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Publisher
Centre de recherche sur les médiations
(CREM)

Electronic version

URL: <http://pratiques.revues.org/3175>
DOI: 10.4000/pratiques.3175
ISSN: 2425-2042

Electronic reference

Marieke Longcamp, Jean-luc Velay, Virginia Wise Berninger and Todd Richards, « Neuroanatomy of Handwriting and Related Reading and Writing Skills in Adults and Children with and without Learning Disabilities: French-American Connections », *Pratiques* [Online], 171-172 | 2016, Online since 07 February 2017, connection on 24 March 2017. URL : <http://pratiques.revues.org/3175> ; DOI : 10.4000/pratiques.3175

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Neuroanatomy of Handwriting and Related Reading and Writing Skills in Adults and Children with and without Learning Disabilities: French-American Connections

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Preparation of this article was supported in part by center grant P50HD071764 from the Eunice Kennedy Shriver National Institute of Child Health and Human Development (NICHD) at the National Institutes of Health (NIH) to the University of Washington Learning Disabilities Research Center.

- 1 Aged only 2, the young child spontaneously produces graphic shapes such as strokes, spirals or ellipses. She will soon learn how to combine those shapes into exquisitely precise gestures aimed at transcribing her ideas while coping with legibility constraints. When one realizes how early the learning starts, how long it develops, and how complex the simultaneous acquisition of the motor and linguistic codes is, one can intuitively understand that learning how to write has consequences not only on the organization of the brain, but also on the functioning of other skills such as reading. See Berninger and Chanquoy (2012) for a review of research on writing development from the first year of life through adolescence.
- 2 Brain imaging is contributing to advancing knowledge of both the brain's role in writing and the relationships of writing to reading. In this article we review what has been learned from brain imaging of individuals with acquired writing disorders, for example, due to disease, stroke, or injury and individuals who are still developing and learning to write. Findings are discussed from both neuroscience and behavioral/cognitive research because both contribute to understanding a complex process such as writing that is a

product of both nature and nurture interactions. Because writing is a complex process, in this article we focus on current understanding of handwriting and its relationship to other skills. We also feature research studies and dissemination efforts that have involved collaborations between French and American researchers.

I. Expert writing depends on specific brain regions

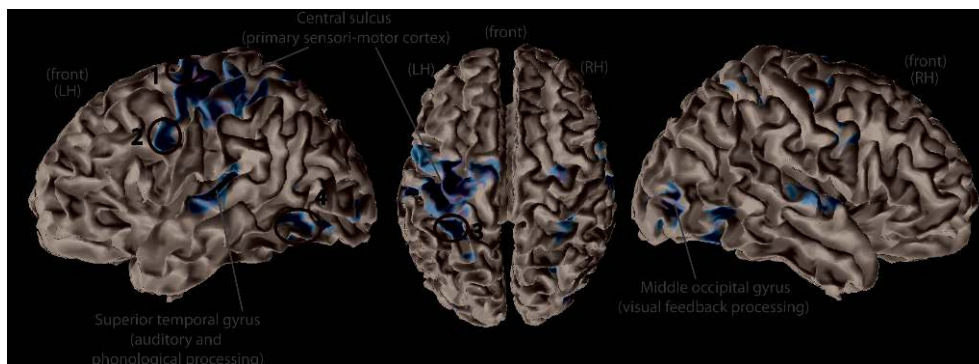
Adults

- 3 The overall shape of a word traced on a piece of paper, on a blackboard, or with a toe on the sand is always preserved despite massive changes in the effectors used to produce the movement. This “motor equivalence”, described by Bernstein in 1967, implies the existence of a memory trace of the sequence of gestures necessary to produce each character (the so-called generalized motor programs). When this memory gets disorganized following a brain lesion, the shaping of the letters becomes difficult for the patients. This condition is termed apraxic agraphia, and typically results from lesions in specific brain regions. Historically, the hypothesis of a brain center containing the “graphic motor images” of letters has been formulated first by Sigmund Exner (1881) based on the observation of some agraphic patients’ brains. Lesions of those patients overlapped in a region close to the junction between the middle frontal and precentral gyri of the left hemisphere that belongs to the premotor cortex (a part of the frontal cortex anterior to the primary motor cortex; see figure 1) and is often referred to as “Exner’s area”.
- 4 The role of a specific left frontal region in writing has later been confirmed by other neuropsychological studies and by several brain imaging studies (see Planton, Jucla, Roux and Démonet, 2013 for a meta-analysis). However, brain imaging results indicate that the exact location of Exner’s area along the precentral gyrus is not that clear, with some studies reporting very dorsal premotor activations at the level of the superior frontal gyrus, and other studies reporting more ventral activations within the dorsal extent Broca’s area. Further investigations are required to refine our understanding of the exact position, and the functional properties and functional specificity of premotor regions in writing.
- 5 In an attempt to do so, Longcamp *et al.* (2014) decided to test whether the response of premotor regions differed when adult participants were writing letters, that is, basic units of written language, compared to digits. Qualitative or quantitative differences in the movements performed to write letters, vs digits or symbols are already detectable in very young children aged around 2 (Yamagata, 2007), undergo a strong dissociation around age six (Adi-Japha & Freeman, 2001), and are still measurable in adults (Delazer, Lochy, Jenner, Domahs & Benke, 2002). These findings indicate that the two representational systems, and their graphomotor functions, develop independently. Longcamp *et al.* (2014) found that indeed, the dorsal part of the premotor cortex, together with two other brain regions involved in phonological processing, was more strongly activated when producing letters than digits. This raises the intriguing possibility that the motor patterns for writing linguistic items such as letters are implemented in a specific brain region and very finely grained functional specificity present in the motor system. Motor representations of letters may be more stable and more strongly anchored in the precentral regions. More stable central representations are likely to impact

execution parameters such as velocity or duration, as shown in the course of handwriting acquisition and practice (Zesiger, Mounoud & Hauert, 1993).

- 6 Of course the so-called Exner's area is not the only part of the brain that gets activated when writing. As shown in figure 1, when the brain signal during writing is compared to a situation where the participants simply hold the pen on the tablet, an extensive network encompassing the primary sensorimotor cortex and various sensory and associative regions supporting visual and auditory processing activates. This activation is due to the fact that writing is not only a finely controlled hand gesture. It also demands neural resources for analyzing the stimulus (for instance auditory processing if the word is dictated) and most importantly for integrating on-line the visual feedback if what is written can be viewed (or "when the word is copied").
- 7 In addition, when one tries to isolate the most consistent activations across studies involving various writing tasks (Planton *et al.*, 2013; Purcell, Turkeltaub, Eden & Rapp, 2011), one observes only a few nodes that seem really crucial independent of the constraints of the task at hand and the control conditions: left superior parietal cortex, the cerebellum, and the left fusiform gyrus (figure 1). The respective functions of these regions are not yet fully understood, but the fusiform gyrus was consistently identified by Purcell *et al.* (2011) as contributing to spelling, and more specifically to the central processes in spelling. More generally, a subpart of the left fusiform gyrus is related to orthographic word-form processing (e.g., Cohen & Dehaene, 2004; Cohen *et al.*, , 2002) and another, more anterior portion to letter-form processing (Joseph, Gathers & Piper 2003).

Figure 1. Brain activations during handwriting, in a group of young healthy right-handed volunteers



- 8 The left panel represents a lateral view of the left hemisphere (LH), the middle panel a top view of the brain, and the right panel a right view of the right hemisphere (RH). Writing to dictation has been contrasted to a situation where the participants were holding the pen on the writing surface without moving. Writing engages a large number of sensory and motor cortical regions, because the dictated stimulus has to be processed by auditory regions and converted into gestures while the visual feedback is processed online by the occipital regions. One can notice the strongly left-lateralized activations, related to the control of the right hand by left hemispheric sensorimotor regions. The 4 brain nodes which activation is consistent among brain imaging studies of handwriting are circled in blue. (1- dorsal premotor cortex, 2- ventral premotor cortex 3- superior parietal cortex, 4- fusiform gyrus). Modified from Longcamp *et al.* (2014). The cerebellum, which is the hind brain, another important center for writing, is not shown in this representation.

Children

- 9 In fact, the writing network evident in many neuroimaging studies of adults also seem to play a pivotal role in children's writing. In an fMRI study involving children between 5th and 6th grade, Richards *et al.* (2011) compared the brain activation resulting from the writing of an unfamiliar letter shape (round shape right above top of an horizontal line), that is a pseudoletter, to a highly practiced letter of comparable shape in ball and stick manuscript format (round shape with vertical line on right). Good writers were more efficient in writing highly practiced letters than poor writers. Interestingly, the premotor and parietal cortices, the cerebellum and the fusiform gyrus were engaged more strongly when the children were writing a new letter (pseudoletter), as if their activation needs to be higher when the memory of the shape is not stable yet in memory. However, this was true only in the case of good writers, because the activation in of those regions did not differ between newly taught and highly practiced letters in poor writers. Another important, and related, result was that efficiency in writing was linked to the involvement of a more restricted and focused brain network. Good writers engaged fewer neural regions to write a newly taught letter than did the poor writers. The component strokes in the novel configuration of the pseudoletter are highly practiced but not in the taught configuration; so the efficiency of production must be related to the overall context or configuration in which those motor strokes are produced. Conversely, poor writers overactivated the visual system, and extra parietal and cerebellar regions. Finally, good and poor writers differed significantly in activation in left fusiform cortex when writing highly practiced letters. This individual fusiform activation correlated significantly with behavioral measures of automatic letter writing and expressive orthographic coding. Multiple regression in which both individual fusiform activation and individual orthographic coding were entered explained significant variance in written composition. Thus, contrary to a popular belief handwriting is not just a motor or movement skill—it also involves written language at the letter form level.
- 10 In sum, the development of a more focused activation of the premotor, parietal and cerebellar regions while learning new letter shapes is associated with good writing skills whereas more widespread activation is associated with poor writing skills. As pointed out by this study, there is also a critical involvement of and interactions between the visual and motor systems in the development of the writing skill, since the efficiency depends both on the focalization of the activation on motor-related brain regions when practicing new shapes and on the fusiform gyrus when writing highly practiced shapes. In addition, this pattern of brain activity significantly predicts other language skills. We will discuss those two statements in turn in the following sections. In addition, not only the level of activation of certain regions differs between good and poor writers, but also the overall pattern of connectivity of the brain (Richards *et al.*, 2011, see below section relationship with other skills).

II. Relationships between writing and other language skills

Letter Recognition and Reading

- 11 In the great majority of clinical observations, alexia, that is the acquired inability to read following a brain lesion, occurs following a lesion of the left occipito-temporal areas (fusiform gyrus). However, deficits in the visual identification of letters can sometimes be associated with inability to write letters (Anderson *et al.*, 1990), an observation hardly compatible with a pure visual deficit. Conversely, visual and sensorimotor cerebral representations of letter shapes could be somehow coupled. In the case described by Anderson *et al.* (1990), the patient in question became alexic and agraphic as the result of a left premotor cortical lesion, corresponding to Exner's area. But is Exner's area also activated when subjects without a brain lesion simply observe visually presented letters? A first answer was given by Japanese researchers, who showed that several associative and motor areas are both activated when ideographic and syllabic Japanese characters are visually presented and written, and when the subjects are instructed to retrieve kanji ideograms (Matsuo *et al.*, 2001). However, ideographic characters are fairly complex stimuli since they represent words and are associated to high level linguistic representations. Roman letters are by far less complex and thus it was important to check whether passively viewing Roman letters may activate some of the sensorimotor cerebral areas also involved in writing movements.
- 12 In a neuroimaging study, (Longcamp *et al.*, 2003) directly assessed this possibility. Using fMRI on a group of right-handed subjects, they observed that a part of the left premotor cortex was activated when letters were being passively observed, and that the same zone was also strongly activated when the subjects were actually writing the letters. Interestingly, this area did not respond to the visual presentation of pseudoletters, to which no predetermined motor program could be associated. Furthermore, in a subsequent study, Longcamp Anton, Roth & Velay (2005a) showed that a symmetrical area of the right premotor cortex was activated when left-handed subjects were passively observing letters, confirming that this visually induced activation is contralateral to the writing hand. They therefore suggested that this premotor activation reflected the involvement of the motor programs for handwriting, corresponding to each letter, in agreement with the conclusions drawn by Anderson *et al.* (1990). These various data indicate that the cerebral representation of letters might not be strictly visual (see also James & Gauthier, 2006). In the framework of an embodied account of cognition (Wilson, 2002), it can be argued that coincident learning of writing movements and visual shapes of letters in early literacy leads to a multimodal representation of letters distributed over the cortical areas that were active when the letters were initially stored in memory.
- 13 This hypothesis can be formally tested in the context of training studies where participants learn how to read and write characters. In fact, several behavioral and brain imaging studies have provided clear evidence that producing characters by hand as opposed to simply viewing or typing them has an impact on the later recognition of these characters (Longcamp, Zerbato-Poudou & Velay, 2005b; Longcamp *et al.*, 2006; Longcamp *et al.*, 2008; James & Atwood, 2009). For instance, Longcamp *et al.* (2006) trained adults how to write characters from an unknown alphabet (Tamil and Bengali fonts) either by

traditional pen-and-paper writing or with a computer keyboard. Following training, they found that characters trained by hand were recognized more accurately and led to stronger activation in several brain regions known to be involved in motor preparation and execution than characters trained by keyboard.

- 14 In children, recent work (James, 2010) has revealed that writing training, but not visual-only training, induces increased activation in bilateral anterior fusiform gyri, a portion of the fusiform gyrus sensitive to visual configuration of single letters, in a letter recognition task in 4 to 5 year old pre-literate children when from pre-training and post-training scans were compared. The stability of the visual representations of letters is therefore strengthened when letters are trained by writing them repeatedly. This was confirmed in a subsequent study, in which a greater effect of freely producing letters by hand compared to tracing or typing them on the fusiform activation during letter perception was observed in the same age-group (James & Englehardt, 2012).
- 15 The positive effect of writing on the cortical network sustaining subsequent visual recognition of the characters can be extended to actual reading. Indeed, at a behavioral level, instructional studies, teaching handwriting has shown transfer to improved word reading even when word reading is not directly taught (e.g., Berninger, Dunn, Lin, & Shimada, 2004; Berninger *et al.*, 1997, 2006; Dunn & Miller, 2009). In a correlational study of Chinese primary school children, Tan, Spinks, Eden, Perfetti, & Siok (2005) found that in both beginning and intermediate readers, the ability to rapidly copy known characters, that is, writing skill, was the best predictor of reading scores. Finally, we previously emphasized the important role of the left fusiform region in the neural network sustaining writing in relation to orthographic word form processing. Several studies indeed show that the fusiform activation is common between reading and spelling and between reading and typing characters on a keyboard, pointing towards shared neural substrates of reading and spelling skills (Purcell, Napoliello and Eden, 2011; Rapp and Lipka, 2011).

Writing Skills : Spelling and Composing

- 16 Letter recognition at the subword level contributes to writing skills at the word level (spelling) and text level (composing) as well as reading (see Berninger, Fayol & Alamargot, 2012). Berninger and her colleagues found positive effects of teaching legible and automatic letter writing close in time with writing activities at other levels of language (word spelling and/or composing): Following such instruction students wrote texts that were longer (Berninger *et al.* 1995) and were completed in less time (Berninger *et al.*, 1997; Berninger *et al.*, 2006; see also Graham, Harris, and Fink, 2000 for similar findings). Writing involves cognitive processes such as idea flow (Kellogg, 1994) and strategic planning for composing (Hayes, 2006) and not just language (Richards *et al.*, 2015). At the brain level, Berninger *et al.* (2009) showed that good and poor writers differed not only in the basic sensorimotor activations specific to handwriting but also on brain regions involved in executive control and working memory during spelling (Richards, Berninger & Fayol, 2009; 2012) and idea generation for composing (Berninger *et al.*, 2008). Brain activation of good and poor writers may therefore also differ during the idea generation process of writing because the good writers are more efficient than poor writers in engaging working memory while generating thoughts.

In a recent study, Richards et al (2015) evaluated several measures of white matter integrity (structural connections within the brain), functional connectivity between regions during two cognitive and two writing-related tasks, and correlations between white matter integrity and functional gray matter connections in typically developing children between grades 4 and 9 and children showing persisting handwriting problems (dysgraphia) or persisting word reading/spelling problems (dyslexia). They showed that the control group differed from dysgraphics but also from a dyslexic group, who differed from each other, in both white matter integrity, fMRI functional connectivity from four seed points for written word production (left occipital temporal, supramarginal gyrus, precuneus, and inferior frontal gyrus), and the white matter integrity–gray matter functional connectivity. Overall, controls tended to have more indicators of structural white matter integrity, and fewer functional connections, consistent with more efficient processing on written language tasks at the subword level (writing a letter than follows a visually displayed letter in the alphabet) and word level (adding a letter in a blank in a letter series to create a word-specific spelling). Functional connection differences were found on (a) letter- and word-level writing tasks, and (b) resting-state (mind wandering which probably underlies flow during writing) and goal-related cognition (planning before composing outside scanner). Thus, even when dysgraphia is defined on the basis of a transcription skill (impaired handwriting which may interfere with spelling), the dysgraphia group differed in cognitive as well as handwriting tasks. Collectively, their findings provide evidence for contrasting neurobiological patterns during two written language tasks and two cognitive tasks for typical writing development, developmental dysgraphia and developmental dyslexia during middle childhood and early adolescence.

Conclusion

- 17 Advances in the understanding of writing organization in the brain are only at their beginning. The basic network sustaining handwriting is now better delineated both in adults and in children. The inclusion of this network in the overall functioning of the brain, and the interconnections between regions during various handwriting and related writing and reading tasks is a promising field of research. Research on the letter level, word level, and text level of the writing brain is fundamental to provide links between brain lessons and teaching tips for educators, to ultimately facilitate writing development (see James, Jao & Berninger, 2015, for an overview). Understanding writing organization in the brain is also crucial to try to understand why handwriting learning is so difficult for some children (with dysgraphia). Finally, it is also highly relevant to anticipate what can possibly change in the writing/reading network if handwriting is replaced by typing teaching at school and hence what would be the impact of such a drastic change in both the writing and reading capacities of children and adults in the next generations.

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ABSTRACTS

- In this article, we present recent neuroimaging studies performed to identify the neural network involved in handwriting. These studies, carried out in adults and in children, suggest that the mastery of handwriting is based on the involvement of a network of brain structures whose involvement and inter-connection are specific to writing alphabet characters. This network is built upon the joint learning of writing and reading and depends on the level of expertise of the writer. In addition, a part of this graphomotor network is also brought into play during the identification letters during visual reading. These skills are also the basis for the development of more complex language activities involving orthographic knowledge and composition of texts. The studies presented cover two perspectives: that of neuroscience and that of cognitive psychology, as both are necessary to understand a complex process of writing and both depend on natural interactions and the influence of educational exposure.

Dans cet article, nous présentons des études récentes de neuro-imagerie destinées à identifier le réseau neuronal impliqué dans l'écriture manuscrite. Ces études, réalisées chez l'adulte et chez l'enfant, montrent que la maîtrise de l'écriture manuscrite repose sur la mise en jeu d'un réseau de structures cérébrales dont l'implication et l'interconnexion sont spécifiques de l'écriture des caractères alphabétiques, se construisent au cours de l'apprentissage conjoint de l'écriture et de la lecture et dépendent du niveau d'expertise du scripteur. En outre, une partie de ce réseau graphomoteur est aussi mise en jeu au cours de l'identification des lettres sur base visuelle et de la lecture. Ces compétences graphomotrices servent également de base au développement d'activités plus complexes impliquant les connaissances orthographiques et la composition de textes. Les études présentées ici sont abordées de deux perspectives, celles des neurosciences et de la recherche en psychologie cognitive, car elles s'avèrent toutes deux nécessaires pour comprendre un processus aussi complexe que l'écriture, à la fois tributaire d'interactions naturelles et éducationnelles.

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Mots-clés: cerveau, écriture, dysgraphie, lecture, neuro-imagerie

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