Hypergraphia: a right hemisphere syndrome

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Summary A new right hemisphere symptom is reported. Five stroke patients with lesions in the perisylvian cortico-subcortical or thalamic region of the right hemisphere produced linguistically correct but semantically loose writing. The behaviour was initiated by subtle prompting and continued semiautomatically. A possible mechanism underlying this hypergraphia is discussed.

Damage to the right cerebral hemisphere has been known to produce a wide range of behavioural symptoms which can tentatively be classified into five groups. The first group includes visuospatial dysfunctions such as constructional impairment or topographical disorders. The second is related to disturbances of the so-called body schema like anosognosia or asomatognosia. The third group is composed of disorders related to attentional mechanisms such as unilateral spatial neglect, motor impersistence or even confusional states. The fourth includes changes of emotion such as aprosodia or indifference to the environment. Yet other symptoms comprise the fifth group which is characterised by positive motor signs such as conjugate eye deviation towards the right,1 or right ipsilateral instinctive grasp reactions.2

We now describe a hitherto unreported behavioural abnormality which appears to belong to the last group. It was observed in five cases who had suffered from right hemisphere stroke. It consists of inappropriate writing behaviour which may most appropriately be termed as hypergraphia, although the term has already been employed for a different symptom seen in temporal lobe epilepsy.3 The symptom is transient, subtle and can easily be overlooked.

Case Reports

Case TN This right handed 50-year-old woman developed sudden left sided weakness on 18 November 1983 and was admitted to the Neurology Service of Hyogo Brain and Heart Center at Himeji (HBHC) immediately. Neurological examination revealed conjugated gaze deviation to the right, left hemiparesis, the leg being more involved than the face and arm, and sensory deficit on the left side. Deep tendon reflexes were exaggerated on the left. Plantar reflex was extensor on the left.

Neuropsychological examination on the third hospital day revealed that the patient was attentive but irritable. Orientation to time and place was preserved. Left unilateral spatial neglect was evident on a line bisection test. Anosodiaphoria for the left hemiparesis was present. Bilateral simultaneous auditory stimuli were received only from the right. Aphasia, apraxia or agnosia was not present. There was no particular change in the amount and quality of her spontaneous speech. Mini-Mental State Test4 which was translated and slightly revised for Japanese patients was administered on the same day yielding a score of 27 out of a total possible score of 30.

On the 12th hospital day when she had been talking with a nurse who had a pencil and a writing pad, she asked for them and started scribbling. On the first sheet of paper she wrote, “It is fine since morning. Twenty-ninth, October. A doctor quoted a haiku for me the other day. (The haiku was fully quoted but was omitted in this translation.) But I wonder if a baby can understand the meaning. I wanted to raise my child with joy.” These were written without hesitation and in a careless manner. The first sentence and the date was written in the right upper half of the sheet starting from the middle and proceeding laterally to the right corner then going down vertically along the right margin of the sheet. The second and third sentences started again from the top middle but this time going down vertically and each vertical line proceeded from right to left until the left margin was reached. Then she turned the sheet 90° to the left and started writing from the left upper corner just below the first sentence. This time the sentence was written laterally, shifting the head of the line slightly to the right each time the line was changed. (Note: In Japanese writing, lateral rightward writing and vertical downward writing are in current use. Generally speaking, the former style is employed for scientific matters and the latter is employed for literary purposes. In handwriting the style depends on personal liking.) Thus spatial arrangement of sentences became chaotic. Letter forms were clumsy and letter sizes were irregular. Some letters were so clumsily written that they could not be identified on later
reading. But there were no omissions or substitutions of letters. Grammar was also without problem, although the meaning is loose and difficult to grasp. She continued writing in a similar fashion consuming two more sheets (fig 1).

But the behaviour was never observed again even with prompting. A tendency for inappropriate use of any other objects was not observed.

A CT scan of the brain revealed a low density area in the white matter of the fronto-parietal lobe and the basal ganglia of the right hemisphere. A right carotid arteriogram revealed an occlusion at the M2 portion of the right middle cerebral artery.

Case CA This right handed 54-year-old woman developed left sided weakness on 5 January, 1984 and was admitted to Neurosurgery Service of HBHC next day. A CT scan of the brain confirmed a haematoma in and around the right internal capsule and lenticular nucleus. Blood was present in the lateral ventricle. The haematoma was removed by immediate operation.

Neurological status after her general condition had stabilised included left hemiparesis, left hemi-sensory deficit, left homonymous hemianopia and the left Babinski sign. When a detailed neuropsychological examination was performed on the 25th postoperative day, she had poor attention span (digit span of four forward), left unilateral spatial neglect and motor impersistence. Aphasia, apraxia or visual agnosia was not observed. Amount and quality of her spontaneous speech was normal.

A hypergraphic tendency was observed for four days from the 22nd to the 25th postoperative days. For instance, one day when the patient was asked to write down her address, she wrote it correctly. But instead of stopping there, she took another sheet of paper and continued writing sentences such as, “My disease is called cerebral palsy. Pain is so severe every day that I feel like crazy. My doctor’s name is so and so (actual name was written). I wish I could be better soon.” These sentences were written without hesitation, not paying much attention to the people present. They were written in vertical direction proceeding first from right to left, then halfway, they were started from left to right. The result was the overlapping of the final sentences in the middle. Lines were directed to the right lower corner obliquely. Slight neglect of the left portion of the sheet was apparent. Even two unidentifiable figures were drawn. When additional papers were provided, she continued writing a series of names of doctors, nurses and physical therapists whom she knew, using three sheets. This writing behaviour was spontaneous and no suggestions were given while she was writing. We did not observe inappropriate use of other objects.

A CT scan obtained on the 32nd hospital day revealed a low density area in the fronto-parieto-temporal lobes and the basal ganglia of the right hemisphere.

Case TK This 64-year-old right handed man had been treated for longstanding mitral valve stenosis and atrial fibrillation. On 25 November 1983, he developed left hemiparesis and dysarthria and was admitted to Neurology Service of HBHC immediately. On neurological examination he was awake and conscious but had left unilateral spatial neglect, left auditory extinction and anosognosia. He had left hemiparesis, left hemisensory deficit, left sided hyperreflexia and the left Babinski sign. Left facial and lingual weakness as well as dysarthria were also present. Aphasia, apraxia or visual agnosia was not observed. Amount and quality of his spontaneous speech was normal. Mini-Mental State Test performed on the fifth hospital day showed a score of 26.

On the fifth hospital day when one of us (YM) asked how he was doing with his left hand, he picked up a pencil nearby and wrote: “Do not suggest. You are unfair. I don’t care if you know you are fair or not. You should use your suggestions for a better purpose.” He did not finish writing until three more sheets were used. Again these were written without hesitation and only the right space of the sheet was used.

On the 10th hospital day when he was given a pencil and asked to write his address, he ignored the command and wrote how he had fallen ill, using three sheets of paper. On the 13th hospital day when he was asked to draw pictures of a rat and a rabbit, he correctly performed these tasks but
added names of these animals on top of the drawing. No inappropriate use of other objects was observed.

A CT scan of the brain revealed a low density area in the perisylvian cortico-subcortical area and in the lenticular nucleus of the right hemisphere.

Case AK This right handed 70-year-old man developed transient left sided weakness on November 16th, 1983. Three days later the left sided weakness recurred and persisted. He was immediately admitted to Neurology Service of HBHC. On neurological examination he showed left homonymous hemianopia, left hemiparesis including the face and left hemisensory deficit. Strong stimulation on the left side was felt on the right (alaesthesia). Anosognosia for the left hemiparesis was present. The left sided weakness rapidly ameliorated and ataxia on the same side became prominent over the next three days.

Neuropsychological examination performed on the 13th hospital day revealed constructional apraxia only. Anosognosia had disappeared. Left unilateral spatial neglect, aphasia, apraxia or visual agnosia was neither detected. No abnormality was noted in his spontaneous speech. Mini-Mental State Test performed on the same day gave a score of 29.

From the 12th to 22nd hospital day, a hypergraphic tendency was clearly present. The patient would start scribbling when a pencil and a sheet of paper was present around him. The content was loose in meaning and difficult to grasp. But grammatically there was nothing wrong in it. Sentences were written laterally with irregular alignment. Letter forms were clumsy and letter sizes irregular. But no paraphasic errors were present. Even an illustration was added to one of the writings (fig 2). Inappropriate use of other utensils was not observed.

A CT scan of the brain revealed a low density area in the centrum semiovale of the right hemisphere. Arteriography revealed an occlusion at M1 portion of the right middle cerebral artery.

Case KH This right handed 65-year-old man developed dysarthria and left sided weakness on 6 February, 1984 and was admitted to Neurology Service of HBHC immediately. On admission he was lethargic and was confused especially deviated to the right. Left hemiparesis including the face, left hemisensory deficit and left Babinski sign were also noted. An immediate CT scan of the brain demonstrated right capsular haemorrhage involving the subthalamus, thalamus and lenticular nucleus. We decided to treat him conservatively. By the third hospital day he became alert and left homonymous hemianopsia, left unilateral spatial neglect and left auditory extinction became manifest. No aphasia, apraxia or visual agnosia was present. Amount and quality of his spontaneous speech was normal.

On the third hospital day when he was asked to write his name, he neglected the command and wrote the following instead: “I have a question to Doctor. Why did it bleed? Pressure was max 150, mi 90.” in vertical fashion in the middle of the paper. Then added several sentences in the lower third of the sheet. Again the spatial arrangement was irregular and the letters were clumsy. Overlapping of letters was also seen. It seemed that he had no intention of communicating with the doctor through this writing. Mini-Mental State Test performed on the next day yielded a score of 25. This hypergraphic tendency was occasionally observed up to the 11th hospital day. No inappropriate use of other objects was noted.

Summary of the cases
Writing behaviour observed in these five patients has several features in common. Of course they were all performed with the unparalysed right hand. First, they started writing spontaneously and once started, they would continue writing without hesitation in semiautomatic fashion not paying much attention to their surroundings. Second, the content of writing was somewhat loose in meaning and had poor value for communication. But no linguistic error was present including grammar and lexical usage. Third, spatial arrangement of sentences was irregular and direction of writing (lateral or vertical) was inconsistent. Letter forms were clumsy and letter sizes were irregular. There was frequent overlapping of letters and sentences. Fourth, conditions for the appearance of this phenomenon were not constant. It could be prompted by a sight of a pencil and a writing pad (Case TN), or by sitting posture at a desk and a sight of a pencil (Case TK, Case AK) or command to write (Case CA, Case KH). But prompting did not necessarily produce the writing behaviour. On one occasion the patient would write but on another he would not pay any attention. It was also a tran-
Hypergraphia: a right hemisphere syndrome

Fig 3 A composite scheme of brain CT of 5 patients. From top to bottom: Case TN, Case CA, Case TK, Case AK, and Case KH.

scent phenomenon. None of the patients showed the symptom beyond the end of four weeks after the onset of the brain damage. All had left hemiparesis and left hemisensory deficit. But no common neuropsychological sign could be picked up in association with hypergraphia. Unilateral spatial neglect which had been seen in all patients at first was absent in Case AK, at the time when his hypergraphia was demonstrated. These features are summarised in the table.

All had right hemisphere damage and all lesions converged in the area supplied by the right middle cerebral artery, except case KH whose lesion involved the thalamus (fig 3). All scored above 25 with Mini-Mental State Test of Folstein.

Discussion

The writing disturbances seen with right hemisphere damage have been characterised by three features: (1) writing on the right part of the paper; (2) inability to maintain a straight line; (3) iteration of strokes and letters. Some of our patients did have components of these features such as rightward writing and difficulty keeping sentences in straight line. However, these were not necessarily the essential feature in the present cases. For example, Case TN, AK, and KH did not show left unilateral spatial neglect in their writings. And above all, the spontaneous and productive nature of our patients’ writing behaviour has never been described as part of this “spatial” agraphia.

Confusional state often produces writing disorders. Chèdru and Geschwind listed five features of confusional agraphia: (1) motor impairment such as tremor, clumsiness, overlapping of letters and so on; (2) spatial disorders; (3) reluctance to write; (4) syntactical disorders; (5) spelling and other linguistic errors. Part of these features, such as clumsy writing or disturbance of spatial arrangement was present in our patients but other features such as syntactical and other linguistic errors were not. The semi-voluntary productive nature is the extreme opposite of the reluctant attitude toward writing observed in confusional. In addition, all of our patients scored above 25 points out of a possible total of 30 with Mini-Mental State Test. With our control study with more than 150 subjects, we regard 23/24 as a cut-off score for cognitive disturbances. Thus it seems unlikely that confusion in the general sense contributed to the present symptom.

A kind of higher order motor disorder has been described by Lhermitte. This so-called utilisation behaviour is characterised by involuntary use of objects presented in front of a patient. A fountain pen was used by one of their patients (Case 2) for writing. This seems to have a superficial similarity to our hypergraphia. However, in utilisation behaviour, the patient only handled the writing utensil appropriately and did not produce a large number of sentences such as seen in the present cases. Also we have not observed this type of behaviour with other objects. We have paid particular attention to this aspect, ever since we described a phenomenon of “compulsive manipulation of tools” in 1982 in which a patient grasped and used a utensil placed in front of him automatically and could not restrain the action even under a command not to use it. Furthermore, utilisation behaviour as well as compulsive use of tools have been claimed to occur in patients with frontal

Table

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Note: A: Aphasia; P: Left sided hemiparesis, An: Anosognosia or Anosodiaphoria for weakness, HH: Left homonymous hemianopia. USN: Left unilateral spatial neglect, S: Left hemisensory deficit, CA: Constructional impairment, MM: Mini-Mental State Test Score.

*Homonymous hemianopsia was absent, but left visual extinction was present.
lesions. This is not the case in the present series. Our patients' behaviour seems to be driven by an urge for expressing something and not by an urge for mere use of an object.

Under the same name "hypergraphia", Waxman and Geschwind reported unusually detailed and copious writing observed in patients with temporal lobe epilepsy. A later study of this interesting syndrome further suggested its possible association with a non-dominant (right) temporal lesion. According to Waxman and Geschwind, the behaviour is probably based on a striking preoccupation with detail, compelling the patient to compulsive writing. Thus they write with extreme attention and care producing almost error free writing. This behaviour is attributed by the authors as a reflection of a deepening of emotional response in the presence of relatively preserved intellectual function. The behaviour persists over years. In contrast our patients' writing was half-voluntary and poorly carried out. It was also transient. Further, none of our patients had a history of temporal lobe seizures. In view of this clear difference except perhaps laterality, a new term should be coined for the present symptom, but lack of an appropriate name compelled us to employ the same term for a different phenomenon.

Thus hypergraphia observed in the present report seems to represent a new phenomenon not described before. It is partly related to spatial disturbance seen in right brain damage and is partly related to attentional derangement seen in acute confusional state. But neither can fully explain the behaviour. It is also important to note that the symptom in not a negative sign in the usual sense.

Location of all the lesions in the present patients was in the perisylvian cortico-subcortical area and the thalamus of the right hemisphere. The area corresponds to the aphasia producing region of the opposite left hemisphere. One tentative hypothesis is that sudden damage in the right hemisphere causes a change in the inter-hemispheric equilibrium. The language-related activity in the left hemisphere may thus become disinhibited. Under these unstable conditions, a subtle prompting such as a sight of a pencil and a writing pad, or sitting in a position with a view of a pencil, or command to write may induce a disinhibited function, that is writing. A question may be raised why concurrent overactivity of the speech does not happen. We do not have a ready answer for this but it may subtly depend on the way facilitation occurs. In this particular series, writing utensils or assumption of an attitude for writing seemed to have played a key role. Although we did not observe any substantial change in the amount of spontaneous speech in the present series, a tendency toward volatility does happen in some patients with right hemisphere damage. This aspect needs further investigation.

In conclusion, we want to emphasise that clinicians should be alert not only to negative signs but also to these soft and evanescent positive signs which will be of value in understanding the dynamic organisation of the higher nervous function.

References


