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A Case of Ankylosing Spinal Hyperostosis with Dysphagia Aggravated Because of Deterioration of Depressive Symptoms

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Abstract

Case report: A 73-year-old male incurred extensive third-degree burns on his abdomen. Although dysphagia had not been identified before the patient sustained burn injuries, VideoFluoroscopy (VF) performed for poor ingestion revealed severe dysphagia after skin grafting surgery and Ankylosing Spinal Hyperostosis (ASH). Although the cause of dysphagia was not identified, a Percutaneous Endoscopic Gastrostomy (PEG) tube was inserted. The patient was subsequently transferred to our hospital for rehabilitation. Direct rehabilitation for dysphagia began with jelly ingestion, and meal consistency was gradually increased. On day 50, he could completely ingest meals. However, beginning on day 70 of hospitalization, the patient began to complain of mental stress because his burns were taking too long to heal, which caused him to lose sleep at night. He was diagnosed with depression and began to choke when eating meals. VF findings again revealed aspiration after swallowing, and the patient's state of deglutition that should have improved deteriorated again. Nutrition and fluid administration required a PEG tube to be re-introduced. After the patient's wounds healed and his mental condition stabilized, he gradually recovered the ability to ingest food. VF findings on day 103 of hospitalization revealed an improvement compared with his previous examination, and aspiration had disappeared.

Discussion: The patient's condition improved temporarily but became aggravated with the deterioration of his depressive symptoms. Subsequently, with an improvement in his depression, dysphagia improved concomitantly. Apart from the deterioration of his depressive symptoms, we did not identify any cause that may have aggravated dysphagia in the patient. We believe that aggravation of dysphagia in the pharyngeal stage could have been caused by the deterioration of the patient's depressive symptoms when he was barely able to swallow, such as in ASH.

Keywords: Ankylosing spinal hyperostosis; Depression; Dysphagia; Video fluoroscopy

Introduction

It has been reported that 9%-42% psychiatric patients suffer from complications such as dysphagia and eating disorders [1], which can result in serious problems such as aspiration pneumonitis and suffocation [2-4]. Psychogenic eating disorders result from specific psychiatric disorders, whereas drug-induced dysphagia results from other causes [5,6]. Pharmacological actions, such as sedation, anticholinergic effects, and extrapyramidal side effects, may impair the deglutition reflex [3,4,7,8]. Here we describe a case of Ankylosing Spinal Hyperostosis (ASH), also known as Forestier's disease [9-14], in which deterioration of depressive symptoms aggravated dysphagia despite not increasing the medication dosage during follow-up, and spontaneous remission occurred because of improvements in depression. We report the clinical course of this case along with a review of the literature.

Case Report

The subject was a 73-year-old male.

Medical history

The patient was under treatment for manic depression and had previously attempted suicide by hanging. He also suffered from colorectal cancer and inflammation of renal calyces. Dysphagia had not been previously identified and the patient consumed normal meals before onset of the current illness.

History of current illness

The patient incurred extensive third-degree burns on his abdomen from a cigarette lighter, for which he was admitted to a hospital emergency room. Skin grafting was performed on day 16. Although the patient's general condition after surgery was believed to be stable, video fluoroscopy (VF) [2,15] performed for poor ingestion revealed severe dysphagia. Although the cause of dysphagia was not identified, the patient's mental condition and postoperative nutritional management were taken into consideration. On day 64, a percutaneous endoscopic gastrostomy (PEG) tube16) was inserted. On day 70, the patient was transferred to our hospital for rehabilitation.

Condition at the time of admission

Glasgow Coma Scale: E4V5M5. The patient's mental condition was stable and he actively participated in his rehabilitation.His burns had completely healed, except those on his left anterior chest, which required treatment with a protective gauze and application of a wound-healing agent. The patient did not complain of pain. The patient had no trouble in oral cavity, teeth, gingival and periodontal health. There was no slant when he stacks his tongue out. But, he could not eat and drank only small amounts of water (3 ml). Although it was occasionally difficult to understand him when he spoke, there were almost no problems in communication.

ROM: No marked limitations were observed for cervical vertebrae, with a cervical flexion of 15°, retroflexion of 10°, left rotation of 15°, and right rotation of 15°.

Functional Independence Measure (FIM)[17]: 52 points (motor: 19, cognition: 33)

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Imaging findings

CT scan of the head: Although an irregular low-attenuation area in the right hemisphere subcortical white matter and general chronic ischemic changes were observed, there were no other changes.

VF findings in the previous emergency room: An osseous growth was observed at the anterior margin of the C5-C6 vertebral bodies, with the bone deformation forming a bridge. These images were taken while a nasogastric tube was in place, revealing incomplete laryngeal elevation, poor pharyngeal constriction, and incomplete opening of the Upper Esophageal Sphincter (UES). A bolus was predominantly passed through the left pyriform fossa, and evident aspiration and pharyngeal residue of pre-thickened liquid barium intake were observed. This anterior protrusion of the vertebral bodies suggested obstructed food passage (Figure 1).

VF upon admission to our hospital: We observed poor pharyngeal constriction, slightly poor laryngeal elevation, and incomplete opening of the UES. Pharyngeal residue of pre-thickened liquid barium intake was also observed; however, aspiration had disappeared. The patient's condition had improved compared with the earlier VF results obtained in the emergency room (Figure 2).

Prescription

Lithium carbonate (Limas®) at 600 mg/day, quetiapine fumarate (Seroquel®) at 75 mg/day, sodium valproate (Depakene®) at 300 mg/day, flunitrazepam (Rohypnol®) at 1 mg/day, and lansoprazole (Takepron®) at 15 mg/day.

Progress after admission

Indirect rehabilitation for dysphagia included oral massage, thermal stimulation, vocal training, breathing exercises, ROM training for the neck, and neck relaxation. Direct rehabilitation for dysphagia began with jelly ingestion, and meal consistency was gradually increased. On day 43 of hospitalization, the patient's mental condition was stable, and quetiapine fumarate was reduced to 50 mg/day, as instructed by the psychiatrist who had taken care of the patient before his burn injuries. On day 50, he could completely ingest meals that included soft vegetables, rice gruel, and thickened water. However, beginning on day 70 of hospitalization, the patient began to complain of mental stress because his burns were taking too long to heal, which caused him to lose sleep at night. He was diagnosed with depression and began to choke when eating meals. Antipsychotic drug administration was not changed, and VF findings on day 84 revealed no changes in

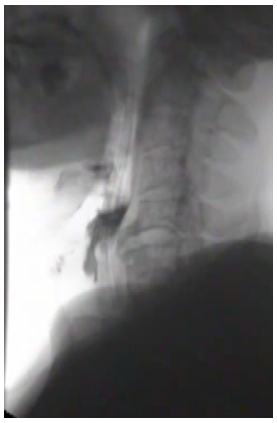

Figure 1: VF findings in the emergency room.
An osseous growth was observed on the anterior margin of the C5–C6 vertebral bodies, with bone deformation that formed a bridge. Aspiration and pharyngeal residue of pre-thickened liquid barium intake were clearly observed. An anterior protrusion of the vertebral bodies suggested obstructed food passage.

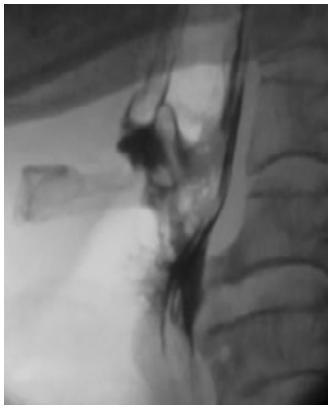

Figure 2: VF findings on hospitalization. Pharyngeal residue of a bolus was observed but aspiration disappeared. The patient's condition improved compared with earlier VF results obtained in the emergency room.

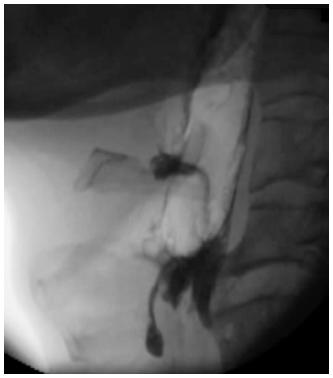

Figure 3: VF findings on day 84 of hospitalization. No changes in the poor laryngeal elevation and pharyngeal constriction were detected. However, aspiration after swallowing was observed again, and the patient's deglutition state that should have improved deteriorated again.

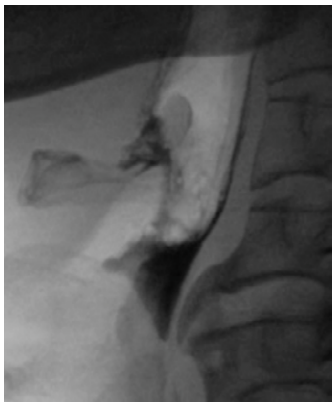

Figure 4: VF findings on day 103 of hospitalization.
No changes in pharyngeal residue were detected. However, aspiration disappeared and an improvement was observed compared with the previous examination.

the poor laryngeal elevation and pharyngeal constriction. However, aspiration after swallowing was observed again, and the patient's state of deglutition that should have improved deteriorated again (Figure 3). Nutrition and fluid administration required a PEG tube to be re-introduced. By day 97, the patient's mental condition stabilized because his wounds had healed, and he was gradually able to ingest food again. During psychiatric examinations, the patient increasingly made positive remarks, and the psychiatrist instructed that the dose of lithium carbonate should be reduced to 400 mg/day starting on day 99. VF findings on day 103 of hospitalization revealed no changes in the pharyngeal residue; however, aspiration had disappeared, and there was an improvement compared with his previous examination (Figure 4). During the hospitalization, there were no newly appeared findings in the head CT. On day 142, the patient could ingest full meals consisting of soft vegetables, rice gruel, and thickened water, and he was discharged from the hospital with an FIM score of 88.

Progress summary

Dysphagia had not been identified before the patient sustained burn injuries. After injury, dysphagia was believed to be caused by ASH. The patient's condition temporarily improved but became aggravated with the deterioration of his depressive symptoms. Subsequently, with an improvement in his depression, dysphagia improved concomitantly (Figure 5).

Discussion

The patient was initially diagnosed with ASH on the basis of the following criteria: (1) ossification on the anterior aspect of the vertebral bodies that formed a bridge between two or more vertebral bodies; (2) absence of any underlying disease that may have caused spondylitis; (3) ossification not attributable to trauma such as compression fractures; and (4) absence of ileosacral arthritis [9-11]. Dysphagia complications in ASH may be caused by the following reasons: (1) obstructed food passage due to mechanical pressure on the pharynx and esophagus; (2) inflammation-induced adhesion of the pharynx and esophagus, fibrillization, and convulsions; (3) traction of the ascending branch of the recurrent laryngeal nerve and abnormal sensitivity of the sympathetic nerve; or (4) psychogenic factors [11-18]. Moreover, the esophageal orifice is anatomically fixed to the C5–C6 vertebral bodies. Therefore, when the maximum protrusion of ASH is at the same level, it restricts ROM and can cause dysphagia [18]. In the patient in this case, the maximum protrusion was found at this site. Intubation was performed while the patient was in the emergency room, and we believe that dysphagia may have been initially caused by acute inflammation of the pharynx. However, it is uncertain whether inflammation aggravated dysphagia during hospitalization because no laboratory tests for enhanced inflammation or other clinical symptoms were performed. The reason why aspiration was observed and then spontaneously ceased cannot be explained by ASH alone.

Subsequently, we examined the possibility of drug-induced dysphagia. The observed reduced pharyngeal clearance suggested drug-induced dysphagia, possibly explaining the obstructed food passage due to ASH. Dysphagia was not diagnosed before hospitalization. In addition, quetiapine fumarate and lithium carbonate dosages were reduced but not increased during hospitalization. Therefore, whether there was a direct causal relationship between these agents and the episodes of dysphagia aggravation during hospitalization remains unknown.

The possibility of psychogenic dysphagia was also investigated [19-25]. Psychogenic dysphagia is a diagnosis of exclusion well described by Buchholz in 1994 [20]. It is characterized by oral apraxia but with intact speech, pharyngeal, and neurological functions. Patients suspected to suffer from psychogenic dysphagia usually undergo detailed neurological evaluation to eliminate the possibility of neurogenic dysphagia. Psychogenic dysphagia is known to be pertinent to psychiatric disorders, such as anxiety, depression, somatoform disorders, hypochondriasis, conversion disorders, or eating disorders

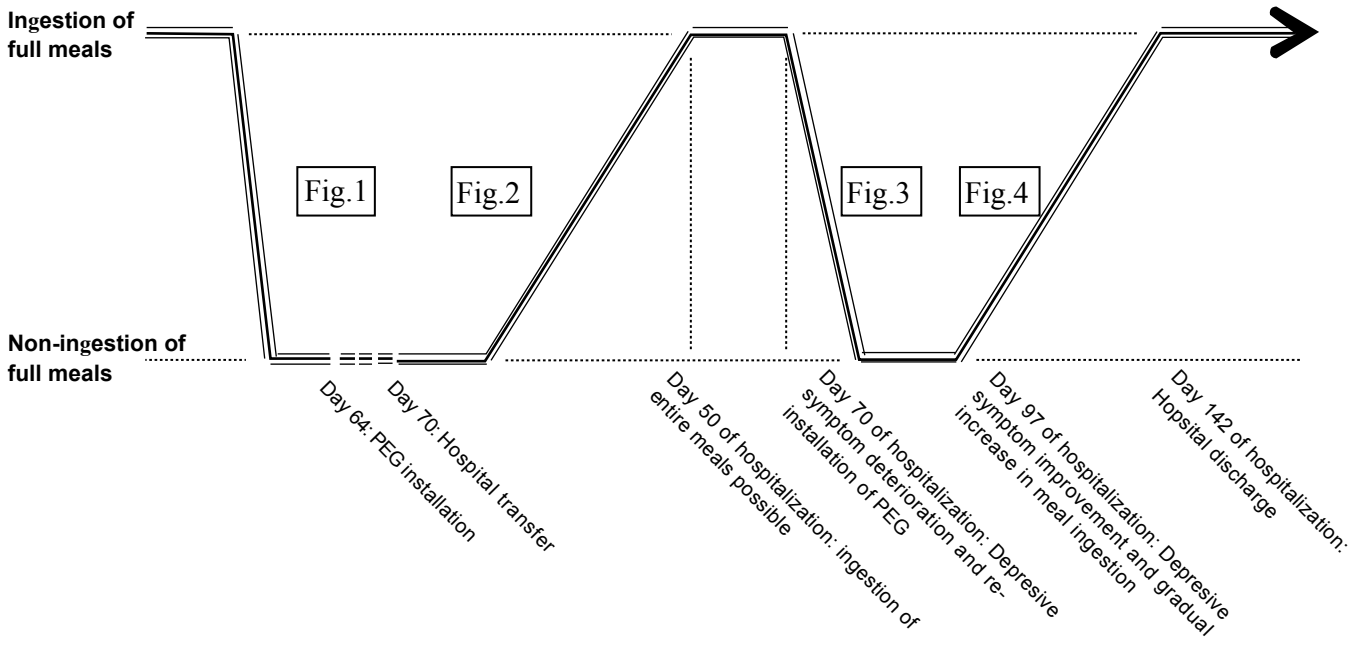

Figure 5: Clinical course during hospitalization.
The patient's condition temporarily improved. However, beginning on day 70 of hospitalization, his depressive symptoms deteriorated and dysphagia re-appeared, which resulted in nutrition management by a PEG tube. Starting on day 97 of hospitalization, his dysphagia improved concomitant with an improvement in his depression.

[20,13]. The term globushystericus was originally used to indicate anxiety-stricken or hysterical patients with dysphagia secondary to a fullness or lump in the throat [22]. Eslick stated that intermittent dysphagia is associated with anxiety, whereas progressive dysphagia is associated with depression [24]. Today, this symptom complex is most commonly associated with gastro esophageal reflux disease. There is some evidence that psychogenic problems are rarely the primary etiological agent of dysphagia [19-22]. Psychogenic issues may trigger organic disorders such as hyperacidity or autoimmune diseases but should not be the only consideration when evaluating patients with dysphagia unless all other possibilities have been eliminated. After a thorough literature search, we could not find any published report mentioning changes in the pharyngeal stage of swallowing function. The only relevant published report was a book by Fujishima, who indicated that "dysphagia may deteriorate during depression (and occasionally in cerebrovascular disease) [25]." According to our understanding of patients believed to develop a certain degree of difficulty with swallowing, such as those with cerebrovascular disease as mentioned above, the deterioration of depressive symptoms may result in aggravation of dysphagia in the pharyngeal stage.However, we believe that there is a lack of general consensus for these cases. In the present case, apart from the deterioration of his depressive symptoms,we could not identify any cause that may have aggravated dysphagia in the patient. After all, we couldn't find out any other physical changes having influence on dysphagia than depression. And, the severity of dysphagia ran parallel with the conditions of depression. Therefore, we supposed that dysphagia is influenced or induced by the depressive state. We believe this is the first detailed report regarding aggravation of dysphagia in the pharyngeal stage caused by the deterioration of depressive symptoms.

Based on the specific progress of the patient, we hypothesize that the impact of cervical spondylosis on swallowing and the effect of antipsychotic agents may have resulted in pharyngeal obstruction and reduced the deglutition reflex before the patient sustained burn injuries. It is highly possible that the initial cause of dysphagia was acute pharyngeal inflammation due to intubation, which was performed in the emergency room to help preserve his ingestion. Aggravation of dysphagia during hospitalization may have been caused by the deterioration of his depressive symptoms due to the delay in the healing of the burn wounds when the patient was barely able to swallow.

In Japan, there are extremely few occasions when hospital physicians who normally diagnose and treat dysphagia, such as rehabilitation specialists and otolaryngologists, are involved in the care of patients with psychiatric disorders [26]. In this case, we obtained advice regarding the patient's prescriptions during routine check-ups by the psychiatrist who had been treating the patient before he sustained burn injuries. This enabled the patient to remain hospitalized in our rehabilitation ward. Consequently, we could follow his clinical course on the basis of VF findings, which showed that the deterioration of his depressive symptoms aggravated dysphagia, whereas an improvement in his depressive symptoms resulted in improvements in dysphagia. In recent years, our understanding of eating disorders and dysphagia has improved, and we anticipate that further clinical research on dysphagia in psychiatric patients will be conducted. We hope that our report will help future clinical studies of dysphagia in psychiatric patients.

Conclusions

Depressive symptoms alone have never been reported to cause aggravation of dysphagia in the pharyngeal stage. However, in rare cases, the deterioration of depressive symptoms can aggravate dysphagia when the patient is barely able to swallow, such as in severe ASH.

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