
DYSEXECUTIVE AGRAPHIA: A MAJOR EXECUTIVE DYSFUNCTION SIGN

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Different types of writing disorders associated with brain pathology have been described. Limited mention the writing disturbances associated with prefrontal pathology, however, is found. Clinical observations of patients not only with focal prefrontal pathology but also with other conditions affecting the frontal system (e.g., traumatic head injury, dementia) confirm the assumption that these patients present an overt decrease in the ability to express ideas in writing. It is proposed that complex aspects of writing, such as planning, narrative coherence, and maintained attention, are significantly disturbed in cases of impairments of executive functions. Frontal lobe patients not only have difficulties in keeping the effort required for writing, but also to organize the ideas in the written texts. The term *dysexecutive agraphia* is proposed to refer to this writing disorder. Three illustrative cases are presented. It is finally suggested that questions regarding the ability to write should be included in dementia questionnaires and executive functioning testing.

Keywords agraphia, dysexecutive syndrome, executive functions, frontal syndrome

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Agraphia can be defined as the partial or total loss of producing written language associated with brain pathology (Benson & Ardila, 1996). The ability to write can be impaired as a result of linguistic defects (aphasia), but other elements not related with language (e.g., motor, spatial, conceptual) also participate in the writing ability (Roeltgen, 2003). To write supposes, at least, knowledge of the language codes (phonemes, words), ability to convert language sounds into graphemes, knowledge of the graphemic system (alphabet), ability to perform fine movements, and appropriate use of the space for distributing, joining, and separating letters. In addition, text writing requires planning, narrative coherence, and maintained attention. It is evident that diverse types of writing disturbances can be found in clinical practice (Benson & Cummings, 1985).

Different attempts to classify writing disturbances are found in the history of neuropsychology. Goldstein (1948) distinguished two major types of agraphia: apractoamnesic and aphasic-amnesic. Luria (1976, 1980) referred to five different types of agraphia, three of them associated with aphasia (sensory agraphia, afferent motor agraphia, and kinetic agraphia) and two associated with visuospatial defects. Hécaen and Albert (1978) distinguished four types of agraphia: pure, apraxic, spatial, and aphasic. During recent decades, psycholinguistic and cognitive models of agraphia have become particularly influential (for a review, see Roeltgen, 2003). A basic distinction between central and peripheral agraphias (dysgraphias) is considered; different subtypes for each major type are distinguished. Regardless of the diversity in the classifications of agraphia, a basic distinction can be established between agraphias due to a language impairment (linguistic, or central, or aphasic agraphias) and agraphias due to other types of impairments (peripheral, or non-aphasic agraphias) disturbing the normal ability to write (Benson & Ardila, 1996).

Agraphia is, in consequence, a complex disorder, and writing disturbances can be observed in cases of damage in different localizations in the brain, including frontal, parietal, and temporal (Anderson et al., 1993). Agraphia can even be associated with right hemisphere pathology (Ardila & Rosselli, 1993).

Chedru and Geschwind (1972) observed that patients in confusional status resulting from diverse etiologies could talk, understand, repeat, name, and read, but were unable to express their ideas in writing. Writing was slow, clumsy, and even with a vague meaning. The authors emphasized the sensibility of written production in case of any diffuse brain condition. Different studies have documented similar writing difficulties in dementia (e.g., Groves-Wright et al., 2004; Henderson et al., 1992; Horner et al., 1988; LaBarge et al., 1992; Luzzatti et al., 2003; Neils, Boller, Gerdeman & Cole, 1989; Platel, Lambert, Eustache, Cadet, Dary & Viader, 1993; Tomoeda & Bayles, 1993). Writing disorders are

considered to represent an early manifestation of Alzheimer's disease (AD), often more severe than oral language difficulties. AD patients produce shorter and less informative written descriptions of a complex picture than controls. These abbreviated texts also include many intrusions, semantic substitutions, and misspellings (Croisile, 1999). Furthermore, significant correlations are observed between narrative writing ability and severity of dementia (Horner et al., 1988). The writing impairments in AD have been interpreted as multi-componential in nature and follow the pattern of cortical deterioration reported in the brains of AD patients (Forbes et al., 2004).

Notably, it has also been documented that elementary-school children with written expression problems perform more poorly in executive functioning tests than children without these difficulties (Hooper et al., 2002). An association between written expression ability and executive functioning is evident.

EXECUTIVE FUNCTIONS

The term "executive function" is relatively new in the neurosciences. "Executive functioning" is a term addressed by many, but coalesced by Lezak (1983) to discriminate cognitive functions from the "how" or "whether" of human behaviors. Lezak emphasized the fluid nature of executive functioning and how dependent the cognitive and emotional aspects of functioning were on the "executive." Baddeley (1986) grouped these behaviors into cognitive domains that included problems in planning, organizing behaviors, disinhibition, perseveration, reduced fluency, and initiation. Baddeley coined the term "dysexecutive syndrome." Each component of executive functioning has added to the array of cognitive processes, which include maintaining a problem-solving set for goal-directed behavior: interference control, flexibility, strategic planning ability, and the ability to anticipate and engage in goal-directed activity (Denckla, 1994). The definition of executive function is encompassed by actions fueled by conceptualizations, such as the ability to filter interference, engage in goal-directed behaviors, anticipate the consequences of one's actions, and the adaptive concept of mental flexibility (Ardila & Surloff, 2004; Denckla, 1996; Goldberg, 2001; Luria, 1969, 1980; Stuss & Benson, 1986; Stuss & Levine, 2002).

AGRAPHIA IN FRONTAL PATHOLOGY

Historically, agraphia has been associated with left premotor area pathology. In 1881 Exner proposed a "writing center" located in the base of the second frontal gyrus ("Exner's area"). Dubois et al. (1969) reported six cases of "pure agraphia," four of them with frontal lesions. Some other reports are found in

the literature indicating that the second frontal gyrus is significantly involved in writing (e.g., Anderson et al., 1990). Nonetheless, agraphia associated with Exner's area damage is not a dysexecutive agraphia.

Patients with prefrontal lobe pathology typically present a preserved ability to read associated with a significant disturbance in the ability to write (a kind of "agraphia without alexia") (Ardila, 2004; Benson & Ardila, 1996). The writing defect is not a primary defect. These patients can convert phonemes into graphemes, appropriately use the writing space, and perform the sequences of writing movements. Patients often have a decreased spontaneous language associated with good comprehension and repetition (transcortical or extrasylvian motor aphasia). They can write words and sentences by dictation, but spontaneous writing is even more impaired than spontaneous language (Berthier, 1999). Reading (perceptual recognition) in these patients is notoriously better than writing (sequential motor production). Copying can be notoriously better (Luria, 1976, 1980; Stuss & Benson, 1986). Perseveration—as in any other motor act, can be observed. Perseveration can include sentences, words, letters, and strokes in letters (Benson & Ardila, 1996; Berthier, 1999).

BRAIN ACTIVATION DURING WRITING

Recently, some articles have been published analyzing the pattern of brain activation observed during writing. Omura et al. (2004) studied the neural substrates of phoneme-to-grapheme conversion in writing to dictation using functional magnetic resonance imaging (fMRI). They employed Japanese as the stimulus language because in Japanese, one phoneme is represented by one grapheme (Kana) and vice versa. fMRI revealed that the left premotor, extending into Broca's area was activated. They concluded that the frontal region is required for the conversion of phonemes to graphemes in writing by dictation. Katanoda et al. (2001) used fMRI, brain hemodynamic activity during three conditions that differentially engaged visual, linguistic, and/or motor functions: (1) writing names of pictures with the right index finger, (2) naming pictures silently, and (3) visually cued finger tapping. Writing without naming comparison and writing minus tapping comparison were performed, and brain regions commonly activated in these two contrasts were detected. The main finding was that such common activation was observed in the anterior part of the left superior parietal lobule, the posterior part of the middle and superior frontal gyri, and the right cerebellum. The parietal and frontal regions were considered to subservise the process of writing as separated from that of naming and finger movements. Matsuo et al. (2003) attempted to discriminate

Exner's area from the frontal eye field using fMRI. They found that saccadic eye movements activated a region defined as the frontal eye field, whereas three language experiments that included translation between grapheme and phoneme activated another region defined as Exner's area. Exner's area was found to be located only 1.5 cm apart from the frontal eye field.

In summary, writing represents a complex task requiring the participation of multiple brain areas. Left frontal areas are crucial in writing.

ILLUSTRATIVE CASES

Three cases are presented as illustrations of dysexecutive agraphia. The first one result from a mild traumatic head injury, the second one to a dementia, and the last one to a cerebrovascular accident involving the frontal lobes. Regardless of the heterogeneity of the cases, all of them present executive disorders associated with some frontal lobe abnormality. In all three cases, writing by dictation was well preserved, but narrative writing was significantly impaired.

First Case

The patient is a 39-year-old right-handed man with a university level of education. After graduating as a physician, he decided to become a catholic priest. As a priest, he devoted most of his time to teaching, religious guidance, and writing activities. His papers usually dealt with religious and philosophic topics, reflections about the meaning of life, ethics, and human behavior. While riding a bicycle, he fell down, briefly lost consciousness, presenting a post-traumatic anterograde amnesia for about one hour. He was taken to a local hospital. A CT scan disclosed a left fronto-temporal fracture with associated edema.

Five months after the head injury he was referred for a neuropsychological exam because he complained about attentional difficulties, mild memory deficits, inability to understand his readings on philosophy, and lack of interest toward his work. A right-handed, well-oriented, and cooperative patient was observed. No motor or sensory deficits were found.

His language production and comprehension were normal for casual conversation. At the Boston Naming Test, long latencies were observed, and the patient presented one semantic verbal paraphasia (score = 50/60). Verbal fluency was normal (FAS = 48; animals = 15), but included derivative words were frequently.

There was not an evident alexia, agraphia, or acalculia on routine reading, writing, and calculation ability testing, but the patient pointed out that he had to

pay too much attention to arithmetical problems, because otherwise, he became confused. He also pointed out that that he had stopped writing; he stated, "*I cannot concentrate and nothing comes to mind.*"

No ideomotor or ideational apraxia were found. No visual, tactile, or auditory agnosia or extinction were observed either. His score in the Rey-Osterrieth Complex Figure was normal (32/36). His scores in the WAIS-R were in the upper range (Full Scale IQ = 119) but potentially decreased for his previous intelligence level. His scores in the Wechsler Memory Scale were also normal (MQ = 116), and only scores in the Digits Backwards and Associative Learning subtest scores had decreased. His performance in the Wisconsin Card Sorting Test was normal.

He described his writing difficulties in the following way: "When I have tried to write, my writing lacks clearness and it is totally confusing. I skip logical steps in the redaction, and additionally, I make many orthographic mistakes. I feel a permanent slowness in all my intellectual activities. When reading or writing, I cannot identify what is the central issue or idea, and how to develop and analyze it." A year and half after the head injury, the writing difficulties persisted unchanged, and as a matter of fact, he had stopped writing, even personal letters.

Second Case

The patient is a 76-year-old, right-handed man with a university level of education. Insidious memory difficulties have been noticed for about five years. Paraphasias in spontaneous language have also been observed. Some recent behavior changes are described.

During the testing session the patient was cooperative and maintained an appropriate social behavior. No evident gross motor defects were observed. Fine movements with the fingers were slow but in general according to his age. No evident auditory or visual difficulties were recorded. He denied any depression. He also denied any memory difficulty or word-finding defects.

In the MMSE the patient obtained a score of 19/30. Spontaneous language was normal, correct in phonology, lexicon, and grammar. Voice volume, speech rate, and prosody were normal for his age. No evident articulation impairments were observed. No phonological or verbal paraphasias were recorded in spontaneous language, but some word-finding difficulties were noticed. Automatic language (e.g., counting, telling the months of the year) was correct. Telling the months backward was impossible.

The patient followed simple and two-step commands, and appropriately recognized right from left in his body and in the examiner's. His score in the Auditory Comprehension of Words and Sentences subtest was 17/18 and his score in the Token Test of the Multilingual Aphasia Examination was 33/44. In the word generation test using phonological (letter) categories he found 6 words beginning with the letter A, 5 words beginning with the letter F, and 6 words beginning with the letter S. When using semantic categories (animals) he produced 8 animal names in 1 minute. Three perseverations were noted. Naming ability had also decreased. His score in the Boston Naming Test was 39/60. Seven semantic paraphasias were recorded. Phonological cueing in most cases resulted in recovering the word. He could name high frequency body-parts (e.g., ear), but failed in naming lower frequency body-parts (e.g., eyelid). Language repetition was normal for 1, 2, and 3-syllable words. He was able to repeat a sentence containing up to nine words. With longer sentences, omissions and changes in word order were noted.

Reading was slow for his educational level. One word omission and one morphological paralexia were recorded in reading a 109-word text. Matching pictures and written words was correct according to the Multilingual Aphasia Examination—Written Comprehension of Words and Sentences. The patient wrote words and simple sentences by dictation. His spontaneous writing was reduced. When required to write any sentence, he wrote his wife's name. When it was insisted that it had to be a complete sentence, he wrote "Everything is possible." In the written description of the Plate #1 from the Boston Diagnostic Aphasia examination, he wrote "A child here the other a lady." According to the patient's wife, about 2–3 years ago he stopped writing letters and cards to his children and grandchildren, as he frequently used to do before. She further pointed out that as a matter of fact, he currently does not write at all.

The ability to perform simple subtractions (e.g., $100 - 7$) orally was decreased. He could only perform simple additions including one digit (e.g., $5 + 6$). He could not solve simple arithmetical problems. He was unable to find how many nickels make a dollar. His scaled score in the WAIS-III Information subtest was 7. He obtained a score 0/3 in the MMSE memory recall item. Two different conditions were used in the Wechsler Memory Scale—Logical Memory subtest: immediate and delayed. His performance in the immediate condition was 9/50. In the delayed recall condition his score was 3/50. Digit span forward was 5, and backward 3. His immediate and delayed recall of the Rey-Osterrieth Complex Figure was abnormal (1st percentile). He simply drew a square with some lines inside. In drawing a clock, spatial

disorganization and inability to place the handles pointing to 11:10 were evident. Performance in the Hooper Visual Organization Test was also decreased (16/30).

Difficulties in finding similarities between words (WAIS-III Similarities subtest) were noted, pointing to abstracting difficulties. Patient's scaled score in the WAIS-III Matrix Reasoning subtest was 6. Patient's scores in the Trail Making Test Part A corresponded to the 20th percentile. His performance in Part B corresponded to about the 1st percentile. One error was recorded. A diagnosis of dementia was proposed.

Third Case

The patient is a 51-year-old, ambidextrous male with a high-school level of education. He was hospitalized on August 28 after developing a sudden onset of severe headache and vomiting. Initial CT scan of the brain showed a right frontal hematoma as well as subarachnoid hemorrhage. Follow-up CT of the brain on August 29 showed bilateral frontal infarctions as well. The patient was found to have an anterior communicating artery aneurysm and underwent coiling of the aneurysm on August 31.

At the neuropsychological exam on September 14, he was alert, and partially oriented in time and place. Gross and fine movements were appropriate, but he presented some pathological reflexes (grasping bilaterally and snout) and paratonia. Tongue protruded in midline. Strength was 4+ to 5-/5 throughout all four extremities. Sensation was grossly intact. Incontinence was also reported. His score in the MMSE was 14/30.

No spontaneous language was observed. In responsive language no errors in phonology, word selection, or grammar were noted. Phrases were short and simple. Voice volume and speech rate were decreased. He could, however, follow simple and two-step commands. He did recognize right from left in his body and could follow crossed commands. He counted forward from 1 to 10, but could not count backward. He could tell the days of the week forward, but failed in telling the days of the week backward.

In the word generation test using phonological (letter) categories he produced one word using the letters F, A, and S. When using semantic categories (animals) he was unable to find any animal name. His score in the Boston Naming Test was 45/60. Four perseverations were recorded. Language repetition was normal for 1, 2, and 3-syllable words. His score in the Sentence Repetition section of the Multilingual Aphasia Examination was average. He could successfully repeat a sentence containing up to 11 words.

Reading aloud was nearly normal. He could read words and sentences, but only after insisting. Writing his name and simple words by command was correct. Letter size was appropriate, and no micrographia or progressive changes in letter size were noted. In the MMSE he failed in spontaneously writing a sentence. He could write two and three digit numbers by dictation, but again, perseverations were noted. No answer was found when requested to make a written description of Plate #1 from the Boston Diagnostic Aphasia Examination

The ability to orally perform subtractions (e.g., $100 - 7$) was decreased. Using simpler tasks (e.g., $20 - 3$) he also failed. Solving simple arithmetical problems was impossible.

Two different conditions were used in the Wechsler Memory Scale—Logical Memory subtest: immediate and delayed. Performance in the immediate condition was 2/50. After some 15–20 min, the patient was unable to recall any idea included in the two stories. A diagnosis of transcortical (extrasyllabic) motor aphasia and dysexecutive (prefrontal) syndrome was proposed

CONCLUSION

Narrative writing represents a complex executive function, highly sensitive to diverse brain pathologies involving the frontal lobes. Patients with focal frontal damage, as well as patients with other conditions impairing the frontal system, present an evident difficulty in expressing ideas in writing. Questions regarding the ability for narrative writing should be included in dementia questionnaires and executive functioning testing.

Writing difficulties in patients with prefrontal lobe pathology may be overlooked because of several reasons: (a) for many people writing is a kind of tangential activity. (b) During formal testing, it is found that the patient can usually write by dictation, and hence, agraphia is ruled out. (c) Writing difficulties may not impair daily life activities, unless the patient requires writing in a continuous way (as in the first case described). Furthermore, frontal lobe patients may present a constellation of cognitive and behavioral defects, obscuring their writing defects.

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