

Development of akinetic mutism and hyperphagia after left thalamic and right hypothalamic lesions

Masaki Shinoda, Atsushi Tsugu, Shinri Oda, Akihiko Masuko, Tsutomu Yamaguchi, Tsuyoshi Yamaguchi, Ryuichi Tsugane, Osamu Sato

Department of Neurosurgery, Tokai University School of Medicine, Bohsei-dai, Isehara, Kanagawa, 259-11 Japan

Received: 25 February 1992

Abstract. A case of childhood post-traumatic akinetic mutism is presented. The patient showed a hyperphagic condition while recovering from akinetic mutism. He had lesions in the left interlaminar nucleus of the thalamus, right globus pallidus, and right dorsomedial nucleus of the hypothalamus. Laboratory data indicated slightly disturbed hypothalamic functions. In general, akinetic mutism can be seen with bilateral destructive lesions, while hyperphagia may occur after destruction of dorsomedial hypothalamic nucleus, but it is very rare. This is the first reported case of akinetic mutism caused by a unilateral lesion.

Key words: Akinetic mutism – Hyperphagia – Interlaminar nucleus of thalamus – Dorsomedial nucleus of hypothalamus – Childhood head injury

Introduction

Akinetic mutism can be seen in association with destructive lesions in the frontal lobe [12], cingulate gyrus [3], or corpus callosum [3]. Recently, however, more attention has been paid to the interlaminar nucleus of the thalamus with regard to the pathogenesis of akinetic mutism [5, 14]. Particularly bilateral lesions of the nucleus have been reported as responsible for this disorder [5, 11].

On the other hand, hyperphagia has seldom been reported following head injury. On these rare occasions it has been described, the cause of this particular phenomenon has been ascribed to a lesion in the dorsomedial nucleus of the hypothalamus [6]. Hyperphagia has also developed after a specific form of limbic lesion known as Klüver-Bucy syndrome [8].

This report presents the first published case of akinetic mutism caused by a unilateral thalamic lesion.

Case report

A 10-year-old boy was injured in an automobile accident on August 10, 1988, when it took 4 h before he was transferred to our hospital. According to ambulance personnel his consciousness was disturbed when he was rescued. On admission, his consciousness level was 5 on the Glasgow Coma Scale; the left pupil was dilated and did not react to light, while the right one remained normal. Head scanning by computed tomography (CT), revealed a mid-line shift to the right of more than 2 cm due to a left frontoparietotemporal epidural hematoma (Fig. 1, left). The patient was immediately taken to the operating room and extensive right frontoparietotemporal craniectomy with removal of the epidural hematoma performed.

A low-density area in the left thalamus and a tiny low-density area in the right globus pallidus were revealed on CT scan on the following day (Fig. 1, right). These low-density lesions disappeared within 7 days after surgery. Instead, high-intensity areas were visible in the paraventricular area of the III ventricle, the anterior and interlaminar nuclei of the left thalamus, the right globus pallidus, and the frontal white matter on the left in Brodmann's areas 8 and 9 on T2-weighted MRI scans on September 6, 1988 (Fig. 2). A month after the accident, the boy still had right severe hemiparesis, and some motor disturbance was recognizable even in the left extremities. He was barely able to respond to noxious stimuli. A Babinski reflex was elicited on the right side but not on the left. The boy was unable to speak or even vocalize any sound. His ocular movement was limited only horizontally, and a diagnosis of akinetic mutism was made.

Two months after the injury, oral intake became feasible when he just swallowed food without chewing. He remained asleep for the most of the day and it was assumed that he was awake only some 8 h a day. Bilateral diffuse slow-wave activity (4–5 Hz) was the basic pattern on the electroencephalogram (EEG) recorded on February 4, 1989. Low-intensity areas were exhibited on the right dorsomedial hypothalamic nucleus, right globus pallidus, and interlaminar nucleus of the left thalamus in T1-weighted MRI on February 14, 1989 (Fig. 3). The patient still remained mute and incontinent 9 months after the accident; nevertheless, some chewing motion was recognized and motor function in the left upper extremity was in the process of recovery. Eighteen months after the accident, he developed oral tendency and corpophagia. At present, 2 years after the injury, he is still mute hemiparetic on the right side, and suffers behavior disturbance. He can write letters and figures with his left hand correctly. Dementia, antegrade amnesia, and hypersexuality are not present. The present laboratory data show hypopituitary function with slight hyperprolactinemia.

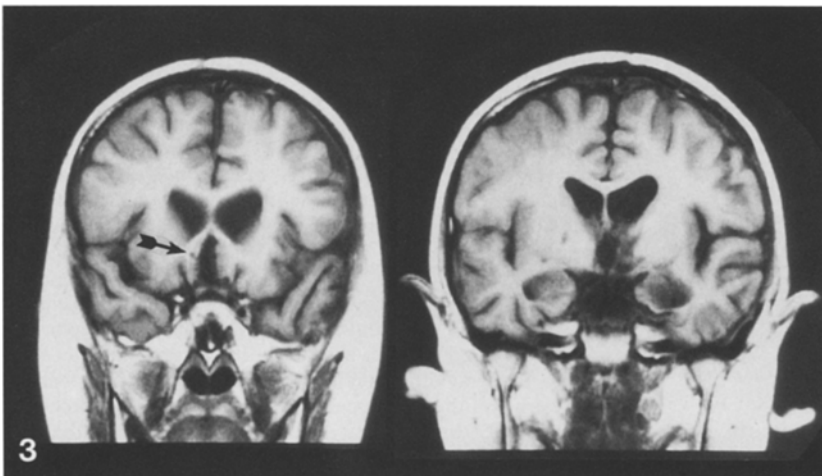
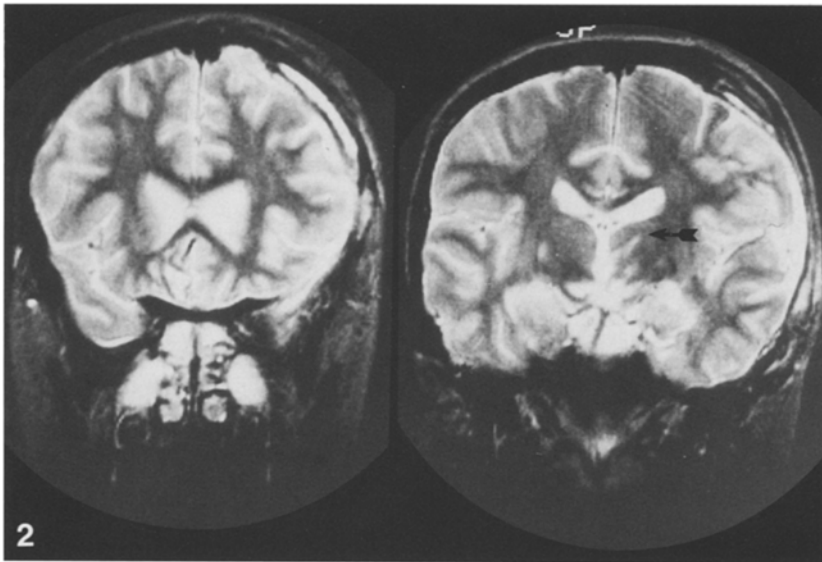
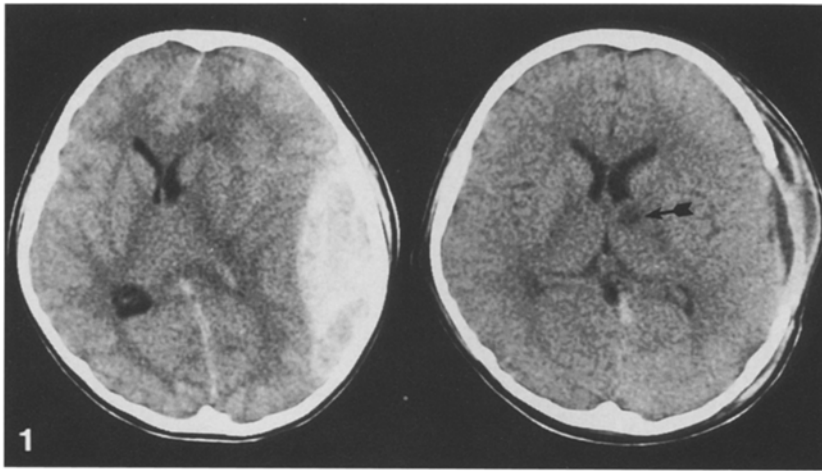


Fig. 1. *Left:* Preoperative CT scan showing epidural hematoma in the left temporoparietal region. *Right:* Postoperative CT scan taken a day after the accident, revealing low-density areas on the anterior portion of the left thalamus (*arrow*) and right globus pallidus

Fig. 2. T2-weighted MRI images taken a month after injury, showing a small contusion on the right side of the III ventricle and a thin left epidural hematoma (*left*). High-intensity areas were demonstrated in white matter beside Brodmann's areas 8 and 9, the right globus pallidus, and the anterior and internal laminal nuclei of the left thalamus (*arrow; right*)

Fig. 3. T1-weighted MRI scans taken 6 months after the accident, showing a small contusional scar of the dorsomedial nucleus of the right hypothalamus (*left; arrow*). Low-intensity areas were demonstrated in the right globus pallidus and interlaminal nucleus of the left thalamus (*right*)

Discussion

A number of lesions have been mentioned in relation to akinetic mutism. Derangements in deep structures of the frontal lobe [12], cingulate gyrus [3], corpus callosum [3], the vicinity of the III ventricle [4, 19], the dorsomedial and centromedial nucleus of the thalamus [5, 14], the

cerebral reticular systems, and the periaqueductal region [7, 9] are known among those. Especially, "hypokinesia" and "mutism" have recently become well-known disadvantageous symptoms after callosotomy [1, 2, 13]. In the developed state of this condition, the sufferer makes no sound and lies inert, except that the eyes regard an observer steadily or follow moving objects, and may be

distracted by sound. Despite the steady gaze, which seems to promise speech, the patient is quite mute. Usually, there are no movements of a voluntary character. Emotional movement also is almost in abeyance. The patient swallows readily, but has to be fed. There is total incontinence of urine and feces.

Our patient became akinetically mute in the process of recovery from ischemic insult to the thalamus after acute epidural hematoma; head CT disclosed a lesion in the anterior portion of the left thalamus, even though only transiently. In addition to this, MRI indicated ischemic changes in the centromedial nucleus and interlaminar nucleus of the left thalamus. Incidentally, these regions are in the perfusional territory of the left posterior thalamo-subthalamic paramedian artery, as pointed out by Percheron [10]. These areas are presumed to be the most cranial portion of the reticular system in which fibers are directed to the thalamus. Since 1970, when Segarra reported so-called "somnolent mutism" caused by bilateral posterior thalamo-subthalamic paramedian artery obstruction [11], a number of cases of akinetic mutism after a thalamic lesion have been reported. In most of the cases there were similar lesions on both sides; only a few authors reported a dominant lesion [3, 5]. Although fiber connections between the centromedial nucleus of the thalamus and the lentiform nucleus have been claimed, no definitive fiber connection between the centromedial thalamus and the globus pallidus has been established. Yamanouchi et al. [14] stated that even in cases of mutism or loss of activity, the left side of the cerebrum is dominant in its function as the left cerebral hemisphere holds dominance in most individuals.

Hyperphagia is well known with lesions of the hypothalamo-pituitary system, and it has been claimed that there are three major cause of obesity which can be elicited after central nervous system disorders: (1) Cushing syndrome, (2) ventromedial hypothalamic and paraventricular nucleus impairment, and (3) disorders of the limbic system. In this reported case, the pathogenesis of the obesity is considered to be hyperphagia developing after dysfunction of the left hypothalamus. A tiny contusion of the left dorsomedial nucleus of the hypothalamus was seen on MRI scan. On analyzing the data, hypopituitary function with slight hyperprolactinemia indicates a decrease of hypothalamic releasing factors, with inhibition of prolactin suppressing the dopaminergic neuron system. No data indicated Cushing syndrome. A patient

with a hypothalamic lesion may often develop a clinical syndrome of life-threatening anorexia or cachexia, but there have been few cases of hyperphagia reported with similar pathology of the hypothalamus [6]. On the other hand, hyperphagia has been often reported in patients with limbic system disorders, and pica and hyperphagia are quite well known in Kluver-Bucy syndrome [8]. This reported case showed oral tendency and corpohagia, and the possibility of some kind of limbic system dysfunction has to be taken into consideration.

References

1. Benes V (1982) Sequelae of transcallosal surgery. *Child's Brain* 9:69–72
2. Benes V (1990) Advantages and disadvantages of the transcallosal approach to the III ventricle. *Child's Nerv Syst* 6:437–439
3. Boetz MI, Lecours AR, Berude L (1983) Speech disturbances related to cingulate lesions, akinetic mutism and mutism without akinesia. In: *Aphasiology*. Ballière Tindall, London, pp 130–132
4. Cairns H, Oldfield RC, Pennybacker JB (1941) Akinetic mutism with an epidermoid cyst of 3rd ventricle. *Brain* 64:273–290
5. Castigne P, Lhermitte F, Buge A (1980) Paramedian thalamic and midbrain infarcts: clinical and neuropathological study. *Ann Neurol* 10:127–148
6. Coffey RJ (1989) Hypothalamic and basal forebrain germinoma presenting with amnesia and hyperphagia. *Surg Neurol* 31:228–233
7. Klee A (1961) Akinetic mutism: review of the literature and report of a case. *J Nerv Ment Dis* 133:526–553
8. Klüver H, Bucy PC (1938) An analysis of certain effect of bilateral temporal lobectomy in the rhesus monkey, with special reference to "Psychic blindness". *J Psychol* 5:33–54
9. Messert B, Henke TK, Langheim W (1966) Syndrome of akinetic mutism associated with obstructive hydrocephalus. *Neurology* 16:635–649
10. Percheron G (1973) The anatomy of arterial supply of the human thalamus and its use for the interpretation of thalamic vascular pathology. *J Neurol* 20:1–13
11. Segarra JM (1970) Cerebral vascular disease and behavior. I. The syndrome of the mesencephalic artery (basilar artery bifurcation). *Arch Neurol* 22:408–418
12. Strub RL, Black FW (1981) *Neurobehavioral disorders: a clinical approach*. Davis, Philadelphia
13. Sussman NM, Gur RC, Gur RE, O'Connor MJ (1983) Mutism as a consequence of callosotomy. *J Neurosurg* 59:514–519
14. Yamanouchi H, Tomonaga M, Toyokura Y (1987) Mutism, lack of initiative and/or dementia in the cerebrovascular thalamic lesion. *Neurol Med (Tokyo)* 27:567–574