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EFORT Open Reviews (2025) **10** 57–63
https://doi.org/10.1530/EOR-24-0110

SPINE

Esophageal perforation more than 10 years after anterior cervical spine surgery: a case report and literature review

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- Esophageal perforation is a rare but serious complication that can occur post-cervical spine surgery. This case report presents the clinical course, diagnostic challenges and management strategies of a patient who had a late-diagnosis esophageal perforation after anterior cervical spine surgery (ACSS).
- A woman in her 50s underwent ACSS for cervical spondylosis. Three months postoperatively, she experienced persistent right neck and shoulder pain. Despite multiple consultations, an esophageal perforation was only diagnosed 10 years later when a neck mass ruptured, discharging food debris. Surgical management included removing the anterior cervical plate and reconstruction with a sternocleidomastoid muscle flap. Postoperatively, she faced wound complications, and the perforation failed to heal despite multiple debridement and stent placements. Ultimately, complete excision of the diverticulum, repair of the perforation and muscle flap reconstruction led to her recovery, with no recurrence over an 8-year follow-up.
- We reviewed the literature on cases with esophageal perforation occurring more than 10 years after anterior cervical surgery and summarized the treatment experiences.
- This case underscores the diagnostic challenges and delayed presentation of esophageal perforation post-ACSS. Early recognition and multidisciplinary management are essential. In cases of late perforation, hardware removal, diverticulum excision and a muscle flap are critical to achieving successful closure of the esophageal lesion, preventing recurrence and ensuring comprehensive repair. Addressing esophageal diverticula during perforation treatment is crucial to prevent recurrence and ensure thorough repair. This highlights the need for high clinical suspicion and a coordinated surgical approach to improve patient outcomes.

Keywords: esophageal perforation; anterior cervical surgery; complication; esophageal diverticula

Introduction

Since the 1950s, physicians and scholars have adopted anterior cervical spine surgery (ACSS) in clinical practice. This surgical approach has gained widespread attention because of its high safety, minimal invasiveness, ease of operation, short operation time, low bleeding and quick recovery (1). With the advancement of clinical practice, anterior cervical surgery has continuously improved and has been widely used in the treatment of cervical trauma, cervical spondylosis, cervical tumors and cervical tuberculosis, making it one of the primary methods for cervical surgery (2, 3). However, as the surgery became more prevalent and the number of cases increased, the

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https://doi.org/10.1530/EOR-24-0110

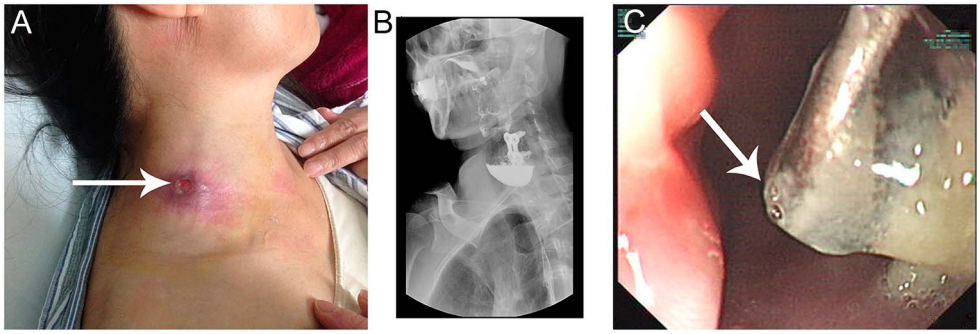

Figure 1

(A) Ruptured neck mass (indicated by white arrows). (B) Esophagogram showing extraluminal contrast extravasation into the cervical spine soft tissues and formation of an esophageal diverticulum. (C) Endoscopic view revealing a 5 cm defect in the posterior wall of the esophagus (indicated by the white arrow), 15 cm from the incisors, with visible hardware and distal esophageal stenosis.

postoperative complications associated with anterior cervical surgery have gradually drawn the attention of medical professionals and been reported (4, 5). Among these, esophageal perforation following anterior cervical surgery is a rare but serious complication, first reported by Balmaseda in 1985, with an incidence rate ranging from 0.04 to 1.62% (6). Although the occurrence rate of this complication is low, its potential consequences can be extremely severe. In mild cases, it may lead to incision infection and difficulty swallowing; in severe cases, it can cause mediastinal infection, pulmonary infection, sepsis and other serious complications, potentially life-threatening, with a mortality rate as high as 20% (7, 8).

This study reviews the literature on esophageal perforation occurring more than 10 years after anterior cervical surgery. Using the keywords 'esophageal perforation' and 'cervical', a search was conducted in the PubMed database for studies published up to June 2024. A total of 12 relevant articles were identified. These articles were evaluated based on the type of anterior cervical surgery performed, the timing of diagnosis, the size of the perforation, treatment methods, duration and treatment outcomes. The summary of the reviewed cases is presented in Supplementary Table 1 (see section on Supplementary materials given at the end of the article).

Case report

A 50-year-old female underwent anterior cervical corpectomy and fusion at the C6 level in 2004 because of cervical spondylosis. Postoperatively, the incision healed well. However, 3 months after the surgery, she began experiencing persistent pain in the right neck and shoulder, accompanied by mild dysphagia, significantly affecting her quality of life. Despite multiple hospital visits, the pain did not improve. Additionally, the patient experienced mild dysphagia, raising concerns about potential esophageal perforation. Six months post-surgery, an esophagogram suggested a possible esophageal perforation, but an upper GI endoscopy did not reveal any esophageal damage, leading to the diagnosis being dismissed. Over the next 10 years, the patient repeatedly sought medical attention for severe right neck and shoulder pain, accompanied by mild dysphagia, yet a definitive diagnosis was never made. Finally, 10 years post-surgery, a neck mass ruptured, discharging food debris (Fig. 1A). An esophagogram revealed extraluminal contrast extravasation into the cervical spine soft tissues and the formation of an esophageal diverticulum (Fig. 1B). Upper GI endoscopy showed a 5 cm defect in the posterior wall of the esophagus, 15 cm from the incisors, with visible hardware and distal esophageal stenosis (Fig. 1C), confirming the diagnosis of an esophageal perforation. The patient was put on nil per os (NPO), and jejunal nutrition was established.

During surgery, a large esophageal diverticulum with a significant rupture was found. The anterior cervical plate was removed, and with the assistance of a thoracic surgeon, a sternocleidomastoid muscle flap with skin transfer was performed, with the skin side sutured to the esophageal mucosa (Fig. 2A and B). Postoperatively, the patient continued the NPO status. On the fifth postoperative day, the wound was swollen and exudative, which improved with dressing changes. One month post-surgery, the patient resumed drinking water and consuming liquids, but the wound became swollen and exudative again. Despite 20 days of continued dressing changes, the wound did not heal. Upper GI endoscopy (Fig. 2C) and an esophagogram (Fig. 2D) still showed the presence of the esophageal diverticulum, necessitating debridement and sinus tract drainage surgery. After two more months of dressing changes, the wound remained unhealed, leading to

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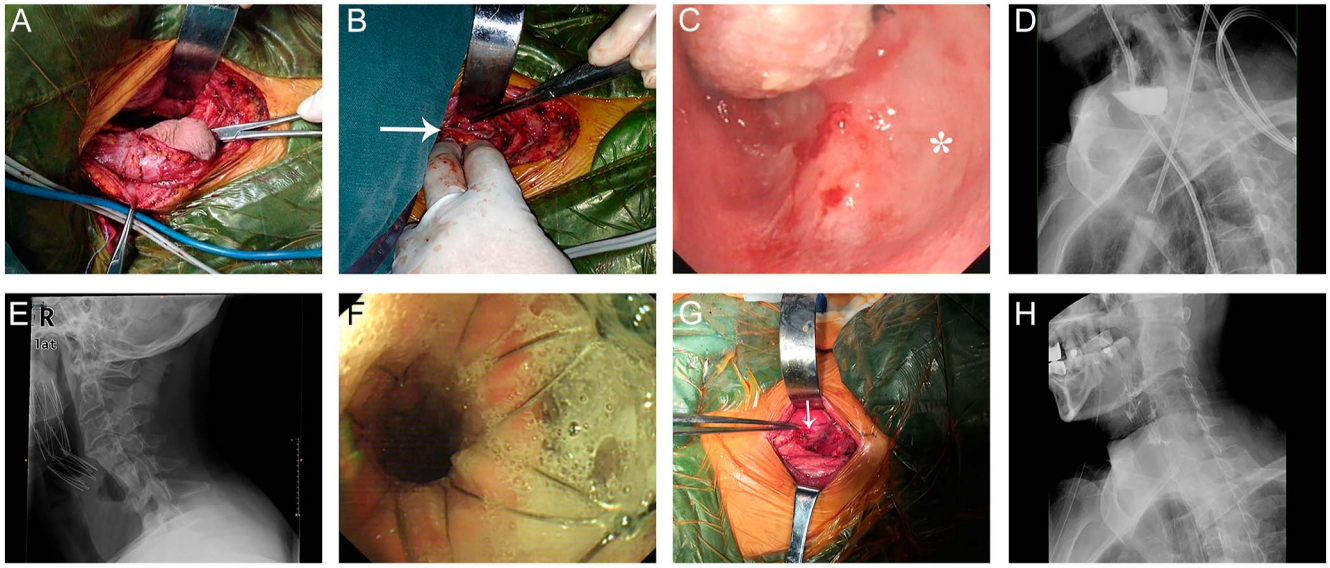

Figure 2

(A and B) Sternocleidomastoid muscle flap transfer with the skin side sutured to the esophageal mucosa (indicated by white arrows). (C) Endoscopic view post-initial surgery showing persistent esophageal diverticulum (indicated by *). (D) Esophagogram post-initial surgery, confirming the presence of the esophageal diverticulum. (E) X-ray after the first esophageal stent placement. (F) Endoscopic view of the second esophageal stent placement. (G) Final surgical intervention involving the excision of the esophageal diverticulum, esophageal reconstruction and SCM insertion between the esophagus and cervical spine (indicated by white arrows). (H) Postoperative esophagogram showing a significant reduction of the esophageal diverticulum.

the placement of an esophageal stent by a senior gastroenterologist (Fig. 2E). One month later, the patient experienced significant throat obstruction. A follow-up upper GI endoscopy revealed that the esophageal stent had dislodged, prompting its removal and continuation of the NPO status. In April 2015, the patient resumed consuming water and liquids, transitioning to porridge after 10 days. However, the neck wound became swollen again, which subsided with fasting. After further symptomatic treatment including dressing changes and fasting, the patient received another esophageal stent on May 27, 2015 (Fig. 2F). Despite 4 months of dressing changes, the neck wound remained swollen and unhealed. Two esophageal repair surgeries were performed by a senior thoracic surgeon on October 26 and November 6, 2015, but both were unsuccessful, resulting in persistent neck swelling, incision breakdown and pain. Finally, in August 2016, the patient underwent excision of the esophageal diverticulum and esophageal reconstruction, and the SCM flap without skin was inserted between the suture site of the esophagus and the cervical spine by a thoracic surgeon (Fig. 2G). The drainage tube was removed after 5 days, and the patient began drinking water 10 days post-surgery, followed by a transition to liquid and soft foods over the next 2 weeks. The wound remained well-healed, and the patient was discharged. A postoperative esophagogram showed significant reduction of the esophageal diverticulum (Fig. 2H). An 8-year follow-up indicated good wound healing with no recurrence of the esophageal fistula, and the patient's right arm pain had resolved.

Discussion

Early esophageal perforations after anterior cervical surgery are often caused by improper placement or displacement of sharp retractors within the esophagus (9). Given the narrow surgical field of anterior cervical procedures, iatrogenic esophageal injuries can be easily overlooked. In contrast, late esophageal perforations are typically induced by chronic compression or contact, leading to necrosis and abscess formation due to graft or screw displacement, sometimes accompanied by plate fractures (10, 11). Unlike early esophageal perforations, which commonly present with pain, dysphagia, coughing, aspiration and voice changes, delayed esophageal erosions may require a high index of suspicion for diagnosis, as many patients may progress over an extended period without exhibiting any signs or symptoms (12). Ikuma and coworkers suggest that the asymptomatic nature of some perforations may be because of chronic inflammation and tissue necrosis caused by microtraumatic pressure ulcers from metal implants on the posterior esophageal wall, leading to strong adhesions around the perforation site (13). These adhesions prevent significant leakage of food or saliva. Over the years, as more metal becomes exposed within the esophagus, the esophageal lumen may become

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almost entirely occupied by the implant, eventually causing dysphagia.

Esophageal perforation has a high mortality rate, making its proper management critically important. For patients with incidentally discovered and asymptomatic perforations, conservative treatment, including NPO, nasogastric or percutaneous endoscopic gastrostomy tube for temporary enteral nutrition and broad-spectrum intravenous antibiotics, may be appropriate. These injuries can spontaneously heal within 4–12 weeks (14). However, this conservative approach is associated with a 20–45% risk of abscess formation (15). Surgical treatment provides definitive management. The SCM flap is considered useful for achieving and maintaining perforation closure because without it, esophageal sutures invariably fail (16). The SCM is highly mobile and well-vascularized, which enhances antibiotic delivery, promotes wound healing and provides a physical barrier between the esophagus and cervical spine (11, 15). Successful management of esophageal perforation depends on the prompt recognition of symptoms and immediate intervention. Intraoperative simple suturing of the perforation can resolve the issue. If the perforation is identified within the first hour or days postoperatively, an urgent return to the operating room for repair is mandatory. Later-diagnosed perforations with abscess formation and compromised esophageal edges can be similarly managed. Larger defects may require reconstruction using muscle flaps such as the SCM or the omohyoid muscle flap. The reconstruction phase involves direct multilayer (at least including mucosal and muscular layers) interrupted inverting sutures using absorbable 2-0 or 3-0 sutures to reduce the risk of pharyngoesophageal stricture post-pharyngoesophageal perforation (PEP). In all cases of delayed PEP, early PEP with large fistulas and intraoperative issues with tissue quality, a muscle flap, such as the SCM or pectoralis major, should be inserted between the sutured pharyngoesophageal wall and the vertebral plane after direct suturing. The SCM flap, commonly used, can be fashioned as an ascending or descending pedicle flap based on branches of the occipital artery or the thyrocervical trunk respectively.

Based on the advice of previous research studies, early esophageal perforation with small openings and mild clinical presentations can achieve healing through fasting and anti-infection treatment (17, 18). Of the 13 patients we included (shown in Supplementary Table 1) (5, 9, 13, 19, 20, 21, 22, 23, 24, 25, 26, 27), only three did not undergo esophageal repair. One case, reported by Park and coworkers, involved a patient who developed a small esophageal perforation 20 years after anterior cervical discectomy and fusion (ACDF) (25). The initial perforation was treated conservatively over a period of 3 months and healed spontaneously although the patient developed mild neurological symptoms. Five years later, the patient experienced rapidly progressive quadriparesis. This was attributed to an epidural abscess caused by the recurrent esophageal perforation, which led to spinal cord compression. A subsequent upper GI endoscopy revealed a recurrent esophageal perforation, necessitating surgical removal of the internal fixation device and esophageal repair. The other two cases, reported by Vrouenraets *et al*. (26) and Yahanda *et al*. (5), achieved healing through conservative management with drainage placement after perforating the esophagus, but both required removal of the internal fixation devices (5, 26). Besides, in the case in Vrouenraets *et al.* (26), the esophageal mucosa was vital, and there was no pus retention. For the other patients, esophageal repair was performed. Among them, six did not have muscle flap coverage after the esophageal repair (9, 13, 19, 21, 25, 27), resulting in two failures and one death due to pneumonia. Therefore, we recommend removing internal fixation devices even for small defects. Prolonged contact between the fixation device and the esophagus can cause further damage, converting small perforations into larger ones, thus increasing treatment complexity and costs. Small esophageal defects, generally less than 1 cm in diameter (28, 29), should be primarily sutured if there is sufficient mucosal tissue. Local flap placement after primary perforation repair is essential to provide adequate healing support (30, 31). Unreinforced primary repair increases healing time and delays the resumption of oral intake (28) while also raising the risk of needing re-operation or secondary esophageal repair (32).

CT and X-ray scans showed that the entire structure had fused well, permitting the safe removal of internal fixation without concerns about spinal instability or the need for anterior or posterior fixation. Although our review included patients who had undergone cervical spine surgery more than 10 years ago, two patients still underwent posterior fusion to further stabilize the spinal structure. In one case, Harman *et al*. performed an ACDF at C4–5 and posterior fixation during the same operation because of adjacent level disease. This was done immediately after removing the internal fixation and suturing the esophagus in a single stage (9). We question the decision to perform ACDF at an adjacent level in an infected wound as this approach undoubtedly increased the surgical trauma and the risk of spreading the infection, even though the patient eventually recovered. In another case reported by Zakko *et al.* (27), during surgery, they found that the interbody cages at C4–5 and C6–7 were loose, and the vertebral bodies of C5 and C7 were eroded by the infection. There was a 14 mm defect in the intervertebral spaces of C4–5 and C6–7. After extensive irrigation and debridement, they placed titanium cages supported by plates between the C4–5 and C6–7 spaces and performed posterior fixation from C4 to C7.

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Zenker's diverticulum is a pulsion pseudodiverticulum caused by the chronic dysfunction of the cricopharyngeus muscle during swallowing. High intraluminal pressure eventually leads to the herniation of the mucosa and submucosa through a defect in Killian's triangle (33). In contrast, diverticula associated with ACSS are considered traction diverticula because of adhesions between the esophageal mucosa and cervical spine hardware. The external traction and shear stress on the layers of the posterior esophageal wall create a true diverticulum adjacent to the spinal hardware (34). Reports of dense fibrosis between some cervical spine hardware and the posterior esophageal wall support the traction diverticulum hypothesis (35, 36). Long-term contact between the spinal hardware and posterior esophageal wall post-ACSS can lead to pressure necrosis and eventually esophageal perforation. Perforations are more likely to occur at Killian's triangle, where the hardware is separated from the posterior esophageal wall only by the buccopharyngeal and prevertebral fascia, making this area relatively weak and more susceptible to instrument-induced injury (37, 38). Pillutla *et al*. (34) describe that exposed cervical hardware can lead to bolus extravasation through a perforation into the retropharyngeal space, resulting in a pseudodiverticulum composed of a thin mucosal wall. This occurs because of chronic bolus extravasation, which leads to expansion and mucosalization posterior to the perforation within the retropharyngeal space. The 'party wall' is the posterior wall of the esophagus, including the cricopharyngeus, which has been displaced anteriorly because of chronic extravasation. This party wall may be diagnosed as a cricopharyngeal bar in barium studies. In some patients, upper GI endoscopy confirmed an intact esophageal wall. These diverticula fall into two categories: true traction diverticula, which can be treated endoscopically without hardware removal, and another type. To minimize the risk of hardware exposure, the authors caution against overly aggressive diverticulotomy and recommend treating only the obstructive bar without opening into the retropharyngeal space.

Pillutla *et al*. (34) suggest that the surgical management of diverticula depends on the presence and exposure of the spinal hardware. If the spinal hardware is present and exposed within the esophageal lumen, it must be removed. This removal is necessary to eliminate sources of pressure necrosis and infection (9). Additionally, the excess diverticular tissue along with the fragile wound should be excised, leaving fresh, healthy mucosal edges for primary esophageal suturing (22). If the hardware is present but not exposed, it can remain in place, and a simple but prudent endoscopic diverticulectomy can be performed to address dysphagia. Similarly, if there is no internal fixation device, endoscopic diverticulectomy can be performed for definitive treatment.

Endoscopic diverticulectomy can effectively treat diverticula both after hardware removal and when hardware removal is not required. While most studies address diverticula during primary open treatment of esophageal perforation, open surgery cannot adequately manage the lateral wall of the diverticulum. Pillutla *et al*. (34) found that ACSS-related diverticula often necessitate staged procedures for comprehensive lateral wall treatment. The staged procedure involves initially removing exposed cervical hardware and repairing the esophageal perforation. Once the esophagus has healed, a definitive endoscopic diverticulectomy is performed. This approach is necessary because the initial repair (including hardware removal, esophageal perforation repair and placement of an SCM flap) does not adequately address the lateral wall or because the diverticulum recurs after repair. Endoscopic diverticulectomy allows for definitive treatment of the lateral wall. In our patient's treatment, certain limitations became apparent. Initially, during the first surgery, although an SCM flap transfer was performed with the skin sutured to the esophageal mucosa, we did not fully recognize the importance of addressing the esophageal diverticulum at that time. This oversight contributed to the failure of the initial esophageal repair. Subsequent attempts involving multiple debridement procedures and the placement of an esophageal stent also failed to achieve healing. Finally, during the secondary surgery, the diverticulum was excised and a new SCM muscle flap was used for coverage, resulting in successful recovery. This case analysis highlights the critical importance of addressing esophageal diverticula concurrently with esophageal perforation to ensure comprehensive treatment. We recommend that in cases where a diverticulum is present, debridement and diverticulum management should be integrated during the initial surgical intervention. Furthermore, if debridement and stenting prove ineffective, timely esophageal reconstruction and muscle flap application should be considered to achieve successful outcomes. This analysis underscores the need for a multidisciplinary approach and provides valuable insight into the management of similar cases in the future.

Conclusions

This case report and literature review emphasize the critical importance of early recognition and a multidisciplinary approach in managing complex cases of esophageal perforation following ACSS. The successful outcomes observed with hardware removal, diverticulum excision and muscle flap reconstruction highlight the efficacy of these surgical techniques. Addressing esophageal diverticula concurrently with esophageal perforation is vital to prevent recurrence and ensure comprehensive repair. In cases of late perforation, hardware removal is essential to achieve

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closure of the esophageal lesion, underscoring its role in the resolution of such complications. This case demonstrates the necessity of high clinical suspicion and coordinated surgical intervention to improve patient prognosis and highlights the significance of considering esophageal complications even many years post-ACSS. Regular follow-up and prompt intervention are key to managing such complications effectively.

Supplementary materials

This is linked to the online version of the paper at https://doi.org/10.1530/EOR-24-0110.

ICMJE statement of interest

The authors declare that there is no conflict of interest that could be perceived as prejudicing the impartiality of the work.

Funding statement

This work was supported by the Enze Medical Center (Group) Scientific Research (No. 23EZA04), Zhejiang Medicine, Taizhou Science and Technology Planning Project (No. 20ywb18) and Health Scientific Research Project (No. 2024KY531). The funder had a specific role in the preparation of the manuscript.

Author contribution statement

All authors made substantial contributions to conception and design, acquisition of data or analysis and interpretation of data. HL prepared the first manuscript draft. All authors contributed to final edits and revisions prior to submission.

Patient consent

Written informed consent was obtained from the patient for the publication of this case report and accompanying images.

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