

Left Alien Hand Sign and Mirror Writing After Left Anterior Cerebral Artery Infarction

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A 43-year-old, right-handed man was admitted complaining of peculiar movements of his left hand. Computed tomography scan and magnetic resonance imaging showed an infarct area in the territory of the left anterior cerebral artery. The abnormal movements of the left hand were diagnosed as so-called alien hand sign. This patient also exhibited mirror writing with his left hand. The clinical manifestations and neuroradiological findings of this case are described in detail, and the possible mechanisms of these extremely rare combinations of alien hand sign and mirror writing are discussed.

KEY WORDS: Alien hand sign; "Diagonistic dyspraxia"; Mirror writing; Cerebral infarction

Infarction of an area of the brain supplied by the anterior cerebral artery is rare, accounting for about 3% of all cerebral infarctions [10]. According to the classic work of Critchley [8], the clinical symptoms of anterior cerebral artery infarction include hemiplegia with crural predominance, indefinite sensory impairment, left-sided apraxia, mental changes, some degree of aphasia in cases with a left hemispheric lesion, and psychomotor phenomena in the affected upper limbs. Recently, cases with so-called alien hand sign due to anterior cerebral artery infarction have been reported by several authors [14,18]. In the present paper the authors report a very unusual case involving a combination of left alien hand sign and mirror writing after left anterior cerebral artery occlusion with infarction.

Case Report

A 43-year-old, right-handed man was found at home in a state of confusion. His condition was diagnosed as

alcoholic delirium and he was admitted to a local hospital, where he remained for 3 weeks. His level of consciousness soon improved and his symptoms, which included aphasia and motor paresis, disappeared. After his discharge, however, the patient noticed that his left hand was behaving in a peculiar fashion. For example, when driving a car and attempting to turn to the right, his left hand turned the steering wheel to the left. He consulted the Department of Neurosurgery of Shizuoka General Hospital 2 months after his original illness. On admission the patient was alert, cooperative, and well oriented. His speech was fluent and there was no aphasia. His mental state was normal and no psychological abnormalities were noticed. During hospitalization no signs of alcoholic delirium were found. The cranial nerve functions were intact. Neither motor paresis nor sensory disturbance was detected. The patient volunteered the information that his left hand sometimes interfered with the actions of his right hand, but no abnormal motor function was observed at the time of admission. During his hospitalization, however, peculiar left-handed behavior was seen by the medical staff on several occasions. For example, when the patient tried to turn on the water with his right hand, his left hand promptly turned it off. When he attempted to remove his shirt with his right hand, his left hand put on the shirt. When he dialed the telephone with his right hand, his left hand immediately replaced the receiver and he was unable to complete the call. The patient tried to control the abnormal movement of his left hand without success, and he was frustrated. These unusual phenomena appeared to constitute the so-called alien hand sign [5,14], or "diagonistic dyspraxia" [1]. Computed tomography scan demonstrated a low-density area in the left hemisphere, predominantly in the medial superior frontal gyrus, cingulate gyrus, subcallosal area, gyrus rectus, and corpus callosum. In other regions neither atrophic changes nor abnormal low-density areas were noticed. The same area of infarction was clearly shown by magnetic resonance imaging (Figure 1). Cerebral angiography revealed a stenotic region in the proximal portion of the left anterior cerebral artery and a dilated segment about 5 cm in length in the distal portion of the artery (Figure 2).

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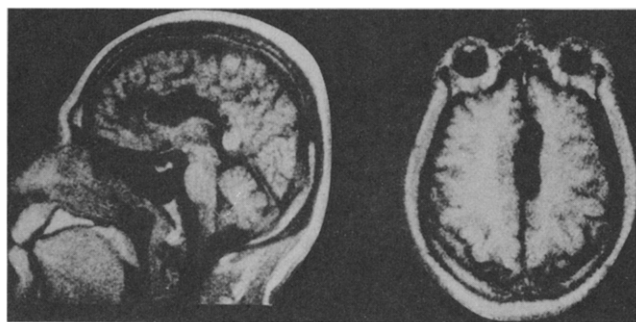


Figure 1. Magnetic resonance imaging (inversion recovery method; Tr 500, Td 300) clearly demonstrating the infarcted area in the left hemisphere, predominantly in the medial superior frontal gyrus, cingulate gyrus, subcallosal area, gyrus rectus, and corpus callosum.

Specific examination of motor function yielded interesting abnormal findings. With his right hand, the patient was able to transcribe copy from a text normally. However, with his left hand, he displayed mirror writing on both dictation and copying (Figure 3 A and B). Written mathematical calculation with his right hand was correct. With his left hand, he wrote mirror-image figures, from right to left, but could calculate fairly accurately (Figure

Figure 2. Left carotid angiogram, oblique view, disclosing a stenotic lesion in the proximal portion of the anterior cerebral artery and localized dilatation in its distal portion.

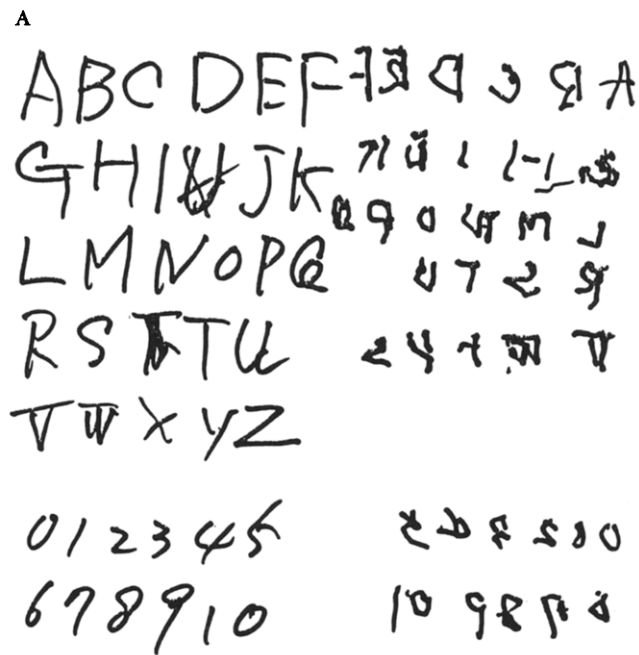
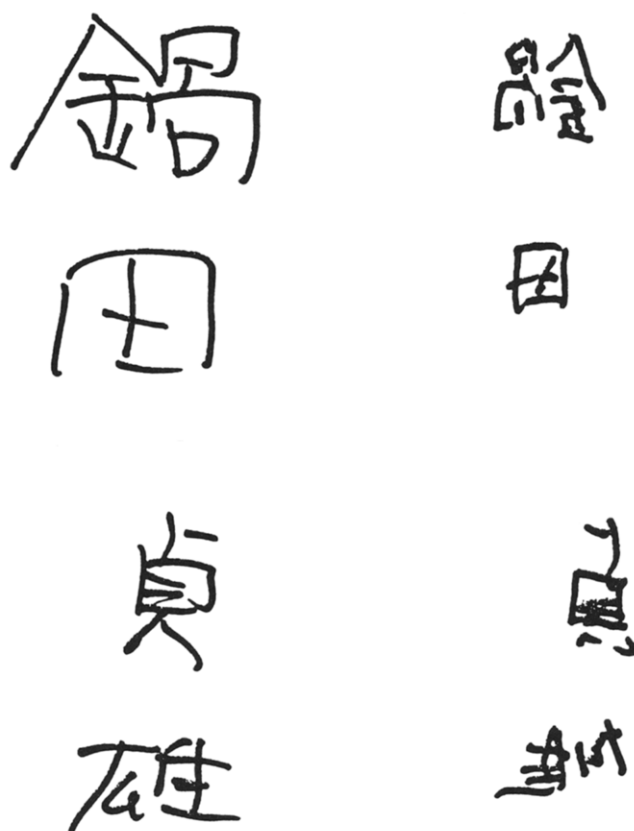


Figure 3. (A) Writing samples of Japanese characters. (Left) Correct writing with the right hand. (Right) Complete mirror writing with the left hand. (B) Samples of English letters and figures. (Left) Correctly written with the right hand. (Right) Mirror written with the left hand.

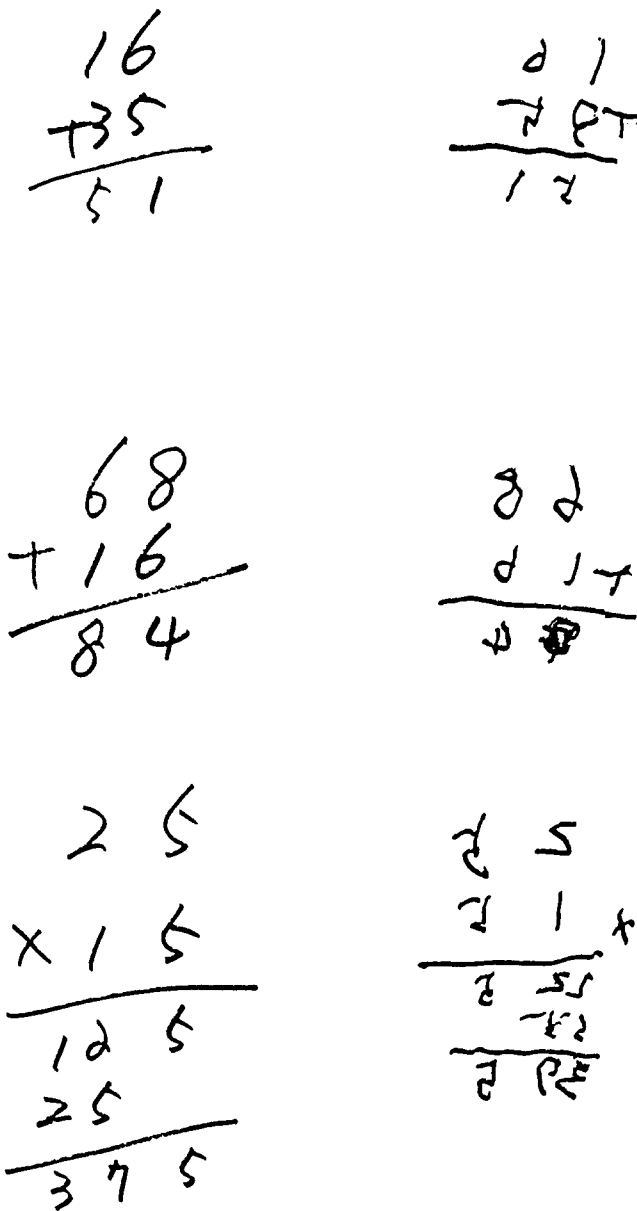


Figure 4. Mathematical calculation. (Left) Correct calculation with the right hand. (Right) Calculation with the left hand. The figures are written right to left in all cases, but most of the answers are correct.

4). Right-left orientation and finger recognition were normal. No signs of tactile agnosia were present in either hand. The abnormal motor behavior of the left hand gradually disappeared over a 6-month period, but mirror writing has persisted for 5 years.

Discussion

The phenomenon that one hand behaves independently of the patient's will or disturbs the motions of the other

hand was first described by Goldstein [15] in 1908. He reported a case of a 57-year-old woman with a resolving left hemiparesis, who complained, "Es muss wohl ein boser Geist in der Hand sein" (There must be a devil in my hand). A similar phenomenon was described by Akelaitis [1] in patients who received commissurotomy for the control of intractable seizures. He named this phenomenon "diagonistic dyspraxia." According to his description, the left hands of the patients disturbed the motions of the right hands, preventing the smooth performance of cooperative movements using two hands. The same symptoms were observed by Brion and Jedynak [5] in patients with tumors of the corpus callosum, and they named this sign "Le signe de la main étrangere." In the English literature, Wilson et al [23] reported a similar sign and translated the French words "la main étrangere" into "the stranger's hand." In recent manuscripts, "alien hand sign," as translated by Goldberg et al [14], has commonly been used. Although the clinical manifestations of the left hand described in the present paper resemble those of Akelaitis's patients, the phenomena described by Goldstein [15], Akelaitis [1], Brion and Jedynak [5], and other recent authors [4,14,18] seem to belong to the same condition.

Three locations of the brain have been postulated as the responsible sites for this phenomenon. The corpus callosum is thought to play an important role in this phenomenon [4,5,11-13,23]. On the contrary, Goldberg et al stated that this peculiar phenomenon may be related to damage to the supplementary motor area, its connecting fibers, or associated structures located on the medial frontal surface of the hemisphere contralateral to its involved extremity. This opinion is in agreement with Brown [6] and McNabb et al [18]. Banks et al [2] postulated that the phenomenon was due to the combination of a partial callosotomy and mesial frontal lesions.

In reviewing the literature, Leiguarda et al [17] classified "the sign of alien hand" into two types. In the patients with the callosal type, the alien hand sign appears in the hand ipsilateral to the language and motor-dominant hemisphere, whereas in the hemisphere type, the sign develops in the hand contralateral to the lesion, regardless of the pattern of cerebral dominance. In the present patient, computed tomography scan and magnetic resonance imaging both clearly indicated infarction in the left hemisphere, particularly in the medial superior frontal gyrus, cingulate gyrus, subcallosal region and gyrus rectus, and corpus callosum. Our present case confirmed the opinion of Banks et al [2] that combined lesions of the corpus callosum and mesial frontal lobe play an important role in the alien hand sign.

In the present case, not only the above-mentioned

phenomena of alien hand but also mirror writing by the left hand was observed. This mirror writing with the left hand was noticed on both dictation and copying. During mathematical calculation, mirror-image figures were written with correct answers. The earliest report of mirror writing, as mentioned by Critchley [9], is that of a left-handed epileptic girl, described by Lentilus in 1698. As to the mechanisms of mirror writing, several theories have been advocated [3,7,9,16,19-22]. Because mirror writing could be learned by daily practice, some authors gave no pathological meaning to this phenomenon [9]. On the contrary, several authors considered it a kind of *agraphia* or *apraxia* [19]. Right-left disturbance and visuospatial disturbance have also been suggested as important in the pathogenesis of mirror writing. Green [16] considered mirror movements as a release phenomenon, and Chaco and Blank [7], in an electromyographic study, showed that muscle contractions equivalent to overt mirror movements are a frequent finding in hemiplegia. A nonorganic psychological disturbance is also postulated as an essential factor. For the location of the lesions, the parietooccipital convexity, the frontotemporal area, and the combined region of the left hemisphere and corpus callosum have been suggested as responsible for mirror writing [20-22]. On the contrary, some authors have denied that there is a specific localized center for mirror writing [3].

In the present patient, although he was diagnosed as alcoholic delirium during his first admission at another hospital, no such delirium was noticed during his hospitalization in our clinic and no psychological symptoms were noticed. According to these findings, the mirror writing of the present patient did not seem to be due to a psychological disturbance. The neuroradiological findings showed a combined lesion of the corpus callosum and mesial frontal lobe. In other regions neither atrophic changes nor low-density areas were noticed. The mirror writing of the present case also seemed due to this combined lesion of the corpus callosum and medial frontal lobe.

In conclusion, the present case showed an extremely rare combination of alien hand sign and mirror writing after anterior cerebral artery infarction. These signs were probably due to a combined lesion of the corpus callosum and left frontal mesial lobe.

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