

Prolonged Dysphagia due to a Combination of Cerebral Hemorrhage and Diffuse Idiopathic Skeletal Hyperostosis: A Case Report

Misa Moriwaki,¹ Hitoshi Hase,² Seiji Fujioka,¹ Noriko Yonekura,¹ Naoko Katao,¹ Kazuhiro Takahashi,³ Masaki Mori,² Tetsuo Koyama,⁴ and Kazuhisa Domen,⁵

A 79-year-old man was diagnosed with left thalamic hemorrhage. On admission, the Functional Independence Measure (FIM) motor score was 13 points, and the Food Intake Level Scale (FILS) was Level 2, with the patient needing enteral nutrition. Six months after stroke onset, the FIM motor score had improved to 38 points and the dysphagia to FILS Level 7. The patient was able to ingest easy-to-swallow food orally three times a day, but only after postural adjustment with rotation of the head. Seven months after stroke onset, the FIM motor score had reached 45 points but without further improvement in swallowing function. Videofluoroscopic swallowing evaluation (VF) revealed that the persistence of dysphagia was due to osteophytes on the cervical vertebrae caused by diffuse idiopathic skeletal hyperostosis. On surgical removal of the osteophytes, swallowing function improved to FILS Level 9; the patient was able to ingest normal food in a seated position without postural adjustment. One year after stroke onset, the patient was discharged with an FIM motor score of 59 points and FILS Level 9. At the 2-year follow-up, there was minimal recurrence of the osteophytes, and both motor and swallowing functions were maintained at the same level as at discharge. This case suggests that dysphagia in elderly patients may be due to multiple disorders, and that surgical intervention may occasionally be effective.

Keywords: Forestier's disease, cerebrovascular, comorbidity, orthopedics, prognosis.

Introduction

The elderly population is increasing worldwide. The proportion of the world's population aged 65 and over was 8% in 2010 and is projected to rise to 16% by 2050 (http://www.who.int/aging/publications/global_health.pdf). The elderly frequently suffer from several diseases at the same time, which decrease their ability to carry out activities of daily living (ADL). The resultant increase in healthcare and social welfare costs is now becoming a serious concern in many

countries. Dysphagia is one such condition that is often seen in the elderly.¹⁾ As it is directly linked to worsening mortality, dysphagia is a topical issue in geriatric health care.

Stroke is one of the commonest diseases in the elderly and its prevalence increases with age.²⁾ About 30%–50% of stroke patients have dysphagia during the acute phase of stroke.^{3,4)} However, most patients with unilateral supratentorial lesions—the most common type of stroke—recover from the dysphagia within a few weeks; consequently, stroke cases with prolonged dysphagia are not common.^{3,4)}

The elderly also have a higher rate of osteoarthritis,⁵⁾ especially diffuse idiopathic skeletal hyperostosis (DISH), which is present in approximately 17% of people aged 50 years or older, and this has been reported to rise with increasing age.⁶⁾ Dysphagia is one of the symptoms of the cervical vertebral lesions caused by DISH.⁷⁾ However, given its high prevalence among the elderly, patients with DISH exhibit a relatively low rate of dysphagia.^{8,9)}

Thus, although both stroke and DISH are common disorders in the elderly, neither disorder on its own causes a high rate of dysphagia. We treated an elderly patient in whom the presence of DISH in addition to a thalamic hemorrhage resulted in prolonged dysphagia, and we obtained good results from dysphagia rehabilitation and surgical removal of the osteophytes.

Case Report

A 79-year-old man suddenly developed a mild disturbance of consciousness and right hemiplegia and was brought to our emergency department. Prior to admission, he had been fully independent in ADL. He had been eating a normal diet within a normal mealtime of approximately 20 min. Although he had been subjectively aware of difficulty in swallowing once or twice a week, he had not felt any particular need to consult a doctor. He suffered from diabetes, which was well controlled on medication with an HbA1c of 5.5%.

On initial examination, he was drowsy but sufficiently conscious to respond to simple verbal instructions. The right hemiplegia was Medical Research Council (MRC) grade 1/5 for upper and lower extremities.¹⁰⁾ A left thalamic hemorrhage was diagnosed on computed tomography (CT) (Fig. 1). Conservative treatment with anti-hypertensive agents and rehabilitation was initiated. Rehabilitation consisted of physiotherapy, occupational therapy, and speech therapy for a joint total of up to 180 min per day. Motor function in ADL was assessed using the Functional Independence Measure

¹Department of Rehabilitation Medicine, ²Spine Center, ³Department of Neurosurgery, Midorigaoka Hospital, 3-13-1 Makami-cho, Takatsuki, Osaka 569-1121, Japan

⁴Department of Rehabilitation Medicine, Nishinomiya Kyoritsu Neurosurgical Hospital, Nishinomiya, Hyogo, 663-8211 Japan

⁵Department of Rehabilitation Medicine, Hyogo College of Medicine, Nishinomiya, Hyogo, Japan

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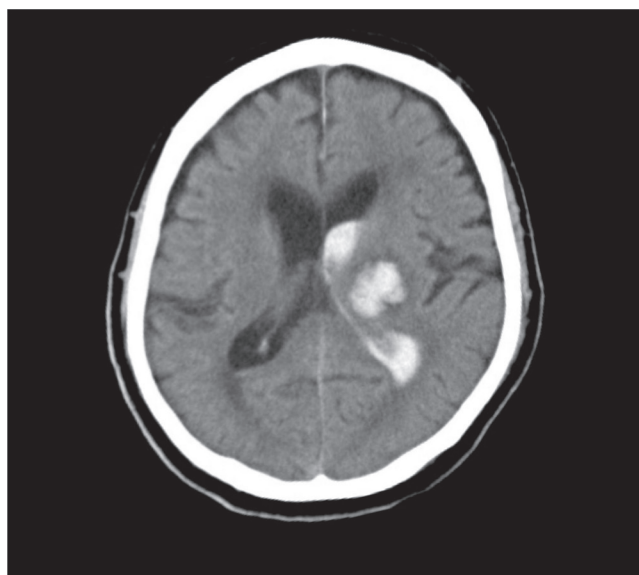


Fig. 1 Brain computerized tomography (CT) image. A thalamic hemorrhage in the left hemisphere was diagnosed.

(FIM) motor score.¹¹⁾ This assessment uses a 7-point scale for the following 13 items: eating, grooming, bathing, dressing upper body, dressing lower body, toileting, bladder and bowel management, transfer to bed/chair/wheelchair, transfers to toilet, transfer to tub/shower, walking or wheelchair propulsion, stair climbing, and total summation ranges from 13 (total dependence) to 91 (full independence). Swallowing function was assessed using the Food Intake Level Scale (FILS)¹²⁾ (see Appendix). On admission, the FIM motor score was 13 points and the swallowing function was FILS Level 2 (Fig. 2). Videofluoroscopic (VF), or videoendoscopic (VE), evaluation of swallowing function was performed approximately every 4–6 weeks.

On the day of admission, the modified water-swallowing test¹³⁾ was administered to assess whether the patient was capable of oral ingestion. The result was Grade 3, and the patient was considered to be incapable of oral ingestion (FILS Level 2). Enteral nutrition via a nasogastric tube was chosen as the nutritional route. Initially, the patient’s dysphagia was thought to be due to pseudobulbar palsy secondary to the thalamic hemorrhage, and the patient was treated with swallowing rehabilitation. This was comprised of improvement in the oral environment and indirect swallowing exercises (such as ice massage)¹⁴⁾ administered by a speech and language pathologist. The patient, however, continued to have FILS Level 2 severe dysphagia 1 month after onset (Fig. 2). VF revealed the presence of osteophytes on the second to fifth cervical vertebrae, and concomitant DISH was diagnosed (Fig. 3A). These osteophytes were particularly severe on the third and fourth cervical vertebrae (C3/4) at the level of the epiglottis, where they projected rostrally in a wedge shape. VF using thickened water revealed a pattern of dysphagia consistent with that exhibited by patients with supratentorial stroke. Clear signs of aspiration were also evident. This was “aspiration before the swallow” in which the bolus was

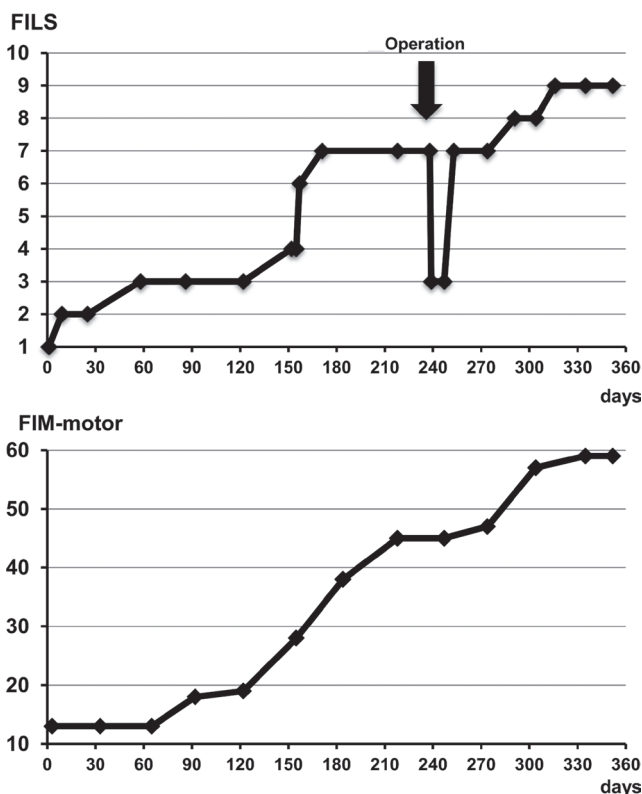


Fig. 2 Time course for Food Intake Level Scale (FILS) (upper panel) and FIM-motor score (lower panel).

induced to enter the trachea before the swallowing reflex by the osteophytes (Fig. 3A). These VF findings showed that the prolonged severe dysphagia in this patient was caused by the combination of pseudobulbar palsy and impaired pharyngeal passage due to DISH osteophytes.

Two months after stroke onset, the patient was transferred to a long-term rehabilitation ward. At approximately 5 months after stroke onset, swallowing function gradually recovered to the point at which oral ingestion became possible (see the FILS Level chart in Fig. 2). At 6.5 months after stroke onset, motor function had improved to an FIM motor score of 38 points. Swallowing function had improved to the point at which the patient was capable of swallowing semi-solid food (FILS Level 7) with postural adjustment maneuvers by rotating the head to the left. This head rotation expanded the pharyngeal cavity, encouraging the bolus passage (Fig. 4). VF at this point (Fig. 3B) showed that the aspiration caused by the wedge-shaped osteophytes (Fig. 5A) was still present. Further examination after 1 month found that although motor function had further improved to an FIM motor score of 45 points, there had been no improvement in swallowing function (Fig. 2). A spinal surgery specialist (the second author of this article) was consulted and determined that removal of the osteophytes was indicated. These findings were explained to the patient who elected to undergo surgery. Removal of the osteophytes was therefore performed. In order to avoid inducing instability of the cervical spine, the portion removed during surgery was

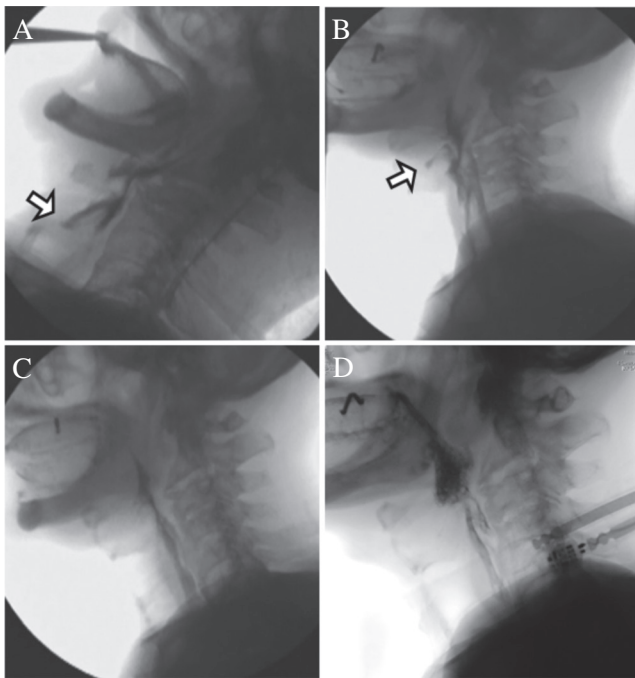


Fig. 3 Images from videofluoroscopy (VF). Arrows heads indicate aspiration. A) One month after stroke onset (thickened water intake): there was no evidence of transport action or bolus formation by the tongue. Aspiration before the swallow was present, with the test substance flowing into the pharynx and induced to enter the airway by the osteophytes. B) At 6.5 months after stroke onset (thickened water intake): although both transport action and bolus formation by the tongue were now apparent, aspiration due to the osteophytes persisted. C) At 9.5 months after stroke onset (post-operative day 52, thickened water intake): no aspiration was evident. D) Two years after stroke onset (cooked rice intake): the ability for oral intake was preserved.

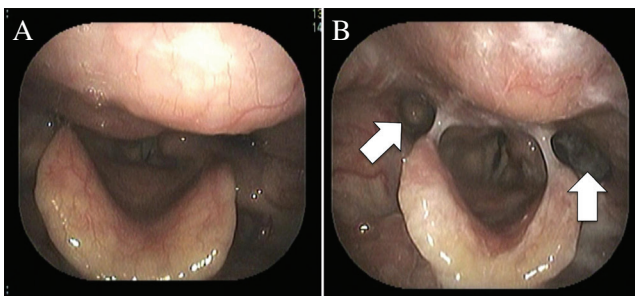


Fig. 4 Images from videoescopy (VE) assessed at 6.5 months after onset: A) the posture is facing forward: B) the posture was adjusted by rotating the head to the left, which expanded the pharyngeal cavity (shown by arrowheads).

restricted to only the protruding tips of the wedge shapes on C3/4 (Fig. 5B, indicated by arrowheads).^{15,16)}

Once the edema at the surgical site had subsided, a custard-consistency diet was started on postoperative day 14 (Fig. 2). Dysphagia rehabilitation was continued until the patient was capable of eating food of close to a normal consistency (Fig. 2). Of note, after the surgery, the patient was able to swallow while facing forward without any postural adjustment. At 9.5 months after stroke onset (postoperative day 52), VF

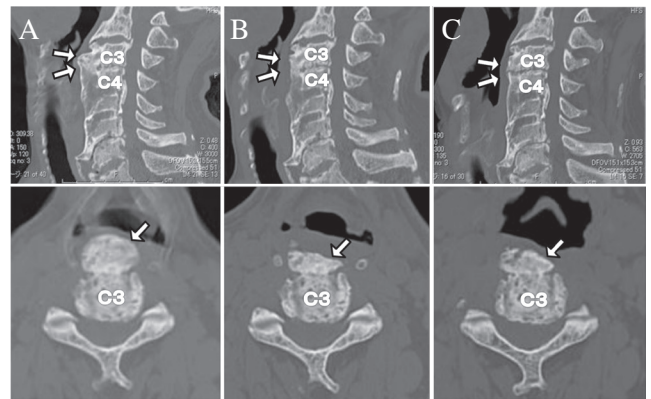


Fig. 5 Neck CT image. A) Pre-surgery. B) Post-surgery: day 14. C) Post-surgery: year two.

with boluses of jelly, thickened water, and thick rice porridge did not reveal any obvious aspiration (Fig. 3C). The patient subsequently proceeded to improve in both motor function to an FIM motor score of 59 points and swallowing function to FILS Level 9 with no occurrence of pneumonia, and he was discharged to home (Fig. 2). VF performed at our outpatient clinic two years after stroke onset did not reveal any aspiration (Fig. 3D). Cervical CT was carried out 2 years postoperatively. Although there had been some growth in the osteophytes compared to the immediate postoperative period, this was only 0.8 mm (Fig. 5C).

Discussion

For stroke patients, particularly those with first-ever supratentorial lesions, dysphagia is often seen; however, swallowing function generally recovers at an early stage.¹⁷⁾ In the case reported here, the pattern of recovery deviated from the norm in that the patient's dysphagia persisted despite an improvement in motor function to an FIM motor score of 45 points.¹⁷⁾ When VF was performed, dysphagia due to concomitant DISH was also found to be present. This suggests that if dysphagia is prolonged in elderly patients, the possibility of the presence of multiple simultaneous conditions should be borne in mind.

If it is clear that dysphagia is caused by osteophytes, their removal offers a better prospect for the improvement in swallowing function. In the present case, the osteophytes were removed more than 6 months after the onset of dysphagia. Before the stroke, this patient had been eating a normal diet while occasionally noticing some subjective difficulty in swallowing, and, therefore, at that point, his swallowing function was FILS Level 9. This suggests that DISH alone had not caused serious dysphagia. Functional impairment caused by stroke generally improves within about 6 months of onset. At that point, VF clearly showed that the dysphagia was caused by osteophytes (Figs. 3, 5). The osteophytes were therefore surgically removed with good results.

Numerous studies have reported that surgery is effective in cases of DISH if dysphagia is serious or if conservative therapy has proven ineffective.^{16,18-22)} Conversely, some other studies have found that aspiration pneumonia may occur

after removal of the osteophytes, implying that surgery is not effective²³⁾ Therefore, as yet, there is no unified consensus. In the present case, the patient improved from FILS Level 2 to Level 7 as a result of conservative intervention with dysphagia rehabilitation, and became capable of eating semi-solid food by means of postural adjustment. However, the patient wanted to relieve the physical and mental stress of having to adjust his posture by rotating his head each time he swallowed a bolus, and to be able to eat food of close to a normal consistency. Surgery was therefore performed and a good outcome was achieved. This case suggests that if dysphagia caused by osteophytes is diagnosed, proactive surgery may be indicated even if enteral nutrition can be discontinued.

When considering removal of osteophytes, the risk of recurrence must be taken into account.^{15,16,24)} Postoperatively, the osteophytes recur in almost all patients with a reported average growth rate of around 1 mm/year.²⁴⁾ In order to prevent recurrence, studies have recommended minimizing the area of bone removed so as not to cause instability of the cervical spine.^{15,16)} In the present case, regrowth was only 0.8 mm 2 years postoperatively (Fig. 5B, C). It is noteworthy that this patient has maintained a good outcome with respect to swallowing function until the age of 81 without osteophyte recurrence.

Conclusion

This case demonstrates the following two points. First, it is possible for the elderly to suffer from multiple conditions that do not cause severe dysphagia individually but may result in severe and prolonged dysphagia in combination. Secondly, a thorough investigation of the causes of dysphagia in such patients may indicate surgical intervention.

Conflict of Interests Disclosure

The authors declare that there is no conflict of interest regarding the publication of this paper.

Appendix

Food Intake Level Scale (FILS)¹²⁾

- No oral intake
 - Level 1: No swallowing training is performed except for oral care.
 - Level 2: Swallowing training not using food is performed.
 - Level 3: Swallowing training using a small quantity of food is performed.
- Oral intake and alternative nutrition
 - Level 4: Easy-to-swallow food less than the quantity of a meal (enjoyment level) is ingested orally.
 - Level 5: Easy-to-swallow food is orally ingested in one to two meals but alternative nutrition is also given.
 - Level 6: The patient is supported primarily by ingestion of easy-to-swallow food in three meals but alternative nutrition is used as a complement.

- Oral intake alone
 - Level 7: Easy-to-swallow food is orally ingested in three meals. No alternative nutrition is given.
 - Level 8: The patient eats three meals by excluding food that is particularly difficult to swallow.
 - Level 9: There is no dietary restriction and the patient ingests three meals orally but medical considerations are given.
 - Level 10: There is no dietary restriction and the patient ingests three meals orally (normal).

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Corresponding author:

Misa Moriwaki, Department of Rehabilitation Medicine, Midorigaoka Hospital, 3-13-1 Makami-cho, Takatsuki, Osaka 569-1121, Japan.

✉ misa.moriwaki@hanamizuki-dreams.org