



## Case Report

# Cervical epidural abscess complicated by a pharyngoesophageal perforation after anterior cervical spine surgery for subaxial spondylodiscitis

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## ABSTRACT

**Background:** The anterior approach to the cervical spine is safe and effective, but not without risks. The pharyngoesophageal perforation (PEP) is a rare but potentially life-threatening complication of this surgical route. A prompt diagnosis and adequate treatment are crucial for the prognosis; nevertheless, there is no unique consent about the best management.

**Case Description:** A 47-year-old woman was referred to our neurosurgical unit for clinical and neuroradiological signs of multilevel cervical spine spondylodiscitis, which was conservatively treated with long-term antibiotic therapy and cervical immobilization after computed tomography-guided biopsy. Nine months later, when the infection was resolved, the patient underwent C3–C6 spinal fusion with anterior plate and screws through anterior approach to the cervical spine for degenerative vertebral changes causing severe myelopathy, and C5–C6 retrolisthesis with instability. Five days after surgical procedure, the patient developed a pharyngoesophageal-cutaneous fistula, detected through wound drainage, and confirmed by swallowing contrast study, without systemic signs of infection. The PEP was conservatively treated, with antibiotic therapy and parenteral nutrition, and it was monitored through serial swallowing contrast and magnetic resonance studies up to the complete resolution.

**Conclusion:** The PEP is a potentially fatal complication of the anterior cervical spine surgery. We suggest an accurate intraoperative control of the pharyngoesophageal's tract integrity at the end of the surgical procedure and a longtime follow-up, because the risk of occurrence is up to several years after surgery.

**Keywords:** Anterior cervical spine surgery, Esophageal fistula, Pharyngoesophageal perforation, Cervical spondylodiscitis

## INTRODUCTION

The anterior cervical spine approach is the most common performed surgical procedure to the anterior cervical spine, mainly for median and neuroforaminal diseases. Although routinely performed, safe and effective, as each other surgical technique, it has its pro and cons, and it is not without risks. Complications related to this surgical route in decreasing order of incidence include dysphagia, postoperative hematoma, worsening of previous myelopathy, symptomatic vocal cord palsy, cerebrospinal fluid fistula, wound infection, worsening of previous radiculopathy, Horner's syndrome, respiratory failure, pharyngoesophageal perforation (PEP), and instrumentation failure<sup>[7,10,31,36]</sup>

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The PEP is a rare (incidence ranges from 0.02% to 1.62%)<sup>[7]</sup> but potentially life-threatening complication of the anterior cervical spine surgery, and it is associated to poor prognosis and high risk of morbidity (range from 13.2% to 19.3%).<sup>[7]</sup>

It may occur during the surgery or in the postoperative period, up to several years after the surgical procedure.

Although its mortality rate is low (range from 12% to 20%),<sup>[7]</sup> if not timely diagnosed and appropriately treated, this complication can lead to several potentially fatal conditions, such as mediastinitis, osteomyelitis, and sepsis.<sup>[10,19]</sup>

Due to the few reported cases in the literature, there are no defined guidelines about the best management of this complication, and strategies range from conservative treatment to the surgery.<sup>[8,21]</sup>

Here, we report a case of cervical epidural abscess complicated by a PEP occurred after cervical anterior stabilization without discectomy through anterior microsurgical approach to the cervical spine, for severe myelopathy due to multilevel degenerative changes after spondylodiscitis and retrolithesis with instability.

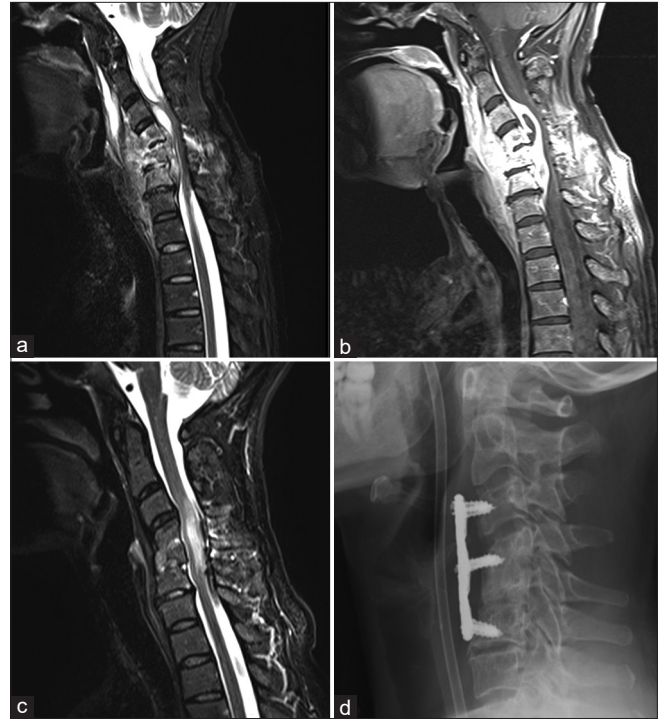
## CASE REPORT

A 47-year-old female complained a month-history of fever, intense neck pain, and progressive motor weakness at upper and lower limbs, with gait disturbance, not responsive to pharmacological conservative treatment. A computed tomography (CT) of the cervical spine showed a median and left paramedian discal herniation at C4–C5 level. A cervical spine magnetic resonance (MRI) showed a severe spondylodiscitis at C4, C5, and C6 levels with mass effect on the spinal cord, where signs of myelopathy were evident, and on the nerve roots.

The neurological examination at admission revealed severe spastic tetraparesis with preservation of proximal shoulder and hip movements, loss of movements of the distal segments of both arms and legs, and impossibility of standing and walking.

Blood cultures, Wright test, and tuber test were negative, while the Widal test was positive for antigen H of *Salmonella paratyphi*, expression of previous infection. Intravenous corticosteroid therapy was administered.

A cervical spine contrast-enhanced MRI contrast showed a marked abnormality at the C4–C5–C6 levels, with fragmentation and destruction of the endplates resulting in focal kyphosis. Dense material on the epidural space, mainly on the ventral side, suggestive of epidural abscess caused severe mass effect on the spinal cord, which appeared markedly compress and posteriorly displaced. Intense and diffuse contrast enhancement of the paravertebral soft tissues from C2 to T1 was also found [Figures 1a and b].



**Figure 1:** Cervical spine magnetic resonance imaging (MRI), sagittal sequences, contrast-enhanced T1-weighted (a), and T2-weighted (b) marked abnormality of the C4–C5–C6 vertebral bodies, with fragmentation and destruction of the endplates resulting in focal kyphosis. Dense material on the epidural space, mainly on the ventral side, suggestive of epidural abscess causing mass effect on the pharyngoesophageal tract posterior wall was detected. The infection also involved the surrounding soft paravertebral tissues. Severe mass effect on the spinal cord, which appears markedly compress and posteriorly displaced, was also evident. Postcontrast cervical spine MRI, T1-weighted sequence (c) 8 months after discharge and after prolonged antibiotic therapy. The inflammatory process was resolved resulting in multilevel degenerative changes. Severe myelopathy signal spanning from C3 to C6 level persisted. Cervical radiogram, sagittal projection (d). The correct position of the stabilization system composed by a titanium plate and six screws was evident.

A CT scan-guided biopsy of the collection resulted positive for Methicillin-Sensitive *Staphylococcus Aureus*. Thus, an adequate antimicrobial therapy with trimethoprim/sulfamethoxazole, 160/800 mg twice a day, was administered. After the initial improvement of the clinical symptoms, the patient was discharged with the indications to continue the antibiotic therapy for 8 weeks, adopting cervical spine immobilization, and performing serial cervical spine contrast-enhanced MRI.

In the following months, the neurological conditions moderately improved, laboratories and neuroradiological examinations showed the progressive resolution of the infectious disease but resulting in multilevel degenerative changes with severe myelopathy [Figure 1c]. Furthermore,

a dynamic XR of the cervical spine (maximal flexion and extension) showed C5-C6 retrolisthesis with instability.

Nine months after the discharge, once the spinal inflammation was resolved, the patient was readmitted to our department, and through anterior approach to the cervical spine, after initial debridement of the subcutaneous tissues, with the pharyngoesophageal complex which appeared tenaciously adherent to the prevertebral fascia and bone and which required careful microsurgical dissection, she underwent C3-C6 anterior cervical stabilization with titanium low profile plate and screws. A postoperative radiogram confirmed the correct position of the implant [Figure 1d]. The immediate postoperative course was uneventful. At 5<sup>th</sup> day after surgery, the patient developed surgical site swelling and drainage of a brownish liquid material from the caudal part of the surgical wound. A barium-swallow study was able to detect PEP, lesser than 1 cm on maximum diameter, through a contrast medium leakage at the pharyngoesophageal junction, close to the cervical plate [Figure 2a]. A MRI of the cervical spine confirmed the presence of the pharyngoesophageal-cutaneous fistula [Figure 2b]. Therefore, feeding was set by parenteral nutrition and systemic broad spectrum antibiotic therapy was administered. In the following days, we appreciated the progressive resolution of the secretion from the surgical wound. Serial barium-swallow studies documented a progressive improvement until the resolution of the fistula [Figure 3]; thus, parenteral nutrition was suspended, and the nasogastric tube was positioned.

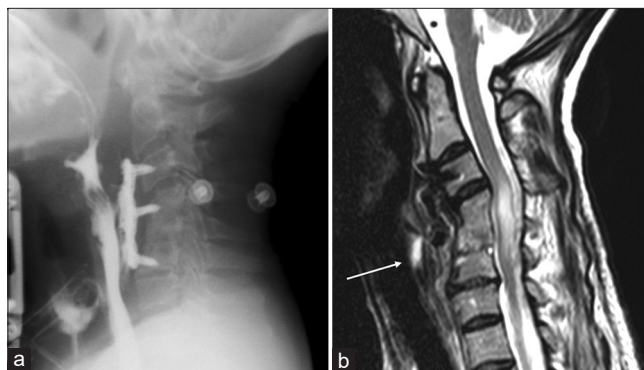
The patient was discharged on POD13 with absence of leak on clinical and radiological evaluation, and with nutrition per os.

## DISCUSSION

The present case is very interesting for several aspects, including the initial choice of a conservative management, despite the large epidural abscess with severe neurological deterioration, secondly, the occurrence of a pharyngoesophageal-cutaneous fistula and its conservative management, and finally the complete remission of both neurological and esophageal symptoms and signs.

Pyogenic spinal infection more often affects the thoracic and lumbar spine, less frequently the cervical region; nevertheless, the involvement of the cervical region is associated to higher mortality rate (21% vs. 3.6%).<sup>[23,26,34]</sup> Several articles report a more rapid neurological deterioration related to the infections of cervical spine compared to the lumbar and thoracic ones.<sup>[2,26,34]</sup> The incidence of epidural empyema ranges from 60 to 100%<sup>[2,9,11,29]</sup> and the presence of an epidural abscess is related to a higher rates of neurological deficits.<sup>[22]</sup>

The spinal cord is very sensitive and small compressions or mass effect from the epidural empyema can lead to severe myelopathy.



**Figure 2:** Barium-swallow study. (a) It was possible to appreciate the pharyngoesophageal perforation through a contrast medium leakage at the pharyngoesophageal junction. Magnetic resonance of the cervical spine, T2-weighted sequence. (b) It is possible to appreciate the persistence of the myelopathy signal and the pharyngoesophageal-cutaneous fistula (white arrow).



**Figure 3:** Barium-swallow study. The resolution of the pharyngoesophageal fistula was detected with the absence of contrast diffusion out of the lumen.

Infections of the cervical spine are rare and treatment options are various;<sup>[13,28]</sup> there is no unanimous consensus about the best timing and modalities of management, both conservative and surgical; nevertheless, an appropriate treatment is mandatory due to the occurrence's risk of a rapid worsening of the clinical conditions, with tetraparesis, sepsis up to death.

Considering the timing of treatment, some authors recommend conservative therapy for spinal infections up to 6 weeks<sup>[1,6,14,25,30,32]</sup> followed or not by surgery; others consider the surgical treatment followed by antibiotic therapy as the first choice due to the risk of a rapid worsening of the patient's clinical conditions.<sup>[3,5,12,20,22]</sup>

In regard of the modality of surgical treatment, several authors suggest the use of titanium mesh cages with or



without posterior instrumentations;<sup>[15,24]</sup> others are in favor of idle cervical spinal fusion;<sup>[18]</sup> others, considering the risk of bacterial colonization of the implant, prefer not using cervical plates.<sup>[27]</sup> A less invasive procedure, as an isolated anterior debridement and stabilization, is associated to a better clinical outcome than other more invasive approaches.<sup>[16]</sup> Mortality and morbidity are similar in patients treated with conservative therapy and those undergone surgery followed by antibiotic therapy.<sup>[35]</sup>

Conservative treatment, comprising long-term antibiotics combined with bed rest and/or an orthosis, seems to be the first line of treatment for pyogenic spondylodiscitis; surgery is indicated in cases of compression of neurological structures, pain worsening, persisting infection on imaging, failure of conservative treatment, spinal deformity, or neurological deteriorating.

In our case, we opted for an initial conservative treatment, consisting in targeted systemic antibiotic therapy for 3 months and cervical spine immobilization. During this period, the patient was monitored through laboratory and radiological examinations, and ambulatory controls. The neurological clinical conditions progressively improved in relationship with the resolution of the spinal infection and the related mass effect from epidural abscess.

The ratio of our strategy to delay the surgery was based on avoiding the risk of microbial colonization of the stabilization system and the consequent persistent infection.

The PEP is a rare (incidence ranges from 0.02% to 1.62%)<sup>[7]</sup> but potentially life-threatening complication related to the anterior cervical spine surgery, and it is associated to poor prognosis and high risk of morbidity (range from 13.2% to 19.3%).<sup>[7]</sup>

Only 159 cases of PEP following anterior cervical spine surgery have been reported in a recent literature review.<sup>[17]</sup> Among them, the most frequent surgical indication was a traumatic injury, followed by degenerative diseases and disk herniation.<sup>[17]</sup>

The cervical spinal segments between C3 and C6 levels, for their anatomical relationship with the cricoid, are the most susceptible to the PEP: the hardness of the cricoid cartilage in fact, exercises some pressure's degree on the posterior wall of the pharyngoesophageal tract during each swallowing act. This predisposing anatomical factor, associated to the incorrect placement of the fixation's system or hardware displacement, may favor the PEP due to decubitus.<sup>[8]</sup> Laimer-Killian's triangle, in the posterior wall of the esophagus, is the most common site of perforation, occurring in 50% of esophageal fistulas in cervical segments.<sup>[8]</sup>

The PEPs may be classified as early and delayed, if the interval between the surgical procedure and the diagnosis of this complication is under or over 30 days, respectively.<sup>[8,17]</sup>

Early perforations account for 40% of the cases and among them, the 12% of cases are due to an intraoperative injury, such as retraction or improper placement of plates, screws or cutting or cauterizing instruments, with immediate identification.<sup>[4,17]</sup> A pharyngoesophageal damage is the most common unrecognized complication during the surgery to the anterior cervical spine.<sup>[33]</sup> In our case, the perforation manifested 5 days after surgery was occurred intraoperatively during the retraction of the esophagus to expose the cervical spine and was due to the adherences resulting from the previous infectious process between the esophagus and the surrounding structures. This complication, the only one of our surgical series of procedures to the cervical spine, was unrecognized, although we routinely verified the integrity of the esophagus at the end of this surgical procedure.

Our patient underwent contrast swallow to detect the site, size, and morphology of PEP and X-ray and MRI to evaluate the spine fusion and stability, and the placement of the implant; the endoscopic exploration was not necessary, because already from the radiological examinations, we could appreciate no penetration of the implant into the esophagus.

Due to the paucity of literature on PEPs, defined guidelines of management are not present; reported options include conservative and surgical treatments. Among the 159 reported cases in literature,<sup>[17]</sup> only 12 were treated in conservative manner, with drainage, nasogastric tube, antibiotic therapy, and enteral and/or parenteral nutrition, nevertheless obtaining the complete resolution of the complication in 83.3% of the cases.

Surgical treatment consists of an initial step, in which wound the debridement and drainage, if infection is present, are made, followed by PEP repair through suture of the defect and reinforcement with muscle flap. If the fusion system is infected or migrate, or plate/screw decubitus is present, the removal of the fixation device is recommended.<sup>[10,17,21]</sup>

Our patient was treated conservatively due to minimal leak (<1 cm) and absence of local and systemic infection signs. We consider that the best management mainly depends on the entity of the perforation and the local condition. Large fistulas with significant inflammatory collection should be operated on. On the other hand, for small perforations, we suggest a conservative management.

## CONCLUSION

The PEP is a rare but life-threatening complication related to the anterior approach to the cervical spine. A tempestive diagnosis and appropriate treatment are mandatories. Defined guidelines of management are not present; the treatment should be tailored according to the size, site, and the morphology of the defect. We suggest an accurate intraoperative control of the pharyngoesophageal's tract

integrity at the end of surgical procedure and a longtime follow-up due to the risk of occurrence up to several years after surgery.

#### Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

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Nil.

#### Conflicts of interest

There are no conflicts of interest.

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