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Analysis of brain activation in teenagers with isolated dysorthography (spelling disorder) and good spellers during a spelling assessment task

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SUMMARY

Background: There has been little research on the neural correlates of writing in children and teenagers. The purpose of the present study was to describe brain activation in a group of teenagers with isolated dysorthography vs. teenagers with good spelling skills, during a task involving the assessment of spelling accuracy in words containing an orthographic difficulty. The results will allow for conclusions relating to neural mechanisms underlying the isolated developmental disorder connected with learning to spell correctly.

Material/ Methods: The participants included 15 subjects with isolated dysorthography and 14 good spellers, aged 13-15. An fMRI examination was performed to obtain data on brain activation prompted by visually presented words (correctly and incorrectly spelled). The participants were asked to judge if the words were spelled correctly or not.

Results: The group with dysorthography was found to activate a total of 18 clusters, while the controls activated 7 clusters. An analysis of the tables shows that the structures activated in the two groups do not overlap. In the group with dysorthography, posterior and anterior cortical areas were activated, while in the control group activation was found mainly in the cerebellum. Both groups showed activation in the middle occipital gyrus; however, in the controls this was in the left hemisphere, while in the subjects with dysorthography it was in the right hemisphere. The cerebellum was active only in the controls.

Conclusions: Cerebellar deficit may be one of the significant mechanisms leading to learning difficulties manifesting as isolated spelling disorder

Key words: fMRI, cerebellum, interior frontal gyrus, parietal lobe, accuracy of spelling, automatization

INTRODUCTION

Despite the fact that difficulties in written language processing include both reading and writing impairments, research on the neurocognitive mechanisms involved has focused mainly on dyslexia (Lipowska & Sajewicz-Radtke, 2012; Krasowicz-Kupis et al., 2009; Pachalska et al., 2009). Studies intended to examine the brain mechanisms related to written language processing include research on reading and writing in normal development, as well as in developmental and acquired pathologies. This includes a search for the critical areas of the brain that are common and specific for various aspects of written language (Richards et al., 2006).

RESEARCH FOCUSING ON HEALTHY SUBJECTS

In terms of its psychological structure, writing is a complex activity, and so activation has been found in various areas of the central nervous system (CNS), depending on which aspect of writing is being analyzed. The most consistent findings involve activation in the left inferior parietal cortex, which is engaged in mapping between phonological and orthographic representations (Booth et al., 2003). Most studies have also demonstrated activation in the left inferior temporal (fusiform) gyrus, which is engaged in orthographic processing (Dehaene et al., 2004), and in the left inferior frontal gyrus, which contributes to the modulation of processes in the posterior regions of the brain (Bitan et al., 2005). Higher accuracy in orthographic decision tasks has been connected with greater activation in the left inferior frontal gyrus (BA 46) and left fusiform gyrus (BA 37), while higher accuracy in auditory spelling tasks correlates with greater activation in the left supramarginal gyrus (BA 40), left angular gyrus (BA 39), and left fusiform gyrus (BA 37; Booth et al., 2003).

Few researchers have focused, however, on finding the neural correlates of writing in children and teenagers (Richards et al., 2005). Three studies have looked for differences between adults and children regarding the assessment of spelling. In one of these, adults demonstrated higher activation than children in the left angular gyrus (BA 39) and left inferior frontal gyrus (BA 9) in a task involving visual spelling, and in the left interior frontal gyrus (BA 44/45/9), left angular gyrus (BA 39) and left superior temporal gyrus (BA 22) in a task involving the auditory assessment of spelling (Booth et al., 2004). Another study reported a modality-independent activation in adults in the left inferior frontal gyrus (BA 9; Booth et al., 2003). When the method of dynamic causal modelling (DCM) was used to examine the effective links between certain areas of the brain, greater top-down control from the left inferior frontal gyrus (BA 45/46/9) to the left inferior parietal lobule/precuneus (BA 40/7; Bitan et al., 2006) was found in adults than in children. Correctness was connected with increased activation in the left inferior frontal gyrus (BA 44/9) and left inferior parietal lobule (BA 40). Given these data, we can assume that the regions of crucial importance in spelling tasks include the left inferior frontal gyrus (BA 44/9), the left inferior parietal cortex (BA

40/39), and the left fusiform gyrus (BA 37), and their importance increases with age and growing ability. Purcell et al. (2011) reported that the perisylvian area, including the inferior frontal gyrus and supramarginal gyrus, appears to play a significant function in phoneme-grapheme mapping in the process of writing.

The left inferior occipitotemporal cortex or the Visual Word Form Area (VWFA), a part of the left ventral visual pathway, is also of importance in the process of writing. It is involved in recognizing a complete word, which is illustrated by more effective processing of familiar vs. unknown words, described as the "orthographic familiarity effect" (Bruno et al., 2008; Kronbichler et al., 2007), and it is engaged in serial, sublexical coding of letter shapes (Schurz et al., 2010; Braet et al., 2012).

Richards et al. (2006) investigated a group of good spellers and readers for activation that would be common and specific for three types of language tasks: phonological, morphological and orthographic. Common areas of activation were found in the left inferior frontal gyrus, the lingual gyrus (bilaterally), the fusiform gyrus (bilaterally), and the left inferior temporal gyrus. Areas with specific activation during an orthographic task included the right middle frontal gyrus, the left superior temporal gyrus, the right middle temporal gyrus and the anterior insula (bilaterally). Only the left cerebellar hemisphere was activated specifically for morphological mapping. In phoneme mapping, the cerebellum was activated bilaterally. Additionally, phonological tasks were found to activate the left orbital frontal gyrus, the thalamus (bilaterally), the right inferior temporal gyrus, and the right cerebellum.

RESEARCH FOCUSING ON CLINICAL GROUPS

Obviously dyslexics do not use entirely different brain systems for language processing, since in various language tasks they show the same activation as in good spellers and readers (Richards et al., 2006). There are also structures which are activated in good spellers and are not activated in dyslexics, and vice versa. Lower activity may indicate difficulties in engaging structures which are necessary for performing a given task. Additional activity may suggest that alternative pathways are being used (compensation; Richards et al., 2006). It appears from this latter study that the right inferior frontal gyrus and right posterior parietal region are particularly significant in orthographic processing (orthographical mapping), and differentiated the group of dyslexics from normal readers; significant change was recorded following treatment. The change was maintained after treatment was completed. Other studies by the same author have reported differences in activation between good and poor readers in the left and right inferior frontal gyrus and bilaterally in the cerebellum (Richards et al., 2008).

Subsequently, Richards et al. (2009) compared children with poor and good spelling skills and reported that good spellers to a greater extent activated the left precentral gyrus, the left post-central gyrus, the inferior frontal gyrus, and the right superior frontal gyrus. In poor spellers, higher activation was found in the left primary motor cortex, the middle frontal gyrus and cuneus in the right hemisphere, and the middle frontal regions.

The only study to compare brain activation in three groups of children (with dyslexia and writing impairments, with isolated spelling disorder, and good spellers) has been conducted by Gebauer et al. (2012). In comparison with the dyslexic children and controls, the subjects with isolated spelling disorder were found to have stronger right hemispheric activation during an orthographic decision task. The children with isolated spelling disorder activated the inferior and middle frontal gyri bilaterally when processing correctly and incorrectly spelled words, while the other two groups showed bilateral activations only in the case of misspelled words. This suggests that additional right frontal activation may generally be connected with the higher requirements of a task and with mental effort during the performance of the task (Gebauer et al., 2012).

Due to the small number of studies focusing on individuals with isolated spelling impairment (e.g. Richards et al., 2009; Angelelli et al. 2010), there are still insufficient data concerning the brain mechanisms underlying isolated spelling disorder. The purpose of the current study was to describe brain activation in a group of teenagers with isolated dysorthography vs. teenagers with good spelling skills, during a task involving assessment of spelling accuracy in words containing an orthographic difficulty. The results will allow for conclusions relating to the neural mechanisms underlying isolated developmental disorder, which is connected with learning to spell correctly.

MATERIAL AND METHODS

Participants

The study included 15 poor spellers (PS) and 14 good spellers (GS), aged 13 -15. The subjects in both groups attended middle schools in Warsaw and Otwock, Poland. The students' assigment to the clinical group was based on their school documentation containing a diagnosis of dysorthography issued by a psychological and pedagogical counselling center, following a double standardized measurement. Afterwards, the selected children's parents were asked for their consent for the examination and interviewed on their children's development. After the parents' consent was obtained, the children's cognitive development was measured with the Wechsler Intelligence Scale, and their skills related to spelling accuracy were assessed with the use of a dictation test developed by Z. Saduś. This test consists of 26 two-word expressions presenting various orthographic difficulties, mainly words with ó-u, rz-ż, ch-h, pairs of Polish graphemes in which each member of the pair is pronounced exactly the same as the other in standard Polish. The score was based on the number of errors in taking dictation. The ultimate criteria for assignment to the clinical group included:

- isolated spelling disorder diagnosed in a psychological and pedagogical counselling center by a team of specialists (psychologist, pedagogue);

Table 1. Descriptive statistics for both groups

Variable	Poor spellers (PS)		Good spellers (GS)		T	P
	Mean	SD	Mean	SD		
Age in months	166.20	7.93	166.86	7.93	-0.946	n.s.
IQ – full scale	107.67	10.63	111.21	13.35	-0.794	n.s.
IQ – verbal scale	103.43	11.40	110.57	11.27	-1.686	n.s.
IQ – non-verbal scale	111.40	10.34	110.29	14.97	0.235	n.s.
Number of errors in dictation	22	7.11	2.42	1.74	10.010	0.000

n.s. – not significant

- educational history showing no impairment in reading skills;
- an IQ greater than 85;
- poor spelling skills.

The controls did not show impairments in writing or reading. None of the participants in our study had neurological disorders or sensory impairments.

The study groups differed significantly in terms of their ability to correctly write words presenting an orthographic difficulty. No differences were found between the groups in terms of age and intelligence in verbal, non-verbal and full scales (see Table 1).

PROCEDURE

An fMRI examination was performed to obtain data on brain activation. The study was conducted in the MRI Lab at the European Health Center in Otwock.

MR imaging was performed on a 3T MR scanner (Achieva 3T TX, Quasar Dual gradients, Philips, Best) with a BOLD Specialist Package, using an 8-channel Sense Head Coil. A single-shot gradient echo, echo planar sequence, was used for fMRI acquisition (FFE-EPI, TE= 30[ms], TR= 3000[ms], TA= 14:30 [min], slice thickness=3[mm], gap=0[mm], matrix=96x96, FOV= 192x192 [mm], number of slices=42, SENSE factor 1.8, dynamics=290).

The participants' task was to judge the correctness of the spelling of Polish words with orthographic difficulties. The experiment used 18 pairs of words with an orthographic difficulty, six in each category: "ó-u", "rz-ż", "ch-h". We chose words with this type of irregularity in Polish because they are the most difficult for children to learn to spell correctly (Polish orthography is otherwise remarkably regular, especially in comparison to English). Brain activation was prompted by visual stimuli: words were the proper stimulus, while a centered cross served as a neutral stimulus. Each sequence of stimuli was preceded by instructions. During the sequences, the subjects were presented with pairs of words, one above the other. Half of the phrases with orthographic difficulties were pairs with two correctly spelled words, while the other half contained one misspelled word. The participants were asked to judge if the words were spelled correctly or not. None of the pairs contained the same word used twice. The words were drawn at random from list A (correctly spelled words) and B (misspelled words). The random factor related to the selection of the word, its position on the screen (top-bottom), and the correctness/incorrectness of its spelling. The subjects were exposed to each pair of words for 6 seconds. Following the presentation of each pair of words, the subject responded YES or NO using a response pad adjusted to operation within the magnetic field. The stimuli were displayed via a projector connected to the computer controlling the experiment. The subjects watched the screen via a mirror placed in front of their eyes. Before the examination each individual adjusted his/her position to make sure the image in the mirror was clear and easy to read.

The examination used the following procedure:

- 30-second display of gaze fixation point (cross);
- 30-second display of a set of words (task AB);
- 30-second display of gaze fixation point (Cross).

Subsequent pairs of words were displayed every 6 seconds, which gave 5 pairs of words in each set. The procedure used a total of 36 word pairs with correct spelling and 36 pairs with one misspelled word.

The results obtained during the fMRI study were analyzed using the SPM8 package (Statistical Parametric Mapping).

RESULTS

The AB vs. CROSS contrast was analyzed for these two groups of teenagers with dysorthography and with good spelling skills. AB vs. CROSS contrast means that the activation in the experimental conditions of rest and no activity (CROSS) was subtracted from the activation during the orthographic decision task (AB). Because of the apparent nature of the lack of activity, such a solution was necessary to measure activation for the primary task. The following threshold values of statistical significance were assumed for computing activations:

- p value = 0.001;
- intensity = 3.852;
- cluster size = 25.

The results obtained for the group with dysorthography and for the group of good spellers are presented in tables 2, 3 and 4. Table 2 contains structures in the posterior region of the brain, Table 3 presents anterior structures, and Table 4 shows cerebellar structures. The group with dysorthography was found to activate a total of 18 clusters (see Fig. 1), and the controls activated 7 clusters (see Fig. 2). An analysis of the tables shows that the structures activating in both groups do not overlap. In the group with dysorthography posterior and anterior cortical areas were activated, while in the control group activation was found mainly in the cerebellum. Both groups were found to show activation in the middle occipital gyrus; however, in the group of good spellers this was in the left

Table 2. Analysis of the one-sample t-test for contrast AB vs. CROSS in both study groups, at p=0.001, activations in the posterior region of the brain

Structure	Cluster size	Peak t-value	MNI x,y,z	GS	PS
Middle Occipital Gyrus	5530	9.0574	28 -90 12		R
Middle Occipital Gyrus, Occipital_Inferior_L	109	4.6566	-48 -72 -16	L	
Precuneus , BA 7, Occipital_Superior_L	41	4.6629	-24 -70 34		L
Supramarginal Gyrus, Angular_R	29	6.7579	40 -50 36		R
Precuneus, Occipital_Mid_R	28	5.4788	32 -74 36		R
Inferior Parietal Lobule, Angular_L	149	6.6232	-38 -62 40		L
Inferior Parietal Lobule, Parietal_Inf_L	29	5.1048	-44 -38 44		L
Inferior Parietal Lobule, Parietal_Inf_R	40	4.7747	50 -46 54		R

R- right hemisphere
L – left hemisphere

Table 3. Analysis of the one-sample t-test for the contrast AB vs. CROSS in both study groups, at p=0.001, activations in the anterior regions of the brain

Structure	Cluster size	Peak t-value	MNI x,y,z	GS	PS
Inferior Frontal Gyrus	457	7.0597	-42 24 2		L
Inferior Frontal Gyrus, Insula_R	99	5.085	38 20 -8		R
Inferior Frontal Gyrus ,BA 9, Precentral_L	667	7.7395	-46 4 30		L
Inferior Frontal Gyrus, Precentral_R	34	4.5338	46 2 24		R
Inferior Frontal Gyrus, Precentral_R	102	5.157	52 12 40		R
Middle Frontal Gyrus, Frontal_Mid_Orb_R	49	5.5648	34 54 -10		R
Middle Frontal Gyrus , Frontal_Mid_L	30	4.4507	-44 24 34		L
Superior Frontal Gyrus, Frontal_Mid_R	64	5.3482	38 50 30		R
Frontal_Sup_Medial_L	40	4.4362	2 34 38	R	
Cingulate Gyrus, Cingulum_Mid_R	288	5.8947	10 24 32		R
Parahippocampal Gyrus, BA 27, Hippocampus_L	41	6.4891	-22 -32 -4		L

R- right hemisphere
L – left hemisphere

Table 4. Analysis of the one-sample t-test for the contrast AB vs. CROSS in both study groups, at p=0.001, cerebellar activations

Structure	Cluster size	Peak t-value	MNI x,y,z	GS	PS
Cerebellum Posterior Lobe, Pyramis	26	5.8813	-32 -70 -44	L	
Cerebellum Posterior Lobe, Declive	342	6.237	0 -64 -26	R	
Cerebellum Posterior Lobe, Tuber	27	5.2367	30 -58 -36	R	
Cerebellum Posterior Lobe, Uvula	27	4.1449	-28 -70 -32	L	
Cerebellum Anterior Lobe, Culmen	30	6.8242	-30 -54 -30	L	

R- right hemisphere
L – left hemisphere

hemisphere, while in individuals with dysorthography it was in the right hemisphere. The cerebellum was active only in the controls.

DISCUSSION

It should be assumed that the activation of specific structures in the group of good spellers is adequate for the requirements of an orthographic decision task, and is an indicator of the normal course of these processes. One characteristic trait in the group of good spellers is the activation of the cerebellum. According to the traditional approach, the cerebellum plays a fundamental role in regulating motor processes. Still, current knowledge concerning the tasks of this structure allows for a much broader understanding of its functions. The cerebellum, including in particular the lateral hemispheres of its posterior lobe, has functional links with association cortical areas involved in complex mental processes. These pathways enable the cerebellum to participate in widespread neural circuits controlling sensorimotor, intellectual and emotional processes. Neurobehavioral deficits resulting from cerebellar disturbances may occur even if there are no motor impairments. When a dysfunction is related to the hemispheric regions of the posterior cerebellar lobes, the impairments most frequently reported are connected with executive functions, such as planning, mental flexibility, verbal fluency, abstract thinking, working memory, or functions of spatial cognition and language processing (Schmahmann & Caplan, 2010).

The concept of cerebellar function related to motor learning includes a hypothesis on the analogous acquisition of rules. Importantly, as a result of decreased attention requirements, automaticity allows a primary task to be performed with little or no susceptibility to interferences caused by other tasks. Balsters et al. (2011) suggest that changes in cerebellar activation are linked with the automatic use of cognitive rules, as a result of the interactions between the anterior lobes of the cerebellum and the prefrontal cortex. Cerebellar dysfunctions, then, may lead to problems with the automaticity of cognitive activities, such as the ability to spell correctly without referring to consciously performed tasks which require effort and executive attention (Balsters & Ramnani, 2011).

The involvement of the anterior cerebellar lobe may suggest that good spellers use automatic motor control. This is consistent with suggestions indicating the existence of such a mechanism. Spelling accuracy is also an effect of motor automaticity developing as a result of multiple repetitions of a specific movement of the hand while writing a given word. Activation in the posterior cerebellar lobe is evidence of automaticity in cognitive processes and language processing in good spellers. Richards (2006) has argued that no or weaker activation of a structure in clinical groups versus controls suggests insufficient involvement of that structure in performing the task, leading to lower effectiveness. In the case investigated by the current study, cerebellar mechanisms which are insufficiently active in the group with dysorthography may contribute to low effectiveness in learning and using the rules of correct spelling. The hypothesis of an automatization deficit connected with cerebellar dysfunction was earlier proposed by Nicolson and Fawcett (Nicolson, Fawcett, & Dean, 2001), who made this assumption taking into account the motor deficits prevailing in dyslexic children and typical for cerebellar impairments, namely discreet impediments to balance, muscle tone and coordination. Although this hypothesis is still subject to discussion, a number of studies suggest that motor deficits result from other developmental problems concurrent with dyslexia, e.g. ADHD (Raberger & Wimmer, 2003).

The subjects with dysorthography activated the middle occipital gyrus, just like the good spellers. However, in the individuals with dysorthography this activation was visible in the right hemisphere, and in the good spellers it occurred in the left hemisphere. In the poor spellers, the activation of the right occipital region may suggest a compensatory mechanism in response to left hemisphere language dysfunctions. No activation in the left hemisphere during the orthographic decision task and activation only in the right hemisphere may suggest that individuals with dysorthography analyze written words in the same manner as drawings, without engaging the language structures of the left hemisphere. The active structures in individuals with dysorthography include the precuneus bilaterally, the right supramarginal gyrus and angular gyrus, and the inferior parietal lobule bilaterally. Their activation suggests that during an orthographic decision task these individuals use the sound-letter relationship and attempt to make a compensatory use of phonology (Booth et al. 2003). Unfortunately, this strategy is not effective for the words used in this study. The selected words with ó-u, rz-ż and ch-h do not differ at the phonological level as a result of the variant spellings. Activation of the right supramarginal gyrus and angular gyrus may serve as an evidence of concentration on the spatial and visual aspects of the letters constituting the written words.

The inferior frontal gyrus in the left hemisphere is frequently indicated as a structure playing an important role in both orthographic and phonological language processing. Activation in this structure has been demonstrated in a number of different language tasks, mainly those which require some effort and decision-making. Differences in the activation of this structure have also been reported in studies comparing dyslexic children with good readers (Richlan, Kronbichler & Wimmer, 2011). Thus it seems that the differences in the activation of the left inferior frontal gyrus are not specific for writing disorders alone, but for language impairments in general. This is because accurate spelling as an activity does not involve visual perception alone, but also the language system (Polk et al., 2002), and so the latter seems to be one of the impaired processes in spelling disorders. Our results would support this hypothesis. Activation of the left medial frontal gyrus in the group of poor spellers may be attributed to the need for these individuals to intentionally employ working memory by (du Boisgueheneuc et al., 2006). Our study contained a task involving a decision on the accuracy of spelling where the participants were required to engage memory processes; these were found to be less functional in poor spellers and demanded greater effort. Such structures as the cingulate gyrus, parahippocampal gyrus (BA 27), and hippocampus are actively involved in searching memory and making a decision; activation of the cingulate gyrus suggests uncertainty and difficulty in making a decision, as well as the existence of an internal conflict.

CONCLUSION

Our subject teenagers with isolated spelling disorder were found to have different brain activation from that of their peers with normal spelling skills in a task involving a judgement of accuracy in the spelling of words presenting an orthographic difficulty (*rz-ż, ó-u, ch-h*). Activation was found in the frontoparietal structures typical for the initial stages of learning to write. The teenagers with good spelling skills mainly activated the cerebellum, which may be an evidence of automaticity in the process of assessing words in terms of spelling accuracy.

Generally, the observed activation indicates that teenagers with dysorthography attempt to cope with a difficult situation by making more effort, applying strategies based on phonology as an intermediary, and excessively analysing words in terms of visual and spatial traits. On the other hand, in good spellers the orthographic decision process is automatized and does not require such measures.

Taking into account our findings we propose that cerebellar deficit may be one of the significant mechanisms leading to learning difficulties manifested in the form of isolated spelling disorder.

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