CASE REPORT

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SPINAL EPIDURAL ABSCESS AFTER CERVICAL PHARYNGOESOPHAGEAL DILATION

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Abstract: Background. Esophageal perforation is an uncommon but known complication of esophageal dilation. Abscess after esophageal tear is rare, especially in the spinal epidural space. This is one case report of such an abscess.

Methods. We present a case of a spinal epidural abscess after cervical pharyngoesophageal dilation.

Results. After surgical decompression and abscess drainage, long-term intravenous antibiotics, and physical therapy, the patient has regained some functional use of her left upper extremity.

Conclusions. Early diagnosis with a gadolinium-enhanced MRI and aggressive surgical treatment are keys to successful management with a good functional outcome after this unfortunate complication. © 2005 Wiley Periodicals, Inc. Head Neck 27: 543–548, 2005

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The patient is an otherwise healthy 56-year-old woman with T2N0 subglottic laryngeal cancer

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who was initially treated with 7680 cGy hyper-fractionated radiation therapy. After the failure of this initial treatment course, the patient underwent a total laryngectomy and bilateral selective neck dissections with radial forearm fascial free tissue flap for soft tissue coverage. No pharyngeal resection or reconstruction was required. The patient subsequently had a pharyngoesophageal stricture develop that was managed with serial esophageal dilations. These dilations occurred every 4 to 12 weeks beginning 4 months after surgical resection.

All dilations were performed over a guidewire placed under direct visualization. During her last dilation, the pharyngoesophagus was dilated from 8 mm (24 F) to 14 mm (42 F). There were no apparent complications from this procedure, although minor mucosal lacerations were observed at the site of the stricture. Sixteen days later, the patient was seen in the emergency department with a 1-week history of throbbing left neck and shoulder pain that radiated to her back. Physical examination revealed an afebrile patient in no acute distress with stable vital signs. Her neu-

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rologic examination was within normal limits. The laboratory examination revealed a white blood cell (WBC) count of 11.8×10^9 /L. Contrast-enhanced CT of the neck was performed to rule out an abscess versus tumor recurrence at the neural foramina. This study showed no obvious acute abnormalities.

On postdilation day 20, the patient was seen in the otolaryngology clinic with persistent left neck and shoulder pain. Physical examination revealed a patient in obvious discomfort. Her neck was slightly asymmetric with left supraclavicular fullness. Neurologic examination revealed an inability to adduct the left shoulder, raising suspicion of a new cranial nerve XI weakness. The rest of the cranial nerves were within normal limits. MRI of the cervical spine was ordered.

Thirty-six hours after the clinic visit (postdilation day 22), the patient returned to the emergency department complaining of progressively worsening neck pain with new-onset headache, vomiting, and mental status changes. Physical examination demonstrated a temperature of 100.5°F and mild tachycardia. The neurologic examination was significant for slightly decreased flexion strength in the left upper extremity, but the remainder of the neurologic examination was normal. Her WBC count was 14.7×10^9 /L, with 92% neutrophils. Basic metabolic panel was normal. Attempts at lumbar puncture were unsuccessful. The patient was admitted to the medicine service for a presumed diagnosis of meningitis and was started empirically on intravenous (IV) ceftriaxone. The otolaryngology and neurosurgery services were urgently consulted, and an MRI of the brain and cervical spine was obtained. The MRI revealed an epidural abscess from C2 to C5-6, as well as abnormal marrow signal involving the vertebral bodies of C4-C5 and C6, suspicious for osteomyelitis (Figures 1 and 2).

While awaiting additional imaging studies to assess for tumor recurrence, the patient had left hemiparesis develop. The patient was then taken to the operating room emergently, where she underwent a posterior approach C3, C4, and C5 hemilaminectomy and drainage of the epidural abscess. Postoperatively, her sensory examination improved more significantly than did her motor examination. She displayed normal response to light touch globally but complete flaccidity of her left upper and lower extremity with normal to near-normal strength of the right upper and lower extremity. She was empirically



FIGURE 1. Sagittal T2-weighted MR image of the neck showing compression of spinal canal by epidural abscess.

placed on IV vancomycin, clindamycin, and ceftriaxone and followed by the infectious disease service throughout her course. Antibiotic therapy was tailored to culture results. She was maintained on a 6-week IV course of antibiotics.

Postoperative MRI showed a persistent epidural mass from C3–C5 predominantly involving the left anterior spinal canal that had been reduced in size compared with preoperative imaging studies. This was thought to be phlegmon rather than an acute abscess, and the decision was made to manage with observation and IV antibiotics rather than further intervention. The patient remained stable and was discharged to rehabilitation on postoperative day 8. The following day, the patient began to experience right-sided (contralateral) shoulder pain and weakness. An urgent MRI of the cervical spine was obtained and revealed recurrence and extension of the epidural abscess (Figure 3).

The patient underwent urgent drainage of the epidural abscess through an anterior approach C4–C5 corpectomy by both neurosurgery and otolaryngology teams, as well as anterior/posterior fusion of C3–C6. During this procedure, a



FIGURE 2. Sagittal MR image, T1 with gadolinium again showing epidural abscess.

linear posterior mucosal tear was observed at the site of the pharyngoesophageal stricture. The prior free tissue transfer was mobilized off the anterior wall of the pharynx and placed posteriorly to create a barrier between the spine and the pharynx. A new anterior pharyngostoma was created, and a salivary bypass tube was placed. Postoperatively, both sensory and motor function of her right side returned to baseline while the left-sided hemiparesis improved slowly. Over many months, the patient regained significant motor function in her left upper extremity and some motor function in her left lower extremity to the point that she could use a walker for mobility.

Eight months after the aforementioned procedure, the patient underwent an elective pharyngocutaneous fistula closure with local turn in flaps and a deltopectoral regional flap for external coverage. One month later, her deltopectoral flap was detached and salivary bypass tube removed. There were no apparent complications during the procedure. Unfortunately, 4 days later, she was readmitted for meningitis with symptoms of headache, neck pain, nausea, vomiting, and fever. A swallow study revealed a tract from

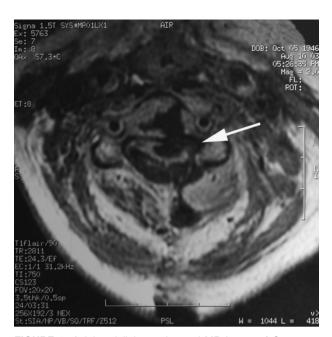


FIGURE 3. Axial gadolinium-enhanced MR image of C4 vertebrae with extension of abscess into spinal canal.

the pharynx to the anterior cervical fusion plate at the C5 level (Figure 4).

She then was taken to the operating room for a pharyngectomy, esophageal closure, and creation of a pharyngostoma as a controlled salivary fistula. She again clinically decompensated with

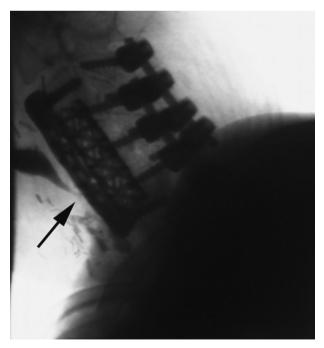


FIGURE 4. Esophagram with contrast communicating between esophagus and anterior spinal plate.

mental status changes a few days later. She was found to have a cerebrospinal fluid (CSF) leak from her anterior neck wound from a pharvngeal-subarachnoid fistula. A neurosurgery/otolaryngology combined team then performed an exploration of the neck wound, removal of anterior cervical plate while keeping the posterior hardware intact, CSF leak repair, lumbar drain placement, and pectoral flap for coverage. The patient recovered with the help of long-term rehabilitation but was left with a significant decrease in left lower extremity strength showing only trace movements. Her left upper extremity showed some finger and hand mobility but only 2 of 5 strength on that side. Fortunately, the right upper and lower extremities were completely normal.

DISCUSSION

Esophageal dilation is a procedure that can greatly enhance the quality of life for patients with larvngeal cancer who have pharvngeal strictures develop and have undergone radiation treatment. Potential complications include those from the use of anesthetic medications, sore throat, infection, bleeding, gastroesophageal reflux, aspiration, and esophageal perforation.1 Esophageal perforation is the most worrisome risk, and, fortunately, its incidence with simple bougies is between 0.04% and 0.40%. Esophageal perforation may lead to serious sequelae such as mediastinitis or sepsis; therefore, postdilation patients are typically warned to contact their physician if they experience severe chest pain, back pain, tachycardia, or fever. To our knowledge, this is the first case reported in the literature of pharyngoesophageal tear presenting as an epidural abscess after esophageal dilation. Factors that may contribute to the likelihood of this complication include pre-existing soft tissue damage (ie, caused by prior radiation therapy), early osteomyelitis or osteoradionecrosis, and the presence of a free flap anteriorly. Our aim is to heighten the awareness of the surgeon to this rare clinical presentation to prevent delays in diagnosis.

The diagnosis of spinal epidural abscess (SEA) is challenging because it is a relatively rare cause of neck or back pain. In addition, the initial symptoms may mimic several other conditions. As a result, 74% of cases of SEA are initially incorrectly diagnosed.² SEA is typically associated with the "classic triad" of localized back pain, progressive neurologic deficit (ie, paresthesias, reflex changes, paraparesis), and fever. However,

this triad occurs in only 37% of patients with SEA, and the combination of back pain and neurologic deficit alone remain the most reliable indicator of an SEA diagnosis.³ Symptoms that commonly indicate progression include bladder or bowel dysfunction, plegia, sepsis, or mental status changes.⁴

Laboratory data in SEA cases is nonspecific. Leukocytosis occurs in 60% of patients with an average value of $13.4 \times 10^9 / L.^3$ One study reported that patients with lower (<12 \times 10 $^9 / L$) WBC at presentation were likely to have a better functional outcome compared with patients seen with higher WBC (>14 \times 10 $^9 / L$) initially. The erythrocyte sedimentation rate (ESR) is nearly always elevated in SEA cases, with an average value of 51 mm/h.

The most helpful tools in diagnosis are imaging studies. Specifically, MRI with gadolinium enhancement is the "gold standard" for diagnosis. This will reveal an isointense or hypointense signal on T1-weighted images and a hyperintense signal on T2-weighted images.3 An SEA will appear as an area of peripheral linear enhancement (inflammatory tissue) surrounding nonenhancing (purulent or necrotic) matter. During the phlegmonous stage, the MRI will show homogenous enhancement representative of granulomatous tissue.4 Advantages of MRI are rapid and accurate location of inflammatory foci, definition of the entire extent of involvement, plan of surgical approach, and usefulness in monitoring therapy with decreased enhancement of a resolving abscess.3 Also, MRI has the ability to detect frequently coexisting infections such as osteomyelitis (76%) or paraspinal abscess (54%).²

The case demonstrated an area on MRI suspicious for osteomyelitis of the vertebral bodies. Radiation was used as a therapeutic modality in this case. Previous radiation in the setting of esophageal dilation along with osteoradionecrosis may have played a role in predisposing to infection. Although the mandible is the most commonly affected site for osteoradionecrosis, there have been case reports of osteoradionecrosis affecting cervical vertebrae. The irradiated bone is predisposed to chronic nonhealing tissue because of cellular injury and fibrosis.

If MRI is not available or is contraindicated, CT myelography is the next best choice. This modality is also highly sensitive but presents a risk of transforming an epidural infection into a subarachnoid space process. In addition, it is not as capable of defining the lower extent of the lesion as a gadolinium-enhanced MRI. Plain spinal radiographs may show erosion of vertebral bodies adjacent to the disk space but do not provide adequate sensitivity for use in the diagnosis of SEA.³

The first-line treatment for SEA is prompt surgical management. This includes spinal cord/thecal sac decompression with drainage of the abscess. The abscess may be approached from either a posterior or anterior direction. The approach may be dictated by the location of the abscess. In addition, the medical and surgical history of the patient must be factored into this decision. The rationale for selecting a posterior approach over an anterior approach in our case was that the patient's anterior neck had been exposed to high-dose radiation and extensive surgical manipulation.

The only patients that should not consider surgical therapy as first-line treatment are those who have (1) underlying medical conditions that make them a poor surgical candidate, (2) complete paraplegia for greater than 48 to 72 hours, (3) extensive spinal involvement prohibiting surgery, or (4) minimal or no neurologic signs or symptoms.^{2–4} These patients are managed medically with antibiotics and require frequent neurologic examinations and serial MRIs. Any signs or symptoms of neurologic deterioration should be followed by immediate re-evaluation for surgical management.^{2–4}

All patients diagnosed with SEA should immediately begin a course of IV antibiotic therapy with empiric coverage. Once the responsible organism is isolated and sensitivities are determined, the antibiotic regimen can be tailored. Anaerobes, such as Fusobacterium, Peptostreptococcus magnus, and Bacteroides fragilis, predominate in infections originating from an esophageal perforation.8 However, a variety of organisms such as Enterococcus, Klebsiella oxytoca, Staphylococcus aureus, and Streptococcus species may also be seen.8 In our case report, culture obtained from surgical drainage showed numerous Fusobacterium nucleatum, Streptococcus intermedius, Enterococcus species, and Hemophilus influenza. Although blood cultures are positive in more than 60% of cases, if possible, the antibiotic choice should be based on direct culture from the abscess. IV antibiotics should be continued for a minimum of 4 weeks and possibly longer, depending on a patient's immunocompetency, presence of coexisting osteomyelitis or paraspinal abscess, results of follow-up MRIs,

levels of inflammatory markers, and presence of systemic bacteremia.³

There has been a fair amount of disagreement regarding prognostic variables in the neurosurgery literature. Most authors agree that patients initially seen with a more severe neurologic deficit have worse outcomes relative to patients with less severe deficits.9 Patients with back pain and/or radiculopathy have a relatively better likelihood of functional improvement than patients with more severe clinical presentation, such as bowel/ bladder dysfunction (53% show improvement), paresis (45.5% show improvement), plegia (33.3% show improvement), or sepsis (0% show improvement). Another factor that may predict outcome is the duration of neurologic symptoms before treatment initiation. In the recent literature, there are both studies supporting the presence of correlation between timing of treatment and outcome and studies showing the absence of this correlation. Most research suggests that treatment of patients initially seen with paresis. incontinence, or plegia within 72 hours is associated with a higher incidence of good outcome than delayed treatment. The degree of thecal sac compression caused by the epidural mass may also affect clinical presentation and future functional status. Neurologic symptoms after the occurrence of SEA are thought to be caused by direct spinal cord and nerve root compression and involvement of the epidural venous plexus, resulting in cord ischemia.9 Surgical decompression of an abscess that is causing severe thecal sac compression (>50%) is likely to be most beneficial within 72 hours after symptoms arise. After this point, infarction may be the predominant cause of neurologic deficit, and thus decompression will not be significantly beneficial.9

Younger patients have shown better outcomes than older patients; however, this may be related to the fact that older patients are more likely to have medical comorbidities. In addition, the spinal cords of younger patients may have more plasticity than those of older patients. Last, there has been no proven advantage of either surgical versus nonsurgical treatment when comparing outcomes between age groups. This comparison is a difficult one to make considering the inherent differences in the two populations of patients.

CONCLUSION

Epidural abscess is an extraordinarily rare complication of pharyngoesophageal dilatation and is widely unrecognized by otolaryngologists. A neurologic deficit in a patient who has recently undergone esophageal dilation should raise suspicion for possible pharyngoesophageal tear resulting in epidural abscess. An MRI with gadolinium enhancement and neurosurgical consultation should be ordered for patients with even the slightest suspicion of epidural abscess. Early diagnosis and treatment are the keys to successful management and good functional outcome. A combination of surgical decompression, abscess drainage, and IV empiric antibiotic coverage remain the first-line treatment recommendations.

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