these tests 8 months later with a detailed writing assessment.

# NEUROPSYCHOLOGICAL ASSESSMENT

Most of the neuropsychological test results (Table 1) were within normal limits, although J.B. had mild difficulties in remembering faces and in executive control. Her language was intact except for difficulties with reading nonwords and with writing.

For the writing part of the assessment, J.B. printed the names of objects shown to her as line drawings (Kaplan et al, 1983), and she printed words and nonwords to dictation (Kay et al, 1992). During these tasks, she spelled aloud spontaneously, speaking the names of the letters that she was trying to write. She printed low dash (underscore) symbols as placeholders for the letters that she could not form (Figure 2A). Occasionally, she produced the first down-stroke of the letter, eg, the "w" in "flower" and "wheelchair" and the "b" in "toothbrush," but not the rest of the letter. She substituted an "m" wherever an "n" belonged, although she said "n" aloud. She was fully aware of her errors and expressed frustration at her inability to write the letters correctly.

Even when she formed letters accurately, her printing was observed to be markedly slowed, and she struggled to make the necessary letter strokes. She completed the writing tasks mostly in lowercase print, with occasional insertions of uppercase letters. We did not consider uppercase versus lowercase in our accuracy counts because we had not asked J.B. to use a specific case.

In addition to her difficulties in writing letters, she had problems spelling aloud, ie, saying the correct component letters as she printed them.

On the Boston Naming Test short form (Kaplan et al, 1983), she printed only 4 of the 30 items without omitting letters, and she correctly spelled aloud only 18 of the items. In printing regular and irregular words to dictation (Psycholinguistic Assessments of Language Processing in Aphasia [PALPA] Subtest 44; Kay et al, 1992), she printed only 7 of 40 words without making letter errors, and her accuracy was independent of the words' regularity. Her spelling aloud was less accurate with irregular (15/20 correct) than regular (19/20 correct) words; however, she made 2 of her irregular word errors with words uncommon in North American English ("aeroplane" and "sledge"), and she did not make any of the regularization type of errors typical of surface dysgraphia.

In writing nonwords to dictation (PALPA Subtest 45), she printed only 1 of the 24 items without letter omissions. Spelling aloud, she was correct on only 13/24 items, below the 21/24 mean of PALPA controls.

With both words and nonwords, the majority of J.B.'s spoken spelling errors were phonologically implausible, in that her spelling did not match the phonology of the item (11/18 word errors and 11/11 nonword errors). For example, for real words she spelled aloud

TABLE 1. J.B. S Neuropsychological rest scores 4 Month's After Initial Evaluation	~		
	Score		
General Intelligence (WAIS-III) <sup>1</sup>			
Verbal IQ	81		
Performance IQ	99		
Learning and Memory			
WMS-R Logical Memory Immediate Recall <sup>2</sup>	25/50 (57th percentile)		
WMS-R Logical Memory Delayed Recall <sup>2</sup>	18/50 (42nd percentile)		
CVLT-II Immediate Free Recall (5 trial total) <sup>3</sup>	43/80 (25th percentile)		
CVLT-II Short-Delay Free Recall <sup>3</sup>	10/16 (32nd percentile)		
CVLT-II Long-Delay Free Recall <sup>3</sup>	11/16 (50th percentile)		
CVLT-II Total Recognition Discriminability (target recall relative to intrusion rate) <sup>3</sup>	3 (50th percentile)		
Recognition Memory Test: Words <sup>4</sup>	46/50 (55th percentile)		
Recognition Memory Test: Faces <sup>4</sup>	38/50 (<10th percentile)		
Visuoperception			
Benton Facial Recognition Test <sup>5</sup>	47/54 (72nd percentile)		
Judgment of Line Orientation Test <sup>5</sup>	27/30 (72nd percentile)		
Sunnybrook Neglect Battery <sup>6</sup>	No neglect		
Rey-Osterrieth Complex Figure (copy)	31.5/36 (~50th percentile)		
Executive Control			
Wisconsin Card Sorting Test <sup>8</sup>	4/6 categories (<16th percentile)		
	23% perseverative errors (12th percentile)		
D-KEFS Color-Word Interference Test (inhibition raw score) <sup>9</sup>	75 (16th percentile)		
D-KEFS Color-Word Interference Test (inhibition/switching raw score) <sup>9</sup>	104 (5th percentile)		
D-KEFS Trail Making Test (switching raw score)	74 (63rd percentile)		
Motor Function			
Pantomime to Verbal Command (right hand) <sup>10</sup>	No apraxia		
Movement Copying (right and left hands) <sup>10</sup>	No apraxia		
Grooved Peg Board Test	66 seconds right ( $\sim 25$ th percentile)		
•	63 seconds left ( $\sim$ 84th percentile)		
Language	20/20		
Boston Naming Test short form <sup>14</sup>	29/30		
Token Test <sup>12</sup>	83/84		
D-KEFS Letter Fluency (sum of " $F'' + "A'' + "S''$ )	27 (16th percentile)		
D-KEFS Category Fluency (sum of 3 categories)	29 (9th percentile)		
WRAI-3 Reading subtest (raw score) <sup>15</sup>	41 (13th percentile)		
Spelling-Sound Regularity and Reading (PALPA Subtest 35) <sup>17</sup>	30/30 regular words		
$\mathbf{N}$ is the second s	27/30 irregular words		
Nonword Reading (PALPA Subtest 36)	14/24 (PALPA mean = 23/24)		
Written Naming (Boston Naming Test short form)	4/30		
Printing words to Dictation (PALPA Subtest 44) <sup>11</sup>	//40		
Printing Nonwords to Dictation (PALPA Subtest 45)	1/24		
Direct Copying of Words (JHDB)	13/21		

#### TABLE 1. J.B.'s Neuropsychological Test Scores 4 Months After Initial Evaluation

<sup>1</sup>Wechsler, 1997. <sup>2</sup>Wechsler, 1987. <sup>3</sup>Delis et al, 2000. <sup>4</sup>Warrington, 1984. <sup>5</sup>Benton et al, 1983. <sup>6</sup>Black et al, 1990. <sup>7</sup>Strauss et al, 2006. <sup>8</sup>Milner, 1963. <sup>9</sup>Delis et al, 2001. <sup>10</sup>Kimura and Archibald, 1974. <sup>11</sup>Kaplan et al, 1983. <sup>12</sup>De Renzi and Vignolo, 1962. <sup>13</sup>Wilkinson, 1993. <sup>14</sup>Kay et al, 1992. <sup>15</sup>Goodman and Caramazza, 1985.

WAIS-III = Wechsler Adult Intelligence Scale. IQ = intelligence quotient. WMS-R = Wechsler Memory Scale. CVLT-II = California Verbal Learning Test. D-KEFS = Delis-Kaplan Executive Function System. WRAT-3 = Wide Range Achievement Test. PALPA = Psycholinguistic Assessments of Language Processing in Aphasia. BNT = Boston Naming Test. JHDB = Johns Hopkins Dysgraphia Battery.

"p-a-r-m-i-d" for "pyramid" and "s-p-r-i-n-k" for "spring"; for nonwords, she spelled "c-l-o-o-p" for "glope" and "t-h-u-r-s-t" for "churse."

J.B. could copy letters better than she could form them spontaneously, although she copied the letters slowly, segment by segment, and she continued to make errors. When asked to copy 21 words presented in lowercase type (half of List 7 from the Johns Hopkins Dysgraphia Battery [JHDB]; Goodman and Caramazza, 1985), she copied only 13 of the words correctly (Figure 2B). She substituted the letter "m" for "n" 5 of 7 possible times, and "g" for "q" in its only appearance. She misformed the letter "f" 1 of 4 possible times, "s" 1 of 8 possible times, and "c" 1 of 3 possible times. Finally, despite reminders to copy the words as presented, she made format errors, substituting lowercase cursive for



FIGURE 2. Examples from patient J.B.'s neuropsychological assessment 4 months after initial evaluation. Panel A: Written naming of line drawings of a house, scissors, flower, toothbrush, helicopter, mushroom, wheelchair, and mask. J.B. marked a low dash (underscore) as a placeholder for each letter that she knew she could not write. Panel B: Direct copying of the lowercase words "bring," "grief," "suspend," "quaint," "strict," "pierce," "hungry," and "length."

print in 3 of 13 spellings using "e" and in 1 of 3 spellings using "l."

This preliminary assessment of J.B.'s writing suggested deficits in both peripheral and central processes. She had a marked peripheral dysgraphia that impaired her ability to form letters in both words and nonwords. Her self-generated strategy of speaking the names of letters while printing them is common in developing writers, as saying the names may facilitate retrieval of letter forms. In addition to her peripheral handwriting deficit, her phonologically implausible oral spellings of words and nonwords were consistent with phonological dysgraphia. This latter diagnosis is in keeping with her problem in reading nonwords since phonological dysgraphia and dyslexia often occur together, likely because of a shared impairment of phonological processing ability (Rapcsak et al, 2009).

#### WRITING ASSESSMENT

Eight months after her neuropsychological assessment (a year after our initial evaluation), we gave J.B. a detailed writing assessment. Over the 8 months since her neuropsychological tests, J.B. had been independently practicing her writing by copying lowercase and uppercase letters from cue cards and words from books. For this assessment of her writing, we adapted tasks from the PALPA (Kay et al, 1992), JHDB (Goodman and Caramazza, 1985), and other studies as referenced below to evaluate her performance in 4 areas:

- 1. Writing in lowercase and uppercase print and in lowercase cursive
- 2. Spelling aloud, in writing, and in typing
- 3. Writing of numbers and symbols
- 4. Visual letter recognition and imagery

Author J.L.I. conducted the testing and recorded J.B.'s responses manually. The tasks were divided over 6 testing sessions, each lasting several hours.

During the writing assessment, J.B. continued to speak spontaneously the letters that she was trying to write, thus allowing us to score her accuracy of spelling aloud separate from her ability to write the letters. For example, when asked to print "build" in lowercase, she orally spelled "b-u-i-l-d" but printed "duiLb." We scored this response as correctly spelled aloud but written with 3 letter formation errors ("b"  $\rightarrow$  "d"; "l"  $\rightarrow$  "L"; "d"  $\rightarrow$  "b"). In contrast, when we asked her to print "virtue" in lowercase, she orally spelled "v-i-r-u-e" and printed "viRuE." We scored this response as incorrectly spelled aloud and with 2 letter formation errors ("r"  $\rightarrow$  "R"; "e"  $\rightarrow$  "E"). We scored letter formation errors only when J.B. had actually spoken a letter aloud. For example, when she left out the "t" in "virtue," we scored an error in spelling aloud, but we did not score a letter formation error because she had not tried to write the "t." For each task, we report analyses of her spoken spelling responses first, and then her handwritten responses.

We used  $\chi^2$  tests to analyze J.B.'s accuracy, and considered a *P* value < 0.05 to be statistically significant.

# 1. Writing in Lowercase and Uppercase Print and in Lowercase Cursive

We compared J.B.'s writing of lowercase print, uppercase print, and lowercase cursive. We assessed each of these formats during a different session because we used the same test items for each format. Although she did not struggle with printing as much as she had during the assessment 8 months earlier, her printing still appeared slow and effortful. We observed that she tended to use the same number of strokes every time she wrote a given letter, but she varied in the direction and order of the strokes. In contrast to her difficulties in printing, her cursive writing was remarkably fluent.

# Writing Words

We dictated 2 lists of words designed to assess the effects of imageability and frequency (PALPA Subtest 40) and grammatical word class (PALPA Subtest 32). We instructed J.B. to write the words in lowercase print, uppercase print, and lowercase cursive. Because of her phonological dysgraphia, we expected the lexical variables (high- versus low-imageability words, high- versus low-frequency words, nouns versus function words) to

affect her accuracy of spelling aloud, and so we analyzed the spoken spelling data for the 2 subtests separately.

In PALPA Subtest 40 (imageability and frequency), J.B.'s accuracy of spelling aloud did not differ significantly among lowercase print (31/40 correct), uppercase print (29/40 correct), and lowercase cursive (32/40 correct;  $\chi_2^2 = 0.65$ , P = 0.72). Therefore, we collapsed the data across formats for the following analyses.

She made many more errors in spelling aloud lowimageability (22/28 errors) than high-imageability (6/28)errors) words; we found a significant imageability effect in uppercase print ( $\chi_1^2 = 6.14$ , P < 0.02) and near-significant effects in lowercase print ( $\chi_1^2 = 3.58$ , P < 0.06) and cursive  $(\chi_1^2 = 3.20, P < 0.07)$ . She made more errors in spelling aloud low-frequency (18/28 errors) than highfrequency (10/28 errors) words, but frequency effects were not significant in any letter format (P > 0.30).

Similarly, in PALPA Subtest 32 (grammatical word class), J.B.'s accuracy of spelling aloud did not differ significantly among lowercase print (59/80 correct), uppercase print (58/80 correct), and lowercase cursive (59/80 correct;  $\chi_2^2 = 0.04$ , P = 0.98). She made slightly more errors in spelling aloud function words (20/64 errors totaled across letter formats) than nouns (15/64 errors), adjectives (14/64 errors), and verbs (15/64 errors), but grammatical class effects were not significant in any letter format (P > 0.5).

We classified J.B.'s errors in spelling words aloud as phonologically plausible (eg, "t-r-a-g-i-c-k" for "tragic"; "s-c-e-a-n" for "scene") or implausible. She made more phonologically implausible than plausible spellings in both word lists (57/92 errors across Subtests 40 and 32). Her letter errors included substitutions (eg, "o-p-i-p-i-o-n" for "opinion") in 21/57 responses, omissions (eg, "s-h-i-n-k" for "shrink") in 21/57 responses, additions (eg, "w-r-a-r-t-h" for "wrath") in 13/57 responses, and transpositions (eg, "h-o-s-i-p-t-a-l" for "hospital") in 10/57 responses. Some responses contained >1 letter error. (Coding adapted from Graham et al, 1997, and Hughes et al, 1997.)

We combined J.B.'s handwriting data for Subtests 32 and 40 because the lexical variables did not affect her accuracy (P > 0.2) and the effects of letter case were similar in the 2 lists. As shown in Figure 3A, J.B. was more impaired at printing lowercase (19/120 words correct) than uppercase (78/120 correct;  $\chi_1^2 = 60.23$ , P < 0.001), and she was most able at writing in lowercase cursive (95/120 correct;  $\chi_1^2 = 5.98$ , P < 0.02).

We found this same impairment gradient at the letter level. Her overall letter accuracy was worse when printing in lowercase (506/660 correct) than uppercase (587/651 correct;  $\chi_1^2 = 43.11$ , P < 0.001), and worse with uppercase print than lowercase cursive writing (623/648 correct;  $\chi_1^2 = 18.15$ , P < 0.001). In these counts, the total number of letters differed between formats because of variations in J.B.'s spellings. We classified each letter error according to its location at the beginning, middle, or end of a word (Caramazza et al, 1987), but the error rates did not differ between letter positions in any format (P > 0.2).



FIGURE 3. Examples from J.B.'s writing assessment 1 year after initial evaluation. Panel A: Writing the words "ability," "build," "hand," and "gentle" to dictation in lowercase print, uppercase print, and lowercase cursive. Panel B: Cross-case transcoding of the words "poem," "brisk," "rigid," and "absent" from uppercase to lowercase print, lowercase to uppercase print, and uppercase print to lowercase cursive.

#### Writing Nonwords

J.B. was asked first to repeat aloud and then to write 24 dictated nonwords (PALPA Subtest 45) in lowercase print, uppercase print, and lowercase cursive. She had no difficulty repeating the nonwords, indicating that her phonological input and output of the items was intact. She made comparable numbers of errors when she spelled the nonwords aloud while printing them in lowercase (11/24 correct), printing them in uppercase (14/24 correct), and writing them in lowercase cursive (13/24 correct; $\chi_2^2 = 0.78$ , P = 0.68). All of her oral spelling errors were phonologically implausible ("g-l-o-b-e" for "glope,"; "d-r-i-n-g-e" for "treng").

J.B. was significantly more impaired at printing lowercase (11/24 correct) than uppercase (20/24 correct);  $\chi_1^2 = 7.38$ , P < 0.01), and more impaired at printing uppercase than writing lowercase cursive (24/24 correct;  $\chi_1^2 = 4.36, P < 0.05$ ), when we scored her accuracy based on her speaking of the letter names. We saw the same pattern at the letter level. Her overall letter accuracy was worse with lowercase (88/104 correct) than uppercase print (103/107;  $\chi_1^2 = 8.35$ , P < 0.01), and worse with uppercase print than lowercase cursive (104/104;  $\chi_1^2 = 3.99, P < 0.05$ ).

#### Direct Comparison of Word and Nonword Writing

To determine whether lexical status (real words versus nonwords) affected J.B.'s ability to write letters in different formats, we asked her to write 36 words and 36 nonwords in lowercase print and lowercase cursive. The words were 4 letters long, high frequency (mean frequency of occurrence in a 1-million-word corpus = 315 [Kucera and Francis, 1967]), and selected such that each letter of the alphabet appeared at least twice in the list and no letters were repeated within a word (Coltheart, 1981). The nonwords were pronounceable and derived from recombining the letters of the selected words, eg, "stum" was derived from "must." In an attempt to bypass J.B.'s spelling difficulties, the examiner spelled the items aloud. Words and nonwords were randomly intermixed.

J.B.'s overall letter accuracy was comparable between words (136/144 correct) and nonwords (130/144 correct) printed in lowercase ( $\chi_1^2 = 1.77$ , P = 0.18) and between words (143/144 correct) and nonwords (142/144 correct) written in cursive ( $\chi_1^2 = 0.34$ , P = 0.56). Collapsing the data on words and nonwords, we found that J.B.'s letter accuracy was worse with print (266/288 correct) than cursive (285/288 correct;  $\chi_1^2 = 15.10$ , P < 0.001).

## Writing Letters

We asked J.B. to write letters to dictation in lowercase print, uppercase print, and lowercase cursive. The examiner said each letter of the alphabet twice, in random order. J.B.'s accuracy was again worse with lowercase (23/52 correct) than uppercase print (36/52;  $\chi_1^2 = 6.62$ , P < 0.01) and worse with uppercase print than lowercase cursive (50/52;  $\chi_1^2 = 13.17$ , P < 0.001).

#### Form Cueing in Written Words

To determine whether word forms aided J.B.'s ability to write letters in different formats, we showed her 148 5-letter words that were missing a letter in the beginning, middle, or end position (adapted from Menichelli et al, 2008). The examiner told her both the word name and the missing letter, and asked her to complete the word by writing in the target letter in the same case as the rest of the word, eg, "MU\_IC"  $\rightarrow$ "MUSIC." We chose the words so that each letter of the alphabet was the target letter twice in each of the 3 positions (with the exceptions of "x" in the beginning position and "j," "q," and "v" in the end position), and the target letter did not appear elsewhere in the word. The words were as high frequency as possible (mean frequency of occurrence in a 1-million-word corpus = 70 [Kucera and Francis, 1967]), and we matched frequencies across each 6-word set containing a specific target letter (eg, the 6 words with "s" as the missing letter). The examiner gave the items in random order, presenting the same stimuli in lowercase print, uppercase print, and lowercase cursive (Brush Script MT font).

J.B.'s overall letter accuracy was again worse with lowercase (125/148 correct) than uppercase print (137/ 148;  $\chi_1^2 = 4.78$ , P < 0.05) and worse with uppercase print than lowercase cursive (146/148;  $\chi_1^2 = 17.35$ , P < 0.001). Her accuracy did not differ significantly among the beginning, middle, and end positions for any format (P > 0.8). Comparing these results to those for her single-letter writing shows that word form cueing helped her to print in lowercase (44% of single letters correct versus 84% correct in word forms) and uppercase (69% of single letters correct versus 93% in word forms). Cueing did not benefit her cursive writing because her accuracy was already close to ceiling levels (96% of single letters correct versus 99% in word forms).

# Cross-Case Letter Transcoding

We asked J.B. to "transcode" letters from 1 format to another. We had her transcribe the 26 single letters of the alphabet, presented in random order, from uppercase to lowercase print, lowercase to uppercase print, and uppercase print to lowercase cursive. There was no difference in accuracy between her transcriptions to lowercase (18/26 correct) and uppercase print (19/26 correct), but both print formats were less accurate than her transcription to lowercase cursive (25/26 correct; P < 0.02).

#### Cross-Case Word Transcoding

We asked J.B. to transcribe 42 words (JHDB List 7) (Goodman and Caramazza, 1985) from uppercase to lowercase print, from lowercase to uppercase print, and from uppercase print to lowercase cursive. As shown in Figure 3B, we found no difference in accuracy between her transcriptions to lowercase (201/234 letters correct) and uppercase print (205/234 letters correct;  $\chi_1^2 = 0.3$ , P = 0.59), but she was less accurate in both print formats than from print to cursive (219/234 letters correct; P < 0.03).

# **Direct** Copying

We asked J.B. to copy the 26 single alphabet letters and 21 words (half of JHDB List 7) (Goodman and Caramazza, 1985), presented in lowercase print, uppercase print, and lowercase cursive (Brush Script MT font). Her copying of the lowercase and uppercase was very slow, but she copied the cursive items at normal speed. She made no errors in any condition.

#### Summary: Analyses of Print and Cursive Letter Writing Tasks

J.B.'s writing showed a consistent impairment gradient across a series of word, nonword, and single-letter tasks. Her printing was impaired, lowercase worse than uppercase, but her cursive writing was relatively intact (Table 2). Her dissociations of case and of print versus cursive were most striking in tasks that required her to write letters singly in response to dictation or in transcoding. Interestingly, her printing of lowercase and uppercase single letters improved when she was given word forms in a cueing task, and when letters were provided as a cue, as in the cross-case transcoding task. Neither lexical status nor letter position within an item influenced her ability to write letters.

Her transcoding from uppercase to lowercase print was better than her printing to dictation. She explicitly knew that the lowercase forms of some letters were

TABLE 2.	J.B.'s	Writing	Assessmer	nt Scores	1	Year	after	Initial
<b>Evaluation</b>								

	% Letters Correct		
	Lowercase	Uppercase	Cursive
Words to dictation	76.7	90.2	96.1
Nonwords to dictation	84.6	96.3	100
Single letters to dictation	44.2	69.0	96.2
Word form cueing	84.5	92.6	98.6
Cross-case transcoding of single letters	69.2	73.0	96.2
Cross-case transcoding of words	85.9	87.6	93.6
Direct copying of single letters	100	100	100
Direct copying of words	100	100	100

smaller versions of their uppercase forms (eg, "P," "S," "W," "Z"), and she transcribed many of them correctly by copying them directly. Unlike her performance 8 months earlier in the neuropsychological assessment, her direct copying of print was now accurate, although it was still markedly slowed. Recall that J.B. had been regularly practicing her letter and word copying since the previous evaluation.

We classified her letter formation errors into 5 categories (adapted from Black et al, 1989; Ellis, 1982; and Weekes, 1994) (Table 3):

- Format errors: incorrect use of uppercase versus lowercase or print versus cursive
- Substitutions: incorrect letters that were well-formed and in the appropriate format
- Reversals: mirror image forms of letters
- Omissions: missing letters
- Misformations: forms that did not represent an English letter

In lowercase and uppercase print, J.B. most often made format errors (eg, "l" $\rightarrow$ "L"), in a wide range of letters. She often substituted "n/N" with "m/M"; we could also classify these as format errors because a cursive

"n" looks similar to a printed "m." Her reversals of letters appeared as well-formed different letters ("b" $\rightarrow$ "d"; "d" $\rightarrow$ "b"; "p" $\rightarrow$ "q") or as nonletters (reversals of "B," "D," "G," "P," and "s/S"). She made few omissions and very few misformations, in contrast to her earlier neuropsychological assessment, during which she had made these types of errors exclusively. As for her cursive writing, she made relatively few errors, most of them being "n" to "m" substitutions.

J.B. was clearly aware of her writing errors. She often underlined her mistakes and joked that she did this to make our "marking easier." When making format errors, she usually said that she was deliberately substituting the wrong case because she could not produce the correct one. Although her premorbid writing sample also contained examples of format substitutions, they were relatively infrequent and not specific to particular formats or letters.

Oftentimes J.B. spontaneously described letter forms that she could not print correctly. For example, after reversing an "s," she commented that she had "made the snake curve in the wrong direction." Many times after reversing a "b" or a "d," she said that she knew the letter was like "a number 1 with a circle" but that she had "put the circle on the wrong side." Similarly, when substituting from "n" to "m" she often said that she knew "n" had "only 1 hump" but that she found it difficult "to stop" the writing pattern.

Figure 4 shows J.B.'s error rates for lowercase, uppercase, and cursive, calculated individually for each letter of the alphabet. We based the percentage of errors on the total number of times she could potentially have written the target letter. For almost all affected letters, J.B. made more errors with print than with cursive. With lowercase letters, she had a > 30% error rate with (from highest to lowest) "n," "l," "f," "r," "q," "b," "j," and "s." With uppercase letters, she had a > 10% error rate with (from highest to lowest) "N," "J," "S," "W," "I," "D," and "T."

	<b>Proportion (%) of Errors and Specific Letters Affected</b>			
	Lowercase	Uppercase	Cursive	
Format	128/222 (57.7%) a, e, f, g, h, i, j, l, q, r, s, t, u, x, y	35/95 (36.8%) A, E, H, I, L, M, N, P, T	0/29 (0%)	
Substitution	50/222 (22.5%) n	33/95 (34.7%) N	27/29 (93.1%) n, v	
Reversal	31/222 (14.0%) b, d, p, s	12/95 (12.6%) B, D, G, S, P	0/29 (0%)	
Omission	12/222 (5.4%) a, b, f, h, j, k, n, p, s, w	15/95 (15.8%) A, B, F, G, H, J, K, N, P, S, W	2/29 (6.9%) a	
Misformation	1/222 (0.4%) z	0/95 (0%)	0/29 (0%)	

**TABLE 3.** J.B.'s Combined Letter Formation Errors in Word, Nonword, and Single-Letter Dictation Tasks and Word Form Cueing

 1 Year After Initial Evaluation



FIGURE 4. Summary results from J.B.'s writing assessment 1 year after initial evaluation: cumulative error rate (%) for each target letter based on the total number of times that J.B. could potentially have written the letter during the lowercase print, uppercase print, and lowercase cursive writing tasks. The error rate combines results from word, nonword, and single-letter dictation tasks and from word form cueing.

We evaluated whether letter-specific factors may have influenced J.B.'s printing accuracy. As seen in Figure 4, the affected letters did not relate to alphabetic order. Although she made many more errors with consonants than vowels in both lowercase print (212/635 consonant errors [33.4%] versus 12/329 vowel errors [3.6%];  $\chi_1^2 = 107.4$ , P < 0.001) and uppercase print (67/636 consonant errors [10.5%] versus 17/332 vowel errors [5.1%];  $\chi_1^2 = 8.11$ , P < 0.01), she made most of these errors on a small cluster of letters that happened to be consonants, rather than clearly dissociating between consonants and vowels.

We assessed the effect of letter frequency by correlating the frequency of letter occurrence in the English language (Baddeley et al, 1960) with J.B.'s individual letter error rates for both lowercase and uppercase printed letters. If letter frequency affected her writing, then letters low in frequency should have higher error rates. The Spearman rank order correlation coefficients ( $r_s$ ) were very small for both J.B.'s lowercase and uppercase errors (P > 0.2), meaning that letter frequency did not influence her printing accuracy. We had established previously that the letter frequencies in our corpus correlated highly with those in the linguistic database ( $r_s = 0.95$ ).

To analyze the relationship between graphomotor complexity and the accuracy of J.B.'s letter writing, we took a stroke segmentation approach (adapted from Black et al, 1989, and Rapp and Caramazza, 1997). We grouped letters according to whether when J.B. wrote them correctly she tended to use 1 stroke (eg, "c," "l," "o," "s"), 2 strokes (eg, "a," "b," "n," "y"), or 3 or more strokes (eg, "k," "m," "w," "z"). We defined a stroke as "a movement segment whose beginning and end corresponded either to points where the pen was lifted off the writing surface or where lifting the pen off the writing surface would not be considered to be an interruption" (Rapp and Caramazza, 1997, page 1140). We categorized lowercase and uppercase letters separately for each of the 3 levels of graphomotor complexity (see Appendix), and then calculated error rates for each level.

We found a significant difference among the 3 complexity levels for lowercase print ( $\chi_2^2 = 15.59$ , P < 0.001). However, graphomotor complexity did not affect J.B.'s writing in the expected direction: She made the most errors with 1-stroke letters (30% error rate), fewer with 2-stroke letters (23%), and fewest with  $\geq$  3-stroke letters (9%). This effect appeared to be explained by her high error rates with the letters "1" (1 stroke) and "n" (2 strokes). We also found a significant difference among the complexity levels for uppercase print ( $\chi_2^2 = 10.76$ , P < 0.01): J.B. made more errors with  $\geq 3$ -stroke letters (12% error rate) than 2-stroke letters (6%) and 1-stroke letters (6%). Although her uppercase results were in the predicted direction, again they appeared attributable to her high error rate with 1 letter, the 3-stroke "N," not to a consistent underlying effect of graphomotor complexity.

As in the initial evaluation, J.B. continued to have problems spelling aloud in addition to her peripheral handwriting deficit. She had particular difficulty spelling nonwords aloud, and the majority of her spoken word misspellings were phonologically implausible. These characteristics, together with the demonstrated imageability effect and slight trends toward grammatical class and frequency effects in her spoken word spellings, are consistent with phonological dysgraphia (Ellis and Young, 1996). In fact, J.B. herself often commented that she was unsure whether her spoken spellings were correct. At 1 point, she remarked that, in addition to her handwriting deficit, she must have "forgotten how to spell, too."

# 2. Spelling Aloud, in Writing, and in Typing

It seemed possible that J.B.'s peripheral handwriting deficit exacerbated her problem in spelling words and nonwords aloud since she might find it difficult to monitor her spelling when she was producing incomplete visual forms of letters. For both words and nonwords, therefore, we compared her spelling in several conditions: oral spelling while printing in lowercase, oral spelling when not writing at all, and typed responses. Because she continued to spell items aloud while printing them, we also scored how accurately she wrote each lowercase letter in relation to her spoken letter name.

# Words to Dictation

The examiner dictated 70 words of 4 to 8 letters, with 14 words of each length (List 6 from the JHDB). In each set of 14 words, 7 were of high frequency and 7 were of low frequency. In different sessions, the examiner asked J.B. to print the words in lowercase, spell them orally, and type them. We found no difference in accuracy among her oral spellings while printing (56/70 correct), oral spellings when not writing (54/70 correct), and typed responses (61/70 correct;  $\chi_2^2 = 2.46$ , P = 0.29).

She made more spelling errors with low-frequency words (35/39 errors totaled across the 3 conditions) than high-frequency words (4/39 errors). We found significant frequency effects in her oral responses while printing (high-frequency: 34/35 correct; low-frequency: 22/35 correct;  $\chi_1^2 = 12.86$ , P < 0.001), oral spelling (high-frequency: 33/35 correct; low-frequency: 21/35 correct;  $\chi_1^2 = 11.67$ , P < 0.001), and typing (high-frequency: 34/35 correct; low-frequency: 34/35 correct; low-frequency: 34/35 correct; low-frequency: 21/35 correct;  $\chi_1^2 = 11.67$ , P < 0.001), and typing (high-frequency: 34/35 correct; low-frequency: 34/35 correct; low-frequ

errors; eg, "p-r-i-t-a-t-e" for "pirate"; "a-v-e-r-n-t" for "avert"; "f-r-e-q-n-u-t" for "frequent").

On this task, J.B.'s handwriting of lowercase print continued to be impaired (28/70 words correct; 358/414 letters correct). Her errors were similar to those shown in Table 3 (ie, 23/27 occurrences of "n" substituted with "m"; 19/24 occurrences of "s" reversed; 14/171 occurrences of "a," "e," "l," "n," and "r" printed in uppercase).

#### Nonwords to Dictation

The examiner dictated a list of 24 nonwords (PALPA Subtest 45) to J.B. in a different condition during each of 3 testing sessions. She had similar difficulties in spelling nonwords aloud while printing them in lowercase (11/24 correct; data from the *Writing Nonwords* section above), oral spelling when not writing (14/24 correct), and typing (12/24 correct;  $\chi_2^2 = 0.78$ , P = 0.68). All of her spelling errors were phonologically implausible ("s-q-u-d" for "squate"; "d-u-b" for "doop"; "b-r-i-t-h" for "pretch").

## Summary: Analyses of Spelling Tasks

J.B.'s consistent difficulties in spelling words and nonwords across output modalities, her frequency effects in word spelling, and her predominance of phonologically implausible spellings confirmed that she had a deficit in the sublexical writing route. This phonological dysgraphia was not caused by her peripheral deficit in handwriting. Because word length did not affect her spelling ability, we did not believe that she had a graphemic buffer impairment.

# 3. Writing of Numbers and Symbols

J.B. was fluent and accurate in writing the Arabic numerals "1" to "10" and in writing 2- to 5-digit Arabic numerals to dictation (10 of each length; 40/40 numerals correct). She made several errors when asked to write Roman numerals from I to X (7/10 correct), but reported premorbid unfamiliarity with writing Roman numerals. She wrote punctuation and mathematical symbols correctly, although she omitted "the 'S' part that looks like a snake" in the dollar symbol (\$).

# 4. Visual Letter Recognition and Imagery

J.B.'s ability to describe the visual characteristics of letters that she could not write suggested some preservation of her allographic representations. The following tasks directly examined her visual letter recognition and imagery. Disturbances in these functions are often assumed to reflect allographic damage.

# Letter Naming

J.B. made no errors in naming letters presented to her in lowercase print (26/26 correct) or uppercase print (26/26 correct).

# Cross-Case Letter Matching

When the examiner showed J.B. the uppercase form of a letter, she had no difficulty distinguishing the

lowercase form from a visually similar distractor, eg, "t" from "f" (PALPA Subtest 19: 26/26). Likewise, when J.B. was shown a lowercase letter, she could easily distinguish the uppercase letter from a visually similar distractor, eg, "N" from "M" (PALPA Subtest 20: 26/26).

# Letter Decision

J.B. made no errors in selecting correctly oriented letters from mirror-reversed forms within an array of intermixed lowercase and uppercase forms (PALPA Subtest 18: 36/36). She also performed perfectly on a task (adapted from Patterson and Wing, 1989) in which she was asked to orient an intermixed series of 26 lowercase and 26 uppercase letters that were presented to her on cards and rotated by 0, 90, 180, or 270 degrees.

# Lowercase Letter Imagery

The examiner spoke the name of each of the 26 letters of the alphabet in random order, and asked J.B. to imagine how each letter looked in lowercase. She was to judge whether the letter shape was relatively flat (eg, "a," "c," "e"), whether it extended upward (eg, "b," "d," "t"), or whether it extended downward (eg, "g," "p," "q") (adapted from Farah et al, 1985). She performed this task perfectly.

# Uppercase Letter Imagery

The examiner spoke the name of each of the 26 letters of the alphabet and the digits "0" through "9," in random order. The items were spoken 3 times. J.B. was asked to imagine each digit and each letter in uppercase, and to respond "yes" or "no" about whether the item contained any straight lines (eg, "B," "4"), any curved lines (eg, "P," "3"), or a vertical line at the far left (eg, "N," "H") (adapted from Shuren et al, 1996). Her only error was not noting the vertical line at the far left of the letter "M" on 1 trial.

## Summary: Analyses of Visual Letter Recognition and Imagery Tasks

J.B.'s good performance on tasks of visual letter recognition and imagery suggests that her lowercase and uppercase allographic representations were relatively preserved. We cannot rule out mild impairment at the allographic level, however, since J.B. may have been able to perform some imagery judgments with only a partially specified mental image. For example, with letters that were prone to visually similar substitutions ("n"  $\rightarrow$  "m") or reversals ("b"  $\rightarrow$  "d"), imagining either the correct letter or the error could have led her to the appropriate response (eg, "n" and "m" are both flat; "b" and "d" both extend upward).

# DISCUSSION

J.B.'s peripheral dysgraphia was unique in that she was impaired in her printing of lowercase and, to a lesser degree, uppercase letters, and yet her cursive writing was reasonably intact. Her writing of numbers was also unaffected. Her handwriting disorder was accompanied by a central phonological dysgraphia, as well as a mild phonological dyslexia. The co-occurrence of phonological dysgraphia and dyslexia is consistent with a model in which spelling and reading share a central phonological system (Rapcsak et al, 2009; Rapp and Lipka, 2011).

Our case study adds to the small number of reported patients who have had deficits in writing specific letter formats. There are previous reports of dissociations between uppercase and lowercase print (Graham et al, 1997; Hughes et al, 1997; Kartsounis, 1992; Patterson and Wing, 1989; Shuren et al, 1996) and between uppercase print and lowercase cursive (Del Grosso Destreri et al, 2000; Menichelli et al, 2008; Silveri, 1996; Trojano and Chiacchio, 1994; Venneri et al, 2002; Weekes, 1994). However, J.B.'s performance pattern was even more specific in that her dissociations were not only between letter cases (lower more impaired than upper) but also between lowercase print and lowercase cursive. Unfortunately, as we did not assess J.B.'s writing of uppercase cursive, we cannot determine if her print and cursive were also dissociated with uppercase letters.

Patient D.N., studied by Hanley and Peters (1996), is the only other person we know of whose writing profile is similar. Like J.B., D.N.'s lowercase writing was substantially better with cursive than print in both word and nonword writing tasks. These tasks, which require sequential production of multiple letters, clearly use different processes in cursive and print. Generation of cursive letters is continuous, and their specific form is determined by what letters they adjoin. In contrast, printed letters are produced as discrete and independent units.

Given these differences, it is perhaps not surprising that the 2 formats can be differentially affected by brain damage. J.B.'s performance is especially interesting in that she continued to demonstrate the cursive-print dissociation when she wrote single letters, both to dictation and in form cueing and transcoding tasks. These findings suggest that the peripheral processes responsible for production of cursive and print are distinct even at the individual letter level.

Conclusions differ about the functional stage that is disrupted in patients with format-specific writing deficits. Most studies have attributed this performance pattern to selective damage at the allographic level (Hanley and Peters, 1996; Hughes et al, 1997; Menichelli et al, 2008; Patterson and Wing, 1989; Shuren et al, 1996; Venneri et al, 2002; Weekes, 1994). However, some have claimed that these dissociations arise more peripherally at the graphomotor stage (Del Grosso Destreri et al, 2000; Kartsounis, 1992; Rapp and Caramazza, 1997; Silveri, 1996). Consistent with this latter proposal, our results suggest that J.B.'s cursive-print dissociation resulted from a graphomotor deficit.

J.B.'s printing appeared labored and she often struggled to make the necessary letter strokes. We saw this most clearly during our initial assessment of her writing, when she made errors even in a copy task (Figure 2B). Although her copying of letters was significantly better than her spontaneous writing, she copied items very slowly, stroke by stroke. Earlier studies of graphomotor impairment have shown that some patients write better when they are copying (Baxter and Warrington, 1986; Grana et al, 2001; Kapur and Lawton, 1983; Kartsounis, 1992; Roeltgen and Heilman, 1983). The explanation may be that copying from a model does not require use of graphomotor patterns (Margolin, 1984).

By the time of her second handwriting assessment 8 months after the first, J.B.'s writing had improved, presumably because of spontaneous recovery and/or her independent letter-copying practice. However, she continued to print all letters slowly, and her direction and order of strokes varied in both copying and spontaneous writing tasks. Unfortunately, we do not have writing velocity data to support our observations. Longitudinal tracking of both her writing accuracy and speed would have been useful. Although kinematic studies of dysgraphia are scarce (Delazer et al, 2002; Patterson and Wing, 1989; Zesiger, 1997), a kinematic analysis may also have helped to differentiate between J.B.'s difficulties in preparing versus executing graphomotor programs.

J.B.'s writing errors included stroke repetitions  $("n"/"N" \rightarrow "m"/"M"$  in print; occasionally, "v"  $\rightarrow$  "w" in cursive) and misplacements causing letter reversals (eg, "b"  $\rightarrow$  "d") that are consistent with a graphomotor impairment. Although she most commonly made format errors (Table 3), which often are associated with allographic deficits (De Bastiani and Barry, 1989; Forbes and Venneri, 2003; Hanley and Peters, 1996; Menichelli et al, 2008; Weekes, 1994), J.B. seemed to make these errors deliberately when she knew that she could not produce the graphomotor pattern for the required case.

J.B. had no problems on our visual letter recognition and imagery tests, suggesting that her allographic representations were relatively preserved. Although the nature of our tasks did not let us confirm that all of her letter shape representations were fully intact (see the Summary: Analyses of Visual Letter Recognition and Imagery Tasks section above), any deficit at the allographic level is likely to be mild and unable to account for the extent of her difficulties in printing. She was fully aware of her writing errors and often spontaneously described the visual features of letters that she could not print. On the transcoding tasks, she explicitly knew that the lowercase and uppercase forms of some letters (eg, "P," "S," "W," "Z") were the same except for their size. Her transcoding to lowercase print was better than her printing to dictation because she transcribed many of the letters correctly by direct copying. These observations provide further evidence for the integrity of her allographic system.

That J.B.'s printing deficit extended beyond tasks of letter and word dictation to transcoding indicates that she had difficulty accessing graphomotor patterns regardless of whether the input was from letter names, word names, or other allographic forms.

Our findings support a model of writing in which graphomotor plans are organized by lowercase versus uppercase and print versus cursive, so that damage at the graphomotor level can differentially impair specific letter formats. We were unsuccessful in identifying other factors that could predict J.B.'s writing accuracy. Neither alphabetic proximity nor letter frequency affected her graphomotor function. Although she had more difficulty printing consonants than vowels, this dissociation appeared to result from high error rates with a small cluster of consonants (see the *Summary: Analyses of Print and Cursive Letter Writing Tasks* section above). Previous patients with consonant-specific dysgraphias have had impairments at the graphemic buffer level (Kay and Hanley, 1994; Miceli et al, 2004). It is unclear why consonant-vowel status would be specified at a later processing stage, ie, the graphomotor level.

Graphomotor complexity also failed to predict J.B.'s printing accuracy, although some of her errors shared graphomotor and/or visuospatial features with the target letters, eg, "n"  $\rightarrow$  "m"; "b"  $\rightarrow$  "d." We cannot differentiate whether graphomotor or visuospatial similarity is the relevant factor predicting J.B.'s substitution errors, because of the limited number of letters that she wrote with nonformat errors and because most of her errors were similar to the targets along both dimensions. Indeed, few studies have independently evaluated the contribution of these 2 factors in their patients' writing errors. Rapp and Caramazza (1997) demonstrated that graphomotor, but not visuospatial, characteristics predicted substitution errors in uppercase print. In contrast, Di Pietro et al (2011) found that graphomotor similarity was unrelated to lowercase and uppercase errors, but visuospatial similarity predicted lowercase errors.

A possible explanation for why graphomotor dysfunction can differentially affect writing formats relates to the extent of premorbid familiarity with the formats (Di Pietro et al, 2011; Patterson and Wing, 1989; Zesiger et al, 1994). Since cursive had been J.B.'s typical writing style, her graphomotor representations for cursive letters might have been more impervious to damage. In fact, cursive and print may develop different types of motor representation because of usage. It has been suggested that letters in more familiar formats may be represented as whole entities, while letters in less familiar formats may be represented as a group of strokes (Di Pietro et al., 2011; Teulings and Schomaker, 1993; Zesiger et al, 1994). This theory was used to account for a dysgraphic error pattern in which patients substituted letters in the familiar format with similarly shaped complete letters, while making stroke errors in letters in the less familiar format (Di Pietro et al, 2011; Zesiger et al, 1994).

Although J.B. did not have this specific error profile, it seems plausible that damage to her graphomotor processes specifying stroke characteristics (number, sequence, position) may have affected her production of printed letters (coded as sets of strokes) more than cursive letters (coded as single units).

Several characteristics of J.B.'s writing performance, however, do not fit with motor familiarity explaining her deficits. First, the theory does not explain her greater difficulty with lowercase than uppercase print. Lowercase is the more commonly used of the 2 formats, and examination of J.B.'s premorbid writing sample, as well as her verbal report, showed her to have preferred lowercase. Also, the graphomotor demands of writing lowercase print are no greater than those of uppercase print, and may even be less because many lowercase letters contain fewer strokes than the uppercase versions (eg, "b" versus "B"; "e" versus "E"; "r" versus "R"). Second, letter frequency, which should serve as a proxy for premorbid familiarity (Patterson and Wing, 1989), was not an important determinant of J.B.'s printing accuracy. Thus, overall, it appears that J.B.'s format-specific writing deficit resulted from selective damage to format-specific graphomotor programs rather than from differences in her premorbid familiarity with the writing formats.

A notable aspect of J.B.'s peripheral dysgraphia was that her format dissociations were most pronounced in tasks of single-letter writing, and she did better at printing lowercase and uppercase single letters when the examiner cued her by giving her word forms. These findings support the interactive nature of writing processes, in which a central component can influence a peripheral level of function. The fact that lexical status did not influence J.B.'s graphomotor function suggests that the top-down effects may originate from the graphemic buffer level shared by the lexical and sublexical routes.

While our report has focused primarily on J.B.'s peripheral handwriting impairment, she also demonstrated a central phonological dysgraphia. We are not aware of other patients in whom these particular deficits appeared together, although an association between peripheral dysgraphia and surface dysgraphia has been documented (Goodman and Caramazza, 1986b; Graham et al, 1997; Shuren et al, 1996; Weekes, 1994). A possible explanation for the co-occurrence of these deficits is that they result from a lesion affecting 2 neuroanatomically proximal but functionally independent systems that are responsible for peripheral and central writing processes, respectively.

Alternatively, a more intuitively appealing mechanism argued by Graham et al (1997) is that this association arises from a single deficit to a highly interactive system. Graham et al's patients had focal neurodegenerative processes in which their surface dysgraphia was consistently followed by the development of peripheral deficits. Because of this progression, the authors suggested that central orthographic deterioration first caused isolated surface dysgraphia, but later disrupted peripheral letter production because of decreased topdown activation.

While it seems unlikely that J.B.'s central phonological deficit was solely responsible for her graphomotor impairment, we did find evidence of top-down effects. We know that J.B.'s phonological dysgraphia was not caused directly by her letter writing deficit because she also had spelling problems in both oral spelling and typing tasks. Perhaps like Graham et al's (1997) study of deficit progression, an evaluation of J.B.'s course of recovery from her 2 dysgraphic syndromes could help us understand the mechanism underlying this association. Clearly, we would also need a more precise delineation of her phonological spelling deficit.

There is a slowly growing group of reported patients with letter-specific writing deficits (Anderson et al, 1990; Davies et al, 1997; Del Grosso Destreri et al, 2000; Delazer et al, 2002; Grana et al, 2001; Hanley and Peters, 1996; Kapur and Lawton, 1983; Macoir et al, 2011; Patterson and Wing, 1989). Like J.B., a few of these patients have also had deficits in activating or executing graphomotor patterns (Anderson et al, 1990; Davies et al, 1997; Delazer et al, 2002; Kapur and Lawton, 1983; Macoir et al, 2011), implying that letters and numbers have functional independence at this level. Although J.B.'s letter-number dissociations might be increased because of her considerable premorbid experience with numbers in her work as a bookkeeper or because of the effects of set size (26 letters versus 10 digits), the extent of her letter-number performance disparity makes it doubtful that these explanations fully account for her results. Letters and numbers are both visually arbitrary, symbolic forms, but their representations are qualitatively different (Delazer et al, 2002). Individual letters do not denote meaning and their positions within a word do not relate to its semantic representation. In contrast, individual digits carry meaning by themselves (eg, "3" signifies a quantity), and their relative positions within multidigit numbers have distinct interpretations. Given such differences, letter and number systems are likely to be at least partially distinct, with the potential to be differentially affected by brain damage.

Interestingly, aspects of J.B.'s performance suggest that her categorical distinctions may be somewhat malleable. While her writing of nonalphabetic items was generally intact, her difficulty writing the dollar sign appeared to be tied to the fact that she represented it, at least overtly, as containing an alphabetic symbol, the letter "S." Also, at the end of our assessment, we were able to help her develop a new writing strategy that involved recoding letter forms as nonalphabetic symbols. For example, we showed her that the letter "S" looks similar to a rounded "5," and the letter "B" looks similar to the right side of a <u>Butterfly</u>. We encouraged her to imagine when printing the letter form that she was, in fact, producing the associated number or object.

Although we did this coaching informally, without systematic training or measuring outcomes, the technique did appear to help J.B. print letters more accurately. Although her overall speed of letter printing remained slow, the delay now seemed to be caused by the extra time that she needed to recall the relevant association. Her actual execution of the graphomotor programs appeared more fluent, presumably because at least to some extent she was using programs for numbers or objects rather than programs for letters.

Given J.B.'s relatively spared ability to write in cursive, and to type and copy letter forms, her functional need for rehabilitation was minimal. Theory-based treatment studies for peripheral dysgraphia are lacking (Beeson and Henry, 2008), but the approach of recoding from alphabetic to other forms might be an avenue to pursue in future research.

J.B.'s presumed left frontoparietal stroke is consistent with studies linking damage in the frontoparietal area with dysgraphia. Peripheral handwriting deficits have been most commonly associated with lesions in the left posterior frontal cortex or in the left parietal cortex, including the superior parietal lobule and the supramarginal and angular gyri (Alexander et al, 1992; Hodges, 1991; Lubrano et al, 2004; Rapcsak and Beeson, 2002; Roux et al, 2010; Sakurai et al, 2007). Peripheral processes are likely subserved by a distributed frontal parietal system that also includes the cerebellum, basal ganglia, and thalamus (De Smet et al, 2011; Denes et al, 2005; Fournier et al, 2011; Marien et al, 2013; Ohno et al, 2000; Pramstaller and Marsden, 1996; Tanridag and Kirshner, 1985). Phonological dysgraphia and dyslexia result from damage in a variety of left perisylvian areas, including the supramarginal gyrus, inferior frontal gyrus, and superior temporal gyrus, which together probably comprise a phonological processing network (Henry et al, 2007; Rapcsak and Beeson, 2002; Rapcsak et al, 2009).

These lesion data agree with a 2011 meta-analysis by Purcell et al that examined normal writing processes with functional neuroimaging. The analyses found the left superior frontal gyrus, precentral gyrus, postcentral gyrus, and superior parietal lobule to be reliably involved in peripheral writing processes. The left supramarginal and superior temporal gyri were shown to be involved in phonological writing processes. Therefore, 1 or more unfortunately placed lesions in J.B.'s left frontal and/or parietal areas could disrupt both her graphomotor and phonological writing processes. A review of cases similar to J.B.'s failed to shed light on the neural substrate underlying her relatively spared writing of cursive and numbers. Many of the other patients had large left-hemisphere lesions (Del Grosso Destreri et al, 2000; Miozzo and De Bastiani, 2002) or diffuse degenerative disease (Macoir et al, 2011; Menichelli et al, 2008), or they were studied before widespread use of magnetic resonance imaging (Patterson and Wing, 1989).

Functional neuroimaging studies in normal participants have not evaluated writing of these different formats, but the studies that have examined visual processing suggest that the right hemisphere is more involved in perception of cursive than print (Hellige and Adamson, 2007) and of numbers than letters (Park et al, 2012; Polk et al, 2002). Tachistoscopic studies also show that normal participants have a right-hemisphere advantage for recognizing cursive (Bryden and Allard, 1976; Hellige and Adamson, 2007) and that patients with disconnection syndromes have a right-hemisphere advantage for recognizing digits over letters (Teng and Sperry, 1973). Because brain areas activated during letter perception are also engaged during writing (James and Gauthier, 2006; Longcamp et al, 2003), the right hemisphere may play a special role in the writing of cursive and numbers as well. If so, then right-hemisphere processing may underlie the relative preservation of J.B.'s ability to write cursive and numbers.

Further study is needed of these dissociations of case, print versus cursive, and letters versus numbers in acquired dysgraphia, as well as in developmental dysgraphia and normal writing. Research using detailed behavioral and neuroimaging protocols will be vital for extending our understanding of written language production.

Letters Printed with 1, 2, or $\geq$ 3 Strokes, as Used in Graphomotor Complexity Analyses			
	Number of Strokes Needed to Write a Letter		
	1	2	$\geq$ 3
Lowercase	c, l, o, s	a, b, d, e, f, g, h, i, j, n, p, r, t, u, v, x, y	k, m, q, w, z
Uppercase	C, O, S	D, G, J, L, P, Q, T, U, V, X	A, B, E, F, H, I, K, M, N, R, W, Y, Z

**APPENDIX** 

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