

Aggravation of dysphagia after surgical removal of anterior cervical osteophytes: a case report

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Young-In Go¹ , Gi-Wook Kim^{1,2} ,
Yu-Hui Won^{1,2} , Sung-Hee Park^{1,2},
Myoung-Hwan Ko^{1,2} , Jeong-Hwan Seo^{1,2} and
Da-Sol Kim^{1,2}

Abstract

Dysphagia induced by anterior cervical osteophytes (ACOs) is frequently reported in older individuals. Surgical resection of ACOs is considered when conservative treatment fails, but its effectiveness is controversial owing to side effects after surgery. We present the case of a 78-year-old man who complained of progressive dysphagia that started 10 months previously. A videofluoroscopic swallow study (VFSS) showed prominent ACOs along C2–C6, which translocated the upper hypopharynx anteriorly, impinging the lumen and impairing epiglottic folding and laryngeal closure. Aspiration of a soft diet was observed. Despite conservative therapy, the symptoms persisted, and ACO resection surgery was performed. Unexpectedly, the patient's dysphagia worsened immediately post-surgery. A VFSS on postoperative day (POD) 2 showed improvement in epiglottic folding. However, prevertebral soft tissue swelling and dysfunction of opening of the upper esophageal sphincter newly arose. Laryngeal aspiration was observed during 5 cc and a large amount of liquid swallowing trials. The patient was provided a modified diet and rehabilitative dysphagia therapy. A VFSS on PODs 6 and 14 showed a gradual improvement in the prevertebral soft tissue swelling. This report suggests that a serial VFSS is effective for evaluating the different mechanisms of dysphagia and for devising an appropriate treatment plan.

¹Department of Physical Medicine & Rehabilitation, Jeonbuk National University Medical School, Jeonju, Republic of Korea

²Research Institute of Clinical Medicine of Jeonbuk National University – Biomedical Research Institute of Jeonbuk National University Hospital, Jeonju, Republic of Korea

Corresponding author:

Da-Sol Kim, Department of Physical Medicine & Rehabilitation, Jeonbuk National University Medical School, 20 Geonjiro, Deokjin-gu, Jeonju, Jeonbuk 54907, Republic of Korea.
Email: munoon@gmail.com



Keywords

Dysphagia, diffuse idiopathic skeletal hyperostosis, osteophyte, videofluoroscopic swallow study, surgery, rehabilitation

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Introduction

Anterior cervical osteophytes (ACOs) are degenerative changes in the cervical spine that frequently occur in 12% to 30% of older patients. ACOs cause ossification of the anterior cervical spine ligament. This condition is also called Forestier's disease and diffuse idiopathic skeletal hyperostosis.¹ In most cases, ACOs do not cause symptoms. However, when bony growth increases, complications such as dysphagia, stridor, and respiratory failure have been reported.¹

Although conservative treatment is the first therapeutic option for ACOs, surgical resection is recommended when conservative treatment fails. Some studies have reported the effectiveness of surgery.^{2,3} However, surgery remains controversial because of the possibility of intraoperative complications, such as prevertebral soft tissue swelling (PSTS) and intraoperative nerve and/or muscle injury.^{4,5} In addition, the recurrence of ACOs has been reported in patients who were followed-up for a long time after resection surgery.⁶

We report the case of a patient who initially complained of dysphagia induced by ACOs and who underwent cervical osteotomy after the failure of conservative treatment. Immediately after the surgery, a new pattern of dysphagia developed that was unusual, and a serial videofluoroscopic swallow study (VFSS) was performed for the diagnosis, treatment, and follow-up. In this report, the diagnostic efficacy of a serial VFSS and the different mechanisms

of dysphagia before and after surgery were analyzed.

Case presentation

This study conforms to the CARE guidelines.⁷ Institutional approval was obtained from the institutional review board (IRB) of Jeonbuk National University Hospital (IRB number: 2021-07-038). The requirement for the patient's consent for publication was waived by the IRB because no information was collected to identify the patient and because the report was based on existing data and documentation. The patient provided consent for evaluation and treatment performed during hospitalization.

A man in his mid-70s visited the Rehabilitation Medicine Department with the chief complaint of progressive dysphagia, which began approximately 10 months prior to his visit. He complained of foreign body sensation in the middle left part of his neck, particularly when swallowing thick materials. He had been diagnosed with tonsillar papilloma 16 years previously and had a history of excision surgery. At the time of the first visit to our department, the patient was alert, and showed no signs of cognitive dysfunction. At a physical examination, sensory and manual muscle test findings were normal, and a neurological examination showed no abnormal findings in cranial nerve function, deep tendon reflex, or pathologic reflex tests. There was no evidence of atrophy or fasciculation

of the tongue or limb muscles, and dysarthria was not observed.

Taking into account the patient's medical history, we suspected a recurrence of tonsillar papilloma and consulted the Otolaryngology Department at our hospital. However, no recurrence or any findings that could cause dysphagia were observed on laryngoscopy. Nevertheless, on a lateral cervical spine radiograph, 13.48-mm-thick ACOs were observed in the anterior portion of the C3–C4 cervical spine. Luminal impingement was induced by severe anterior translocation of the upper portions of the oropharynx and hypopharynx (Figure 1a). Cervical computed tomography was then conducted. ACOs were identified across the C2–C6 levels, and they were the most prominent at C3–C4 (Figure 1b and 1c). In a VFSS using diluted barium, in the pharyngeal phase, epiglottic folding was insufficient owing to the ACOs, and a grade 2 vallecular residue was observed (Figure 3a). The bolus did not completely enter the esophagus and passed through the

airway owing to luminal impingement at the entrance of the esophagus. The pharyngeal transit time was delayed to 5 s in the steamed rice swallow task (regular, International Dysphagia Diet Standardization Initiative [IDDSI] Level 7). Laryngeal aspiration was observed with rice porridge (minced and moist, IDDSI Level 5) and a large amount of thin liquid barium (slightly thin, IDDSI Level 1). Laryngeal penetration was observed with curd-type yoghurt (liquidized, IDDSI Level 3) and with 2 cc of thin liquid barium (Table 1).

Conservative treatment was administered, but the patient complained of progressively worsening dysphagia and a poor oral intake. We consulted with the Neurosurgery Department for an operation to perform ACO removal. The patient's C2–C6 ACOs were resected using an anterior approach. However, his dysphagia was aggravated immediately after surgery. He complained that the bolus was stuck in his throat and that he could not swallow.

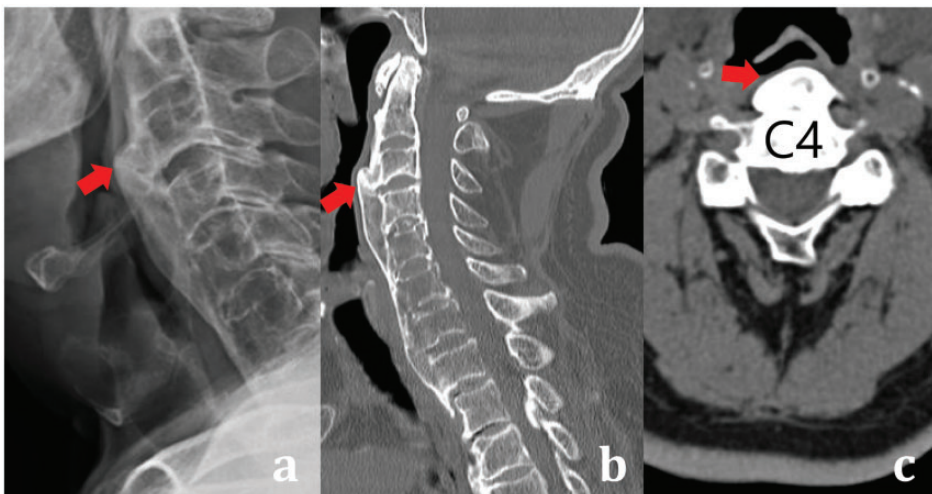


Figure 1. (a) Lateral cervical spine radiograph and (b) sagittal cervical spine computed tomography (CT) showing anterior cervical osteophytes from C2 to C6. The anterior cervical osteophytes at the C3 and C4 levels are the most prominent (arrows) and (c) Axial CT slices through C4 show that the posterior wall of the pharynx is anteriorly translocated by the anterior cervical osteophytes (arrow).

Table 1. Summary of results of the videofluoroscopic swallowing study.

Study time	PAS ^{a)}				Vallecular residue ^{b)}				Pyriform sinus residue ^{b)}			
	Pre	P2	P6	P14	Pre	P2	P6	P14	Pre	P2	P6	P14
Pudding	1	3	1	1	2	2	1	1	1	3	3	2
Rice porridge	5	3	2	1	2	1	1	1	1	3	2	1
Curd-type yogurt	2	3	2	1	1	1	1	1	1	2	1	1
Steamed rice	2	3	3	1	2	1	1	1	1	3	3	2
Thin liquid (2 cc)	2	3	3	2	1	1	1	1	1	1	1	1
Thin liquid (5 cc)	1	5	3	5	2	1	1	1	1	2	1	2
Thin liquid (large amount)	5	8	3	5	2	1	1	1	1	3	1	1

PAS, Penetration–Aspiration Scale; Pre, pre-operation; P2, postoperative day 2; P6, postoperative day 6; P14, postoperative day 14.

^{a)}Grade 1, material does not enter the airway; grade 2, material enters the airway, remains above the vocal folds, and is ejected from the airway; grade 3, material enters the airway, remains above the vocal folds, and is not ejected from the airway; grade 4, material enters the airway, contacts the vocal folds, and is ejected from the airway; grade 5, material enters the airway, contacts the vocal folds, and is not ejected from the airway; grade 6, material enters the airway, passes below the vocal folds, and is ejected into the larynx or out of the airway; grade 7, material enters the airway, passes below the vocal folds, and is not ejected from the trachea despite effort; and grade 8, material enters the airway, passes below the vocal folds, and no effort is made to eject.¹⁸

^{b)}Grade 0, no residue; grade 1, residue occupying < 25% of the height of the space; grade 2, 25% to 50% of the space; grade 3, > 50% of the space.¹⁹

No abnormal findings were observed on physical or neurological examinations. The previously observed ACOs on the lateral cervical spine radiograph had been removed. However, prominent PSTS with a thickness of 17.59 mm was observed at the C5–C6 levels (Figure 2a and b). A follow-up VFSS was performed on postoperative day (POD) 2. The previously observed narrowing of the oropharynx was resolved. However, narrowing of the hypopharynx due to the marked PSTS and dysfunction of opening of the upper esophageal sphincter were observed at the C5–C6 levels. The pharyngeal transit time during the steamed rice swallowing task increased to 9 s. Previously observed impairment of epiglottic folding showed improvement, and the vallecular residue improved to grade 1 with all materials. However, the pyriform sinus residue was aggravated at grades 2 to 3 (Figure 3b and Table 1). Laryngeal aspiration was observed with 5 cc of thin liquid, and a large amount of thin liquid barium, and

penetration was observed with other test materials (Table 1). The patient received rehabilitative dysphagia therapy for 15 minutes each day for 5 days and was recommended to take pureed, homogenous, and cohesive foods, as well as thickened liquid. A follow-up VFSS was performed on POD 6. Only penetration was observed with 5 cc of thin liquid and a large amount of thin liquid, with which massive aspiration was previously observed. The pyriform sinus residue decreased, particularly when a large volume of thin liquid barium was used (Figure 3c and Table 1). In a VFSS performed on POD 14, the laryngeal penetration previously seen with rice porridge, curd-type yoghurt, and steamed rice was improved (Table 1). In addition, the pyriform sinus residue was improved to grades 1 to 2 with all materials (Figure 3d and Table 1). The pharyngeal transit time was markedly improved to 1.2 s, and PSTS was reduced to 11.49 mm (Figure 2c). One month after discharge, the patient underwent a follow-up VFSS while

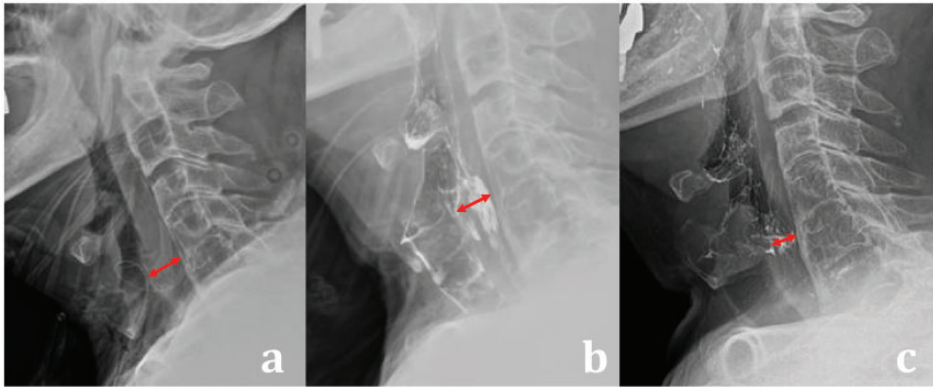


Figure 2. Lateral cervical spine radiographs obtained on postoperative days 1 (a), 2 (b), and 14 (c). Prevertebral soft tissue swelling (bidirectional arrows) shows improvement in panel c compared with panels a and b.

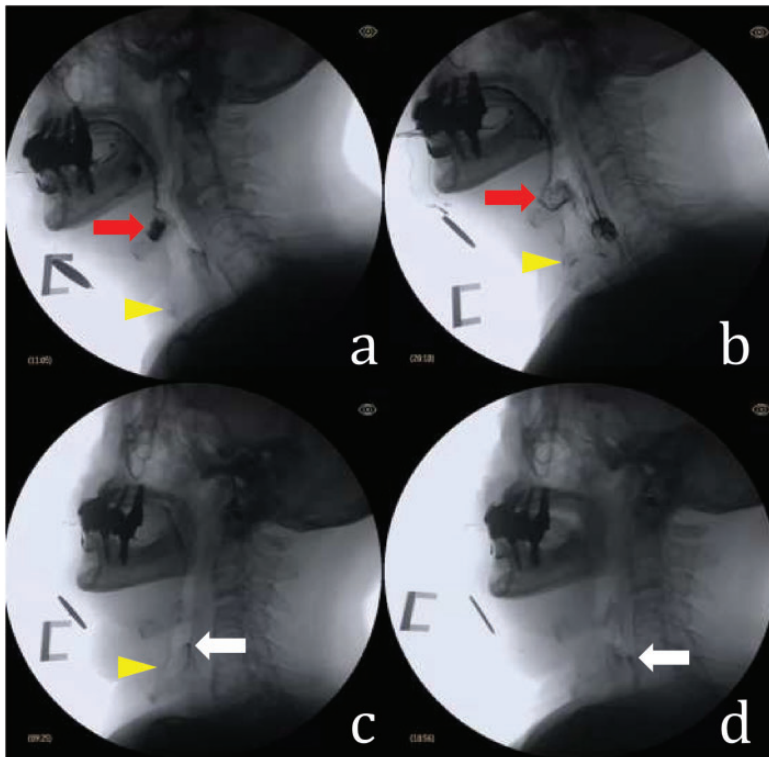


Figure 3. Videofluoroscopic swallowing study performed before (a) and 2 (b), 6 (c), and 14 (d) days after surgery. (a) Grade 2 vallecular residue (red arrow) and laryngeal aspiration (arrowhead) of a large amount of thin liquid is seen. (b) Improvement of vallecular residue (red arrow) and laryngeal aspiration (arrowhead), and aggravation of pyriform sinus residue (white arrow). (c) Improvement of pyriform sinus residue (white arrow) and laryngeal penetration (arrowhead) and (d) Improvement of pyriform sinus residue (white arrow).

maintaining a minced and moist diet. However, he was lost to follow-up in the rehabilitation medicine outpatient clinic thereafter. He visited the neurosurgery outpatient clinic 6 months later and his clinical symptoms were improved.

Discussion

A main complaint of symptomatic patients with ACOs is dysphagia. Dysphagia occurs in 92% of symptomatic patients and is the most frequent complaint, followed by dyspnea (20%) and neck pain (12%).⁸ Dysphagia occurs most frequently at the C3–C4 levels, followed by the C4–C5 and C5–C6 levels because at the C5–C6 or lower levels, the soft tissue between the anterior cervical spine and the esophagus is wide and has free space. Therefore, the mass effect is less on the surrounding structures despite ACO growth.² C3–C5 level ACOs mainly interfere with epiglottic folding and cause incomplete laryngeal closure. Therefore, direct aspiration occurs as the bolus enters the open larynx along the inclined surface of osteophytes. C5–C7 level ACOs induce post-swallowing retention of residue, resulting in delayed aspiration.^{2,9} In addition, esophageal fixation occurs when small osteophytes are generated at the level where the cricoid cartilage is located. Inflammation of periesophageal or peripharyngeal tissue due to repeated irritation by osteophytes, and cricopharyngeal and esophageal spasms due to pain caused by osteophytes are the main pathological mechanism of dysphagia.^{3,10,11} In the present case, the osteophytes protruded most prominently at the C3–C4 levels. The consequent impairment of epiglottic folding and the direct influx of the bolus into the open larynx might have been the primary cause of the patient's dysphagia. Conservative treatment is preferred for ACOs. Anti-inflammatory agents, steroids, analgics, and muscle relaxants can be used

in combination with a modified diet for ACOs.^{11,12} However, our patient showed severe aspiration during the VFSS, his clinical symptoms continued to worsen, and his oral intake was reduced. Therefore, we decided to refer the patient to a neurosurgeon for removal of the ACOs.

The incidence of dysphagia after anterior cervical spine surgery has been reported to reach 29.8%, 6.9%, and 6.6% at 3, 6, and 24 months, respectively.¹³ The causes of postoperative dysphagia include recurrent laryngeal nerve and superior laryngeal nerve injury, soft tissue edema secondary to intraoperative retraction of the pharynx or esophagus, ischemic injury, and reperfusion injury of the pharynx and esophagus due to prolonged retraction followed by release.^{4,5} According to previous reports, the prognosis after surgical resection for ACOs is positive. Von der Hoeh et al. reported that dysphagia in six patients improved after surgery without any complications.¹⁴ Ruetten et al. reported that 14 patients who underwent ACO resection surgery showed a large improvement in dysphagia symptoms, without recurrence or complications during a 50-month follow-up.³ However, our patient experienced postoperative aggravation of dysphagia. The surgery was successful because the incomplete epiglottic closure and retention of vallecular residue appeared to have improved in the postoperative VFSS. However, the postoperative VFSS newly showed PSTS at the hypopharynx level. The retention of pyriform sinus residue was caused by impairment of opening of the upper esophageal sphincter, and laryngeal aspiration was caused by reflux of this residue. This newly detected PSTS due to retraction of the trachea and esophagus during surgery might have caused postoperative aggravation of dysphagia.

Treatments of PSTS include the following: (i) modifying the diet, (ii) thermal

tactile stimulation before swallowing if there is a delay in triggering the pharyngeal swallow, (iii) the supraglottic swallow and/or effortful swallowing, (iv) postures such as chin tuck, head tilt, or rotation to the relatively intact side of the pharynx, and (v) exercises to improve pharyngeal function (e.g., Mendelson maneuver and Masako maneuver).^{4,15} Immediately after surgery, the patient was provided a modified diet, taught to perform a chin tuck when swallowing, and had tactile stimulation therapy for 15 minutes/day for 5 days. Additionally, he was provided training on the effortful swallow, Mendelson maneuver, and Masako maneuver.

According to Sanfilippo et al.,¹⁶ PSTS persists for up to 2 weeks after surgery and shows improvement after 6 weeks. Kim et al.¹⁷ investigated 160 patients who underwent anterior cervical spine surgery. They found that the mean thickness of PSTS at the C6 level was 14.43 mm (13.96–14.91 mm) before surgery, 19.18 mm (18.59–19.77 mm) immediately after surgery, and 17.92 mm (17.37–18.47 mm) at 2 weeks after surgery. In these studies, rehabilitative dysphagia therapy was not administered. However, dysphagia therapy was performed in our patient. Consequently, the patient had PSTS of 17.59 mm on POD 2, which was markedly improved to 11.49 mm on POD 14. In addition, the follow-up VFSS performed on POD 14 showed that the pyriform sinus residue had markedly improved. However, laryngeal aspiration was still observed during the thin liquid barium task. The subjective symptoms that the patient initially complained of were also considerably improved.

This report has the following limitation. A follow-up examination was scheduled within 1 month, at which we planned to adjust the patient's diet on the basis of the results. However, he did not show up to our rehabilitation medicine outpatient clinic,

and the treatment could not be terminated. However, after 6 months, the patient's subjective symptoms of dysphagia were improved as shown by neurosurgery outpatient records, and an improvement in prevertebral soft tissue was shown by a lateral cervical radiograph.

Conclusion

In this patient, dysphagia induced by ACOs was characterized by a reduction in epiglottic folding and by space narrowing of the oropharynx and hypopharynx by mechanical compression. Additionally, elongation of the pharyngeal transit time due to PSTS and an upper esophageal sphincter opening disorder were observed after surgery. By performing a serial VFSS over a short period, we were able to objectively and radiologically confirm the altered symptoms of dysphagia before and after the surgery. On the basis of the study results, we were able to provide the patient with an appropriate diet and rehabilitative dysphagia therapy guidelines. We were also able to further follow-up on the patient's clinical course.

Data availability

The data that support the findings in this study are available from the corresponding author on request.

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Declaration of conflicting interest

The authors declare that there is no conflict of interest.

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Author contributions

YI Go performed the videofluoroscopic swallow study and contributed to writing a draft of the manuscript; GW Kim, YH Won, and SH Park contributed to data analysis; YI Go, MH Ko, JH Seo, and DS Kim contributed to the overall diagnosis and treatment of the patient; DS Kim was responsible for revision of the manuscript and intellectual content. All authors approved the final version of the manuscript to be submitted.

ORCID iDs

Young-In Go  <https://orcid.org/0000-0002-5453-218X>

Gi-Wook Kim  <https://orcid.org/0000-0002-1628-8382>

Yu-Hui Won  <https://orcid.org/0000-0003-2007-9652>

Myoung-Hwan Ko  <https://orcid.org/0000-0002-0566-3677>

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