



# Treatment considerations for cervical and cervicothoracic spondylodiscitis associated with esophageal fistula due to cancer history or accidental injury: a 9-patient case series

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## Abstract

**Background** The combination of cervical spondylodiscitis and esophageal fistula is rare but life-threatening. Due to both the rarity of these conditions' coincidence and the complexity and heterogeneity of individual cases, there is no optimal treatment or management approach. The aims of this study are to obtain an overview of patients' outcomes and to discuss treatment options.

**Method** This study is a retrospective analysis of patients who presented with cervical spondylodiscitis and associated esophageal fistula between January 2010 and November 2018. We examined reports of 59 patients who suffered from cervical spondylodiscitis and included nine patients (15.25%) who had an esophageal fistula as the underlying cause. We assessed clinical findings, treatment, and outcome.

**Results** Three of the nine patients were female, and the mean age of the sample was 64.56 years. Six of the patients had a history of esophagopharyngeal cancer and had undergone tumor resection followed by radiotherapy. Two of the remaining patients' fistulas were caused by an iatrogenic injury during cervical spine surgery and a swallowed toothpick; in the final case, the origin remained unclear. Five patients presented with tetraparesis or tetraplegia, and the other four patients were neurologically intact. In seven cases, dorsal instrumentation was initially performed. Three patients secondarily received a ventral approach for debridement, and one received explantation of the ventral implants. Two patients died during the hospital stay, and three were transferred to a palliative care unit. Thus, the spondylodiscitis and esophageal fistula were cured in only four patients. At discharge, two patients were neurologically intact, two others remained in tetraparesis.

**Conclusions** Cervical spondylodiscitis in association with an esophageal fistula carries high morbidity and high mortality. Because patients whose infections are not cured have high morbidity, we recommend using interdisciplinary and individual management, including definite surgical treatment of the discitis and fistula, in every case.

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**Keywords** Cervical spondylodiscitis · Esophageal fistula · Epidural abscess · Retropharyngeal abscess · Esophageal injury · Neck cancer

## Abbreviation

ENT Ear, nose, and throat

## Introduction

The association of cervical spondylodiscitis with an esophageal fistula is rare but life-threatening. The development of an esophageal fistula is a common complication after multimodal treatment for head and neck cancer involving radionecrosis; it is also a less frequent result of insufficient anastomosis or infection after surgery [6]. An esophageal fistula may also occur after cervical spine surgery, but this is extremely rare, with an incidence of 0.04–0.25% [25]. Spinal infection usually occurs via hematogenous spreading as a result of distant pyogenic foci or following spine surgery [4, 10, 15, 16, 22]. Cervical spondylodiscitis is particularly associated with severe neurological deficits and rapid neurologic deterioration [9]. The treatment of spondylodiscitis is still a matter of some controversy. In the absence of neurological deficits or spinal instability, many experts advocate for a conservative approach [2]. However, a paradigm shift has occurred in the last decade; more spinal surgeons are now recommending spinal instrumentation for spondylodiscitis patients [22, 23]. Both the surgical and conservative treatment options also include intravenous antibiotic treatment for at least 2 weeks, followed by oral antibiotics for an additional 10 weeks [22]. Furthermore, the pyogenic focus of the inflammation should be detected and eradicated [22].

Depending on the extent of the esophageal fistula, several options may be possible: conservative treatment, surgical excision of the fistula, and primary closure or complex tissue reconstruction [18, 24, 25]. However, in cases with acute inflammation and a local pyogenic infection, some of these treatment options are not optimal. Flap reconstruction is generally performed within the first week after an acute infection, when the local tissue inflammation has begun to resolve [11]. However, definite surgical treatment of the affected cervical segment is usually impossible when an esophageal fistula is present; thus, both the fistula and the discitis remain a source of infection, which constitutes a kind of medical standoff. Despite existing treatment strategies in which each disease is treated separately, there is no gold standard for the optimal treatment and management of these problems in combination.

In this study, we present a case series of 9 patients with cervical or cervicothoracic spondylodiscitis and concomitant esophageal fistulas. The study's aims are to obtain an overview of the patients' outcomes and to discuss the treatment options.

## Materials and methods

We performed a retrospective analysis of a prospective clinical database and included patients who presented with cervical or cervicothoracic spondylodiscitis due to an esophageal fistula. We assessed the clinical findings, treatment strategies, and outcomes. We also performed a review of the literature.

The Ethics Committee approved this study (No. 238/17S). Patient consent was not necessary because all patients signed forms consenting to allow the clinic to store and analyze their data, tissue and blood samples for scientific purposes.

## Results

### Patient histories

Out of 59 patients who presented with cervical or cervicothoracic spondylodiscitis between January 2010 and November 2018, 9 (15.25%; 3 female) had an associated esophageal fistula as the underlying cause. These 9 patients' mean ( $\pm$  standard deviation) age at presentation was 64.56  $\pm$  14.88 years (range 32–81 years).

Six patients had a history of esophageal, laryngeal, or pharyngeal cancer; the cancers occurred between 2 and 30 years prior to admission, which explain their vulnerability to developing esophageal lesions. All 6 of these patients had undergone tumor resection followed by radiotherapy, and 5 of them had a permanent tracheostoma. Out of the 6 patients with a history of cancer, 4 patients had undergone prior treatment for an esophageal fistula (Table 1, cases 3–5 and 9), and 2 of those had received surgical treatment in which a muscle flap covered the fistula within 2 years prior to their presentation with the spine infection (Table 1, cases 3 and 4) (Fig. 1). Another patient with a cancer history—a 71-year-old woman—had received surgical treatment for cervical discitis 3 years prior in the form of an anterior cervical discectomy and fusion with the ventral plating. At that time, doctors used a conservative treatment to heal an existing fistula (Table 1, case 5; Fig. 2a, b). This patient later sustained a relapse of both the fistula and the spondylodiscitis (Fig. 2d); in an endoscopic examination, the ventral plate was visible through the esophagus (Fig. 2e). A 75-year-old male patient with a cancer history had received an esophageal stent due to a fistula some months before presentation; this required several revision surgeries due to complications (Table 1, case 9). In the fifth case of a patient with a cancer history, a 64-year-old female had undergone a balloon dilatation due to esophageal stenosis resulting from scar tissue some months before; this procedure

**Table 1** Demographics and overview of the individual cases including level of discitis, symptoms, medical history, imaging and diagnosis, treatment of discitis and fistula, complications, and outcome

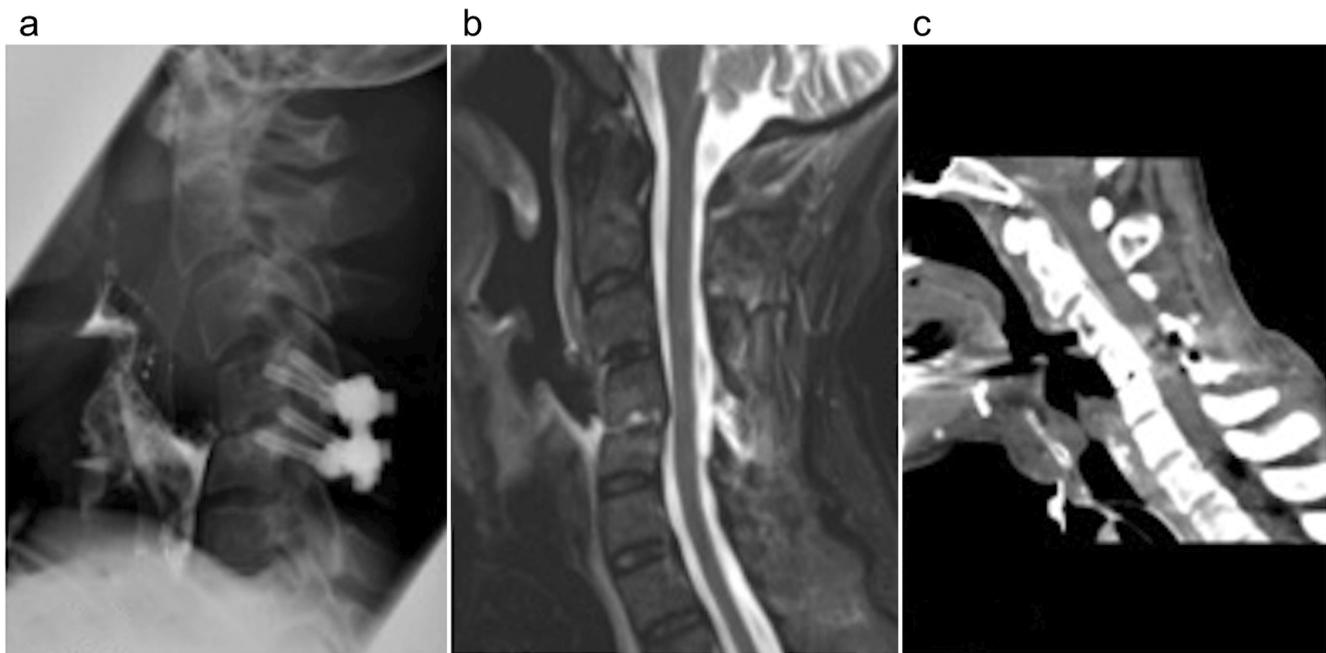
Case/ patient	Case 1	Case 2	Case 3	Case 4	Case 5	Case 6	Case 7	Case 8	Case 9
	female, 64 years	male, 81 years	male, 61 years (Fig. 1)	male, 79 years	female, 71 years (Fig. 2)	female, 61 years	male, 57 years	male, 32 years	male, 75 years
Level of discitis	C4-7 + epidural empyema + prevertebral abscess	C3-5 + epidural empyema + prevertebral abscess	C4-5	C6-7, T3-4	C4-7 + epidural abscess + prevertebral abscess	C2-3 + epidural abscess + prevertebral abscess	C3-6 + epidural abscess + prevertebral abscess	C4-7 + prevertebral abscess	T1-3 + epidural abscess
Symptoms	- tetraparesis ASIA B	- tetraparesis ASIA B - dysphagia	- cervical pain - no neurological deficit	no neurological deficit	- ataxia	- ataxia	- tetraparesis ASIA B - sepsis	- tetraplegia ASIA A	- tetraplegia ASIA A
Medical history	- esophageal cancer in 2005 and chemotherapy <i>complicated by:</i> - 2016 esophageal stenosis resulting from scar tissue treated three times by balloon dilatation	- larynx carcinoma 30 years ago - surgery and radio- therapy - permanent tracheo- stoma	- hypopharynx carcinoma 2 years ago - surgery, radio- and chemotherapy <i>complicated by:</i> - radionecrosis of the cervical soft-tissue treated with pectoralis major muscle flap - permanent tracheostoma	- hypopharynx carcinoma 3 years ago - surgery, radio- and chemotherapy <i>complicated by:</i> - esophagotracheal fistula, salivary leakage, perforation of an esophageal stent - covering of fistula by sternocleido- mastoid muscle flap, - pectoralis muscle flap one year later - permanent tracheostoma	- larynx carcinoma in 2014 - surgery, radio- and chemotherapy - cervical discitis surgically treated by ACDF C4-7 + ventral plate in 2014 - pharyngo- and esophagotracheal fistula detected in 2014 - conservative treatment of fistula 2014 - permanent tracheo- stoma - in 2017 three times balloon dilatation for treatment of esophageal stenosis	- no cancer - had swallowed a toothpick two month ago	- no cancer - tetraparesis ASIA B - respiratory insufficiency - no cancer - iatrogenic injury of pharynx during neuro- surgical surgery for cervical disc herniation by an anterior approach	- no cancer - larynx carcinoma diagnosed in 2013 - surgery, radio- and chemotherapy - tracheotomy due to tumor progression in 2017 - again radio- and chemotherapy in 2017 - development of chronic esophagotracheal fistula - 12/2017 implantation of esophageal stent with several revision surgeries due to complications	- larynx carcinoma diagnosed in 2013 - surgery, radio- and chemotherapy - tracheotomy due to tumor progression in 2017 - again radio- and chemotherapy in 2017 - development of chronic esophagotracheal fistula - 12/2017 implantation of esophageal stent with several revision surgeries due to complications
Diagnostic of fistula	- <i>Esophagogra- phy:</i>	- <i>CT with oral contrast:</i>	- <i>Dynamic pharyngo- graphy:</i> suspicious <i>spine:</i>	- <i>CT scan/ cervical spine:</i>	- <i>Pharyngograp- hy:</i>	- no active fistula or fissure shown in imaging but suspicion	- <i>Intraoperat- ive</i>	- <i>CT of the neck:</i> retropharyngeal air and	- <i>Endoscopic exploration</i>

Table 1 (continued)

Case/ patient	Case 1	Case 2	Case 3	Case 4	Case 5	Case 6	Case 7	Case 8	Case 9
	female, 64 years	male, 81 years	male, 61 years (Fig. 1)	male, 79 years	female, 71 years (Fig. 2)	female, 61 years	male, 57 years	male, 32 years	male, 75 years
	suspicion of fistula at level C6	prevertebral leakage at level C3-5 with fistula at level C4-5 (Fig. 2b)	contact of the posterior wall of pharynx to the intervertebral space between C4-5; - Endoscopically no existing leak or a fistula found	destruction of vertebrae C6-7 and T3-4	leakage of contrast between plate and vertebra at level C7	of an already healed esophageal lesion	exploration: no fistula seen	fluid collection, collapsed pharynx	esophageotracheal fistula, stent visible through trachea
	- Intraoperative endoscopic exploration: the disconnection of the esophagus at the level of hypopharynx	- Esophagoscopy: fistula 9 cm from tooththrow	- Endoscopically no existing leak or a fistula found	- Dynamic pharyngography: relapse of esophageotracheal fistula	- Endoscopic exploration: fissure of pharynx		- Endoscopic exploration: showed pharyngeal fistula	- Intraoperative exploration: pharynx-fistula to cervical spine and cervical vessels	
		- Surgical and endoscopic exploration: lesion of the pharyngeal posterior wall, fistula also to the neurovascular bundle			- CT of the neck: also showed contact between esophagus and ventral plate in terms of esophagovertebral fistula in				
Surgical treatment of discitis	1) Spondylosis C4-6 (massa lateralis screws) C4-6, pedicle screws T1-2 + laminectomy, empyem 2) ACD C4-7 for debridement, no fusion 3) Revision of Spondylosis due to screw pullout	1) Spondylosis C2, pedicle screws C4-7), laminectomy C3-6 2) ACD C3-6 + ENT for pharyngoscopy	Spondylosis C4-5 (massa lateralis screws)	Spondylosis C5-T1-3-5 (pedicle screws)	1) Spondylosis C3-5-T1-2 (pedicle screws) 2) explantation of ventral plate	Spondylosis C1-2-3 + hemilaminectomy C2+3	Spondylosis C2 (isthmus screws)-7 (pedicle screws)+ laminectomy C4-7 2) ACD C3-6 + ENT for exploration	1) Re-ACDF C4-7+ ventral plating 2) Explantation of ventral plate 3) Spondylosis C4-7 (C4-5 massa lateralis screws, C6-7 pedicle screws)	1) Hemi-laminectomy T1-3, evacuation of empyema 2) Dorsal spondylosis T1-2-3-4 + laminectomy T1-3 3) Resection of esophagus and debridement of the level T2-3 via thoracotomy
Treatment of fistula	- PEG - tracheotomy - implantation of	- placement of tamponade due to a	- PEG - no treatment due to tumorprogression	- reposition of esophagealstent, - intraoperative	- conservativ treatment of fistula in 2014	- surgical abcess evacuation and drainage	- tracheotomy - clipping of esophageal	- tracheotomy, - daily lavage of pharynxfistula and	- thoracotomy and resection of esophagus

**Table 1** (continued)

Case/ patient	Case 1	Case 2	Case 3	Case 4	Case 5	Case 6	Case 7	Case 8	Case 9
	female, 64 years	male, 81 years	male, 61 years (Fig. 1)	male, 79 years	female, 71 years (Fig. 2)	female, 61 years	male, 57 years	male, 32 years	male, 75 years
	transcutaneous tube to bypass the perforation	bleeding from the pharynx	without surgical or oncological treatment concept	esophagoscopy and bougenage of a stenosis	- 2017 suture of pharynx fissure		fissure	easy-flow-drainage	
	- no definite surgical management by ENT possible	- PEG		- colon interposition for esophageal reparation	- in esophagography regressive fistula		- several endoscopic explorations due to pharyngeal bleeding	- no final surgery possible by ENT	
	- success of conservative treatment not expected	- patient refused further treatment		- salivary fistula not possible			- PEG	- botox injection for inhibition of salivation	
							- decreasing fistula in imaging, but remaining infection		
Complications and Outcome	- septic shock	- spontaneous respiration	- tumorprogression	- worsening ventilation/hypoxia	- no deficit at discharge	- no deficit at discharge	- intraoperative injury of A. vertebralis	- patient died to circualtion insufficiency after reanimation due to cervical bleeding of vessels - dissolved by infection	- recovery after first surgery
	- peritonitis (laparotomy)	- tetraparesis ASIA B	- palliativ care unit	- acute abdomen	- cured fistula and discitis	- cured fistula and discitis	- persisting tetraparesis at discharge		- remaining ASIA A after relapse
	- mechanical ventilation	- transfer to pallive care unit due to patients wish		- died on ICU after unsuccessful reanimation					- cured fistula and discitis
	- palliativ care unit in her home country								



**Fig. 1** (Case 3): Dynamic pharyngography (a) and computer tomography of the neck (b) showed a contact of the posterior wall of the pharynx to the intervertebral space C4/5 but an existent leak or a fistula could not be proven. Cervical Magnetic Resonance Imaging showed discitis at level C4/5 (c).

was the assumed reason for the esophageal lesion (Table 1, case 1). Finally, an 81-year-old man had experienced neck cancer 30 years ago but had experienced no complications before presentation of the fistula.

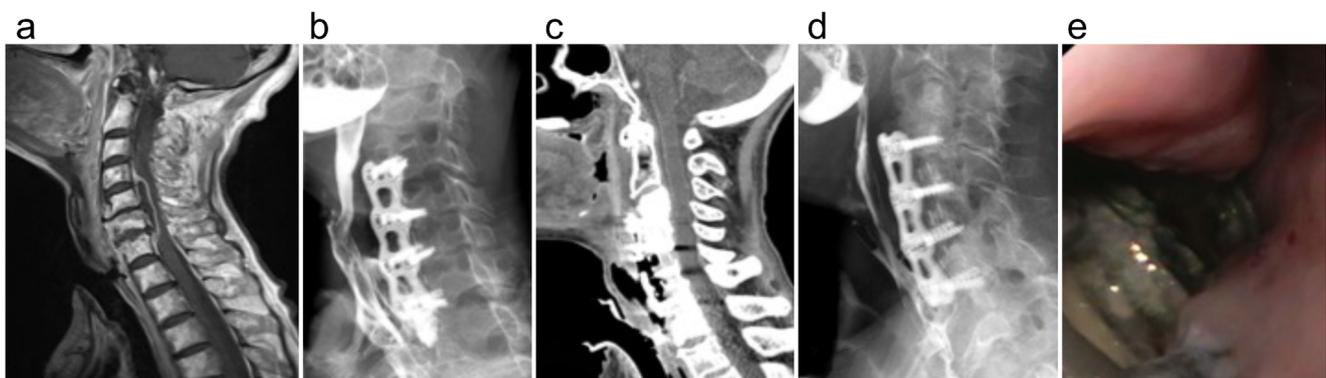
The remaining 3 patients had no history of cancer. In one case, ENT doctors detected an already-healed esophageal lesion in a 61-year-old woman (Table 1, case 6) during an endoscopic examination. She had, 2 months prior to presentation, swallowed a toothpick and then successfully removed it. In another case, a 32-year-old patient who presented with tetraparesis (Table 1, case 8) had sustained an undetected iatrogenic injury of the pharynx during anterior cervical spine surgery. Finally, for a 57-year-old male (Table 1, case 7), the

causes of both the discitis and the fistula remained unclear; this patient had no cancer history and no prior cervical surgery.

At time of their admission to our department, 5 of the 9 patients presented with tetraparesis or tetraplegia, and 2 had gait ataxia. The remaining 2 patients had neck pain but were neurologically intact, and one of these cases involved kyphosis of the cervical spine.

### Diagnostics

We diagnosed the spondylodiscitis via magnetic resonance imaging (MRI) scans with or without contrast, as well as with short-tau inversion recovery (STIR) sequences. The diagnosed discitis



**Fig. 2** (Case 5): Cervical magnetic resonance imaging (a) showed discitis C4–7 and an epidural abscess when presenting the first time, esophageal fistula was seen in pharyngography (b). Three years later, the computer tomography of the cervical spine showed a destruction of the vertebral

bodies and a connection between esophagus and ventral plate (c). Pharyngography showed a relapse of the fistula (d), and in an esophagogastroduodenoscopy, the plate was visible through the esophagus (e).

is at one level for 2 patients and at multiple levels for the other 7. A retropharyngeal or paravertebral abscess was present in 7 cases, and an epidural abscess was found in 6 cases. For the diagnoses of esophageal fistula, we first attempted dynamic esophagography and pharyngography. When a patient had a poor overall health or when dynamic pharyngography was not possible for other reasons, we instead performed a computer tomography (CT) of the neck using oral contrast. In some cases, either endoscopic examination or surgical exploration was necessary as part of an interdisciplinary surgical approach or by an ear, nose, and throat (ENT) doctor alone (Table 2).

## Treatment

To treat their discitis, all patients received surgery, intravenous antibiotic treatment for at least 2 weeks, and long-term oral treatment. The treatment of the esophageal fistula differed in each case, but every patient received either parenteral nutrition or a percutaneous endoscopic gastrostomy. Furthermore, they received pharmacological inhibition of salivation.

For treatment of spondylodiscitis, the majority of patients (7) initially received dorsal instrumentation based on the ventral scar tissue due to prior neck-cancer surgery or spinal instability. However, the main cause was an assumed or already-diagnosed esophageal fistula because the dorsal fixation of the spine provided the possibility of avoiding ventral implants, which can interfere with the fistula-healing process. Three

patients secondarily received a ventral approach for debridement, and one received the explantation of the ventral implants. In case 9, a 75-year-old man with a predominant epidural abscess first received a posterior approach via hemilaminectomy for the evacuation of the epidural empyema without instrumentation. Due to concomitant spondylodiscitis, we discussed a further surgery for posterior fixation, but according to the ENT doctors, no definite treatment was possible for the fistula. Due to the remaining source of infection, no instrumentation was used. The patient was transferred back to the department after several weeks due to a relapse of empyema and an increasing disc infection. At that time, we completed a posterior fixation to treat the patient's discitis. Only one patient (Table 1, case 8) received an anterior approach first for the revision of the prior cervical spine surgery (which was initially the assumed source of the infection). One week thereafter, this patient received posterior instrumentation; furthermore, the patient underwent a third surgery for explantation of the ventral implants. In case 1, revision was necessary after a screw came loose from eroded bone.

In 2 cases, the ENT doctors joined in the surgery, using a ventral approach to explore the esophageal fistula. Doctors performed definite operative treatment of the esophageal fistula in 4 cases, whereas in 2 cases, a primary suture and clipping of the existing fissure was possible. In one case, a colon interposition for esophageal reparation was done (Table 1, cases 4 and 9). In case 9 (Table 1), we performed a resection

**Table 2** Microbiological findings and antibiotic treatment

Case	Germs	Origin of isolation	Duration of antibiotic treatment	Antibiotics
Case 1	<i>Enterococcus faecalis</i> , <i>Escherichia coli</i> (3- MRGN), <i>Streptococcus angiosus</i>	Cervical abscess	Intravenous: 54 days until transfer to palliative care unit	Intravenous: ceftriaxone, ciprofloxacin, imipenem, metronidazole, linezolid, levofloxacin, micafungin, aciclovir, meropenem, daptomycin, caspofungin
Case 2	<i>Staphylococcus epidermidis</i> , <i>Candida albicans</i>	Disc space	Intravenous: 43 days until transfer to palliative care unit	Intravenous: levofloxacin, metronidazole
Case 3	<i>Staphylococcus epidermidis</i>	Blood culture	Intravenous: 22 days Oral: until transfer to palliative care unit	Intravenous: vancomycin, piperacillin/tazobactam Oral: ciprofloxacin, cefuroxime
Case 4	No isolation	–	Intravenous: 27 days until dead	Intravenous: vancomycin, meropenem
Case 5	<i>Streptococcus anginosus</i> , <i>Candida albicans</i> , <i>Serratia marcescens</i>	Disc space	Intravenous: 2 weeks Oral: 10 weeks	Intravenous: metronidazole, cefuroxime Oral: metronidazole, cefuroxime
Case 6	No isolation	–	Intravenous: 20 days Oral: 10 weeks	Intravenous: vancomycin, meropenem oral: Clindamycin
Case 7	<i>Staphylococcus aureus</i> subfascial	Subfascial anterior	Intravenous: 47 Oral: 10 weeks	Intravenous: cefuroxime, ciprofloxacin, flucloxacillin, rifampicin, caspofungin Oral: levofloxacin
Case 8	<i>Pseudomonas aeruginosa</i>	Cervical abscess	Intravenous: 26 days intravenous until dead	Intravenous: daptomycin, meropenem
Case 9	<i>Pseudomonas aeruginosa</i> (4 MRGN), <i>Candida</i>	Cervical abscess	Intravenous: 35 days intravenous Oral: 14 weeks/lifelong	Intravenous: meropenem Oral: amoxicillin, ceftazidime, fluconazole lifelong

of the esophagus (thoracotomy) in collaboration with thoracic surgeons. During this surgery, we also debrided the T2/3 segment.

In the other cases, the ENT doctors deemed definite treatment of the fistula impossible. Instead, the patients received conservative treatment and—in case 1—the implantation of a transcutaneous tube to bypass salivation.

We could identify the underlying germs in 7 cases: 2 in the disc space, 4 in the cervical abscess, and 1 in the blood culture. Nine patients received intravenous antibiotic treatment for a mean of 34.35 days (range 20–54 days), and 5 patients received oral treatment for at least 10 weeks afterwards (Table 2).

## Outcomes

Early morbidity and mortality was high in this case series. During a treatment period of 3 months, 4 patients died. Two patients died during their hospital stays: 1 due to bleeding from a cervical artery that had been eroded due to the ongoing inflammation, saliva, and superinfection, and 1 due to multiorgan failure as a consequence of a severe systemic infection. Of the 3 patients who were transferred to a palliative care unit, only one presented with tumor progression. For 1 patient, the ENT doctors advised that neither a definite surgical treatment nor conservative management was possible due to the extent of the defect (case 1, Table 1), and the patient refused further treatment. Two of the 3 palliative care patients died within the first month after discharge, and 1 was lost from further follow-ups after she returned to her home country. Thus, only 4 patients had their spondylodiscitis and esophageal fistula cured. At discharge, 2 of these patients were neurologically intact, but the other 2 were still suffering from tetraparesis ASIA B.

## Illustrative case: case 9

A 75-year-old man was transferred to our clinic after developing ASIA A paraplegia due to an epidural empyema of level T1–3, in addition to signs of spinal infection (e.g., spondylodiscitis). In 2012, the patient underwent laryngectomy and radiochemotherapy due to a laryngeal carcinoma. In 2017, he received a tracheotomy due to tumor recurrence. He had developed an esophago-tracheal fistula, which the ENT doctors treated through the implantation of an esophageal stent. He underwent several revision surgeries due to dislocations of that stent, but the fistula remained. At time of admission, we performed an emergency hemilaminectomy (T1–3) to evacuate the empyema, with the intention of further treatment in terms of posterior instrumentation, depending on the tumor's state and prognosis. We detected no tumor progression, but the ENT doctors deemed definite treatment of the fistula to be impossible. The patient was discharged to a

rehabilitation center, where he recovered very well until he experienced increasing spondylodiscitis and a relapse of empyema. At the time of readmission, he was paraplegic again. He then received both dorsal spondylodesis (T1–2–3–4) and re-decompression (T1–3). Due to the necessity of curing the remaining source of infection, we resected the patient's esophagus by thoracotomy in collaboration with thoracic surgeons. During the same surgery, we debrided segment T2/3; this time, he did not recover from his neurological deficits.

## Discussion

Our case series illustrates that cervical spondylodiscitis with a concomitant esophageal fistula is a clinical problem with very high morbidity and mortality. During the treatment period of 3 months, 4 of 9 patients died. Two patients died during the hospital stay, and of the 3 patients who transferred to a palliative care unit, 2 died within the first few months after discharge. The third palliative care patient was lost from further follow-ups after she moved back to her home country. In this case series, 6 patients had histories of neck cancer and had undergone surgery and radiation, which explains their vulnerability to developing esophageal fistulas. However, the patients without cancer histories also experienced esophageal fistulas with fatal outcomes. The tumor progressed in only 1 patient; in the other patients who died, the deaths were clearly caused by ongoing infections. Only 4 patients' spondylodiscitis and esophageal fistulas were cured. We treated all the patients' discitis surgically and with long-term antibiotic therapy, and the fistulas healed through various methods: conservative treatment (case 6), endoscopic sutures, and the clipping of small esophageal fissures (cases 5 and 7), or (more drastically) the resection of the esophagus (case 9). At discharge, 2 patients were neurologically intact, and 2 others still suffered from tetraparesis.

In 1963, Sealy described esophageal perforation as “the most rapidly fatal and serious perforation of the gastrointestinal tract” [21]. An esophageal fistula can occur as a late-onset complication of treatment for head or neck cancer, due to an iatrogenic injury incurred during cervical spine surgery, or even as a result of a traumatic spinal or neck injury [6, 12, 14]. Patients who have undergone neck dissection and radiotherapy are also at risk of cervical spine infection [6]. Both radiotherapy fibrosis and tissue ischemia may promote a weakening of the cervical fasciae and the formation of a fistulous tract [7]. Furthermore, damage to the peripheral lymphoid tissues may lead to deficiencies in regional infection resistance [5, 6]. Also of note is that the incidence of such cases is increasing due to better and more aggressive multimodal oncological treatments for advanced neck cancer.

An esophageal fistula may also occur as a complication of spinal surgery using an anterior approach; this has an

incidence of 0.04–0.25% and a mortality rate of 9–45% [25]. Early diagnosis of an esophageal fistula is important to prevent catastrophic and potentially life-threatening complications and to decrease the mortality rate [25]. In the early stage, without a manifest infection, a primary suture—if necessary, with the interposition of vital tissue—is sufficient for complete healing [24]. Otherwise, a retropharyngeal or epidural abscess can form, and spondylodiscitis, mediastinitis, sepsis, and even death can occur [12]. Depending on the extent of the fistula, conservative treatment, simple fistula repair, or complex tissue reconstruction can be used to treat an esophageal fistula [18, 24, 25]. Conservative treatment includes the avoidance of oral food ingestion, the provision of intravenous, high-dose, broad-spectrum antibiotics, and nutritional support. This is, however, associated with a failure rate of 20%, in which a spinal infection develops [19, 25]. Benazzo [1] reported good results in 3 patients when using a sternocleidomastoid flap for esophageal repair following injuries due to cervical spinal surgery; the patients' fistulas were cured without further complications. Hanwright [11] described how using surgical intervention in addition to conservative treatment could reduce mortality to 12% and argued that this should be the gold standard. Hanwright [11] also mentioned the restoration of gastrointestinal integrity, infectious control, and spinal stability as the 3 key factors in the management of esophageal fistulas.

Frequently described as an additional tool in management of wound healing disorders is hyperbaric oxygen therapy (HBOT), which has also shown benefit in treating patients with delayed radiation injury [8, 13]. Regarding that especially patients with a history of neck cancer are at a high risk of developing a complicated esophageal fistula due to ischemic and hypoxic tissue after radiation, hyperbaric oxygen therapy could be considered to be added to the multidisciplinary protocol especially in the early state of fistula treatment. In a case of a 12-month-old child, hyperbaric oxygen therapy was successfully used to treat an esophagotracheal fistula conservatively [20]. However, the patients should be carefully selected and the doctors should be aware of contraindications and feasibility regarding the clinical state.

The management of patients with cervical discitis and an underlying esophageal fistula is particularly challenging; however, no systematic reviews for the management of the coincidence of these conditions have been published, and no robust evidence is available. Decisions, therefore, need to be made on a case-by-case basis, and should involve an interdisciplinary team of neurosurgeons, ENT doctors, gastroenterologists, and general or thoracic surgeons, who should customize the treatment to every patient. With only a few case reports in the literature [3, 15, 17] and a high heterogeneity in the studied patients, no generalized treatment recommendation exists due to a lack of evidence. Nevertheless, we underwent a learning curve towards a treatment more aggressive regarding the surgical treatment of the fistula. Case 9 was the case

most recently. The drastic decision for resection of the esophagus was made in the interdisciplinary setting. In this case, we succeeded in a definitive healing of fistula and discitis in a progressed state of inflammatory disease. Researchers' aim should be to develop an early definite treatment of discitis in combination with a fistula in order to prevent neurological deterioration and preserve acceptable quality of life, especially for patients with limited life expectancy.

As patients who do not receive active treatment often experience fatal outcomes, we recommend the following:

#### Posterior approach for the treatment of spondylodiscitis

Other than in case of isolated cervical spondylodiscitis (in which surgical treatment usually consists of anterior cervical discectomy and fusion), the surgical treatment of spondylodiscitis with a concomitant esophageal fistula should be done via posterior instrumentation (with or without decompression) in order to avoid the use of ventral implants, which apparently disturb the healing of the fistula.

#### No ventral implants

For relevant prevertebral and/or epidural abscesses, ventral debridement only—without ventral implants—may be necessary. In cases of existing ventral instrumentation, surgery to remove the implants should be performed.

#### Antibiotics and nutrition

Every patient should receive intravenous antibiotics for at least 2 weeks, followed by oral treatment for at least 10 weeks. The individual treatment duration depends on the kind of germ, the treatment effect (controlled by inflammatory markers), the imaging results, and the patient's clinical status. Treatments should be discussed with the Department of Infectology.

If parenteral nutrition is not provided, then a feeding tube must be inserted, or a percutaneous endoscopic gastrostomy must be performed.

#### Curing of fistula as source of infection

If the esophageal fistula continues for some time after the conservative treatment, a staged fistula closure should be performed to provide definite treatment; if the inflammatory signs are decreasing, a muscle flap can be used, depending on the microvascular conditions; however, a vascularized sternocleidomastoid flap should be preferred. In the case of a persistent fistula with a recurring or chronic infection, drastic surgical treatment (e.g., resection of the esophagus, as shown in case 9) must be considered.

## Compliance with ethical standards

**Conflict of interest** The authors declare that they have no conflict of interest.

**Ethical approval** The Ethics Committee approved this study (No. 238/17S). For this type of study, formal consent is not required, because all patients signed forms consenting to allow the clinic to store and analyze their data, tissue and blood samples for scientific purposes.

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