A clinical study of hypergraphia in epilepsy

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Abstract

Fifteen patients with epilepsy and hypergraphia were compared with 32 patients with epilepsy but without hypergraphia. The number of previous psychiatric episodes, the number of Washington Psychosocial Seizure Inventory (WPSI) items indicating emotional maladjustment, and the number of CT scan abnormalities were significantly greater in the hypergraphic patients than in the non-hypergraphic patients. Cognitive performance, EEG laterality and the scores of WPSI items related to the psychological stress of seizures did not differ significantly between the two groups. Hypergraphia reflects changes in emotional responsiveness secondary to organic temporal lobe lesions.

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Hypergraphia is an extensive and compulsive writing tendency sometimes coupled with hyposexuality and hyperreligiosity. This syndrome was first described by Waxman and Geschwind[1] in 1974 as characteristic behaviour seen during an interictal phase in patients with temporal lobe epilepsy (TLE). Several hypotheses have been proposed to explain the mechanism of hypergraphia. Waxman and Geschwind[1] suggested that hypergraphia represents a deepening of emotional responses in a TLE patient with intact intelligence. Bear[2-3] described the sensory-limbic hyperconnection which forms an underlying mechanism of hypergraphia. Blumer and Benson[4] assumed that hypometamorphosis and hyperemotionality are important factors associated with hypergraphia in patients with epilepsy. Hypergraphia is often associated with right sided non-dominant temporal lesions; Roberts *et al*[5] and Trimble[6] therefore suggested that disturbance of the right hemisphere function leads to an alteration of the writing process. Hermann *et al*[7] considered hypergraphia to be multifactorial and emphasised the importance of using a standardised interview method for its assessment.

A number of studies have been carried out to clarify the mechanism of hypergraphia,[8-11] but the results are still inconclusive. There are at least three methodological problems: 1) the assessment and conceptualisation of hypergraphia are not standardised;[7] 2) the samples of studies are biased, for example, the patients had psychiatric disorders, the seizures were poorly controlled, and the written samples were not properly evaluated;[12] 3) few studies have dealt with the multiaetiological factors related to hypergraphia.

In our study, we compared the clinical characteristics of patients with and without hypergraphia to clarify its mechanism.

Subjects and methods

Between January 1989 and December 1990, we studied 100 consecutively enrolled ambulatory patients with epilepsy at the epilepsy clinic in Osaka Medical College Hospital. Among these patients, 53 agreed to participate in the study and gave informed consent. All patients met the criteria of International Classification of Epilepsies and Epileptic Syndrome.[13] None of the patients exhibited overt psychiatric symptoms or neurological abnormalities. All patients spoke Japanese as their native tongue.

The 53 patients were interviewed using the clinical interview procedure of Mungas.[7] Hypergraphia was defined as a rating of 3, 4 or 5 and non-hypergraphia as a rating of 0, 1 or 2 on Mungas' score. Two of the authors (Okamura and Fukai) independently interviewed the patients and graded their responses. Moreover, both authors rated the written samples, for example, diaries, novels, poems, records of seizures, or letters. The diagnosis of hypergraphia was made only when the two authors agreed.

The following factors were analysed in the hypergraphic and non-hypergraphic groups: 1) Neurological factors: age of onset, duration of epilepsy, hand preference, history of organic brain disease which could have been aetiologically related to their epilepsy and the total number of seizures. 2) Neuroradiological factors: findings of brain CT examination. 3) Neurophysiological factors: localisations of electrical abnormalities of scalp EEG in all patients and the nature of P300 component of auditory event-related potential in 12 HG patients, and 30 non-HG patients.[14] 4) Psychosocial factors: psychiatric history, scores on the Japanese edition of the WPSI[15 16] and scores on the Japanese version of the Mini-Mental State Test (MMS).[17 18]

Statistical analyses comparing the two groups were carried out using Student's *t* test for comparison of means and Fisher's exact test for frequency data. Scores of WPSI were evaluated with Wilcoxson's rank sum test. All mean values are given with SD in brackets.

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Table 1 Neurological factors

	HG group (N = 15)	n-HG group (N = 32)
Age of onset (years)	17·7 (8·7)	12·2 (6·8)*
Duration of epilepsy (years)	18·4 (12·8)	20·2 (12·4)
Hand preference		
Right/Left	14/1	31/1
History of organic brain disease	6	1**
Number of seizures/year	10·8 (17·6)	6·4 (10·5)
Temporal lobe epilepsy	14	19*
Focal non-temporal lobe epilepsy	1	2
Idiopathic generalised epilepsy	0	11**

HG group = hypergraphic group. n-HG group = non-hypergraphic group.
*P < 0·05; **P < 0·01.
(SD in brackets).

Results

Fifteen patients (7 men and 8 women) who scored 4 or 5 on the rating scale were diagnosed as having hypergraphia (table 1). The mean age was 36·1(13·2). Their mean number of years spent in formal education was 12·2(2·3). Thirty two patients (19 men and 13 women) with a rating of 1 or 2 were graded non-hypergraphic. The mean age of these patients was 32·4(11·5). Their mean number of years spent in formal education was 13·1(2·2). Two patients who scored 3 on the rating scale were excluded, because their written samples were not extensive. Four other patients were also excluded, because the two authors did not agree upon the rating of 2 or 3.

Twenty five of these 47 patients were treated with single anti-epileptic drugs, and the remaining 22 were given 2-4 different anti-epileptic drugs. The mean number of drugs administered was 2·1(0·3) in the hypergraphic group and 1·5(0·4) in the non-hypergraphic group. No patients showed stigmata of intoxication. The serum concentrations of drugs were within or below the therapeutic range in all patients.

Table 1 shows neurological factors. No difference was found between the hypergraphic and non-hypergraphic groups in duration of epilepsy. All patients had less than 2 seizures per week. The age of onset was later in the hypergraphic group than in the non-hypergraphic group (p < 0·05). The number of histories indicating organic brain disease was greater in the hypergraphic group than in the non-hypergraphic group (p < 0·01). There was one patient with left hand preference in

Table 2 Findings of CT scan

	HG group (N = 15)	n-HG group (N = 32)
Abnormal findings	6	1**
Dominant enlargement of the right posterior horn†	1	0
Low density in the left temporo-parietal lobe	1	0
Small low density in the right basal temporal lobe	1	0
Small low density in the bi-temporal lobe	1	0
Enlargement of cavum septi pellucidi, cavum Vergae	2	1

†Table 2 shows a major finding related to the temporal lobe region. The enlargement of bi-lateral ventricles with a small calcified nodule was found in this patient.
HG group = hypergraphic group. n-HG group = non-hypergraphic group.
**P < 0·01.

Table 3 Neurophysiological factors

	HG group (N = 15)	n-HG group (N = 32)
EEG focus		
Right temporal	3	4
Left temporal	3	8
Bi-temporal		
R > L/R < L/R = L	2/3/2	2/5/0
Focal non-temporal	1	2
Diffuse	0	11
Unknown	1*	0
P300†		
Age corrected latency (mean ms)		
Cz	363·3 (27·6)	345·7 (32·8)
Pz	365·0 (26·1)	345·7 (32·0)
Amplitude (mean μv)		
Cz	13·2 (8·2)	14·2 (7·0)
Pz	12·4 (7·2)	16·1 (7·3)

*This patient showed a normal EEG but her ictal symptoms were typical for complex partial seizure.
†P300 was performed in 12 hypergraphic patients and 30 non-hypergraphic patients.
HG group = hypergraphic group.
n-HG group = non-hypergraphic group.
(SD in brackets).

each group. The proportion of TLE patients was greater in the hypergraphic group than in the non-hypergraphic group (p < 0·05). The hypergraphic group had no IGE patients. The hypergraphic patients took more drugs than the non-hypergraphic patients.

Table 2 shows the findings of the CT scan. There were 6 patients with CT abnormalities in the hypergraphic group and one in the non-hypergraphic group. Abnormal CT findings involving the temporal lobe were found in 4 hypergraphic patients. There was one patient who showed CT abnormalities without a history of organic brain syndrome. There was another patient who had normal CT scan but gave a history of organic brain disease.

Table 3 shows neurophysiological factors. In the hypergraphic group, there was one case of left hand preference. No relationship was found between hypergraphia and the laterality of EEG foci. No differences were found in the age-corrected latency and in the amplitude of P300 between the two groups.

Table 4 shows psychosocial factors. The incidence of psychiatric history in the hypergraphic group was higher than that in the non-hypergraphic group (p < 0·01). Three patients with neurotic state, one with heightened sensitivity and two with conversion hysteria, were found in the hypergraphic group.

Table 4 Psychosocial factors

	HG group (N = 15)	n-HG group (N = 32)
Psychiatric history	6	2**
MMS (scores)	28·5 (1·9)	29·2 (1·3)
WPSI (scores)†		
Family background	2·3 (2·0)	1·8 (2·0)
Emotional adjustment	16·3 (6·2)	11·2 (6·0)*
Interpersonal adjustment	10·0 (4·8)	7·0 (4·5)
Vocational adjustment	6·1 (2·5)	3·6 (3·3)**
Financial status	3·4 (1·9)	2·1 (1·8)**
Adjustment to seizure	8·2 (3·6)	6·9 (2·8)
Medicine and medical management	1·9 (1·2)	2·1 (1·5)
Overall psychosocial	26·4 (9·9)	17·0 (9·9)**

† One patient in the HG group and six patients in the n-HG group were excluded, because their validity scales were questionable.
HG group = hypergraphic group.
n-HG group = non-hypergraphic group.
*P < 0·05; ** P < 0·01.
(SD in brackets).

They were treated with anxiolytics and psychotherapy. In the non-hypergraphic group, one patient showed an episodic psychotic state and one patient conversion hysteria. No difference was found between the two groups in the MMS score. In WPSI scores, the hypergraphic group had more difficulties than the non-hypergraphic group on the items of emotional adjustment ($p < 0.05$), vocational adjustment ($p < 0.01$), financial status ($p < 0.01$) and overall psychosocial functioning ($p < 0.01$).

Discussion

Hypergraphia has been diagnosed using various different procedures in previous studies.[1 5] [11 19] Hermann *et al*[7] found a correlation between the result of a study with open-ended stimulus questions and that with the Mungas' procedure. They recommended use of the Mungas procedure to standardise the study method. They found 30% of their patients to be hypergraphic using this procedure. A total of 15% of our patients were diagnosed as hypergraphic. The possible presence of hypergraphia should be considered when examining a patient with epilepsy. Once hypergraphia is suspected, the patient should be examined further to confirm the diagnosis.

Forty seven patients did not consent to participate in the study because of work commitments. The mean age at onset of IGE patients was younger than that of TLE patients. The mean age at onset of the hypergraphic patients was older, because the non-hypergraphic group contained more IGE patients.

The frequency of patients with psychiatric histories was greater in the hypergraphic group than in the non-hypergraphic group. The hypergraphic patients had more difficulties than the non-hypergraphic patients in emotional adjustment as measured by WPSI. These results suggest a close relationship between emotional difficulties and hypergraphia. According to Geschwind,[20] the interictal symptoms are associated with an intermittent spike focus in the temporal lobe, leading to altered responsiveness of the limbic system. This alteration is manifested as deepened emotionality and hypergraphia. Blumer and Benson[4] observed that hyperemotionality in patients with complex partial seizure is expressed as depression, anxiety and heightened sensitivity to the point of paranoid reaction. Our data are consistent with both of these arguments.

The number of patients with histories that suggested organic brain disease was greater in the hypergraphic group than in the non-hypergraphic group. In addition, CT abnormalities were found exclusively in the hypergraphic group. Waxman and Geschwind,[1 21] and Roberts *et al*[5] reported 15 hypergraphic cases which included 6 patients with organic brain syndrome. In our study, 4 of 6 cases with CT abnormalities had temporal lobe lesions. More TLE patients were found in the hypergraphic group than in the non-hypergraphic group. There were no IGE patients in the hypergraphic group. These results imply that hypergraphia is linked with temporal lobe dysfunction. Organic brain lesions may be a pre-existing condition for hypergraphia.

Edeh and Toone[22] reported that various emotional and psychiatric morbidities were related to all types of focal epilepsies and CT abnormalities. Their observation agrees with our finding that CT abnormalities of the hypergraphic patients are related to emotional difficulties.

We found no relationship between hypergraphia and laterality of scalp EEG. A study by Hermann *et al*[7] showed similar results using sphenoidal, nasopharyngeal, and/or subdural EEG. We previously reported that hypergraphic patients having suffered right-sided cerebral vascular accident had lesions in the right cerebral hemisphere, corresponding to the aphasia producing region of the opposite hemisphere.[23 24] A possible cause of hypergraphia in these stroke patients was considered to be a disturbance in the right hemisphere. This hypothesis of interhemispheric imbalance as a cause of hypergraphia may be valid for stroke patients but may not apply to patients with epilepsy.

No differences were found between the two groups in MMS score and P300 characteristics. These results suggest the lack of difference in cognitive function between the two groups. One of our patients explained that she wrote because she was forgetful. Waxman and Geschwind[1] denied memory dysfunction as a possible cause of hypergraphia; nonetheless, this possibility requires further attention.

Could psychological stress be a cause of hypergraphia? The hypergraphic group did show high scores on WPSI items related to seizures, but not significantly higher than the non-hypergraphic group. In agreement with Geschwind,[25] we do not consider the stress of seizure itself as a cause of hypergraphia. Instead, we assume that social stress which is a source of emotional instability is indirectly related to hypergraphia. Our findings that the hypergraphic group showed higher scores than the non-hypergraphic group on the items of social stress in vocational adjustment and financial status support this assumption.

The results of our study would indicate: 1) hypergraphia reflects changes in emotional responsiveness secondary to organic temporal lobe lesions; 2) social stress such as vocational maladjustment or financial difficulties may be indirectly related to hypergraphia; 3) cognitive impairment, EEG laterality and psychological stress due to seizure are not shown to be related to hypergraphia.

We intend to identify whether any of these factors are independently related to hypergraphia in a further study.

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