Published in final edited form as:

Cogn Neuropsychol. 2017 ; 34(3-4): 119–143. doi:10.1080/02643294.2017.1375904.

Do reading and spelling share orthographic representations? Evidence from developmental dysgraphia

Christopher Hepner^a, Michael McCloskey^b, and Brenda Rapp^b

Christopher Hepner: chepner3@jhu.edu; Michael McCloskey: Michael.McCloskey@jhu.edu; Brenda Rapp: brapp1@jhu.edu

^aDepartment of Neurology, Johns Hopkins University, Baltimore, USA

^bDepartment of Cognitive Science, Johns Hopkins University, Baltimore, USA

Abstract

Both spelling and reading depend on knowledge of the spellings of words. Despite this commonality, observed dissociations between spelling and reading in cases of acquired and developmental deficits suggest some degree of independence between the cognitive mechanisms involved in these skills. In this paper, we examine the relationship between spelling and reading in two children with developmental dysgraphia. For both children, we identified significant deficits in spelling that affected the processing of orthographic long-term memory representations of words. We then examined their reading skills for similar difficulties. Even with extensive testing, we found no evidence of a reading deficit for one of the children. We propose that there may be an underlying difficulty that specifically affects the learning of orthographic word representations for spelling. These results lead us to conclude that at least some components of lexical orthographic representation and processing develop with considerable independence in spelling and reading.

Keywords

spelling; developmental dysgraphia; developmental cognitive neuropsychology; orthographic knowledge

The relationship between perception and action has been investigated in a variety of domains (e.g., vision, spoken language processing). In the written language domain, the issues concern the relationship between reading (perception) and writing (action). Researchers have asked such questions as: Do we use the same knowledge about the spellings of words when we read and write?; Does knowledge about how letters are written influence the ability to recognize letters or words?; and, Do visual representations of letters play a role in writing? Questions of this sort have been asked not only with regard to adult expert users of written language, but also in the context of literacy acquisition. Improving our understanding of the relationship between learning to read and learning to write is important not only for enhancing theoretical knowledge concerning the structure and development of cognitive

reading and writing mechanisms, but also for development of effective teaching methods and strategies for remediating deficits.

In the research reported here, we examine the relationship between reading and spelling in two children with developmental dysgraphia. As a foundation for discussion of these cases, we first provide an overview of the cognitive representations and processes making up the adult writing system. We then summarize evidence from adults regarding shared representations and processes in reading and spelling, as well as evidence from studies of developmental dyslexia and dysgraphia. It is important to note that the term *dysgraphia* has different senses in the literature, sometimes referring to impaired spelling and sometimes to deficits affecting the motor planning or production processes required for handwriting. In this article we focus on spelling, although we also consider and discuss handwriting. By *developmental dysgraphia* we mean difficulties in the acquisition of writing (spelling, handwriting, or both), despite adequate learning opportunities, and the absence of obvious neuropathology or gross sensory-motor dysfunction.

The adult reading and spelling systems

Figure 1A depicts the adult cognitive mechanisms for single-word reading. When a visual stimulus such as STEAK is presented, letter identification processes map the visual letter shapes onto abstract representations specifying the identity and ordering of letters (e.g., Caramazza & Hillis, 1990b, 1990a; McCloskey, Fischer-Baum, & Schubert, 2013; Rapp & Caramazza, 1989; Schubert & McCloskey, 2013). The letter identity/order representations are processed in *Orthographic Working Memory* (O-WM; sometimes referred to as the graphemic buffer) pending subsequent operations.¹

If the stimulus is a familiar word, the representation of letter identity and order will activate a stored lexical-orthographic representation in *Orthographic Long-Term Memory* (O-LTM; sometimes referred to as the orthographic input lexicon). The activated lexical representation in turn activates the meaning of the word in the *Lexical Semantic System*. If the word is to be read aloud, a lexical-phonological representation is activated in *Phonological Long-Term Memory* (P-LTM; also referred to as the phonological output lexicon). The phonological representation is activated from the lexical-semantic representation; according to many theorists, it can also be activated via direct links between O-LTM and P-LTM (e.g., Schwartz, Saffran, & Marin, 1980).

In addition to whole-word lexical representations, reading also draws upon knowledge about the relationships between sublexical orthographic units (letters and letter groups) and their corresponding sounds. These spelling-sound mappings (sometimes referred to as grapheme-phoneme mappings or correspondences) form the basis of the *Spelling-to-Sound Conversion*

¹The role of O-WM in reading is a rather under-researched topic. However, it has been argued (Caramazza, Capasso, & Miceli, 1996; Tainturier & Rapp, 2003) that the degree to which O-WM is engaged is determined by task demands. While spelling always places demands on O-WM, “normal” horizontal word reading is typically carried out in parallel with few demands placed on O-WM. In contrast, vertical word reading, reading words with letters presented serially, or even pseudoword reading place greater demands on O-WM. In support of the hypothesis of shared O-WM processes in reading and spelling, Caramazza et al. (1996) and Tainturier & Rapp (2003) reported cases of individuals with acquired dysgraphia with deficits affecting O-WM in spelling who showed similar patterns of errors and length effects in spelling, pseudoword reading, and reading with alternative formats (e.g., mirror reversed and orally presented letters).

System that takes as input the letter identity/order representation (e.g., P-L-O-F-E), and generates a plausible pronunciation (e.g., /ploʊf/). This sublexical reading process is critical for pronouncing unfamiliar letter strings (e.g., words not previously encountered or invented pseudowords). The sublexical process may also contribute to oral reading of familiar words, but is not adequate for consistently generating correct word-reading responses. Because most letters and letter groups have multiple alternative mappings to phonemes or phoneme sequences (e.g., EA may correspond to /i:/ or /e/), the sublexical reading process generates pronunciations that, although plausible, may be incorrect (e.g., STEAK → /sti:k/). The likelihood of error is especially high for words that instantiate uncommon spelling-to-sound mappings (e.g., CHUTE, with the infrequent CH → /ʃ/ mapping).

As depicted in Figure 1B, the cognitive processes for spelling and writing are in many respects the converse of those implicated in reading. Orthographic production can be initiated by hearing words (e.g., taking notes in a lecture, spelling to dictation), from word meanings (e.g., generating a grocery list, writing a letter) or from seen objects (e.g., writing picture names). Consider the case of hearing a spoken word (e.g., /wʌns/). After auditory and phonological speech processing, the P-LTM store of familiar word sounds is addressed, followed by access to the Lexical Semantic System. Access to semantic representations forms the basis for the retrieval of the lexical orthographic representations in O-LTM that specify the letter identities and ordering constituting the word's spelling (e.g., O-N-C-E). In addition to the semantically mediated lexical route, a direct, non-semantic lexical route between P-LTM and O-LTM has been posited (Bub, Cancelliere, & Kertesz, 1985; Funnell, 1983; Sartori, Masterson, & Job, 1987; Schwartz et al., 1980). Parallel to reading, there is also a sublexical system, the *Sound-to-Spelling-Conversion System*, that uses knowledge about the systematic relations between speech sounds and letters or letter groups. This sublexical spelling system generates phonologically plausible spellings for unfamiliar words. If a word has a (regular) spelling that consists of a few common sound-spelling correspondences, it will likely be spelled correctly via the Sound-to-Spelling Conversion System, but if it contains uncommon correspondences or letters/letter groups with multiple possible spellings (e.g., /jʌt/), an incorrect spelling is likely to be generated (e.g., YOT instead of YACHT). (For more detailed reviews, see: Rapp & Fischer-Baum, 2015; Tainturier & Rapp, 2001).

Orthographic information generated by either the lexical or sublexical system is then processed by O-WM. This information consists of symbolic letter representations and their order. O-WM maintains the activity of the representations and carries out the serial selection of the constituent letter identities for subsequent production. Orthographic knowledge can be expressed in a variety of formats (e.g., written spelling, oral spelling, typing, drawing with a toe in the sand). For written spelling, abstract, effector-independent motor plans of letter strokes are retrieved to direct the generation of effector-specific motor programs that drive production of the written response. For oral spelling, the names of the letters must be selected and produced on the basis of the same abstract orthographic representations used for written spelling.

Shared representations and processes in reading and spelling? Evidence from adults

In the literature on adult orthographic processing, discussion of shared cognitive mechanisms for reading and spelling has focused largely on O-LTM representations, although a small amount of work has considered shared processes for O-WM (e.g., Caramazza, Capasso, & Miceli, 1996; Tainturier & Rapp, 2003; see footnote 1), allograph selection (e.g., Rapp & Caramazza, 1997), and sound-spelling/spelling-sound conversion processes (e.g., Beauvois & Dérouesné, 1979, 1981). In this paper we also focus on the question of shared O-LTM representations and processes in reading and spelling.

It may seem intuitively plausible that when we encounter word spellings during reading, we form long-term memory representations that would be available for both recognizing the word on subsequent encounters (reading) or for producing the spelling (writing). Empirical evidence bearing on this intuition of shared O-LTM representations comes from behavioral, cognitive neuropsychological, and neuroimaging studies. Several studies of neurotypical adults have reported evidence consistent with shared lexical orthographic representations. Monsell (1987) reported significant repetition priming in reading words that had been spelled earlier (without visual feedback). In addition, Holmes and Carruthers (1998) and Burt and Tate (2002) reported that words that individuals could not spell correctly also elicited slower or less accurate responses in reading tasks (see also Holmes & Davis, 2002; Holmes, Malone, & Redenbach, 2008). These findings are consistent with the proposal that reading and spelling share lexical orthographic representations in LTM. However, this interpretation has not gone unchallenged (e.g., Beauvois & Dérouesné, 1979, 1981, Frith, 1978, 1980). For example, priming effects might arise not from shared representations for reading and spelling, but rather from episodic memories that are created in one task (e.g., spelling) which then influence performance on the other (e.g., reading). In terms of cross-task associations of performance speed or accuracy, one alternative account is that similarities across tasks are driven by some factor involved in both reading and spelling rather than by shared orthographic representations; another is that representations activated for reading trigger automatic “updates” of spelling representations, and vice versa.

In the cognitive neuropsychological literature, several cases have been reported of associated reading and spelling impairments that seem to originate in O-LTM (Behrmann & Bub, 1992; Coltheart & Funnell, 1987; Philipose et al., 2007; Rapcsak & Beeson, 2004). However, the interpretation of these associations is uncertain because associated deficits could indicate either a shared process or, alternatively, damage that coincidentally affects neurally-adjacent O-LTM components for reading and spelling. Furthermore, in addition to the associations, dissociations have been reported in which either reading or spelling is intact in the context of O-LTM impairment in the other modality (Hillis, Rapp, & Caramazza, 1999). Dissociations such as these are also subject to multiple interpretations as they could result from damage to distinct O-LTM memory components for reading and spelling, but could alternatively occur if lesions affect modality-specific access to a shared O-LTM system.

Possibly the strongest evidence of shared O-LTM processes and representations comes from neuroimaging studies probing whether reading and spelling recruit the same neural

substrates for O-LTM. The small number of fMRI studies that have examined the neural substrates of both reading and spelling in the same individuals (Purcell, Napoliello, & Eden, 2011; Rapp & Dufor, 2011; Rapp & Lipka, 2011) have all found co-activation for the two tasks in the left mid-fusiform and inferior frontal gyri. Evidence that these areas are involved in O-LTM comes from Rapp and colleagues (Rapp & Dufor, 2011; Rapp & Lipka, 2011), who found both areas to be sensitive to the lexical frequency of the words that were read and spelled. Overlapping activation for reading and spelling is, nonetheless, consistent with the possibility of distinct O-LTM components for reading and spelling supported by different subpopulations of neurons within the region. Purcell, Jiang, and Eden (2017) challenged this interpretation using the neural adaptation approach, which is based on the assumption that repeated activation of a neural population leads to a decreasing neural response. Purcell et al. (2017) examined neural adaptation for words that were first spelled and then read, or first repeated and then read. They found neural adaptation in a left ventral fusiform area (VWFA-visual word form area) when words were spelled and then read, but not when they were repeated and then read.

In sum, while the preponderance of evidence from behavioral and neuroimaging studies of neurotypical adults favors the hypothesis of shared O-LTM for reading and spelling, the issue is certainly not resolved, as there is considerable room for alternative interpretations of the existing evidence. Furthermore, the findings from the adult system do not necessarily extend in a straightforward manner to the developing system. Conceivably, for example, O-LTM processes may initially develop independently for reading and spelling, becoming unified only later in the developmental trajectory.

Shared representations and processes in reading and spelling? Evidence from developmental dyslexia/dysgraphia

In research with typically developing children, a close relationship has been documented between reading and spelling skill levels, with Ehri (2000) reporting correlations of reading and spelling scores of .68-.86 across six studies involving first graders to adults. However, drawing conclusions about shared processes from these findings is problematic, given that there are many possible reasons for the strong correlations other than shared processes and representations. Similarly, in studies of developmental deficits, dysgraphia is very commonly found to co-occur with developmental dyslexia (Ehri, 2000; Greenberg, Ehri, & Perin, 1997). However, here too there could be many reasons for high co-occurrence rates and, furthermore (as we discuss in more detail below), some cases of dissociations between developmental dyslexia and dysgraphia have also been reported.

In discussing specific patterns of developmental dysgraphia and dyslexia, it is useful to consider the distinction made by Coltheart (2015) between *proximal* and *distal* causes of developmental disorders. Proximal causes refer to abnormalities in the cognitive system mediating the skill of interest. In the case of developmental dysgraphia, for example, proximal causes are deficiencies in developing one or more components of the cognitive spelling/writing system (Figure 1B). Distal causes, often referred to as underlying causes, are the factors responsible for proximal deficits. Distal causes arise outside the cognitive

system of interest and may include general cognitive dysfunctions (e.g., a phonological awareness deficit that impairs learning of sound-spelling correspondences) or external factors such as poor teaching (for further discussion, see McCloskey & Rapp, 2017).

Researchers studying proximal causes of developmental dysgraphia have reported several performance patterns that appear to reflect deficiencies in developing specific components of the spelling system (for a review of similar claims regarding developmental dyslexia, see Castles & Coltheart, 1993). In one such pattern, referred to as developmental surface dysgraphia, spelling of irregular words is impaired (with errors taking the form of phonologically plausible misspellings) but spelling of pseudowords/unfamiliar words is largely intact. This pattern, which is attributable to deficient development of O-LTM, has been reported in multiple languages, including English (Brunsdon, Coltheart, & Nickels, 2005; Coltheart, Masterson, Byng, Prior, & Riddoch, 1983; Goulandris & Snowling, 1991; Hanley & Gard, 1995; Hanley & Kay, 1992; Romani, Ward, & Olson, 1999; Seymour, 1986; Seymour & Evans, 1993; Temple, 1984, 1985), German (Bergmann & Wimmer, 2008; Cholewa, Mantey, Heber, & Hollweg, 2010), Italian (Angelelli, Judica, Spinelli, Zoccolotti, & Luzzatti, 2004; Angelelli, Marinelli, & Zoccolotti, 2010), and Greek (Douklias, Masterson, & Hanley, 2009). The complementary pattern, labelled developmental phonological dysgraphia, involves better spelling of words (including irregular words) than pseudowords, and is attributed to difficulty in developing the sound-to-spelling conversion system (Campbell & Butterworth, 1985; Funnell & Davison, 1989; Snowling, Goulandris, Bowlby, & Howell, 1986; Temple, 1986; Temple & Marshall, 1983). In addition, recent reports have described children with spelling difficulties suggesting selective impairment in the development of O-WM (Barisic, Kohnen, & Nickels, 2017; Gvion & Friedmann, 2010; Roncoli & Masterson, 2016; Yachini & Friedmann, 2010).

Cases such as these—of developmental deficits selectively affecting a specific processing component—provide precisely the type of opportunity needed to examine the relationship between spelling and reading during development. To our knowledge, however, only a very few studies have evaluated whether individuals show the same proximal deficit, affecting the same cognitive component, in both reading and spelling (e.g., Roncoli & Masterson, 2016; Sotiropoulos & Hanley, 2017), with these providing some evidence in support of shared O-WM and O-LTM systems in reading and spelling.

When considering *distal causes* of developmental dysgraphia (and dyslexia), it is key to keep in mind that, because written language is a recent human invention, there has not been enough evolutionary time to develop a genetic blueprint for neural networks that carry out written language processing. Developmental deficits in reading and spelling are, therefore, generally assumed to stem from underlying difficulties in evolutionarily older domains such as spoken language, vision, attention, and motor processes. A number of hypotheses have been proposed regarding the underlying causes of developmental dysgraphia, most of which have also been posited as underlying causes of developmental dyslexia. In this investigation, we examine phonological awareness and processing, motor processing, visual attention span, visual learning and memory, verbal learning and memory, letter order encoding and memory, and orthographic learning. A better understanding of the underlying deficits will allow us to evaluate the extent to which developmental reading and spelling deficits have

common underlying causes. In turn, this will further our understanding of the complex trajectory involved in the acquisition of reading and spelling in normally developing individuals.

The current investigation

To make progress on the issue of whether developmental dysgraphia and dyslexia affect shared or separate cognitive representations and processes, we need detailed investigations that evaluate the integrity of specific components in both reading and spelling. In this article, we report on an investigation guided by the theoretical framework described above, involving two children with developmental dysgraphia. We first investigated proximal causes of the dysgraphias, identifying clear difficulties in the acquisition of O-LTM. Based on this understanding of the children's spelling deficits, we carried out a detailed examination of their reading skills to assess the possibility of shared O-LTM between reading and spelling. Finally, we considered a range of potential distal deficits to develop a more in-depth understanding of the genesis of the developmental dysgraphia in these cases.

Our results shed light on the proximal and distal causes of impaired spelling for both children. However, the most striking result from our study is the finding of a clear dissociation in one of the children between superior reading skills and impaired spelling. This finding leads us to conclude that at least some aspects of lexical orthographic representation and processing can develop independently in reading and spelling.

Case histories

PJT is a right-handed Canadian boy who was enrolled in a French immersion program for two years (kindergarten and first grade) and thereafter was enrolled in an English language program. He was 8 years 4 months old and had finished the second grade at the time of most of the testing carried out for this study (July 2015), although some additional testing was carried out until July 2017. When we began our testing with him, PJT had attended an all-English program for one full school year. We additionally report on testing administered by a clinical psychologist prior to our study, when PJT was 6 years 11 months old. The clinical testing was initiated by the parents who had noticed that PJT was having academic difficulties during his time in the French immersion program. On the basis of that testing, a discrepancy between reading and spelling performance was reported and PJT was diagnosed with Attention Deficit Hyperactivity Disorder–Combined Type (ADHD-C).

AKR is a right-handed American girl with a history of articulation delay related to motor planning difficulty that had resolved by the time of this investigation. She was 10 years 5 months old and enrolled in the fifth grade at the time most of the testing was carried out (November 2015), although some additional testing was carried out until March 2017. In addition, an evaluation was conducted by an occupational therapist in October 2015 and an assessment of general cognitive functions and academic achievement was carried out by her school in November 2015.

General cognitive function

General intelligence, spoken language, and academic achievement were evaluated with the following tests: Wechsler Intelligence Scale for Children (PJT: WISC-IV [Wechsler, 2003], 2/2014; AKR: WISC-V [Wechsler, 2014], 9/25/2015), Peabody Picture Vocabulary Test (PJT: PPVT-4 [Dunn & Dunn, 2007], 2/2014; AKR: PPVT-R [Dunn & Dunn, 1981], 11/09/2015), Wechsler Individual Achievement Tests (WIAT-III [Psychological Corporation, 2009]: PJT, 2/2014; AKR: 11/12/2015) and Expressive Vocabulary Test (EVT-2 [Williams, 2007]: PJT, 2/2014). Because some of these tests were administered in outside evaluations, there are some differences in the test versions and subtests administered to the two children.

WISC results (see Table 1) indicate very high cognitive ability levels for both children, with a General Ability Index of 160 (99th percentile) for PJT and a Full Scale IQ of 123 (94th percentile) for AKR. Both PJT and AKR obtained high scores in Verbal Comprehension subtests (Similarities and Vocabulary), scoring below the 50th percentile only in Letter-Number Sequencing and Coding (PJT) and Matrix Reasoning (AKR).

Spoken language tests on the WISC and WIAT (Table 2) revealed high levels of functioning, consistent with school reports and our own observations. Also, on the Peabody Picture Vocabulary Test (PPVT), which evaluates comprehension of aurally presented words, both children scored at the 99th percentile with standard scores of 160. In addition, PJT scored at the 95th percentile (standard score 125) on the EVT, a test of expressive word vocabulary.

The WIAT evaluates academic achievement, providing a useful overview of performance in basic skill areas. Scores for both PJT and AKR were well above average (and often very high) on most of the language, reading, and math measures. Notably, the lowest scores were on the Spelling subtest, with both children scoring at the 23rd percentile. Reading subtest scores, in contrast, ranged from the 50th to the 99th percentile.

In summary, both children have very high intellectual capacity and excellent spoken language skills. Academic achievement ranged from average to far above average in all areas except spelling. These results confirm the impression of the parents, teachers, and the children themselves that spelling was an area of particular difficulty for both children, in striking contrast with their otherwise high levels of ability and achievement. More specifically, the results are consistent with the impression that the children's spelling difficulties are at odds with their reading abilities and interests, as exemplified by their extensive reading for pleasure. Figures 2 and 3 present writing samples from PJT and AKR, respectively. The samples, produced shortly before our testing of each child (PJT: age 8, AKR: age 10), illustrate the difficulty in spelling experienced by these children despite their high level of intellectual ability and language skills.

Spelling: proximal sources of impairment

For both children we carried out testing aimed at identifying the proximal cause(s) of their impaired spelling—that is, the specific components of the cognitive spelling system that had not developed adequately.

Written vs. oral spelling

Deficits affecting central components of the spelling system—orthographic long-term memory, sound-to-spelling conversion, and orthographic working memory—should manifest in both written and oral spelling (see Figure 1B), whereas deficits at subsequent levels should affect output modalities selectively. For example, an impairment in handwriting processes should affect written but not oral spelling.

For both PJT and AKR, performance was comparable on written and oral spelling tasks, pointing to a central locus (or loci) of impairment. Specifically, for PJT, 309 words from the Dolch (1948) word sets (words that children are expected to read by “sight” without sounding them out, at pre-primer through third grade levels) were tested for written spelling and, in separate sessions, for oral spelling. He was 53% correct in written spelling (165/309) and 46% correct (141/309) in oral spelling, $\chi^2(1, N = 618) = 3.42, p > .05$. For AKR, both forms of the WRAT3 (Wide Range Achievement Test, 3rd edition; Wilkinson, 1993) Spelling test were administered, one for written and one for oral spelling. She was 35% correct in written spelling (14/40) and 33% correct in oral spelling (13/40), $\chi^2 < 1$.

Direct copy transcoding

Given that comparable performance in written and oral spelling suggests that the children’s spelling difficulties arise from central components of the spelling system, we would expect good performance in direct copy transcoding, a task in which words and pseudowords are presented in either upper or lower case and the participant is asked to copy transcode them in the other case. This task can be performed without recruiting central spelling processes (although one cannot prevent participants from engaging these processes). Consistent with our expectation, PJT was 100% correct in copy transcoding (20/20), and AKR was 93% correct (25/27). AKR’s two errors were letter transpositions: AFRAID → AFIARD and QUAINIT → QUANIT.

Pseudoword spelling

For purposes of evaluating sublexical sound-to-spelling conversion processes, a pseudoword spelling-to-dictation task was administered to both PJT and AKR. The Diagnostic Spelling Test–Nonwords (DiSTn; Kohnen, Colenbrander, Krajenbrink, & Nickels, 2013) consists of 74 pseudoword stimuli (e.g., YARSH) with a wide variety of phoneme-grapheme correspondences. Norms are available by grade level based on a sample of over 600 Australian children 6–14 years of age.

AKR scored at the 46th percentile (raw score 46), suggesting adequate development of sublexical spelling processes. Examination of her pseudoword spelling responses supported this conclusion. According to the scoring criteria for the test, AKR misspelled 28 of the 74 pseudowords; in other words, she failed to produce phonologically plausible spellings for these items. In assessing AKR’s sublexical spelling ability it may be useful to consider that 6 of her responses, although not included among the correct spellings listed in the scoring criteria, are not grossly incorrect. In fact, they would be considered phonologically plausible if they were evaluated against possible English spellings for their component sounds (for a given syllable position) according to Hanna, Hanna, Hodges, and Rudorf (1966). This is a

more lenient criterion that is sometimes used in scoring spelling (e.g., Bruck & Treiman, 1990; McCann & Besner, 1987; Rapp, Epstein, & Tainturier, 2002). Examples from AKR's responses include /zɛl/ → ZEL (cf. GEL), /ku:v/ → KOVE (cf. MOVE), and /ʃʌs/ → SHOUSS (cf. ROUGH). The remaining responses scored incorrect were rather subtle vowel errors (e.g., /lɛŋ/ → LANG), and 4 instances of /kw/ realized as QW rather than QU (e.g., /kwɪ:d/ → QWEED). The latter error suggests that AKR has not learned the sound-spelling correspondence rule /kw/ → QU, but her sublexical spelling otherwise appears entirely adequate. This assessment is also supported by the observation that in spelling tasks with word stimuli AKR typically produced plausible spellings even for long, polysyllabic words that were presumably unfamiliar to her (e.g., VICISSITUDE → VISISATTUDE, IRIDESCENCE → EARADESANCE).

PJT, in contrast to AKR, exhibited clear difficulties in pseudoword spelling, scoring at the 6th percentile on the DiSTn when tested with written responses (raw score 13), and at the 8th percentile (raw score 14) when the test was administered for oral spelling. Examination of his responses indicated deficient knowledge of sound-to-spelling correspondence rules. PJT consistently produced incorrect mappings for /tʃ/, /ð/, /ʃ/, /θ/, and /kw/. For example, in 18 attempts to spell /θ/ and /ð/, he never produced TH, instead producing T 14 times, H twice, and S twice (e.g., /θɪf/ → TIF, /flɪθ/ → FLAS). In addition, he consistently realized /ɜ:/ as R (rather than ER, IR, or UR), yielding orthographically illegal spellings (e.g., /zɜ:n/ → ZRN). He had similar difficulties producing the highest probability mappings for /i:/ and /u:/, which were always realized by a single E or O, respectively, and never by EE, E_E, or OO. As these examples illustrate, PJT had particular difficulty with sound-spelling correspondence rules in which phonemes are mapped to multiple letters (e.g., /θ/ → TH), although he also made occasional errors involving one-to-one phoneme-grapheme mappings.

Pseudoword spelling requires not only sound-to-spelling conversion processes, but also orthographic working memory: the spellings assembled by the sublexical process must be held in working memory while the processes required for generation of overt responses (e.g., handwriting processes) are carried out (see Figure 1). Accordingly, AKR's normal performance in pseudoword spelling suggests that her orthographic working memory is intact, whereas PJT's impaired pseudoword spelling raises the question of whether he may suffer from a deficient orthographic working memory in addition to under-developed sound-to-spelling conversion processes.

To obtain additional evidence concerning PJT's orthographic working memory we generated a new list of pseudowords. This list consisted of 40 1- and 2-syllable pseudowords constructed to avoid as much as possible the sound-spelling mappings that proved problematic for PJT in the DiSTn task (e.g., θ → TH). In this way, we hoped to evaluate PJT's orthographic working memory without complications introduced by his deficient knowledge of sound-to-spelling correspondences.

The list was administered twice, once for written and once for oral spelling. PJT produced a phonologically plausible spelling for 73 of the 80 stimuli (91%). Two of his seven phonologically implausible responses (/dʒi:vək/ spelled GVOK twice) apparently stemmed

from a sound-spelling conversion error often observed in children learning to spell, in which a letter name (in this case, /dʒi:/ for G) is used as the basis for mapping from sound to spelling (e.g., Treiman, 1993). Hence, PJT made errors not readily attributable to deficient sound-spelling correspondence knowledge for only 5 of the 80 stimuli (e.g., /sæfzeɪm/ → SAFTAIZ, /tɒnɪd/ → TOND). This 6% error rate seems unexceptional for an 8-year-old spelling pseudowords. Consequently, we suggest that PJT's orthographic working memory is normal, and that his spelling errors do not reflect dysfunction in working memory processes.

Spelling irregular words

The Diagnostic Spelling Test–Irregular Words (DiSTi; Kohnen, Colenbrander, Krajenbrink, & Nickels, 2015) was administered to both PJT and AKR. The DiSTi consists of 74 words with low-probability phoneme-grapheme mappings (e.g., BISCUIT). Accurate spelling of these words requires orthographic long-term memory (i.e., stored knowledge of whole-word spellings), because sublexical spelling processes will not reliably generate the correct spelling. AKR was tested on the DiSTi for written spelling. For PJT, the test was split into even- and odd-numbered items, which were administered separately for written and oral spelling.

Both children performed well below average for their age and grade level (PJT: raw score 6, 15th percentile; AKR: raw score 23, 14th percentile). Their very high levels of performance on tests of auditory word comprehension (e.g., PPVT) rule out difficulties in phonological and semantic processing of the dictated stimulus words. Furthermore, we have presented evidence that their spelling difficulties do not arise from deficits in orthographic working memory or peripheral production processes (e.g., handwriting processes). Consequently, a deficiency in orthographic long-term memory is the most likely proximal cause of their difficulties in spelling irregular words. That is, both PJT and AKR show impairment in learning the spellings of words.

Evaluation of the children's spelling errors supports the conclusion of deficient lexical-orthographic knowledge. AKR's errors were examined on the 153 words she spelled from the DiSTi and the two forms of the WRAT3 Spelling task. Her error rate on these words was 67% (103/153)². The vast majority of her errors (83/103, or 81%) were phonologically plausible misspellings (e.g., PURCHASE → PURCHES, BISCUIT → BISCKET). These errors may be interpreted by assuming that AKR had not learned the spelling of the word and relied instead on her well-developed sublexical sound-to-spelling conversion processes. Most of the remaining errors involved letter omissions, transpositions or insertions in long, low-frequency words (e.g., ENTHUSIASM → ENTHUSAM, OPPORTUNITY → OPPORTINY, CACOPHONY → CONCAFONY). These errors may reflect occasional (and unremarkable) difficulty in spelling long words sublexically, and/or partial knowledge of the word's spelling.

²Note that both the DiSTi and the WRAT include words that a child of AKR's age would not be expected to spell correctly (e.g., CACOPHONY, IRIDESCENCE). Accordingly, although her 67% error rate is higher than expected for a child of her age, it does not reflect catastrophic spelling impairment. Note also that the spelling corpus for PJT, described below, includes a larger proportion of words a child of his age would be expected to spell correctly, and hence his somewhat lower error rate (53%) does not imply a less severe spelling impairment.

For PJT, we considered word stimuli from the WRAT3, DiSTi, and Dolch Word tasks. Across these tasks, his error rate was 53% (354/672). Phonologically plausible misspellings (e.g., BOX → BOCS, RAIN → RANE) accounted for 60% of the errors (211/354). The phonologically plausible errors presumably represent instances in which PJT had not learned the spelling of the word, but succeeded in spelling it sublexically using his sound-to-spelling correspondence knowledge. The remainder of his errors were implausible spellings. Most of these errors apparently reflected the deficient sublexical spelling processes evident in the pseudoword spelling tasks. For example, PJT mapped /θ/ to T rather than TH in spelling TOGETHER as TEGETER and THINK as TINGK; he realized /ʒ/ as R in BIRD → BRD, UNDER → UNDR, and NEVER → NEVR; and he spelled /ɑʊ/ variously as O, U, AW, and AU in errors such as GROUND → GROND, ABOUT → ABUT, COW → CAW, and DOWN → DAUN.

PJT also made errors that appeared to reflect partial learning of a word's spelling. Examples include ELEPHANT → ELAPINT, in which /f/ is mapped to P, and KNOW → KOW, in which /n/ is mapped to K. These mappings never occur in English words, and PJT never produced the mappings in spelling pseudowords—for example, he always realized /f/ as F in pseudoword spelling tasks. Consequently, it is likely that the errors reflect partial knowledge of the spellings for ELEPHANT and KNOW. Partial lexical knowledge may also underlie a number of errors in which PJT produced the correct letters but in the wrong order (e.g., MOTHER → MHTOER, THEY → THYE, BLUE → BULE). He almost never made letter transposition errors in pseudoword spelling, indicating that these errors in spelling words are unlikely to reflect deficient sublexical sound-to-spelling conversion processes or orthographic working memory failures. Rather the ordering errors probably represent cases in which PJT had learned the letters in a word's spelling, but had not entirely mastered the ordering.

Proximal sources of impairment: conclusions

PJT and AKR are both significantly impaired in spelling. The results we have presented indicate that the spelling deficits do not originate in word comprehension, orthographic working memory, or peripheral production processes. For AKR, sublexical sound-to-spelling conversion processes are also normal, but for PJT these processes are deficient. Finally, both children show clear impairment in orthographic long-term memory: with respect to stored knowledge of word spellings PJT and AKR are well below the level expected given their ages and grade levels. Furthermore, the home-training results for PJT (discussed in a later section) suggest continued difficulty in learning of word spellings. This deficiency in learning the spellings of words is especially striking in light of their very strong cognitive abilities and apparently excellent reading skills.

These conclusions raise two important sets of questions. The first concerns the relationship between spelling and reading in PJT and AKR. If the same lexical-orthographic knowledge underlies both reading and spelling, we might expect the children to show orthographic long-term memory impairment in reading tasks; yet both PJT and AKR appear to be excellent readers. The second set of questions concerns distal causes of the children's spelling impairments: Why do PJT and AKR have difficulty learning the spellings of words,

and why does PJT also have difficulty acquiring knowledge of sound-to-spelling correspondences? We take up these questions in the following sections.

Reading

Sublexical reading processes were evaluated via pseudoword reading tasks, and lexical reading processes were assessed with several tasks involving word stimuli.

Reading pseudowords

Both PJT and AKR read aloud the 40 pseudowords on the Castles and Coltheart Reading Test 2 (CC2; Castles et al., 2009) and the 45 pseudowords making up the Word Attack subtest of the Woodcock Reading Mastery Test-Revised (WRMT-R; Woodcock, 1987). On both tasks AKR scored slightly below the mean for her age and grade level, but within the average range (27th percentile on the CC2 pseudowords, and 37th percentile on the WRMT Word Attack subtest). Examination of her responses revealed that most of her errors were rather subtle (e.g., CEDGE → /kɛdʒ/, in which she read C as /k/ despite the following E), and that she frequently read long, polysyllabic pseudowords correctly (e.g., SPOLTCHURB, HOPDALHUP). Note also that AKR scored at the 97th percentile on the WIAT Pseudoword Decoding task. Hence, AKR's sublexical reading processes, like her sublexical spelling processes, appear to be adequate.

PJT's pseudoword reading performance was average to superior: He scored at the 47th percentile for the CC2 pseudowords, and at the 84th percentile on the WRMT Word Attack subtest. These results stand in clear contrast to his deficient performance (below the 10th percentile) on pseudoword spelling tasks. Examination of PJT's pseudoword reading performance revealed that he was usually able to perform accurate spelling-to-sound conversion even for the correspondences between single phonemes and multiple graphemes that proved so difficult for him in pseudoword spelling. For example, he successfully read the pseudowords ZOATH, BRINTH, MORSHAB, PHET, and PLEECH, despite his inability to produce TH, SH, PH, or CH correctly in spelling. PJT, then, evidenced strong sublexical reading skills despite his impairment in sublexical spelling processes.

Reading regular and irregular words

Both PJT and AKR read aloud the 40 regular and 40 irregular words on the CC2, and both children showed high levels of performance for both sets of words: PJT scored at the 88th and 97th percentiles for regular and irregular words, respectively, and AKR scored at the 98th and 87th percentiles. These results demonstrate very strong orthographic long-term memory representations and processes for reading in both children. That is, both PJT and AKR have strong stored knowledge of the orthographic forms of words, and can access this knowledge when reading. Their scores for irregular words are especially diagnostic, given that reliably accurate reading of irregular words can be accomplished only by accessing stored knowledge of the words in orthographic long-term memory.

Discriminating words from pseudohomophones

Additional evidence of strong lexical reading processes emerged from the Test of Orthographic Choice (Kohnen, Anandakumar, McArthur, & Castles, 2012). In this task, 30 word-pseudohomophone pairs (e.g., GLANCE-GLANSE) are presented, and the examinee is asked to select the real word in each pair. This task requires knowledge of the spellings of words, because the decisions cannot be made on the basis of the pronunciation of the word. Both PJT and AKR showed excellent performance, with percentile ranks of 91 and 85, respectively.

Defining homophones

In this task PJT and AKR were asked to provide definitions for homophones presented visually (e.g., MANE, PEACE). AKR was tested with 46 homophones, and PJT with 30. Both children showed excellent performance: 45/46 (98%) for AKR, and 26/30 (87%) for PJT. In a separate homophone discrimination task, PJT was asked to define paired homophones (e.g. PLANE and PLAIN) and indicate which definition went with each spelling. He defined 29/30 (97%) of the homophones correctly and matched each of those definitions to the correct spelling. Like the word-pseudohomophone discrimination task, the homophone definition task requires knowledge of the spellings of words. For example, to give the correct definition for PEACE, one must be able to distinguish this word from PIECE, and this distinction can only be made via knowledge stored in orthographic long-term memory. The children's definitions demonstrated not only their strong lexical-orthographic knowledge, but also their exceptional intellectual ability. For instance, PJT defined BEE as "the insect that most people are afraid of that stings but also makes honey," and AKR defined WRITE as "using an agreed on set of symbols to create messages that others can understand."

Reading migratable words

The Letter Position Test (Kohnen, Marinus, et al., 2012) was administered to both PJT and AKR. Stimuli are 30 word pairs in which the two words in each pair are anagrams (e.g., LOTS-LOST, BOLT-BLOT, THERE-THREE). The words are presented one at a time for reading aloud, with the first 30 trials consisting of one word from each pair, and the second 30 trials including the remaining word from each pair. High rates of letter migration errors (e.g., reading LOTS as LOST) are taken to suggest difficulty in perceiving or encoding letter order. Errors are categorized as migration errors, other word errors, or other errors.

Results are presented in Table 3. PJT clearly had no difficulty with the migratable words, scoring at the 95th percentile for overall accuracy and making very few letter migration errors. AKR's performance was in the low-average range, with accuracy at the 27th percentile and migration errors at the 21st percentile. She made 7 letter migration errors (e.g., WARP → WRAP), whereas PJT (who is two years younger) produced only 2. Although AKR's performance was not severely impaired, these results raise the possibility of a mild deficit in encoding or retaining the ordering of letters. This test was administered for a second time to AKR in March 2017 to determine if the difficulty with migratable words was reliable; these results are also reported on Table 3 and indicate a continued, consistent difficulty with migratable word reading.

Reading: conclusions

Whereas PJT demonstrated clear lexical (orthographic long-term memory) and sublexical deficits in spelling, we were not able to find any evidence of lexical or sublexical reading difficulties. His performance was well above average in reading regular words, irregular words, migratable words, and pseudowords (the only exception being an average score on one of the two pseudoword reading tasks). In addition, he showed excellent performance in defining homophones and distinguishing homophones from pseudohomophones. We conclude that PJT shows a striking dissociation between significantly impaired spelling and very strong reading ability.

AKR also performed very well on most of the reading tasks, including reading regular and irregular words, distinguishing homophones from pseudohomophones, and defining homophones. Her performance on pseudoword reading tasks was more variable, ranging from low average to superior, suggesting little if any deficiency in sublexical reading processes. However, a potential reading deficiency is indicated by her frequent letter migration errors (e.g., FILES → “flies”) on the letter position test. While her performance at the 21st percentile does not indicate a severe impairment, it is striking in the context of her otherwise high levels of word reading. This result raised the possibility of impairment in encoding or retaining letter order information. In discussing distal sources of impairment in the following section, we report some additional relevant evidence.

Spelling: distal sources of impairment

We next explore possible distal (underlying) causes of the developmental spelling deficiencies evidenced by PJT and AKR. Specifically, we consider potential deficits in phonological processing, motor skills, visual attention span, learning and memory (visual and verbal), letter order representation, and orthographic learning.

Phonological processing

Deficits in phonological awareness and/or phonological processing are widely thought to be prominent underlying causes of developmental dyslexia (e.g., Bradley & Bryant, 1978, 1983; Bruck, 1993; Liberman, 1973; Stanovich, 1988; Wagner & Torgesen, 1987), and many theorists have suggested that phonological deficits are also implicated in developmental dysgraphia (e.g., Campbell & Butterworth, 1985; Snowling et al., 1986; but see Castles & Coltheart, 2004). For PJT and AKR the possibility of phonological impairment was assessed with the Comprehensive Test of Phonological Processing (PJT: CTOPP [Wagner, Torgesen, & Rashotte, 1999]; AKR: CTOPP-2 [Wagner, Torgesen, Rashotte, & Pearson, 2013]). The CTOPP includes multiple tasks probing phonological awareness, phonological memory, and other aspects of phonological processing. The results, presented in Table 4, indicate normal levels of performance for both children. Therefore, despite the fact that impaired phonological processing/awareness is the most widely held and entrenched hypothesis regarding underlying causes of developmental dyslexia and dysgraphia, we find no evidence of phonological deficits in PJT or AKR.

Handwriting and motor skills

Both families expressed concern about the children's poor and effortful handwriting. The writing samples in Figures 2 and 3 illustrate that both children's handwriting is often unclear. Our observations indicated that PJT had especial difficulty and experienced considerable frustration with handwriting. However, both children showed normal performance on tests probing the general motor skills required for writing. On the VMI Motor Coordination Test (Beery & Beery, 2010), which assesses fine motor control in drawing geometric figures, PJT achieved a standard score of 103, placing him in the 58th percentile. On the Fine Motor Precision, Fine Motor Integration, and Manual Dexterity subtests of the Bruininks-Oseretsky Test of Motor Proficiency (Bruininks & Bruininks, 2005) AKR's scaled scores were 22, 15, and 20 respectively, all in the average to above-average range. Also, on the Test of Handwriting Skills – Revised (Milone, 2007), all of her skill scores were average or better and her overall standard score was 115.

Also relevant is that both PJT and AKR showed comparable spelling impairment in written and oral spelling tasks, suggesting that handwriting difficulties were not playing a significant role in their spelling performance. Conceivably, struggles with handwriting could have contributed to difficulty in learning the spellings of words, especially for PJT, who expended much more effort with handwriting, and experienced much more frustration, than AKR. The need to focus on mechanical aspects of writing could perhaps have reduced the amount of attention available for learning to spell. Nevertheless, it seems unlikely that handwriting difficulties can entirely account for the spelling deficiency in either PJT or AKR.

Visual attention span

Valdois and colleagues (Ans, Carbonnel, & Valdois, 1998; Bosse, Tainturier, & Valdois, 2007; Bosse & Valdois, 2009; Valdois, 1996; Valdois et al., 2003) have proposed that reduced visual attention span is the underlying cause in some cases of developmental dyslexia. According to their argument, difficulty in processing multiple letters in parallel impairs the ability to learn the orthographic forms of words, as well as the ability to recognize words rapidly during reading. To the extent that reduced visual attention span impairs orthographic learning, one might expect visual attention span deficiencies to affect the development of spelling as well. Accordingly, we evaluated visual attention span in PJT and AKR via two tasks developed by Valdois et al. (2003; see also Bosse & Valdois, 2009). The task of principal interest was a global report task, in which strings of 5 consonants are presented briefly (200 ms), and the participant is asked to report as many of the letters as possible, in any order. This task requires distribution of visual attention across multiple letters. The second task was a letter identification control task designed to assess whether weak performance in global report could be due to deficient processing of individual letters. Single letters are presented at one of 5 exposure durations ranging from 33–101 ms, and the participant is asked to name each letter. Bosse and Valdois (2009) report control data from French children in grades 1, 3, and 5.

Both PJT and AKR showed entirely normal performance on both the letter identification and global report tasks. In letter identification, their scores were 142 (PJT) and 150 (AKR), which are well above average relative to the Bosse and Valdois (2009) control children. On

the global report task PJT reported 76% of the letters correctly, and was entirely correct on 20% of the strings. On both measures his performance fell between the means for Grade 1 and Grade 3 (Bosse & Valdois, 2009), consistent with his second grade placement. AKR correctly reported 98% of the letters and 90% of the whole strings, well above average for her grade level. We conclude that reduced visual attention span is not a distal cause of the spelling deficiency for either PJT or AKR.

Encoding and representing letter order

Romani et al. (1999) proposed that an impairment in processing letter order is the distal cause in some cases of developmental surface dysgraphia. As discussed above, PJT's spelling errors included some letter transpositions (e.g., BLUE → BULE), although he evidenced no difficulty with letter order in reading. AKR did not make large numbers of letter transposition errors in spelling, but in reading migratable words she made letter migration errors at a higher than normal rate (e.g., reading WARP as WRAP).

Romani et al. (1999) specifically proposed a deficit in the learning and recall of letter order information. We were not able to examine these specific skills with PJT and AKR, although we did briefly examine their perception of letter order in a letter string reading task. PJT and AKR were presented with 12 strings of 12 consonants each, and asked to name the consonants in order. AKR read 99.3% of the letters correctly (143/144), and PJT was correct on 98.6% (142/144). Neither child made any letter order errors. As an informal basis for comparison, a college student with a letter order encoding deficit tested in our lab read correctly only 83% of the letters in 12-letter strings, making frequent letter transpositions.

These results do not rule out some forms of letter order impairment (e.g., selective impairment in retaining information about letter order in orthographic long-term memory). However, the absence of letter order errors in reading aloud long consonant strings does suggest that neither child is significantly impaired in the perception of letter order.

Visual and verbal learning and memory

Deficits affecting O-LTM in spelling could result from an underlying deficit in learning and long-term retention of verbal and/or visual information. These possibilities were assessed with three tests.

WRMT-R Visual-Auditory Learning Task—The Visual-Auditory Learning Task (Woodcock, 1987) assesses short-term learning of associations between written symbols and spoken words. The participant is shown a small set of symbols and told that each symbol corresponds to a particular word or morpheme (e.g., a triangle inside a circle = BOB, and a small circle on top of a big circle = ON). The participant is then shown strings made up of the symbols and asked to “read” them as phrases or sentences. This process repeats, adding a few more symbols with each iteration until there are a total of 28 symbols to be remembered. Both PJT and AKR showed excellent performance, scoring at the 97th and 71st percentiles, respectively.

Children's Memory Scale—PJT's visual and verbal short-term learning and memory were also evaluated with the Children's Memory Scale (CMS; Cohen, 1997). Visual tests include dot locations and faces and verbal tests include stories and word pairs. The delayed conditions evaluate retention after 30 minutes. PJT performed above the 75th percentile on all subtests (see Table 5).

Gascoigne et al. (2014) Visual Learning Task—Because the orthographic learning required for skilled reading and spelling involves memory over a longer time scale than evaluated by either the WRMT or the CMS, we administered a visual learning task from Gascoigne et al. (2014) that included assessment of learning following a one-week delay. The task was based on a spatial memory task from Hepworth and Smith (2002), and evaluated retention after 30 minutes (short delay) and 7 days (long delay).

The to-be-learned stimuli consist of 10 visual designs that occupy specific positions within a 6×4 grid. In the initial learning phase, the participant views the designs in the grid. Then an empty grid is presented with the 10 designs off to one side, and the participant is asked to drag and drop each design into the appropriate grid location. The study/test process is then repeated until perfect performance is achieved on two consecutive learning trials, up to a maximum of 12 trials. Recall is then tested (using the same procedure as in the learning phase) at short (thirty minute) and long (seven day) delays; the seven-day assessment also includes a recognition test. Results are reported in Table 6. No data were recorded for PJT's learning and short-delay recall phases due to accidental premature closure of the online program (although his mother estimates that three or four learning trials were required to reach the learning criterion). Both PJT and AKR show normal performance on all measures (trials needed for initial learning and memory at all delays).

Orthographic learning

In PJT's case, given the lack of evidence for the distal deficits that were examined, we considered the possibility of a specific impairment of lexical orthographic learning (Hanley, Hastie, & Kay, 1992). To examine this possibility, we developed a home training program for PJT in which his mother worked with him to help him learn word spellings. The program was principally aimed at remediation, but also allowed us to assess informally the extent to which PJT continued to struggle in learning to spell words. Training stimuli were 76 words from the Dolch List (Dolch, 1948) that PJT had spelled incorrectly in both written and oral spelling. It is important to point out that these are all words that PJT can easily read. Training sessions were carried out several days per week. For the first session, 6 of the 76 training stimuli were selected as the initial training set. In each training session, training words were presented for writing to dictation. If PJT spelled a word incorrectly, he copied it three times and then was asked to spell it to dictation again. A training word that was spelled correctly on the first attempt in three consecutive training sessions was moved to a "learned" set, and a new word was added to the training set. Three words from the learned set were retested in each training session. When a learned word was correct at retest, it remained in the learned set; however, if it was spelled incorrectly the word was returned to the training set and underwent the training process again. Three to five words were trained on each session.

Over a period of 15 months (82 training sessions) PJT reached the learning criterion (correct spelling in three consecutive sessions) for 68 of the 76 training words. He took an average of 4.8 training sessions to learn the spelling of a word. However, some words required many more sessions. For example, BLUE, PLEASE, and BETTER each required more than 12 sessions to reach the learning criterion. Furthermore, 17 of the 68 words that reached criterion were subsequently misspelled during retesting.

The following examples illustrate the progression of training and retraining. The spellings shown are those PJT produced when the training word was first tested in each session.

- “blue”: BULE $\times 2 \rightarrow$ BULY $\rightarrow$ BULE $\rightarrow$ BLUE $\times 3 \rightarrow$ BULE $\times 2 \rightarrow$ BLUE $\times 4$
- “floor”: FLORE $\rightarrow$ FLOR $\rightarrow$ FLUR $\rightarrow$ FLOUR $\rightarrow$ FOORE $\rightarrow$ FLOOR $\times 5 \rightarrow$ FOOR $\rightarrow$ FLOOR $\times 4$
- “brown”: BROWN $\rightarrow$ BROMN $\rightarrow$ BRON $\rightarrow$ BRAWN $\rightarrow$ BROWN $\rightarrow$ BROUN $\rightarrow$ BRAWN $\rightarrow$ BROWN $\times 4$
- “does”: DUZE $\rightarrow$ DOSE $\times 6 \rightarrow$ DOES $\times 4 \rightarrow$ DOSE $\times 3 \rightarrow$ DOES $\times 4 \rightarrow$ DOSE $\rightarrow$ DOES $\times 2$

To formally assess training effects, a spelling to dictation task was administered with 37 trained and 37 untrained control words matched for length, frequency, and phoneme-grapheme mapping probability. PJT’s accuracy on the trained words increased from 64.9% on the first training attempt to 75.7% after training, a difference that was not statistically significant, $\chi^2(1, N = 74) = 1.03, p = 0.31$. Further, the accuracy on the trained words was not statistically different from the accuracy on the matched untrained words (62.2%), $\chi^2(1, N = 74) = 1.58, p = 0.21$. Similar results were observed when we scored PJT’s spelling responses in terms of letter accuracy: Accuracy on trained words increased by an average of 4.3% from pre- to post-training (89.8% to 94.0%), $t(36) = 1.39, p = 0.17$, and post-training letter accuracy on trained words did not differ statistically from accuracy on untrained words (90.5%), $t(36) = 1.09, p = 0.28$.

The very limited improvement in PJT’s spelling accuracy for trained words shows a very striking level of difficulty with orthographic learning. Although we do not have the necessary control data to establish that PJT’s learning performance is impaired, the results seem at odds with his strong learning and memory abilities, and especially with his superior learning of the lexical orthographic knowledge required for reading. Hence, the results suggest that PJT continues to experience significant difficulty in acquiring the lexical orthographic knowledge necessary for successful spelling of words.

Probing orthographic word knowledge

With the goal of developing a better understanding of the difficulties PJT faces in learning lexical orthographic representations, we administered a spelling-probe task in which words were dictated and PJT was asked whether specific letters were present in the spellings of the words. This task tests knowledge about the letter identities in a word’s spelling independent of knowledge about letter order. In studies of acquired dysgraphia, spelling-probe

performance has been shown to generally mirror spelling performance (Rapp & Kong, 2002). Furthermore, the spelling-probe task has been shown to recruit the same neural network that is activated by writing (Rapp & Dufor, 2011). Given PJT's frustration with producing written or oral spelling, we hoped that this task would provide a less stressful means of collecting detailed and targeted spelling data.

We tested PJT in two probe conditions, both of which involved asking, on each trial, about two letters. In the dual-letter condition a word was dictated and PJT was asked, for each of the two letters, whether the letter was present in the word—for example, the word TEN was dictated, followed by the questions, “Is there a T in TEN?”, and “Is there an N?” In the repeated-letter condition the same letter was queried twice—for example, dictated stimulus DID with questions “Is there a D in DID?”, and (contingent upon a “yes” response) “Is there another D?”

Stimuli were taken from the pool of words PJT had already spelled in prior testing. For the repeated-letter condition we constructed sets of four words: one with a repeated letter (e.g., DID), two controls with a single instance of the letter (DAY and HAD), and one foil which did not contain the letter (HIM). The words in each set were matched in length and lexical frequency. The first and second control words included the probed letter in the same positions as the first and second occurrences of that letter in the repeated letter word. For the dual-letter condition the four-word sets consisted of one word with both probed letters (e.g., TEN for probe letters T and N), two controls with one of the letters (TOP and RAN), and a foil with neither letter (BOX). We created 15 sets of 4 items for each condition, and randomized the ordering of items within conditions.

To allow for meaningful comparisons between letter-probe and spelling results, we scored PJT's prior spellings of the probed words as if the probe questions had been answered on the basis of those spellings. For example, in the repeated-letter condition the word AGAIN was presented with the probe letter A. PJT had previously spelled AGAIN as AGGANE and AGEN. For the first probe question (“AGAIN, is there an A in AGAIN?”) the corresponding scores for both prior spellings would be 1 (i.e., correct), because both AGGANE and AGEN include an A. For the second probe question (“Is there another A?”), AGGANE was scored 1 because it has two As, but AGEN, which includes only one A, was scored 0.

Collapsing across conditions and probe questions (first and second) PJT's accuracy in the letter probe task was almost identical to his spelling accuracy. Probe-task accuracy was 90.6% (203/224), compared to 89.3% (393/440) for prior spelling responses scored for comparability with probe responses, $\chi^2(1, N = 664) = 0.15, p = 0.70$.

In both repeated and dual-probe conditions, the control words showed lower accuracy for letters occurring later in the word: Probe-task accuracy decreased from 100% (30/30) for the earlier letter (or instance of a repeated letter) to 80% (24/30) for the later letter, $p = 0.024$ by Fisher's exact test. For spelling, the accuracy difference between earlier (89.3%) and later (74.1%) letters was marginally significant; $\chi^2(1, N = 114) = 3.40, p = 0.065$.

In dual-letter words (i.e., words containing both of the probed letters), the decrease in accuracy from the earlier to the later letter was the similar to that observed for the control

words (100% to 80% in the letter-probe task, 93.5% to 83.9% for spelling). Probably as a result of the small sample sizes (15 observations per cell for the letter-probe task, 31 per cell for spelling), these differences did not reach significance: probe task $p = 0.22$, Fisher's exact test; spelling $\chi^2(1, N = 62) = 0.64, p = 0.42$.

For repeated-letter words (e.g., DID) the accuracy decrease between the questions probing the first and second occurrence of the letter was substantially larger and highly reliable. In the letter probe task, PJT's accuracy decreased from 100% (15/15) on the first probe to 53.3% (8/15) on the second; $p = 0.006$, Fisher's exact test. His spelling performance was nearly identical, dropping from 96.7% correct (29/30) on the first instance of the letter to 53.3% (16/30) on the second instance; $p < 0.001$, Fisher's exact test.

These results support several conclusions. First, the strong similarity between PJT's spelling-probe and written spelling performance suggests that spelling-probe task is a viable alternative to traditional spelling tasks, and one that may provide a less stressful means for evaluating spelling representations and processes. Second, PJT's poorer performance on later than earlier letters in a word suggests that his difficulty in learning the letter identities making up the spelling of a word is affected by their position in the word. This conclusion is confirmed by the results shown in Figure 4, which depicts accuracy by normalized position (as in Wing & Baddeley, 1980) for PJT's spelling data. The position effects raise a number of questions that could be investigated in future work. Finally, PJT's surprising difficulty with repeated letters suggests that he may have some specific difficulty in learning or processing representations involving multiple tokens of the same abstract letter identity. However, additional testing would be required to clarify the nature of the difficulty.

General discussion

We have reported on an investigation of two children with developmental dysgraphia, PJT and AKR. The aims of the study were to identify causes of the children's spelling impairments, and to examine the relationship between their spelling and reading. The design, selection of tasks, and interpretation of results were guided by a theoretical framework specifying the representations and processes involved in adult spelling and reading. We first evaluated a number of potential proximal causes of the spelling impairments—that is, potential deficiencies within the spelling system. This evaluation identified clear and significant deficits in orthographic long-term memory in both children, as well as an additional impairment in the sound-to-spelling conversion system in PJT. Based on these findings, we then examined orthographic long-term memory and sublexical spelling-to-sound conversion processes in reading. We found no evidence of any reading impairment for PJT, indicating a striking dissociation between impaired spelling and superior reading. For AKR we found evidence of mild difficulty with letter order in reading, revealed by errors in reading migratable words (e.g., reading TRIED as TIRED). Whether this letter order difficulty is related to AKR's spelling impairment is, however, uncertain, especially given that letter order errors were not prominent in her misspellings.

Finally, we explored potential distal deficits that could have led to the children's proximal deficits within the spelling system. We considered phonological processing and awareness,

motor skills, visual attention span, encoding and representing letter order, and visual and verbal learning and memory. Neither child exhibited distal deficits that could clearly account for their difficulty in learning word spellings or (in the case of PJT) sound-to-spelling conversion. AKR's difficulty with migratable words in reading may indicate an underlying difficulty with letter order that affects both reading and spelling. Conceivably, the letter order difficulty could reflect a more general underlying deficit in sequence processing. However, based on the limited testing results we are not able to reach firm conclusions regarding the distal cause of her difficulty in reading migratable words, nor about whether this distal cause would also explain some of her difficulties in spelling.

Distal causes for developmental dysgraphia

Although we evaluated PJT and AKR for the deficits most commonly proposed as distal causes of developmental dysgraphia (e.g., phonological processing deficit, impaired visual attention span), we failed to find distal deficits that would account for the spelling impairment in either child. This outcome allows several possible interpretations. First, and most obvious, our testing clearly did not examine every cognitive process that could conceivably contribute to acquisition of orthographic long-term memory representations (or, for PJT, sound-spelling correspondence knowledge). Hence, the distal deficits in PJT and AKR may affect cognitive processes we did not examine.

A second possibility is that the distal deficits in PJT and AKR lay in processes that were deficient at an earlier period, but that have now "caught up" in their development and are functioning normally. On this account, the earlier distal deficiencies delayed the acquisition of spelling knowledge, with effects on spelling performance that were clearly detectable in our study. However, the distal deficiencies could not themselves be detected, because they had resolved by the time of our study. For example, for PJT one might consider whether his early literacy experiences in the French immersion program could have delayed his acquisition of English spelling. This possibility cannot be ruled out entirely, although we note that the French program clearly did not prevent him from attaining a high degree of proficiency in English reading. More generally, we cannot exclude for either child the possibility of an earlier distal deficit that had resolved by the time of our testing, but at least for PJT this account seems rather implausible, given his continuing struggle to learn spellings in the home training program.

A final possibility, and perhaps the most interesting one, is that the underlying cause of the spelling impairment in PJT and/or AKR is not a deficiency in some general cognitive function (e.g., visual attention span), but rather a highly selective deficit that affects only the acquisition of orthographic knowledge and perhaps (at least for PJT) only the orthographic knowledge required for spelling. Given such a deficit we would not expect to see difficulty in tasks probing more general cognitive processes. In support of the plausibility of a selective deficit in orthographic learning is the notion that although spelling and reading must rely on evolutionarily older underlying skills, they cannot be reduced to these skills. In other words, something new must be learned when literacy is acquired. This new learning may involve tuning evolutionarily older processes and representations in ways that are specifically appropriate for written language processing and creating connections that are

needed to support orthographic knowledge and processing. It is conceivable that the specific type of learning that is required to tune and interconnect brain areas is disrupted in certain individuals while the underlying skills themselves are intact.

Further research and theory development will be important for advancing our understanding of distal causes in developmental dysgraphia (and dyslexia). For example, advances in theory concerning the cognitive processes implicated in acquiring lexical orthographic representations would be helpful for systematically defining the set of general cognitive deficits that could potentially act as distal causes of developmental dysgraphia, and for evaluating the plausibility of hypotheses positing highly specific orthographic learning deficits.

Implications of PJT's spelling-reading dissociation

The most striking result from the present study is the dramatic dissociation between PJT's impaired orthographic long-term memory processes in spelling, and superior orthographic long-term memory processes in reading. This dissociation reveals that at least certain aspects of the system for processing lexical orthographic representations are not shared between reading and spelling.

There are two general classes of interpretation for this dissociation. The first posits distinct sets of lexical orthographic representations for reading and spelling, whereas the second assumes distinct processes for activation/retrieval of information from a single orthographic long-term memory system shared between reading and spelling. We discuss each of these in turn and suggest a specific interpretation that builds on theoretical proposals in the domain of spoken word production.

In the adult neuropsychological literature, dissociations in orthographic long-term memory between reading and spelling are often described as surface dyslexia without surface dysgraphia, and vice versa (Behrmann & Bub, 1992; Coltheart & Funnell, 1987). To account for these findings, some researchers have posited different orthographic long-term memory stores for reading and spelling (often referred to as the orthographic input and output lexicons, respectively; see, e.g., Allport & Funnell, 1981; Caramazza, Miceli, Villa, & Romani, 1987; Hanley & McDonnell, 1997; Margolin, 1984; Patterson & Shewell, 1987). We refer to this account as the *modality-specific orthographic representations hypothesis* (here using *modality* to refer to reading vs. spelling). According to this hypothesis, PJT has a well-developed reading O-LTM system while his spelling O-LTM system is deficient.

A modality-specific representations hypothesis could assume either that the orthographic representations for reading and spelling have identical, duplicated content, or that the two sets of representations differ in some way. A proposal of duplicated content appears somewhat unappealing on grounds of parsimony, and because independent motivation for positing such duplication is lacking. The possibility of different types of orthographic lexical representations for reading and spelling is more interesting, but until specific proposals are put forth regarding the nature of the differences, this potential version of the modality-specific representations hypothesis is little more than a promissory note.

Also worth noting is that neither version of the modality-specific hypothesis provides a straightforward account of the various reported associations between reading and spelling we discussed in the introduction, and neither version straightforwardly allows for the generalization of orthographic lexical knowledge from reading to spelling.

A second approach to interpreting PJT's reading-spelling dissociation is to posit lexical orthographic representations that are shared between reading and spelling, with modality-specific processes for accessing and/or processing the stored orthographic knowledge. In reading, lexical orthographic information is accessed from representations of letter identities and order (see Figure 5), whereas in spelling the lexical orthographic knowledge is accessed from phonological or semantic representations. This *shared orthographic representations hypothesis* would interpret dissociations between reading and spelling by positing deficits to procedures that are specific to reading or spelling. For PJT the interpretation would be as follows: The shared lexical orthographic representations were well learned, as were the processes for accessing these representations in reading; however, PJT had difficulty developing the processes required for accessing the lexical orthographic representations from phonological LTM and/or the semantic system. As a consequence, when PJT attempts to spell a word, the process of activating the lexical-orthographic representation may yield only weak, fragmentary activation, or may fail entirely, leading to a spelling error.

One challenge for this hypothesis concerns the difficulty of testing the critical assumption that lexical-orthographic representations are intact in cases of impairment attributed to access deficits (see Rapp & Caramazza, 1989). For example, how could we find evidence that the lexical-orthographic representations that mediate spelling are intact in PJT?

Progress in evaluating these theoretical alternatives empirically, and even in assessing their plausibility, will depend on advances in our thinking about the nature of lexical orthographic representations and computations. For example, implicit in the preceding discussion is the notion that orthographic long-term memory in some sense contains information about the orthographic forms of words (e.g., the information that the word DOG corresponds to the letter sequence D-O-G). Given this implicit assumption, the hypothesis of separate lexical orthographic representations for reading and spelling runs the risk of positing duplicate representations of this information, as we have discussed. Consider, however, the somewhat different conceptualization illustrated in Figure 6. According to this view, orthographic lexical representations consist of word nodes that do not "contain" information about letter sequences; rather the word nodes are linked to letter nodes, and thereby provide a basis for activating (in the case of spelling) or being activated by (in the case of reading) the corresponding sequence of letter representations.

Within this general perspective, we could formulate both modality-specific and shared orthographic representations hypotheses. In fact, we could develop a broad range of hypotheses, depending upon whether we assume that the word nodes, the letters nodes, and the word-letter connections are shared or separate for reading and spelling. Figure 6 illustrates a version in which lexical nodes are shared between reading and spelling, but the letter nodes, and the links between letter and word nodes, are separate for reading and spelling. This nodes-and-links perspective is consistent with a widely adopted theoretical

framework for spoken word production (Dell, 1986; Levelt, Roelofs, & Meyer, 1999) which posits that production involves two stages: activation of a word node (lexical selection), leading in turn to activation of the nodes for the constituent phonological segments (segmental encoding).

Applied to reading and spelling, the nodes-and-links perspective is not radically different from the conceptual framework in which we think of the orthographic LTM system as “containing” knowledge about letter sequences. Indeed, in Figure 6 we could draw a box encompassing the word nodes, the letter nodes for spelling, and the word-letter connections, and we could then label this box *orthographic LTM system for spelling*. Nevertheless, the nodes-and-links perspective is potentially valuable, for a number of reasons. First, as we have just mentioned, this perspective is helpful in articulating potential versions of the modality-specific and shared orthographic representations hypotheses. Second, this perspective may encourage us to rethink the distinction between separate representations and separate procedures. For example, does Figure 6 illustrate a modality-specific orthographic representations hypothesis, or a modality-specific procedures hypothesis? If we consider the orthographic representations to consist solely of the word nodes, then the figure shows representations that are shared between reading and spelling, with modality-specific processes for accessing and using these representations. However, if we conceive of orthographic representations as encompassing the word nodes, the letter nodes, and the links between them, then Figure 6 shows largely (although not entirely) separate representations for reading and spelling.

The nodes-and-links perspective may also affect our views on the plausibility of various theoretical alternatives. If we conceive of orthographic LTM as a container holding knowledge of letter sequences, then positing two separate orthographic LTM systems may seem implausible. However, the hypothesis illustrated in Figure 6 may appear more plausible, despite positing largely separate orthographic knowledge (embodied in the word-letter links) for reading and spelling.

We do not intend to suggest that theorists should abandon the lexicon-as-knowledge-container perspective in favor of a nodes-and-links perspective. Rather, our principal point is that contrasting these conceptual frameworks reveals the extent to which our formulation of theoretical alternatives, our assessment of their plausibility, and perhaps even our arguments relating empirical evidence to theoretical claims, rest upon implicit assumptions that have not been cashed out. Considerable theoretical development is needed if we are to make progress in understanding the relationship between reading and spelling in a way that accounts for the detailed patterns of data available in the literature. Our hope is that this theoretical work, in conjunction with the documentation of increasingly detailed and distinct performance patterns such as those reported here, will move us towards a position in which we will be able to develop proposals with predictions that would adjudicate between them.

Summary and conclusions

We have presented a strong case of a dissociation between reading and spelling that clearly reveals that at least certain representations and/or processes involved in reading and spelling

develop independently from one another. These findings provide important additional constraints on theories of reading and spelling. We have suggested that the full set of available findings seem to be well accommodated in a system in which reading and spelling share at least some components of orthographic long-term memory with modality-specific access and segmental encoding processes. This provides a reasonably parsimonious framework for understanding both the associations and the dissociations between reading and spelling that have been reported to date.

Acknowledgments

We are grateful to PJT, AKR, and their families for their support for this project and their cheerful participation in long hours of testing. We are also grateful to Michael Gascoigne for his help with testing materials and normative data for the visual learning task.

Preparation of this article was supported by a Johns Hopkins Science of Learning Institute grant to Michael McCloskey, by National Institutes of Health Grant DC006740 to Brenda Rapp, and by a generous gift from a donor to the Johns Hopkins University.

References

- Allport DA, & Funnell E (1981). Components of the mental lexicon. *Philosophical Transactions of the Royal Society B: Biological Sciences*, 295(1077), 397–410. 10.1098/rstb.1981.0148
- Angelelli P, Judica A, Spinelli D, Zoccolotti P, & Luzzatti C (2004). Characteristics of writing disorders in Italian dyslexic children. *Cognitive and Behavioral Neurology*, 17(1), 18–31. [PubMed: 15209222]
- Angelelli P, Marinelli CV, & Zoccolotti P (2010). Single or dual orthographic representations for reading and spelling? A study of Italian dyslexic-dysgraphic and normal children. *Cognitive Neuropsychology*, 27(4), 305–333. 10.1080/02643294.2010.543539 [PubMed: 21229408]
- Ans B, Carbonnel S, & Valdois S (1998). A connectionist multiple-trace memory model for polysyllabic word reading. *Psychological Review*, 105(4), 678–723. 10.1037/0033-295X.105.4.678-723 [PubMed: 9830376]
- Barisic K, Kohnen S, & Nickels L (2017). Developmental graphemic buffer dysgraphia in English: A single case study. *Cognitive Neuropsychology*.
- Beauvois M-F, & Dérouesné J (1979). Phonological alexia: Three dissociations. *Journal of Neurology, Neurosurgery & Psychiatry*, 42(12), 1115–1124. 10.1136/jnnp.42.12.1115
- Beauvois M-F, & Dérouesné J (1981). Lexical or orthographic agraphia. *Brain*, 104(1), 21–49. 10.1093/brain/104.1.21 [PubMed: 7470843]
- Beery KE, & Beery NA (2010). The Beery-Buktenica developmental test of visual-motor integration (Beery VMI) with supplemental developmental tests of visual perception and motor coordination and stepping stones age norms, from birth to age six: Administration, scoring, and teaching manual (6th ed.). Bloomington, MN: Pearson.
- Behrmann M, & Bub D (1992). Surface dyslexia and dysgraphia: Dual routes, single lexicon. *Cognitive Neuropsychology*, 9(3), 209–251. 10.1080/02643299208252059
- Bergmann J, & Wimmer H (2008). A dual-route perspective on poor reading in a regular orthography: Evidence from phonological and orthographic lexical decisions. *Cognitive Neuropsychology*, 25(5), 653–676. 10.1080/02643290802221404 [PubMed: 18642138]
- Bosse M-L, Tainturier MJ, & Valdois S (2007). Developmental dyslexia: The visual attention span deficit hypothesis. *Cognition*, 104(2), 198–230. 10.1016/j.cognition.2006.05.009 [PubMed: 16859667]
- Bosse M-L, & Valdois S (2009). Influence of the visual attention span on child reading performance: A cross-sectional study. *Journal of Research in Reading*, 32(2), 230–253. 10.1111/j.1467-9817.2008.01387.x

- Bradley L, & Bryant PE (1978). Difficulties in auditory organisation as a possible cause of reading backwardness. *Nature*, 271(5647), 746–747. 10.1038/271746a0 [PubMed: 625341]
- Bradley L, & Bryant PE (1983). Categorizing sounds and learning to read—a causal connection. *Nature*, 301(5899), 419–421. 10.1038/301419a0
- Bruck M (1993). Word recognition and component phonological processing skills of adults with childhood diagnosis of dyslexia. *Developmental Review*, 13(3), 258–268. 10.1006/drev.1993.1012
- Bruck M, & Treiman R (1990). Phonological awareness and spelling in normal children and dyslexics: The case of initial consonant clusters. *Journal of Experimental Child Psychology*, 50(1), 156–178. 10.1016/0022-0965(90)90037-9 [PubMed: 2398331]
- Bruininks RH, & Bruininks BD (2005). *Bruininks-Oseretsky test of motor proficiency* (2nd ed.). Circle Pines, MN: AGS Publishing.
- Brunsdon R, Coltheart M, & Nickels L (2005). Treatment of irregular word spelling in developmental surface dysgraphia. *Cognitive Neuropsychology*, 22(2), 213–251. 10.1080/02643290442000077 [PubMed: 21038247]
- Bub D, Cancelliere A, & Kertesz A (1985). Whole-word and analytic translation of spelling-to-sound in a non-semantic reader In Patterson K, Marshall JC, & Coltheart M (Eds.), *Surface dyslexia: neuropsychological and cognitive studies of phonological reading* (pp. 15–34). London, England: Lawrence Erlbaum Associates.
- Burt JS, & Tate H (2002). Does a reading lexicon provide orthographic representations for spelling? *Journal of Memory and Language*, 46(3), 518–543. 10.1006/jmla.2001.2818
- Campbell R, & Butterworth B (1985). Phonological dyslexia and dysgraphia in a highly literate subject: A developmental case with associated deficits of phonemic processing and awareness. *The Quarterly Journal of Experimental Psychology Section A*, 37(3), 435–475. 10.1080/14640748508400944
- Caramazza A, Capasso R, & Miceli G (1996). The role of the graphemic buffer in reading. *Cognitive Neuropsychology*, 13(5), 673–698. 10.1080/026432996381881
- Caramazza A, & Hillis AE (1990a). Levels of representation, co-ordinate frames, and unilateral neglect. *Cognitive Neuropsychology*, 7(5–6), 391–445. 10.1080/02643299008253450
- Caramazza A, & Hillis AE (1990b). Spatial representation of words in the brain implied by studies of a unilateral neglect patient. *Nature*, 346(6281), 267–269. 10.1038/346267a0 [PubMed: 2374591]
- Caramazza A, Miceli G, Villa G, & Romani C (1987). The role of the graphemic buffer in spelling: Evidence from a case of acquired dysgraphia. *Cognition*, 26(1), 59–85. 10.1016/0010-0277(87)90014-X [PubMed: 3608396]
- Castles A, & Coltheart M (1993). Varieties of developmental dyslexia. *Cognition*, 47(2), 149–180. 10.1016/0010-0277(93)90003-E [PubMed: 8324999]
- Castles A, & Coltheart M (2004). Is there a causal link from phonological awareness to success in learning to read? *Cognition*, 91(1), 77–111. 10.1016/S0010-0277(03)00164-1 [PubMed: 14711492]
- Castles A, Coltheart M, Larsen L, Jones P, Saunders S, & McArthur G (2009). Assessing the basic components of reading: A revision of the Castles and Coltheart test with new norms (CC2). Retrieved from <http://www.motif.org.au/>
- Cholewa J, Mantey S, Heber S, & Hollweg W (2010). Developmental surface and phonological dysgraphia in German 3rd graders. *Reading & Writing*, 23(1), 97–127. 10.1007/s11145-008-9153-7
- Cohen MJ (1997). *Children's memory scale*. San Antonio, TX: The Psychological Corporation.
- Coltheart M (2015). What kinds of things cause children's reading difficulties? *Australian Journal of Learning Difficulties*, 20(2), 103–112. 10.1080/19404158.2015.1114000
- Coltheart M, & Funnell E (1987). Reading and writing: One lexicon or two? In Allport A, MacKay DG, & Prinz W (Eds.), *Language perception and production: Relationships between listening, speaking, reading and writing* (pp. 313–339). San Diego, CA: Academic Press.
- Coltheart M, Masterson J, Byng S, Prior M, & Riddoch J (1983). Surface dyslexia. *The Quarterly Journal of Experimental Psychology Section A*, 35(3), 469–495. 10.1080/14640748308402483
- Dell GS (1986). A spreading-activation theory of retrieval in sentence production. *Psychological Review*, 93(3), 283–321. 10.1037/0033-295X.93.3.283 [PubMed: 3749399]

- Dolch EW (1948). Problems in reading. Champaign, IL: Garrard Press.
- Douklias SD, Masterson J, & Hanley JR (2009). Surface and phonological developmental dyslexia in Greek. *Cognitive Neuropsychology*, 26(8), 705–723. 10.1080/02643291003691106 [PubMed: 20407965]
- Dunn LM, & Dunn DM (2007). Peabody picture vocabulary test (4th ed.). Minneapolis, MN: Pearson Assessments.
- Dunn LM, & Dunn LM (1981). Peabody picture vocabulary test, revised (Rev. ed.). Circle Pines, MN: American Guidance Service.
- Ehri LC (2000). Learning to read and learning to spell: Two sides of a coin. *Topics in Language Disorders*, 20(3), 19–36.
- Frith U (1978). From print to meaning and from print to sound, or how to read without knowing how to spell. *Visible Language*, 12(1), 43–54.
- Frith U (1980). Unexpected spelling problems In Frith U (Ed.), *Cognitive processes in spelling* (pp. 495–515). London, England: Academic Press.
- Funnell E (1983). Phonological processes in reading: New evidence from acquired dyslexia. *British Journal of Psychology*, 74(2), 159–180. 10.1111/j.2044-8295.1983.tb01851.x [PubMed: 6883008]
- Funnell E, & Davison M (1989). Lexical capture: A developmental disorder of reading and spelling. *The Quarterly Journal of Experimental Psychology Section A*, 41(3), 471–487. 10.1080/14640748908402377
- Gascoigne MB, Smith ML, Barton B, Webster R, Gill D, & Lah S (2014). Accelerated long-term forgetting in children with temporal lobe epilepsy. *Neuropsychologia*, 59, 93–102. 10.1016/j.neuropsychologia.2014.04.012 [PubMed: 24784007]
- Goulandris NK, & Snowling M (1991). Visual memory deficits: a plausible cause of developmental dyslexia? Evidence from a single case study. *Cognitive Neuropsychology*, 8(2), 127–154. 10.1080/02643299108253369
- Greenberg D, Ehri LC, & Perin D (1997). Are word-reading processes the same or different in adult literacy students and third–fifth graders matched for reading level? *Journal of Educational Psychology*, 89(2), 262–275. 10.1037/0022-0663.89.2.262
- Gvion A, & Friedmann N (2010). Letter position dysgraphia. *Cortex*, 46(9), 1100–1113. 10.1016/j.cortex.2009.09.008 [PubMed: 19846074]
- Hanley JR, & Gard F (1995). A dissociation between developmental surface and phonological dyslexia in two undergraduate students. *Neuropsychologia*, 33(7), 909–914. 10.1016/0028-3932(95)00038-5 [PubMed: 7477816]
- Hanley JR, Hastie K, & Kay J (1992). Developmental surface dyslexia and dysgraphia: An orthographic processing impairment. *The Quarterly Journal of Experimental Psychology Section A*, 44(2), 285–319. 10.1080/02724989243000046
- Hanley JR, & Kay J (1992). Does letter-by-letter reading involve the spelling system? *Neuropsychologia*, 30(3), 237–256. 10.1016/0028-3932(92)90003-5 [PubMed: 1574160]
- Hanley JR, & McDonnell V (1997). Are reading and spelling phonologically mediated? Evidence from a patient with a speech production impairment. *Cognitive Neuropsychology*, 14(1), 3–33. 10.1080/026432997381600
- Hanna PR, Hanna JS, Hodges RE, & Rudolf EH Jr. (1966). Phoneme-grapheme correspondences as cues to spelling improvement. Retrieved from <http://eric.ed.gov/?id=ED128835>
- Hepworth S, & Smith ML (2002). Learning and recall of story content and spatial location after unilateral temporal-lobe excision in children and adolescents. *Child Neuropsychology*, 8(1), 16–26. 10.1076/chin.8.1.16.8718 [PubMed: 12610773]
- Hillis AE, Rapp B, & Caramazza A (1999). When a rose is a rose in speech but a tulip in writing. *Cortex*, 35(3), 337–356. 10.1016/S0010-9452(08)70804-9 [PubMed: 10440073]
- Holmes VM, & Carruthers J (1998). The relation between reading and spelling in skilled adult readers. *Journal of Memory and Language*, 39(2), 264–289. 10.1006/jmla.1998.2583
- Holmes VM, & Davis CW (2002). Orthographic representation and spelling knowledge. *Language and Cognitive Processes*, 17(4), 345–370. 10.1080/01690960143000263a

- Holmes VM, Malone AM, & Redenbach H (2008). Orthographic processing and visual sequential memory in unexpectedly poor spellers. *Journal of Research in Reading*, 31(1), 136–156. 10.1111/j.1467-9817.2007.00364.x
- Kohnen S, Anandakumar T, McArthur G, & Castles A (2012). The test of orthographic choice (TOC). Retrieved from <http://www.motif.org.au/>
- Kohnen S, Colenbrander D, Krajenbrink T, & Nickels L (2013). Diagnostic spelling test - nonwords with normative data. Retrieved from <http://www.motif.org.au/>
- Kohnen S, Colenbrander D, Krajenbrink T, & Nickels L (2015). Assessment of lexical and non-lexical spelling in students in grades 1–7. *Australian Journal of Learning Difficulties*, 20(1), 15–38. 10.1080/19404158.2015.1023209
- Kohnen S, Marinus E, Friedmann N, Anandakumar T, Nickels L, McArthur G, & Castles A (2012). Letter position test. Retrieved from <http://www.motif.org.au/>
- Levelt WJM, Roelofs A, & Meyer AS (1999). A theory of lexical access in speech production. *Behavioral and Brain Sciences*, 22(1), 1–38. [PubMed: 11301520]
- Lieberman IY (1973). Segmentation of the spoken word and reading acquisition. *Bulletin of the Orton Society*, 23(1), 64–77. 10.1007/BF02653842
- Margolin DI (1984). The neuropsychology of writing and spelling: Semantic, phonological, motor, and perceptual processes. *The Quarterly Journal of Experimental Psychology Section A*, 36(3), 459–489. 10.1080/14640748408402172
- McCann RS, & Besner D (1987). Reading pseudohomophones: Implications for models of pronunciation assembly and the locus of word-frequency effects in naming. *Journal of Experimental Psychology: Human Perception and Performance*, 13(1), 14–24. 10.1037/0096-1523.13.1.14
- McCloskey M, Fischer-Baum S, & Schubert T (2013). Representation of letter position in single-word reading: Evidence from acquired dyslexia. *Cognitive Neuropsychology*, 30(6), 396–428. 10.1080/02643294.2014.880675 [PubMed: 24512595]
- McCloskey M, & Rapp B (2017). Developmental dysgraphia: An overview and framework for research. *Cognitive Neuropsychology*.
- Milone M (2007). *Test of handwriting skills, revised* (Rev ed.). Novato, CA: Academic Therapy Publications.
- Monsell S (1987). On the relation between lexical input and output pathways for speech In Allport A, MacKay DG, & Prinz W (Eds.), *Language perception and production: Relationships between listening, speaking, reading and writing* (pp. 273–311). San Diego, CA: Academic Press.
- Patterson K, & Shewell C (1987). Speak and spell: Dissociations and word-class effects In Coltheart M, Sartori G, & Job R (Eds.), *The cognitive neuropsychology of language* (pp. 273–294). London, England: Lawrence Erlbaum Associates.
- Philipose LE, Gottesman RF, Newhart M, Kleinman JT, Herskovits EH, Pawlak MA, ... Hillis AE (2007). Neural regions essential for reading and spelling of words and pseudowords. *Annals of Neurology*, 62(5), 481–492. 10.1002/ana.21182 [PubMed: 17702036]
- Psychological Corporation. (2009). *Wechsler individual achievement test* (3rd ed.). San Antonio, TX: Psychological Corporation.
- Purcell JJ, Jiang X, & Eden GF (2017). Shared orthographic neuronal representations for spelling and reading. *NeuroImage*, 147, 554–567. 10.1016/j.neuroimage.2016.12.054 [PubMed: 28011250]
- Purcell JJ, Napoliello EM, & Eden GF (2011). A combined fMRI study of typed spelling and reading. *NeuroImage*, 55(2), 750–762. 10.1016/j.neuroimage.2010.11.042 [PubMed: 21109009]
- Rapcsak SZ, & Beeson PM (2004). The role of left posterior inferior temporal cortex in spelling. *Neurology*, 62(12), 2221–2229. 10.1212/01.WNL.0000130169.60752.C5 [PubMed: 15210886]
- Rapp B, & Caramazza A (1989). Letter processing in reading and spelling: Some dissociations. *Reading and Writing*, 1(1), 3–23. 10.1007/BF00178834
- Rapp B, & Caramazza A (1997). From graphemes to abstract letter shapes: Levels of representation in written spelling. *Journal of Experimental Psychology: Human Perception and Performance*, 23(4), 1130–1152. 10.1037/0096-1523.23.4.1130 [PubMed: 9269731]

- Rapp B, & Dufor O (2011). The neurotopography of written word production: An fMRI investigation of the distribution of sensitivity to length and frequency. *Journal of Cognitive Neuroscience*, 23(12), 4067–4081. 10.1162/jocn_a_00109 [PubMed: 21812571]
- Rapp B, Epstein C, & Tainturier M-J (2002). The integration of information across lexical and sublexical processes in spelling. *Cognitive Neuropsychology*, 19(1), 1–29. 10.1080/0264329014300060 [PubMed: 20957529]
- Rapp B, & Fischer-Baum S (2015). Uncovering the cognitive architecture of spelling In Hillis AE (Ed.), *The handbook of adult language disorders* (2nd Ed., pp. 47–69). New York, NY: Psychology Press.
- Rapp B, & Kong DL (2002). Revealing the component functions of the graphemic buffer. *Brain and Language*, 83(1), 112–114.
- Rapp B, & Lipka K (2011). The literate brain: The relationship between spelling and reading. *Journal of Cognitive Neuroscience*, 23(5), 1180–1197. 10.1162/jocn.2010.21507 [PubMed: 20433242]
- Romani C, Ward J, & Olson A (1999). Developmental surface dysgraphia: What is the underlying cognitive impairment? *Quarterly Journal of Experimental Psychology: Section A*, 52(1), 97–128. 10.1080/027249899391241
- Roncoli S, & Masterson J (2016). “Unexpected” spelling difficulty in a 10-year-old child with good reading skills: An intervention case study. *Writing Systems Research*, 8(2), 143–166. 10.1080/17586801.2016.1159539
- Sartori G, Masterson J, & Job R (1987). Direct route reading and the locus of lexical decision In Coltheart M, Sartori G, & Job R (Eds.), *The cognitive neuropsychology of language* (pp. 59–78). London, England: Lawrence Erlbaum Associates.
- Schubert T, & McCloskey M (2013). Prelexical representations and processes in reading: Evidence from acquired dyslexia. *Cognitive Neuropsychology*, 30(6), 360–395. 10.1080/02643294.2014.880677 [PubMed: 24512594]
- Schwartz MF, Saffran EM, & Marin OSM (1980). Fractionating the reading process in dementia: Evidence for word-specific print-to-sound associations In Coltheart M, Marshall JC, & Patterson K (Eds.), *Deep dyslexia* (pp. 259–269). London, England: Routledge & Kegan Paul.
- Seymour PHK (1986). *Cognitive analysis of dyslexia*. London, England: Routledge & Kegan Paul.
- Seymour PHK, & Evans HM (1993). The visual (orthographic) processor and developmental dyslexia In Willows DM, Kruk RS, & Corcos E (Eds.), *Visual processes in reading and reading disabilities* (pp. 347–376). Hillsdale, NJ: Lawrence Erlbaum Associates.
- Snowling M, Goulandris N, Bowlby M, & Howell P (1986). Segmentation and speech perception in relation to reading skill: A developmental analysis. *Journal of Experimental Child Psychology*, 41(3), 489–507. 10.1016/0022-0965(86)90006-8 [PubMed: 3734692]
- Sotiropoulos A, & Hanley JR (2017). Developmental surface and phonological dyslexia in both Greek and English. *Cognition*, 168, 205–216. 10.1016/j.cognition.2017.06.024 [PubMed: 28710937]
- Stanovich KE (1988). The right and wrong places to look for the cognitive locus of reading disability. *Annals of Dyslexia*, 38(1), 154–177. 10.1007/BF02648254 [PubMed: 24235039]
- Tainturier M-J, & Rapp B (2001). The spelling process In Rapp B (Ed.), *The handbook of cognitive neuropsychology: What deficits reveal about the human mind* (pp. 263–289). Philadelphia, PA: Psychology Press.
- Tainturier M-J, & Rapp B (2003). Is a single graphemic buffer used in reading and spelling? *Aphasiology*, 17(6/7), 537.
- Temple CM (1984). Surface dyslexia in a child with epilepsy. *Neuropsychologia*, 22(5), 569–576. 10.1016/0028-3932(84)90021-6 [PubMed: 6504297]
- Temple CM (1985). Developmental surface dysgraphia: A case report. *Applied Psycholinguistics*, 6(4), 391–405. 10.1017/S0142716400006329
- Temple CM (1986). Developmental dysgraphias. *The Quarterly Journal of Experimental Psychology Section A*, 38(1), 77–110. 10.1080/14640748608401586
- Temple CM, & Marshall JC (1983). A case study of developmental phonological dyslexia. *British Journal of Psychology*, 74(4), 517–533. 10.1111/j.2044-8295.1983.tb01883.x [PubMed: 6640236]
- Valdois S (1996). A case study of developmental surface dyslexia and dysgraphia. *Brain and Cognition*, 32(2), 229–231. 10.1006/brcg.1996.0065

- Valdois S, Bosse M-L, Ans B, Carbonnel S, Zorman M, David D, & Pellat J (2003). Phonological and visual processing deficits can dissociate in developmental dyslexia: Evidence from two case studies. *Reading & Writing*, 16(6), 541–572.
- Wagner RK, & Torgesen JK (1987). The nature of phonological processing and its causal role in the acquisition of reading skills. *Psychological Bulletin*, 101(2), 192–212. 10.1037/0033-2909.101.2.192
- Wagner RK, Torgesen JK, & Rashotte CA (1999). *Comprehensive test of phonological processing* (1st ed.). Austin, TX: Pro-Ed.
- Wagner RK, Torgesen JK, Rashotte CA, & Pearson NA (2013). *Comprehensive test of phonological processing* (2nd ed.). Austin, TX: Pro-Ed.
- Wechsler D (2003). *Wechsler intelligence scale for children* (4th ed.). San Antonio, TX: Psychological Corporation.
- Wechsler D (2014). *Wechsler intelligence scale for children* (5th ed.). San Antonio, TX: Psychological Corporation.
- Wilkinson GS (1993). *Wide range achievement test*. Wilmington, DE: Wide Range, Inc.
- Williams KT (2007). *Expressive vocabulary test* (2nd ed.). Minneapolis, MN: Pearson Assessments.
- Wing AM, & Baddeley AD (1980). Spelling errors in handwriting: A corpus and a distributional analysis. In Frith U (Ed.), *Cognitive processes in spelling* (pp. 251–285). Retrieved from https://catalyst.library.jhu.edu/catalog/bib_383930
- Woodcock RW (1987). *Woodcock reading mastery tests, revised* (Rev. ed.). Circle Pines, MN: American Guidance Service.
- Yachini M, & Friedmann N (2010). Developmental graphemic buffer dysgraphia. *Procedia - Social and Behavioral Sciences*, 6, 148–149. 10.1016/j.sbspro.2010.08.073

Figure 1.
 Depictions of the cognitive architectures for (A) reading and (B) spelling in adults.

8. Name one dangerous liquid and one dangerous solid.

BleYcs Neyoclr wasd

b) Explain why it is important to keep dangerous solids and liquids in their original containers.

because it coode look layks
somsing als and smw
on miyt drinkt / eta

A. Writing Sample

Bleycs

Neyoclr wasd

because it coode look layks
 somsing als and smw
 on miyt drinkt / eta

B. Transcription

Bleach

Nuclear waste

because it could look like
 something else and some
 one might drink / eat

C. Intended Text

Figure 2.

A. PJT writing sample. B. Literal transcription. C. Text PJT intended to write.

A. Writing Sample

The local restrant with an unknow name got a very bad reivew today. Well trying to acomadate a custerms allergys the custormer was give a very bland plate. It was not clorful and delitful. In the custorms worts "(It) was more like a lage snak plate than a dinner." The cholclate coverd bannas at the end hardly made up for it. Plus the the custormor was tainted by the smelles of a s?in-ff of one of her favrote dishes wich she can not eat due to allergys.

B. Transcription

The local restaurant with an unknown name got a very bad review today. While trying to accommodate a customer's allergies the customer was given a very bland plate. It was not colorful and delightful. In the customer's words, "(It) was more like a large snack plate than a dinner." The chocolate covered bananas at the end hardly made up for it. Plus the customer was taunted by the smells of a sniff of one of her favorite dishes which she cannot eat due to allergies.

C. Intended Text

Figure 3.

A. AKR writing sample. B. Literal transcription (ignoring crossed-out letters and occasional use of upper-case in place of lower-case letter forms). C. Text AKR intended to write.

Figure 4.
Accuracy by normalized position in PJT's spelling data.

Figure 5.

A model of a cognitive architecture for written language processing with modality-independent lexical orthographic representations. Sublexical processes are omitted. Solid lines are used for processes and representations involved in spelling and dashed lines for those involved in reading. Bold indicates processes and representations involved specifically in lexical orthographic processing in reading and spelling.

Figure 6.

A model of a cognitive architecture for written language processing from a nodes-and-links perspective. Labels on nodes are only for clarification, and do not indicate content "inside" a node.

Table 1.

Performance on the WISC subtests for PJT and AKR. Standard scores are reported with percentile ranks in parentheses.

		PJT	AKR
Verbal Comprehension	Similarities	19 (99)	16 (98)
	Vocabulary	19 (99)	18 (99)
	Comprehension	15 (95)	-
Perceptual Reasoning	Block Design	16 (98)	10 (50)
	Picture Concepts	19 (99)	-
	Matrix Reasoning	19 (99)	8 (25)
Working Memory	Digit Span	13 (84)	15 (95)
	Letter-Number Sequencing	8 (25)	-
Processing Speed	Coding	9 (37)	14 (91)
	Symbol Search	11 (63)	18 (99)
Other	Spatial Span Forward	14 (91)	-
	Spatial Span Backward	11 (63)	-
	Visual Puzzles	-	10 (50)
	Figure Weights	-	11 (63)

Table 2.

Results from WIAT-III (Wechsler Individual Achievement Test – 3rd Edition). The table reports standard scores with percentiles in parentheses.

	PJT	AKR
Listening Comprehension	-	132 (98)
<i>Receptive Vocabulary</i>	-	122 (93)
<i>Oral Discourse Comprehension</i>	145 (99)	130 (98)
Reading Comprehension	126 (96)	111 (77)
Math Problem Solving	125 (95)	127 (96)
Sentence Composition	122 (93)	113 (81)
<i>Sentence Building</i>	-	124 (95)
<i>Sentence Combining</i>	-	100 (50)
Word Reading	133 (99)	101 (53)
Pseudoword Decoding	100 (50)	129 (97)
Spelling	89 (23)	89 (23)
Essay Composition	-	159 (99)
<i>Word Count</i>	-	147 (99)
<i>Theme Development and Text Organization</i>	-	154 (99)
<i>Grammar and Mechanics</i>	-	156 (99)
Numerical Operations	111 (77)	122 (93)
Oral Expression	-	155 (99)
<i>Expressive Vocabulary</i>	-	135 (99)
<i>Oral Word Fluency</i>	-	139 (99)
<i>Sentence Repetition</i>	-	157 (99)
Oral Reading Fluency	-	116 (86)
<i>Oral Reading Accuracy</i>	-	117 (87)
<i>Oral Reading Rate</i>	-	112 (79)
Math Fluency – Addition	-	101 (53)
Math Fluency – Subtraction	-	105 (63)
Math Fluency – Multiplication	-	94 (34)

Table 3.

Results from the Letter Position Test (Kohnen, Marinus, et al., 2012).

	PJT			AKR			AKR Retest		
	Score	Percentile	z-Score	Score	Percentile	z-Score	Score	Percentile	z-Score
Total Errors	5	95	1.64	11	27	-0.60	7	30	-0.54
Migration Errors	2	92	1.43	7	21	-0.80	6	21	-0.82
Word Errors	1	86	1.08	3	27	-0.60	1	41	-0.23
Other Errors	2	66	0.41	1	27	-0.60	0	44	-0.16

Table 4.

Performance of PJT and AKR on the Comprehensive Test of Phonological Processing. Standard scores and percentiles are reported.

Composite Measure	PJT		AKR	
	Score	Percentile	Score	Percentile
Phonological Awareness	105	63	112	79
Phonological Memory	113	81	112	79
Rapid Naming (Symbolic)	104	61	106	65
Alternative Phonological Awareness	-	-	118	89
Alternative Rapid Naming (Non-Symbolic)	98	45	97	42

Table 5.

PJT's performance on the CMS (Children's Memory Scale; Cohen, 1997).

	Index Score	Percentile Rank
Visual Immediate	112	79
Visual Delayed	115	84
Verbal Immediate	137	99
Verbal Delayed	137	99
General Memory Index	136	99
Learning Index	125	95
Delayed Recognition	115	84

Table 6.

Performance of PJT and AKR on the Gascoigne et al. (2014) visual learning task. The table shows means for controls with standard deviations in parentheses.

	PJT	Controls (N = 12)	AKR	Controls (N = 8)
Age	9.8	10.1 (0.8)	11.4	10.5 (0.4)
Learning Trials	-	8.3 (2.6)	8	8.7 (2.8)
Short-Delay Recall	-	95% (8%)	100%	96% (8%)
Long-Delay Recall	60%	50% (36%)	50%	35% (31%)
Long-Delay Recognition (hits/false alarms)	3/1	2.9 (1.6)/2.0 (1.8)	4/1	2.6 (1.7)/2.4 (1.7)