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Rapidly Progressing Dysphagia After Thoracic Spinal Cord Injury in a Patient With Ankylosing Spondylitis: A Case Report

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Abstract

Introduction: Ankylosing spondylitis (AS) is a chronic systemic inflammatory disease affecting the axial skeleton, including the sacroiliac joint, which causes vertebral fusion in the advanced stage. However, reports of anterior cervical osteophytes compressing the esophagus and causing dysphagia in patients with AS are rare. Here, we present the case of a patient with AS and anterior cervical osteophytes who exhibited rapidly progressing dysphagia after thoracic spinal cord injury (SCI). **Case Presentation:** The patient, a 79-year-old man, was previously diagnosed with AS and had syndesmophytes at C2-C7 without dysphagia for several years. In 2020, he began to experience paraplegia, hypesthesia, and bladder and bowel dysfunction after a fall. He also had T9 SCI American Spinal Injury Association Impairment Scale grade A due to a T10 transverse fracture. Four months after SCI, he developed aspiration pneumonia, and a videofluoroscopic swallowing study indicated dysphagia with epiglottic closing problems due to syndesmophytes at the C2-C3 and C3-C4 levels. He received treatment for dysphagia and VitalStim therapy thrice (once daily); however, the recurrent pneumonia and fever continued. He further underwent bedside physical therapy and functional electrical stimulation once daily. However, he died from atelectasis and exacerbation of sepsis. **Discussion and Conclusion:** General deterioration of the patient's physical condition due to SCI, sarcopenic dysphagia, and compression of cervical osteophytes seemed to be involved in rapid exacerbation following SCI. Early screening for dysphagia is vital in bedridden patients with AS or SCI. Additionally, assessment and follow-up are important if the number of rehabilitation treatments or the out-of-bed movement activity decreases because of pressure ulcers.

Keywords

deglutition disorders, spondylitis, ankylosing, osteophyte, spinal cord injuries

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Introduction

Ankylosing spondylitis (AS) is a chronic systemic inflammatory disease that predominantly affects the axial skeleton, including the sacroiliac joint.[1] AS can also affect the extra-articular organs, causing pericarditis, uveitis, pulmonary fibrosis, chronic inflammation, and new bone formation. This results in a radiologic bamboo

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spine appearance, osteophyte and syndesmophyte formation, and ligament ossification.[2,3] Although HLA-B27 seropositivity is not essential for diagnosis, >85% of AS patients are HLA-B27 positive.[4] Anterior cervical osteophytes in patients with AS may compress the esophagus and cause dysphagia; although such cases are rare, they are observed occasionally.[5-7] Herein, we present a case of rapidly progressing dysphagia after thoracic spinal cord injury (SCI) in a patient with AS and anterior cervical osteophytes.

Case Presentation

A 79-year-old man diagnosed with AS visited our hospital on October 25, 2020, complaining of back pain which began after a fall on October 19, 2020, with subsequent paraplegia, hypesthesia, and bladder and bowel dysfunction. The patient was diagnosed with T10 vertebral body transverse fracture with thoracic myelopathy and subsequently underwent decompressive laminotomy at T9-T10 and posterior spinal fusion at T8-T12 on November 2, 2020.

Physical examination revealed grade I motor weakness in the lower extremities and sensory deficits below T9. The cervical spine, bilateral shoulder, hip, and knee showed limited range of motion, and radiography indicated osteoarthritis and multiple spur changes. He tested negative for HLA-B27 in 2015, but pelvic radiography revealed sacroiliitis.

C-spine sagittal magnetic resonance imaging (MRI) performed after admission revealed syndesmophytes at the C2-C7 level that were pressing on the hypopharynx, pyriform sinus, and esophagus. These syndesmophytes were only slightly increased in size, with no notable difference in the nearby structure compression, compared with that previously observed in 2014 (Figure 1). However, the patient did not complain of dysphagia, and he had no signs of aspiration and no history of aspiration pneumonia. He was transferred to a different hospital after rehabilitation for paraplegia because of T9 SCI American Spinal Injury Association Impairment Scale grade A. Rehabilitation treatment included neurodevelopmental therapy, functional electrical stimulation twice daily, ergometry and tilting once daily, and occupational therapy once daily.

Following treatment at a different hospital for approximately 1 month, exacerbation of the coccygeal pressure ulcer was noted, and he was readmitted to our hospital after 3 months of SCI in 2021. At this time, the patient complained of symptoms of "rice coming up from the throat to the mouth." A videofluoroscopic swallowing study (VFSS) confirmed significant amounts of residue in the vallecular space and pyriform sinus in the dysphagia diet level 3, which comprises a combination of moist foods in bite-sized pieces and a regular diet, but showed no definite aspiration signs for all diet forms. Thus, the patient and caregivers were provided diet and self-exercise education through a single session of dysphagia treatment. During rehabilitation in our hospital, neurodevelopmental therapy was conducted once or twice daily, and tilting and functional electrical stimulation were performed twice daily. As the patient had pressure ulcers, wheelchair ambulation was restrained, the number of rehabilitation treatments was reduced, and treatment was performed in the side-lying and prone positions.

The patient showed recurrent fever and pneumonia at 4 months after SCI onset, and another VFSS was performed. Larger amounts of residue were noted during this VFSS than during the previous VFSS. Penetration and aspiration were observed for all diets and during water intake, and an epiglottic closing problem due to syndesmophytes at the C2-C3 and C3-C4 levels was observed intermittently (Figure 2;

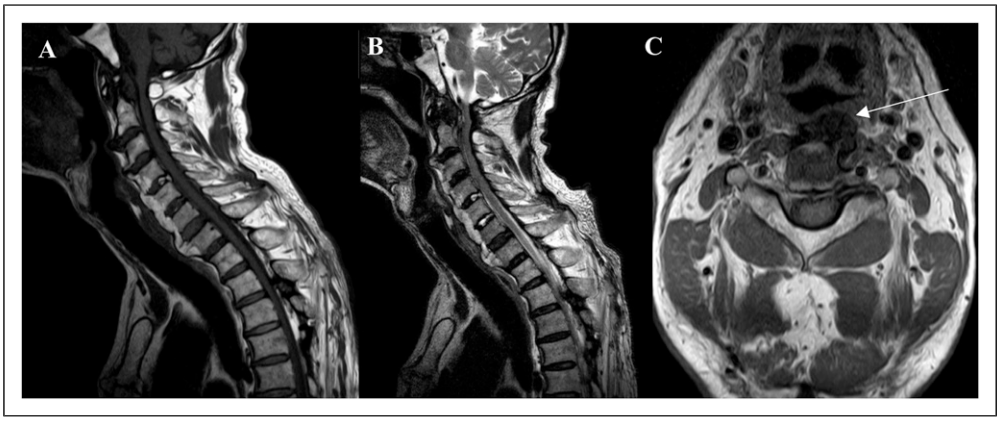

Figure 1. (A) MRI T1-weighted sagittal image (2014) of the patient's cervical spine showing C2-C7 syndesmophytes. (B) MRI T2-weighted sagittal image (2020) of the patient's cervical spine. (C) MRI T1-weighted axial image (2014) of the cervical spine at the C2-C3 level showing C2-C3 syndesmophytes compressing the hypopharynx (arrow). MRI: magnetic resonance imaging.

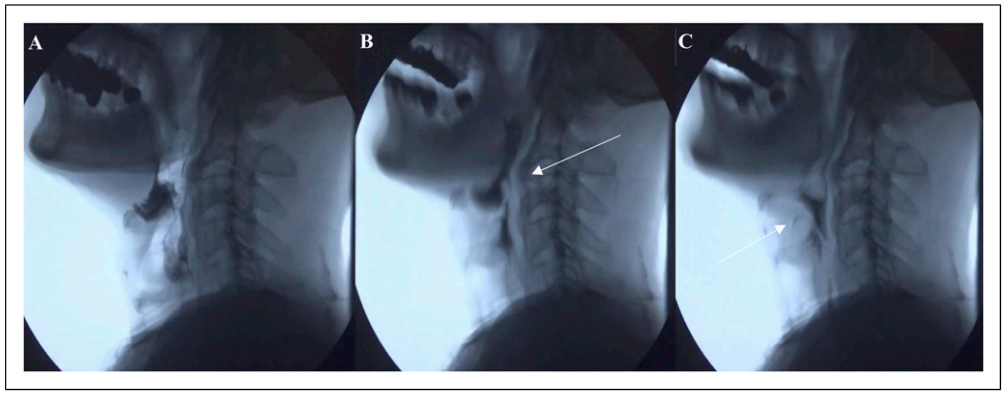

Figure 2. Images taken during the videofluoroscopic swallowing study performed 4 months after spinal cord injury onset. (A) A large amount of residue is visible in the vallecular space and pyriform sinus after swallowing. (B) An epiglottic closing problem due to syndesmophytes at the C2-C3 level (arrow) can be seen. (C) Penetration during swallowing (arrow).

Table 1. Comparison of VFSS at 3 and 4 months after SCI occurrence.

VDS Parameters	3 months after SCI Onset	4 months after SCI Onset
Oral phase		
Lip closure	0	0
Bolus formation	3	3
Chewing	0	0
Tongue control	1.5	1.5
Tongue to palate contact	5	5
Premature bolus loss	1.5	1.5
Oral transit time	3	3
Pharyngeal phase		
Residue in the valleculae	6	6
Reduced laryngeal elevation and epiglottic closure	9	9
Residue in the pyriform sinuses	13.5	13.5
Coating of pharyngeal walls	9	9
Pharyngeal delay time	0	0
Pharyngeal transit time	0	0
Aspiration	0	12
Total score	51.5	63.5

VFSS, videofluoroscopic swallowing study; SCI, spinal cord injury; VDS, videofluoroscopic dysphagia scale.

Table 1). As such, dysphagia treatment and VitalStim therapy were performed once daily. The pressure ulcers became severe because of repeated pneumonia and fever, and thus the treatment was discontinued. Other rehabilitation treatments included bedside physical therapy and functional electrical stimulation once daily. His aspiration pneumonia became severe, and he developed atelectasis. The patient subsequently died from exacerbation of sepsis.

The Institutional Review Board of Jeju National University Hospital waived the requirement for obtaining patient informed consent as no personal information was divulged (no.2021-09-001).

Discussion and Conclusion

To the best of our knowledge, this is the first case of dysphagia that progressed rapidly after SCI in a patient with AS who had no dysphagia. Despite reports of dysphagia due to anterior cervical osteophytes,[5-7] there have been no reports of newly developed dysphagia in patients with pre-existing osteophytes following SCI.

Our patient had multilevel syndesmophytes at C2-C7; therefore, diffuse idiopathic skeletal hyperostosis (DISH) was also suspected, but the patient did not meet the diagnostic criteria for DISH due to sacroiliitis.[8] AS and DISH commonly occur simultaneously;[9] hence, it is not

meaningful to distinguish them. In this case, the C4-C7 syndesmophytes compressed the esophagus, and, particularly, the C2-C3 and C3-C4 syndesmophytes affected epiglottis tilt and closure, thereby worsening dysphagia.

Although the cause of syndesmophyte formation in AS is unclear, the predominant hypothesis is that inflammatory and mechanical components, such as microdamage, play an essential role in local and new bone formation.[10] In AS, syndesmophytes can occur in all vertebrae. This is commonly observed in the anterior cervical region, and dysphagia due to the mechanical compression of cervical osteophytes and syndesmophytes has been described in the literature.[5,11]

There are several mechanisms by which the anterior cervical pathological bone may cause dysphagia, including the following:[12] (1) large osteophytes may mechanically trigger blockage of the esophagus; (2) small osteophytes may compress and obstruct the esophageal segment attached to the cricoid cartilage, most commonly at C5-C6; (3) esophagus-compressing osteophytes coexisting with a decreased closing ability of the larynx may additively worsen dysphagia; (4) osteophytes at C3-C4 may impair tilting of the epiglottis over the laryngeal inlet; and (5) inflammation in the regions surrounding the osteophytes may lead to esophagitis or pharyngitis, eventually worsening dysphagia. In this case, we believe that from among the second to fifth mechanisms, the fourth mechanism was the main cause of dysphagia.

However, despite the presence of osteophytes, our patient lived without dysphagia for at least 6 years and showed rapid progression and deterioration following SCI. Therefore, multifactorial effects are expected. In the present case, the second possible cause is the occurrence of bony callus and fragments due to new bone formation observed following T10 vertebral body fracture, which may cause dysphagia through compression of the nearby sympathetic trunk, right greater splanchnic nerve, or esophagus.[12,13]

General deterioration of the patient's physical condition due to SCI and old age may have further contributed to the development of dysphagia. Recently, the concept of sarcopenic dysphagia has been established;[14,15] and it is also possible that the patient may have had muscle loss due to his bedridden state. In fact, he lost 4 kg in 1 month, equivalent to a 5% loss. However, handgrip strength, gait speed, and swallowing muscle strength tests were not performed to diagnose sarcopenic dysphagia in this patient. Additionally, dual X-ray absorptiometry or bioimpedance analysis to measure muscle mass were not performed due to deconditioning and poor cooperation. Accordingly, it was difficult to confirm whether the patient's dysphagia was sarcopenic. However, considering that he had lived without apparent dysphagia until admission, it is logical to assume that sarcopenia may have contributed to dysphagia in this patient.

Expectedly, the disuse of the neck muscle in our patient due to the limited range of motion of the neck led to pharyngeal muscle weakness and atrophy, which was also related to dysphagia. Dysphagia therapy, including the shaker exercise, was attempted, but could not be performed correctly because of the patient's limited range of motion of the neck.

A bedridden state is expected to cause muscle loss. Our patient was completely bedridden for nearly 1-2 months, and dysphagia only developed at 3-4 months after SCI onset. He underwent rehabilitation treatment at the same frequency as other patients until 2 months after SCI. After admission to another hospital for approximately 20 days, the pressure ulcers worsened, and he was treated with antibiotics for a urinary tract infection. Thus, the patient was bedridden with little rehabilitation treatment. He received rehabilitation treatment at 3 months after SCI onset. However, by 4 months after onset he was completely bedridden due to worsened pressure ulcers, recurrent pneumonia, and fever. Therefore, it seems that the dysphagia rapidly exacerbated within approximately 4 months of partial bedrest after SCI onset, or during the month that the patient was completely bedridden due to pressure ulcers and medical complications. In a study by Maeda et al.,[16] sarcopenic dysphagia developed within 2 months in patients without dysphagia; thus, sarcopenia may have played a significant role in our patient.

Unfortunately, our study is limited in that, although various mechanisms were suggested, the exact cause of the dysphagia could not be identified because the diagnostic test for the accurate diagnosis of sarcopenic dysphagia suggested as a multifactorial effect could not be performed.

In conclusion, the findings of this case suggest that early screening for dysphagia may be necessary in bedridden patients with severe weakness after SCI, particularly in patients with AS and anterior cervical osteophytes. Even if mild signs and symptoms of aspiration are present, evaluation and follow-up of dysphagia are necessary. In addition, assessments for sarcopenic dysphagia, such as bioimpedance analysis and the grip strength test, may help to differentiate the cause.

Declaration of Conflicting Interests

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Informed consent

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