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Case Reports & Case Series

Cognitive outcome in a patient with poor grade aneurysmal subarachnoid hemorrhage: Focus on aphasia

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A B S T R A C T

Background: Recently, better outcomes have been reported in patients with poor grade subarachnoid hemorrhage (SAH) after rupture of a cerebral aneurysm, such as improvement of complications and recovery of cognitive function, but there have been no reports on the recovery of language in patients with aphasia after aneurysmal SAH.

Case presentation: A male patient developed grade V SAH (Hunt and Kosnik classification) due to a ruptured cerebral aneurysm, but was able to return to work after 16 months. This patient is reported with a focus on recovery from aphasia.

Conclusion: In patients with poor grade SAH on admission, both medical/surgical treatment and cognitive rehabilitation should be performed, and individualized decision-making is necessary.

1. Introduction

On admission to hospital with subarachnoid hemorrhage (SAH), its grade is determined according to the presence or absence of complications such as loss of consciousness, headache, and neurological signs. The grade of SAH not only influences decisions about appropriate treatment, but is also a determinant of the functional outcome [1,2]. The Hunt and Hess classification of SAH [3], the Hunt and Kosnik grade [4], and the World Federation of Neurological Surgeon (WFNS) scale for clinical assessment of SAH [5] are methods for grading the clinical status of patients with SAH that are utilized internationally (Table 1). In addition, the Fisher Grading Scale [6] is known to predict the risk of cerebral vasospasm after SAH based on the amount of blood in the subarachnoid space on the initial computed tomography (CT) scan (Table 2).

The Japanese Guideline for the Management of Stroke (2015 revision) [7] provides recommendations for treating stroke patients based on a 5-grade classification of SAH (Table 3). According to the Guideline, treatment to prevent rebleeding is recommended within 72 h after the onset of SAH if it is mild (grades I–III) and there are no other complications (grade B recommendation). In patients with grade IV SAH, treatment can be considered depending on the patient's age and the location of the aneurysm (grade C1 recommendation). On the other hand, in patients with the most severe SAH (grade V), treatment might be considered if recovery is expected, but there is insufficient evidence to recommend active measures (grade C1 recommendation).

In spite of these negative recommendations, better outcomes have been reported in some patients with poor grade SAH secondary to a ruptured aneurysm, such as improvement of complications and recovery of cognitive function [8–12]. However, to the best of our knowledge, there have been no reports that focused on the recovery of language in patients with aphasia and grade V SAH.

Accordingly, we investigated the time course of language recovery in a patient with aphasia due to poor grade SAH caused by a ruptured cerebral aneurysm. The patient was eventually able to return to work.

This study was approved by the ethics committee of Chiba Emergency Medical Center.

Abbreviations: CTA, computed tomography angiography; FAB, Frontal Assessment Battery; GCS, Glasgow Coma Scale; H & K, Hunt and Kosnik; HRQOL, health-related quality of life; IQ, intelligence quotient; LP shunt, lumboperitoneal shunt; mRS, modified Rankin Scale; NPH, normal pressure hydrocephalus; RCPM, Raven's Coloured Progressive Matrices; SAH, subarachnoid hemorrhage; SLTA, Standard Language Test of Aphasia; TMT, Trail Making Test; VP shunt, ventriculoperitoneal shunt; WAIS III, Wechler Adult Intelligence Scale – III; WFNS, World Federation of Neurological Surgeon

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Table 1
Clinical grading scales for aneurysmal SAH.

Grade	Hunt and Hess [3]	Hunt and Kosnik [4]	WFNS [5]
0		Unruptured aneurysms	
1	Asymptomatic, or minimal headache and slight nuchal rigidity.	Asymptomatic or minimal headache, slight nuchal rigidity	GCS 15, no motor deficit
1a		No acute meningeal reaction but with fixed neurological deficit	
2	Moderate to severe headache, nuchal rigidity, no neurological deficit other than cranial nerve palsy.	Moderate to severe headache nuchal rigidity, no neurological deficit other than cranial nerve palsy	GCS 13 to 14, no motor deficit
3	Drowsiness, confusion, or mild focal deficit.	Drowsiness, confusion or mild focal deficit	GCS 13 to 14 with motor deficit
4	Stupor, moderate to severe hemiparesis, possibly early decerebrate rigidity and vegetative disturbances.	Stupor, moderate to severe hemiparesis, possibly early decerebrate rigidity and vegetative disturbances	GCS 7 to 12, with or without motor deficit
5	Deep coma, decerebrate rigidity, moribund appearance.	Deep coma, decerebrate rigidity, moribund appearance	GCS 3 to 6, with or without motor deficit

SAH, subarachnoid hemorrhage.
GCS, Grasgow Coma Scale.

Table 2
The Fisher scales for grading SAH on admission CT scan [6].

Grade	CT scan
1	No blood visualized
2	A diffuse deposition or thin layer with all vertical layers of blood (interhemispheric fissure, insular cistern, ambient cistern) < 1 mm thick
3	Localized clots and/or vertical layers of blood 1 mm or greater in thickness
4	Diffuse or no subarachoid blood, but with intracerebral or intraventicular clots

Table 3
Classification of grades of recommendation.
(The Japanese Guideline for the Management of Stroke [7]).

Grade of recommendations	Type of recommendations
A	Strongly recommended
B	Recommended
C1	Although might be considered, no sufficient evidence
C2	Not recommended because of lack of scientific evidence
D	Recommended not to do

Translated from Japanese.

2. Case presentation

2.1. Present Illness

The patient was a left-handed male university graduate working at a stockbroking company. He was on antihypertensive therapy with Telmisartan for hypertension. At the age of 52, he suddenly developed stertorous breathing and incontinence, and was brought to our hospital by ambulance. On arrival, the level of consciousness was E1V1M2 according to the Glasgow Coma Scale (GCS). His pupils showed downward deviation without anisocoria, and left hemiparesis was observed. A diagnosis of grade V SAH (H & K classification) was made. Head CT showed diffuse SAH (Fig. 1), and computed tomography angiography (CTA) demonstrated a ruptured aneurysm on the right middle cerebral artery (Fig. 2). After 1 h, his GCS improved to E3V1M4, deviation of the pupils gradually normalized, and the H & K classification also improved to grade III. He underwent clipping of the aneurysm within 9 h of the onset. Cranioplasty was performed at 1 month after the onset. At 1 month and 20 days after the onset, a left ventriculoperitoneal (VP) shunt was placed as a treatment for normal pressure hydrocephalus (NPH). The VP shunt was subsequently removed due to postoperative infection, and a right lumboperitoneal (LP) shunt was placed instead at 4 months after the onset.

3. Clinical course (up to 4 months)

3.1. Mental state and cognitive function

Disturbance of consciousness was prolonged, but he gradually became alert after insertion of the LP shunt at 4 months after the occurrence of SAH. However, in addition to fatigue, he had persistent cognitive dysfunction with inattention, incontinence of speech, and a euphoric tendency. Neuropsychological assessment using Reven's Coloured Progressive Matrices (RCPM) [13] (maximum score: 36) showed an increase from a score of 19 at 2 months after SAH to a score of 30 (which is in the normal range) at 4 months. His intelligence quotient (IQ) measured by Kohs Block Design Test increased from 77.6 at 2 months after SAH to 79.7 at 4 months. When the Trail Making Test (TMT) was used to assess attention, speed, and mental flexibility, his practice time for Part A was 153 s (cutoff < 180 s) [14], but he could not complete part B which is more complex than part A and requires sequential alternation of attention.

3.2. Language

He showed fluent aphasia with lexical/semantic disorders (transcortical sensory aphasia). Although, he could only repeat meaningless sounds (“ho ho”) in the acute stage, his speech gradually improved. The following samples of the patient's speech were translated from Japanese. At 1 month after occurrence of SAH, he made spontaneous statements such as “What can I say…”, “Me? Am I like that?” At 1.5 months after SAH, he was able to talk about rehabilitation (“I understand I must do, but…”) and about visits from his wife (“She actually comes”). However, his conversation was one-sided and the contents were sparse compared to the volume of speech, which not only included verbal paraphasia, e.g., “shamisen (Japanese string-instrument)” for shinbun (newspaper), but also phonological paraphasia, e.g., “kora” for koma (spinning top). When the examiner asked him to repeat what he had just said, he often made no answer or did not pay attention to the questions. Although his ability to repeat words was well preserved, confrontation naming, oral reading, and writing all showed severe impairment.

The Standard Language Test of Aphasia (SLTA) [15] is one of the most widely administered aphasia tests in Japan, so it was used to

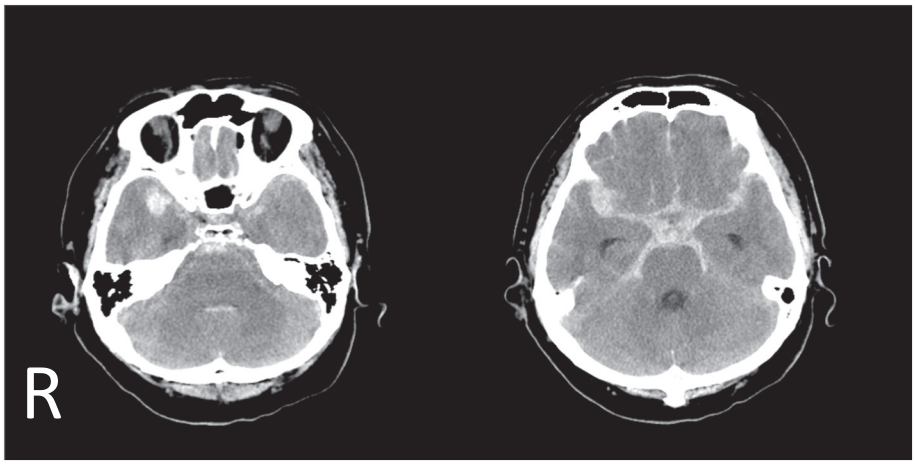

Fig. 1. CT scans obtained on admission show diffuse SAH.

assess the language function of this patient. The SLTA covers five major areas, including auditory comprehension, speech, visual comprehension, writing and arithmetic, with the tests for each area incorporating several subtests so that there are a total of 26 subtests. The total score of this test is classified into 10 grades (grade 1 is the worst performance and grade 10 in the best) [16]. The patient's total SLTA score improved from grade 2 at 2 months after SAH to grade 6 at 4 months. In subtest 8 of the SLTA ("Explanation of a comic strip"), patients are required to orally explain the story of a comic strip in which a man is walking with a stick and his hat is suddenly blown off by the wind. He chases his hat and finally catches it using the grip of his stick (Fig. 3). At 2 months after SAH, the patient's explanation was as follows:

> "Impossible, impossible? It's impossible…yeah, thirty years old, how this changes, it's really tough…"

At 4 months after SAH, the explanation improved to:

> "Once he is walking. When he is walking, the wind…here, marching, he is walking. When he is walking, the wind and, the wind and, and what is this?, hat? To hat, being blowed. Then, next, picking up the hat, at the next point, the thing picked up, went into the car."

At 5 months after the occurrence of SAH, he was transferred to a rehabilitation hospital. His activities of daily living (ADL) improved to the level of independence at 10 months after SAH and he was discharged. After that, he was followed-up at our hospital to receive ongoing treatment for residual cognitive dysfunction and support for returning to work.

4. Progress during outpatient follow-up (10–16 months after SAH)

4.1. Mental state and cognitive function

At 12 months after occurrence of SAH, his verbal intelligence quotient (IQ) was 97, performance IQ was 97, and full IQ was 96 according to the Wechsler Adult Intelligence Scale - III (WAIS III)test [17] (Japanese translation). At 14 months after SAH, his maximum scores for the mini-mental state examination (MMSE) [18] (Japanese translation) and the Frontal Assessment Battery (FAB) [19] (Japanese translation) were 30 and 18, respectively. Although he still showed slight inattention, incontinence of speech, and a euphoric tendency, these neuropsychological assessments suggested that his cognitive state was almost normal.

4.2. Language

At this stage, his total SLTA score reached the maximum of grade 10. There was marked improvement of both auditory and visual comprehension, and he had no difficulty in daily conversation. Language production also showed further improvement, but difficulty finding words and redundancy of speech still persisted. His performance in the SLTA subtest 8 at 10 months after the occurrence of SAH was as follows:

> "Well… a man, umm… with a hat, with a stick, was walking. When walking, since the wind blew, the hat he wore flew away. The man was surprised. The… the man walked to the hat to get back the hat which flew away. Well…the place was, like a quay by the water. The walking man…the hat… (Oh, my mistake), the hat went into the sea. And he tried to pick up the hat which went into the sea using the top of the stick, tried to pick up, using the top of the stick, he

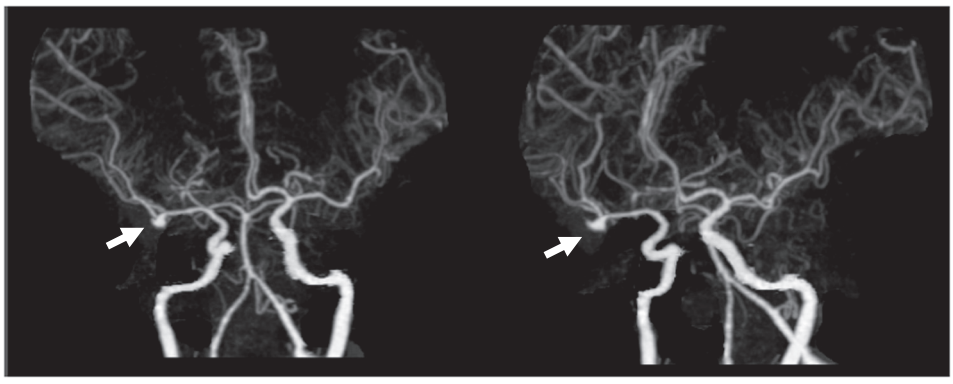

Fig. 2. CTA performed on admission suggests a raptured aneurysm on the right middle cerebral artery (arrowheads).

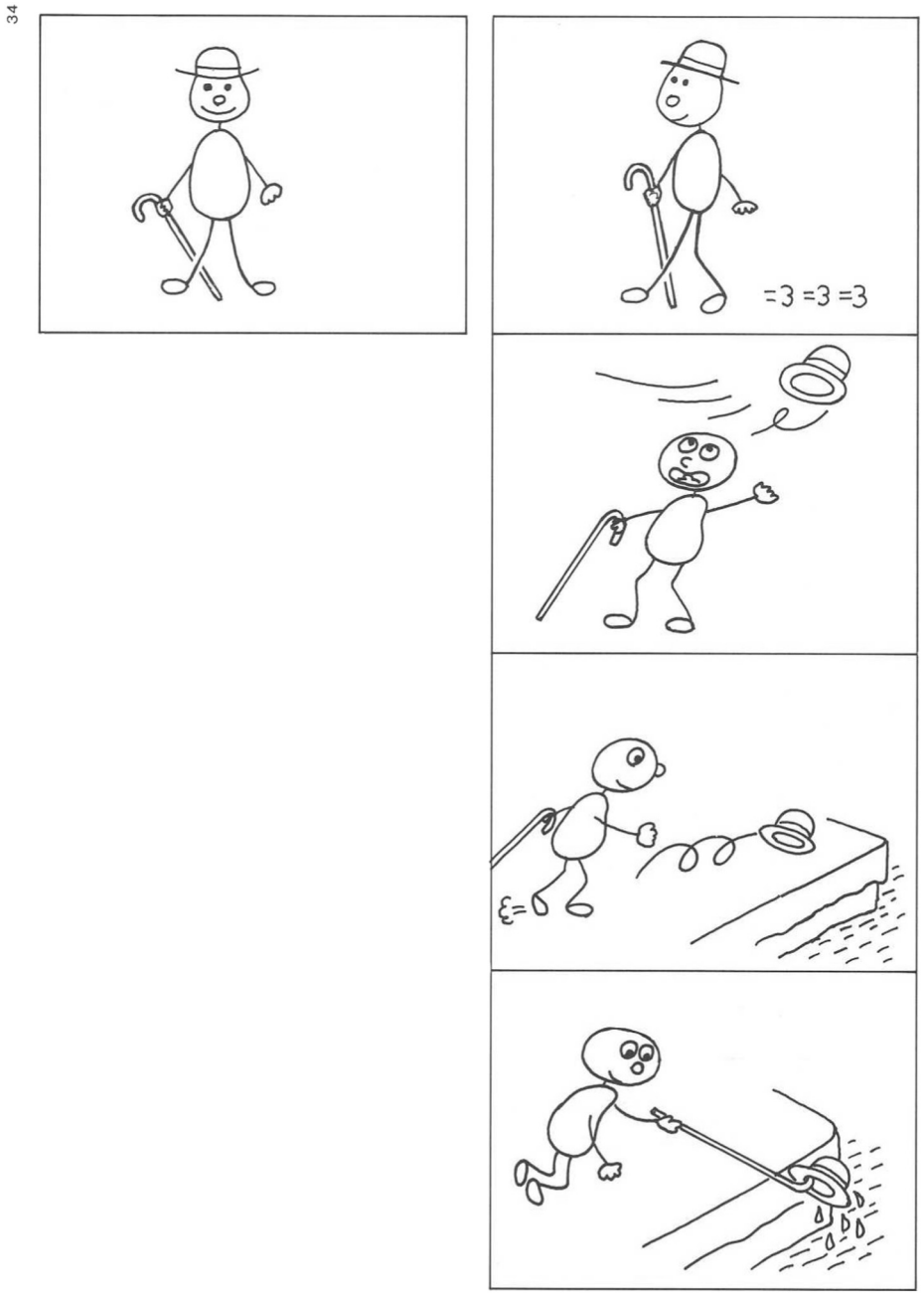

Fig. 3. SLTA subtest 8 "Explanation of a comic strip". Reprinted with permission.

tried to hook up and get the hat."

His explanation of the same task at 13 months was as follows:

"A man, wearing a hat, umm, taking a stick, is walking. Umm then, the wind blew, and his hat was blown away by the wind. The man tried to find the hat and walked to the hat. The hat went into, umm, the water, and, he tried to hook the hat which came into… went into, by the top of his stick, umm hooking, and he tried to take it."

At 16 months after SAH, he talked about his plans for visiting the hospital outpatient clinic after discharge:

"At the point of returning to work, first, I will do an outpatient visit. In regard to that, because I underwent shunt surgery, about how often a year, whether I do it on a weekday or not…, in addition, there is a possibility to go to that private office for higher brain dysfunction…that is, on a weekday, or the weekend?…"

As noted above, he had effectively grasped his own physical and mental condition at this stage, and his language had improved sufficiently to explain that he needed ongoing follow-up at the hospital. At 16 months after occurrence of SAH, he returned to work with a minor change of position. Three years have passed since then and his progress has been favorable.

5. Discussion

We reported the profile of recovery for a patient with poor grade aneurysmal SAH who succeeded in returning to work, specifically focusing on the changes of aphasia. In this patient, disturbance of consciousness persisted for about 6 months after the onset of SAH. Although we initiated therapy for aphasia from an early stage and continued to treat his lexical/semantic disorders, no improvement was obtained. Lack of improvement of his language during this period was

considered to be due to impairment of consciousness and the general mental state, which is common in patients with acute stroke, rather than his aphasia per se. His speech increased after shunt surgery improved overall brain function, although the content was sparse and communication remained poor. At 10 months after occurrence of SAH when he was discharged to home from the rehabilitation hospital, he still had aphasic symptoms such as word finding difficulty and lexical/semantic disorders besides mild cognitive impairment causing symptoms like fatigue and a euphoric state. However, in contrast to the early stage after SAH, therapy for his aphasia was so effective during this period that the materials had to be upgraded for more sophisticated ones at every session. It seems that deterioration of general brain function (so-called “Durchgangssyndrom”) resolved during this period and his aphasic syndrome became more obvious as a “core” sequel of SAH. In addition, functional reorganization of the brain for language production was occurring at this time.

Schuss et al. [20] assessed 242 patients with poor grade SAH (WFNS grades IV–V on admission), including their clinical characteristics, treatment, radiologic features, and functional outcomes. The patients were stratified into favorable and unfavorable outcome groups at 6 months after SAH according to the modified Rankin Scale (mRS) [21], with the favorable group having an mRS score of 0–2 and the unfavorable group having a score of 3–6. It was clarified that a favorable outcome was achieved in 24% of patients with poor grade SAH who underwent aneurysm treatment. On the basis of these results, Schuss et al. stressed that treatment of patients with poor grade SAH should not be abandoned and indicated careful individualized decision-making is necessary.

According to information from the Japan stroke databank [22], when a favorable outcome was defined as an mRS score of 0–2 and an unfavorable outcome was defined as a score of 3–6, the proportion of favorable outcome cases was 14.05 times large for patients with grade IV SAH by the H & K classification and 32.1 times large for those with grade V SAH. These results suggest that, although treating patients with poor grade SAH (grades IV and V) is not strongly recommended, it should be considered while taking into account various complications, such as intracerebral hemorrhage, acute hydrocephalus causing delayed impairment of consciousness, or hypoxic brain damage due to cardiac arrest.

Haug et al. [10] investigated 26 patients with aneurysmal SAH who were comatose on arrival at hospital (Hunt and Hess Grade V), using a comprehensive battery of neuropsychological tests and 2 health-related quality of life (HRQOL) questionnaires at 1 year after SAH. They found that the patients could be divided into groups with good or poor cognitive function. Patients with poor function were older, had fewer years of education, a higher preoperative ventricular score, and a higher frequency of shunt surgery. There were also differences between the 2 groups with regard to the GCS score, mRS, and employment status, while HRQOL was lower in the patients with poor cognitive function.

Despite his age and the large amount of bleeding at the occurrence of SAH, our patient did not remain comatose for long and treatment to prevent rebleeding was in initiated soon after admission. He did not develop complications that can worsen the outcome of SAH, including intracerebral hemorrhage, delayed vasospasm, cerebral infarction, or cerebral herniation. In addition, he received early shunt surgery for postoperative NPH. Although shunt removal was required due to infection, a new shunt was placed after infection had been controlled. These interventions are considered to have contributed to the favorable outcome in this case. The factor of handedness might have also played an important role in the good recovery of aphasia.

6. Conclusion

In agreement with Schuss, et al. [20], we wish to emphasize that even patients with poor grade SAH should receive active treatment, and individualized decision-making is important. It is also important to provide adequate rehabilitation for improvement of cognitive dysfunction, including aphasia, from the perspective of achieving return to the pre-SAH functional level, depending on the patient's circumstances.

Declaration of Competing Interest

The authors declare that the article content was composed in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

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