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A Case of Aqueduct Stenosis in Adults with Various Neurological and Psychiatric Symptoms

Nobutaka Motohashi, Yoshikazu Ishizuka, Takashi Asada, Koichi Shiraishi, Hitoshi Fukuzawa, and Tetsuhiko Kariya

Department of Neuropsychiatry, Yamanashi Medical College, Tamaho, Yamanashi 409-38, Japan

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Summary. A case of aqueduct stenosis in an adult with several depressive episodes and the clinical features of delusional depression is presented. The patient complained of a strange feeling in the throat. She also had had several falls with loss of consciousness. The relationship between aqueduct stenosis and diencephalic dysfunction is discussed.

Key words: Aqueduct stenosis – Delusional depression – Fall – Diencephalon

Introduction

Aqueduct stenosis in adults is a rare developmental anomaly. Although it was once thought to be psychically asymptomatic (Busch 1940), many cases with various psychiatric manifestations have been reported (Beckett et al. 1950; Petit-Dutaillis et al. 1950; Elvidge 1966; Little et al. 1975). Recently, a relationship between aqueduct stenosis and schizophrenic disorders has been suggested (Reveley and Reveley 1983; Roberts et al. 1983). We now report a case of aqueduct stenosis with various neuropsychiatric symptoms.

Case Report

A 44-year-old housewife, who had been born by normal delivery and in whom hydrocephalus had not been detected, graduated from a junior high school; she then worked at a factory for several years. She was married when aged 24 years. At the age of 26, after delivery of her first child, she became depressed and thought of committing suicide. She received several series of electroconvulsive therapy, which resulted in remission. Soon after the abortion of the third child, when she was 31, she had sleep disturbance, loss of appetite, psychomotor retardation and again considered committing suicide. She complained of a dry, aching throat and a lack of saliva, because she had spat too much. She was admitted to a mental hospital and received fluphenazine and electroconvulsive therapy. She stayed there for a week; again a period of remission followed. In November, 1986, at the age of 42, she was admitted to the neuropsychiatric ward of Yamanashi Medical College Hospital. For several days, she had had sleep disturbance, loss of appetite and depressed mood. She complained of ache in the throat. The day before admission to our hospital, she attempted suicide by jumping from the roof of her house. She suffered a fracture of the spine and was admitted to an orthopaedic hospital. The next morning she attempted suicide again by jumping from the second floor verandah of the hospital. She lost consciousness and was taken to the neurosurgical outpatient department of our hospital. Neurological examination revealed no abnormality. Because she appeared determined to commit suicide, she was admitted to the neuropsychiatric ward. She insisted that she was not crazy and that something was wrong with her throat. She said that her throat dried up because she had spat too much. She wanted to consult an otolaryngologist. She was treated with sulpiride and diazepam. Otolaryngological examination revealed no abnormality. Serological tests for autoimmune diseases were also negative. Although her mental state soon improved, she sometimes giggled and could not keep still in bed. After a month, when her fractures had healed, she firmly wished to be discharged from the hospital, and was discharged. At this time she did not mention her throat of her own accord. Thereafter she attended the neuropsychiatric outpatient department regularly for 2 months. Then she stopped taking medicine. From the end of April 1987, she was depressed and thinking of suicide. She insisted again that she had throatache and attempted suicide by carbon monoxide poisoning. She was admitted to the neuropsychiatric ward. Her consciousness was clear. Although she was mute, she sometimes tried to leave the ward. The electroencephalogram (EEG) was slightly abnormal. The resting record was normal. Following hyperventilation, delta bursts appeared and lasted for 1 min. Brain computed tomography (CT) revealed dilatation of the lateral and third ventricles with the size of the fourth ventricle normal (Fig. 1A–C). No tumour was detected. The cerebrospinal fluid (CSF) was normal. Radioisotope cisternography using [^{111}In]diethyl-triamine-penta-acetic acid showed a slight delay in CSF flow but no ventricular reflux. Single photon emission tomogaphy with *N*-isopropyl-*p*-[^{123}I]iodoamphetamine demonstrated normal cerebral blood flow. Psychological testing showed that she had a Wechsler Adult Intelligence Scale verbal intelligence quotient (IQ) of 67 and a performance IQ of 72. She was treated with sulpiride, levomepromazine and amitriptyline. Her recovery was gradual. She was discharged in October 1987. Thereafter she has attended the outpatient department regularly. Her mood has been kept stable by treatment with maprotiline. From November 1987, she had fallen several times with loss of consciousness. Each of these episodes had lasted for less than 5 min. CT scan showed no remarkable changes. Magnetic resonance im-

Offprint requests to: N. Motohashi

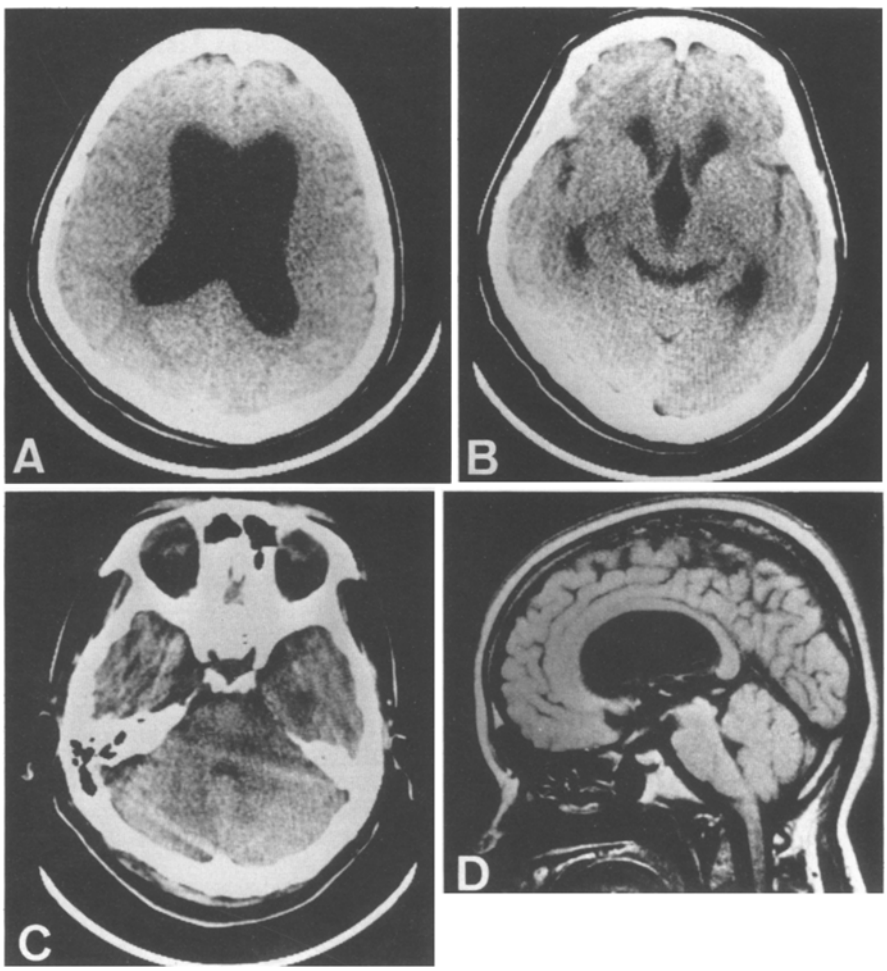

Fig. 1. Computed tomographic scan (**A–C**) and magnetic resonance imaging (**D**) of this patient. Note marked dilatation of the lateral (**A**) and the third (**B**) ventricles and the normal size of the fourth ventricle (**C**). Part of the aqueduct of Sylvius is obscure (**D**)

aging demonstrated dilatation of the lateral and third ventricles. Part of the sylvian aqueduct was obscure (Fig. 1D). Since EEG recording showed diffuse theta bursts and temporal sharp waves, carbamazepine was introduced instead of maprotiline. She has not fallen since then.

Discussion

Although there are many reports of neurological symptoms in aqueduct stenosis in adults (Harrison et al. 1974; Little et al. 1975; McMillan and Williams 1977), few cases with psychiatric manifestations have been described. These include behavioural abnormalities (Beckett et al. 1950; Petit-Dutaillis et al. 1950; Elvidge 1966), character changes (Petit-Dutaillis et al. 1950) and endogenous depression (Little et al. 1975). Recently, an association of aqueduct stenosis and schizophrenias has been described. Reveley and Reveley (1983) reported three schizophrenic patients with aqueduct stenosis. All of them showed auditory hallucination and EEG abnormalities. Five cases of schizophrenic psychosis associated with aqueduct stenosis have also been reported (Roberts et al. 1983). Their prominent symptoms were delusions, hallucinations or thought disorder. Two of them also had depressive episodes.

The present report describes a case of aqueduct stenosis in adults with several depressive episodes. The clinical features of each episode were almost the same. Strange feelings in the throat were repeatedly insisted on. The patient interpreted the cause of the feelings delusionally. The idea of committing suicide was strong enough to lead to several attempts. Moreover, the patient had fallen with loss of consciousness several times. EEG recordings were abnormal. Neurological symptoms are in accordance with previous reports (Harrison et al. 1974; Little et al. 1975; McMillan and Williams 1977).

Are depressive episodes really related to aqueduct stenosis? Roberts and colleagues (1983) suggested that the periodicity of psychotic symptoms might be derived from the valve-like phenomenon which raised pressure in the third ventricle and disturbed diencephalic function. It is interesting in this regard that the association of psychotic symptoms and diencephalic function has long been described (Stoerring 1938). Moreover, depressive states with or without delusion have been reported in patients with third ventricle colloid cysts (Burkle and Lipowski 1978; Lobosky et al. 1984; Upadhyaya and Sud 1988). Huber (1957) proposed the fourth category of schizophrenia, *coenaesthetische Schizophrenie*, which was characterized by coenaesthetic, delusional and depressive symptoms with vegetative, motor and sensory

disturbances. Enlargement of the third and lateral ventricles was observed by pneumoencephalography in most of the patients. He suggested a causal relationship between this disorder and diencephalic dysfunction. Although he did not comment on aqueduct stenosis, some of his patients might have had a narrowed aqueduct of Sylvius. Our patient's coenaesthetic delusion, which had occurred repeatedly, may have been associated with diencephalic dysfunction.

As suggested by the present case report, adult patients with aqueduct stenosis may show various neuropsychiatric manifestations. Careful follow-up is necessary when confronted with such cases.

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