



Recurrent esophageal perforation after anterior cervical spine surgery: case report

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Abstract

Purpose Delayed esophageal perforation after anterior cervical discectomy and fusion (ACDF) is an extremely rare cause of infection such as spondylodiscitis. We present a rare case in which a patient had two delayed esophageal perforations occurring 20 and 25 years after ACDF. By sharing our experience of this rare case, we hope to provide new information related to delayed esophageal perforation.

Methods We present the case of a 72-year-old patient who underwent ACDF due to cervical spondylosis 25 years ago. Delayed esophageal perforation occurred 20 years postoperatively and healed spontaneously with conservative treatment.

Results Five years later, a second esophageal perforation occurred, which required surgical intervention and involved recurrent infection.

Conclusions We suggest that it is important to consider follow-up in patients with spontaneously healed esophageal perforations. Furthermore, any patient with symptoms subsequent to a spontaneously healed esophageal perforation, even after an interval of several years, should receive a thorough evaluation for possible recurrent esophageal perforation.

Keywords Anterior cervical discectomy and fusion · Esophageal perforation · Infection · Late complication

Introduction

Complications of anterior cervical discectomy and fusion (ACDF) include injuries to the trachea, esophagus, recurrent laryngeal nerve and ca-rotid artery [1, 2]. Esophageal perforation is a potentially life-threatening complication and may develop either in the early postoperative period or years after surgery. Delayed esophageal perforation after ACDF is an extremely rare cause of infection such as spondylodiscitis [3–7]. By sharing our experience of this rare case, we hope to provide new information related to delayed esophageal perforation.

Case report

A 66-year-old male patient was admitted to our hospital with a 3-month history of shoulder pain and progressive quadriplegia. He had undergone ACDF on C4–6 at another hospital in 1992, 20 years prior to admission. Neurological examination revealed that muscle strength of the upper and lower extremities was 4/5. Cervical radiographs showed an absence of the right C6 screw (Fig. 1a). MRI revealed spondylitis between spinal levels C4 and T1, with associated epidural abscess (Fig. 1b). Because spondylitis can be caused by a dislodged screw, an esophagogram and esophagoscopy were performed to confirm esophageal perforation by a dislodged screw. An esophagogram using gastrografin contrast showed no sign of contrast leakage (Fig. 2b). However, during esophagoscopy, a small opening was noted in the scar from the previously healed esophageal perforation below the upper esophageal sphincter at the C5–6 level where the missing screw was supposed to be located (Fig. 2b). We theorized that a delayed esophageal perforation occurred due to the loosened right C6 screw, leading to spondylitis. Because his paralysis was not severe and the fistula was healed, we decided to monitor his progress. Conservative treatment was

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Fig. 1 Cervical imaging of a patient who had undergone anterior cervical spinal fixation 20 years earlier. **a** Cervical radiographs showed an absence of the right C6 screw. **b** Gadolinium-enhanced MRI demonstrates vertebral body enhancement and peripheral enhancement of the C6/7 disc space. And Gadolinium-enhanced MRI shows homogeneous epidural phlegmon enhancement extending from C4 to T1 and prevertebral space enhancement

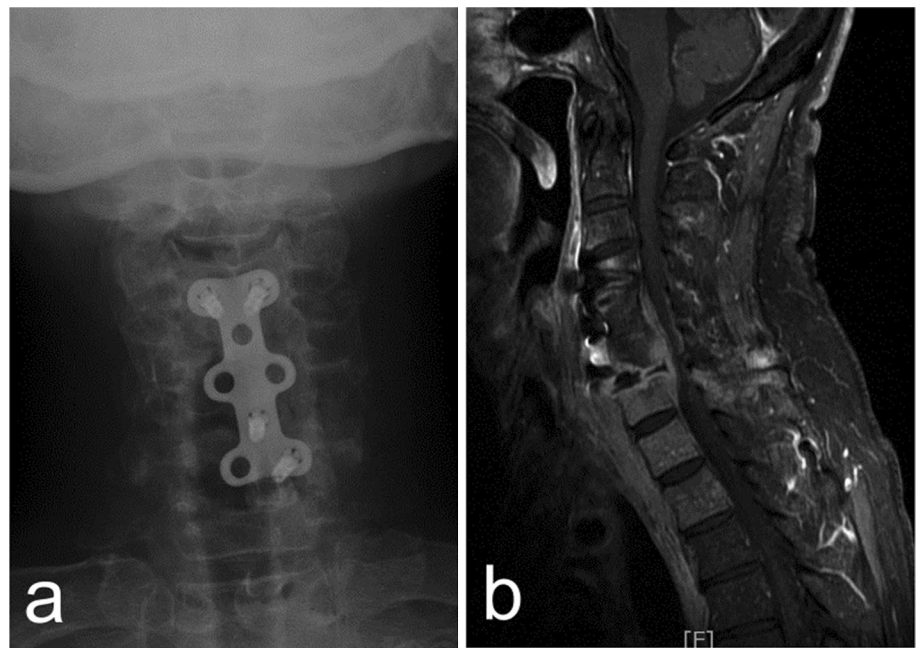
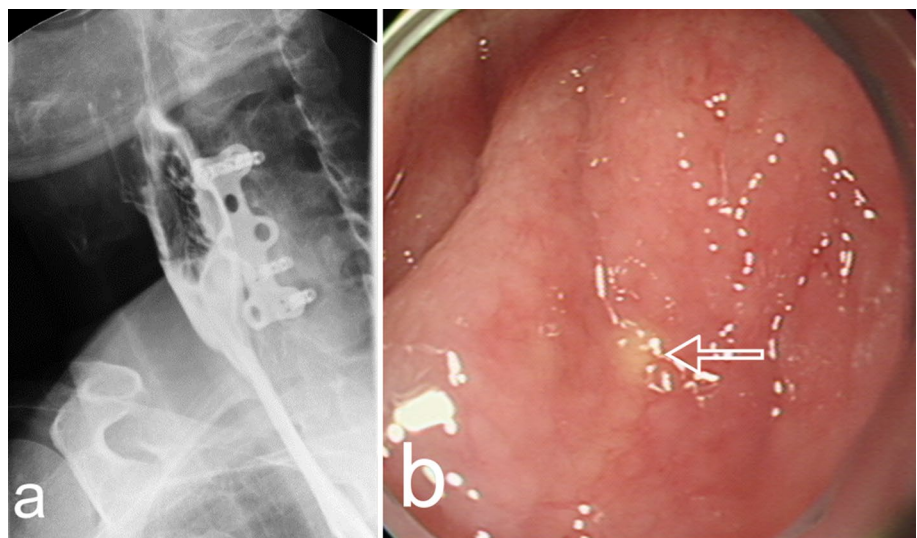


Fig. 2 Radiological studies were carried out to investigate esophageal perforation. **a** Esophagogram using gastrografin contrast showed no sign of contrast leakage. **b** Esophagoscopy showed that a small opening in the scar from the previously healed esophageal perforation was observed below the upper esophageal sphincter, at the C5–6 level, where the missing screw is supposed to be located (arrow)



started with intravenous antibiotics and 3 weeks later follow-up cervical MRI showed improvement of the spondylitis.

Five years later, the patient was again admitted to our emergency department with a 2-day history of rapidly progressive quadriparesis. Physical examination revealed upper and lower extremity strength to be 1/5. Cervical MRI revealed a protruding disc at the C3/C4 causing indentation of the cervical spinal cord (Fig. 3a). On admission, the patient had a low-grade fever and lab work showed a markedly increased erythrocyte sedimentation rate (ESR) of 59, and a C-reactive protein (CRP) of 15.64. Initially, we felt these findings were due to aspiration pneumonia. Emergency surgery was planned and no other studies were done prior to surgery. The patient underwent ACDF at the

C3–4 level. During surgery, when we opened the posterior longitudinal ligament (PLL), whitish pus gushed out. After discectomy and thorough abscess debridement, a Cervios cage was placed in the disc space. Surgical specimen cultures revealed *Streptococcus alactolyticus*, and the patient was treated with appropriate antibiotics. Contrast-enhanced cervical MRI was performed postoperatively and showed an epidural and paravertebral abscess associated with osteomyelitis at the C3–4 (Fig. 3b). An esophagogram showed an extravasation of the contrast at C6 level (Fig. 4a). Esophagoscopy revealed a defect of the posterior wall of the esophagus with exposure of the plate, and an esophageal perforation was found just below the upper esophageal sphincter, which was the location of the previous perforation (Fig. 4b).

Fig. 3 **a** On cervical spine MRI, a protruding intervertebral disc, from the C3/C4 intervertebral space, causing indentation of the cord was detected. **b** Contrast-enhanced cervical spine MRI exhibited an epidural and paravertebral abscess associated with osteomyelitis at the C3–4 level

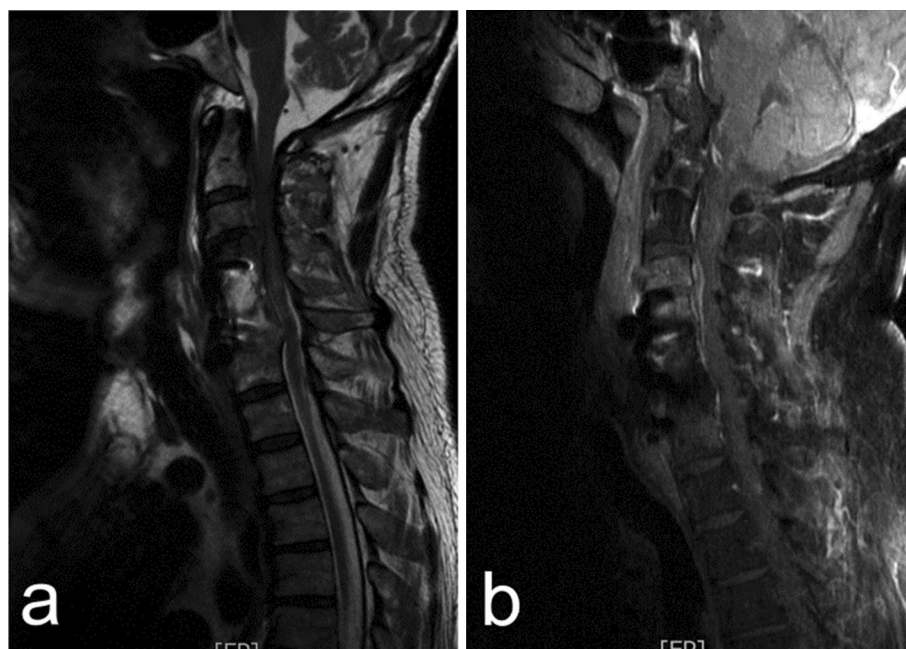
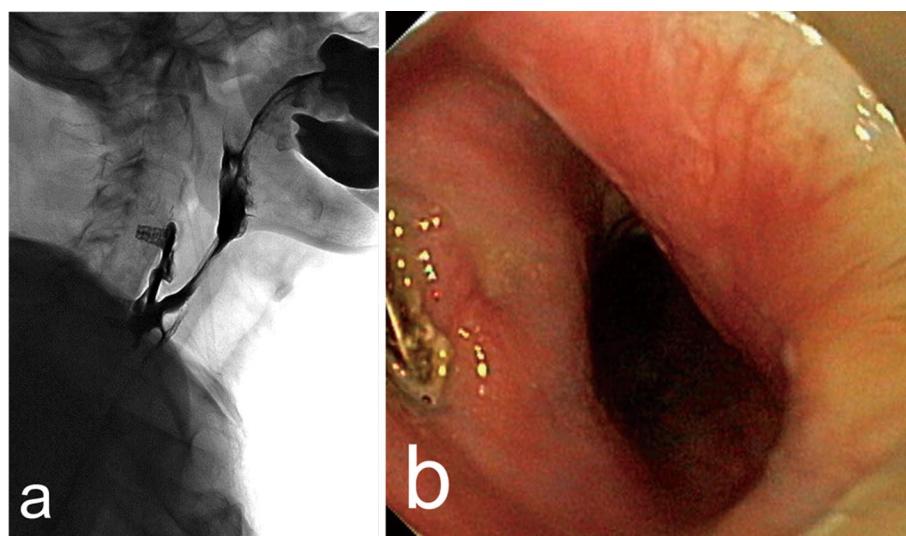


Fig. 4 **a** Esophagogram showed esophageal extravasation of the contrast at the level of the C6 vertebra. **b** Esophagoscopy revealed a defect of the posterior wall of the esophagus with exposure of the ventral plate (from previous surgery), and an esophageal perforation was found just below the upper esophageal sphincter, which was the location of the previous perforation



Based on the diagnosis of recurrent esophageal perforation, the patient was scheduled for surgery. A neck dissection were performed by the ENT surgeon. An esophageal perforation was found and a direct primary closure using 4-0 vicryl suture was done by a thoracic surgery team (Fig. 5). Removal of the previous plate at C4–6 followed in a second step. We then performed an iliac bone graft with anterior plating at C3–4, instead of the previous cage. No esophageal leakage could be found on either the postoperative esophagogram or on endoscopy done on postoperative day 2. Two months post-operative MRI showed epidural and prevertebral space enhancement improved compared with previous MRI (Fig. 6a). Postoperative cervical spine radiographs at 8 months demonstrated no loosening sign of instrumentation

and no complications (Fig. 6b). At the 1-year follow-up, the patient had no complaints, and he could walk without assistance.

Discussion

Delayed esophageal perforation may occur due to various mechanisms. In one review article, delayed esophageal perforation following ACDF was found to be primarily associated with hardware failure, such as plate or screw migrations. The next most common cause of perforation was chronic compression and repetitive friction between the posterior wall of the esophagus and adjacent surgical devices [7]. In

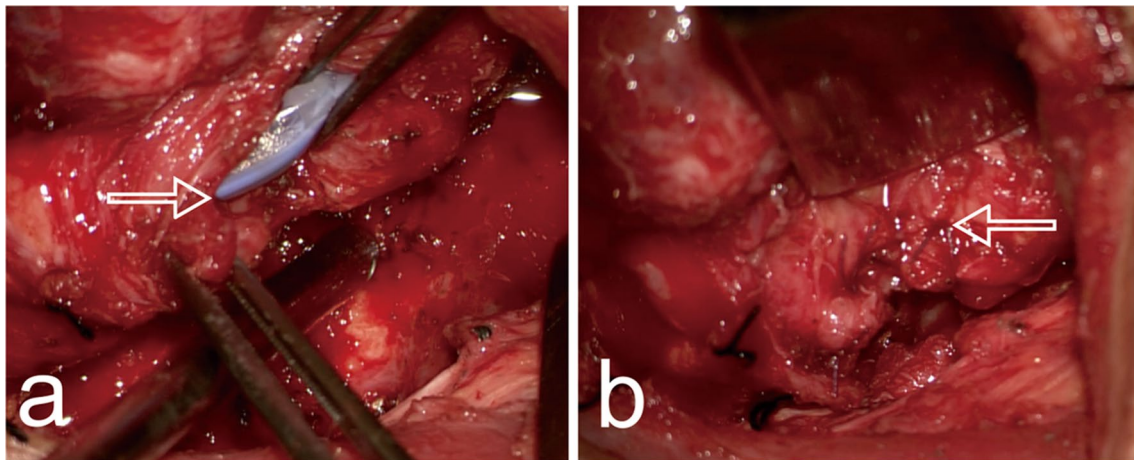


Fig. 5 **a** Esophageal perforation and nasogastric tube are revealed in the intraoperative view. **b** Primary closure of the esophageal mucosa and overlying muscle layer is performed with absorbable and non-absorbable interrupted suture

Fig. 6 **a** Gadolinium-enhanced MRI at the 2-month follow-up showed epidural and prevertebral enhancement improved compared with previous MRI. **b** Postoperative cervical spine radiographs at 8 months



our report, the patient's first delayed esophageal perforation, due to a dislodged screw, was diagnosed 20 years after ACDF. Migrated screws often follow a benign course, due to the small diameter of the screws and the slow migration of the screw from the external to the internal mucosa of the esophagus, which permits spontaneous healing of the defect [8]. We theorize that the reason for our patient's first delayed esophageal perforation might have been due to direct injury by a migrated screw and that the migrated screw might have been eliminated through the gastrointestinal tract. In contrast, we suggest that the reason for his second esophageal perforation might have been chronic esophageal compression by a plate at the point of previous healing. Therefore,

we discussed possible mechanisms for the cause of recurrent delayed esophageal perforation, and theorize that different mechanisms were responsible for the two recurrences in our patient.

To date, there are no obvious standard treatment strategies for delayed esophageal perforation, requiring case by case evaluation to choose either conservative treatment or primary closure with complex reconstruction [9, 10]. In this report, we present a rare case of a patient with two delayed esophageal perforations occurring 20 and 25 years after ACDF. In our patient's first perforation episode, despite the presence of spondylodiscitis with abscess formation, the perforation healed, and he had mild neurological symptoms

which improved. Therefore, we chose conservative treatment. In our report, the patient's second esophageal perforation occurred 5 years after the first esophageal perforation was healed. The emergent nature of his presentation prevented us from performing any further evaluations prior to surgery. In addition, we considered the fever to be a symptom of pneumonia, and did not recognize spondylitis caused by a repeated esophageal perforation. This is a reminder that even if esophageal perforation has healed spontaneously, there is a possibility of perforation recurrence years later. We recommend that conservative management is a reasonable option for patients with healed esophageal perforation. However, surgeons should keep in mind that healed esophageal perforation can recur subsequently.

Conclusions

We present a rare case of a patient with two delayed esophageal perforations occurring 20 and 25 years after ACDF. In light of our experience with this patient, we suggest that it is important to consider follow-up in patients with spontaneously healed esophageal perforations. Furthermore, any patient with symptoms subsequent to a spontaneously healed esophageal perforation, even after an interval of several years, should receive a thorough evaluation for possible recurrent esophageal perforation.

Compliance with ethical standards

Conflict of interest None of the authors has any potential conflict of interest.

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