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Cervical Epidural Abscess Secondary to Aorto-Duodenal Fistula : A Case Report

Although cervical epidural abscess is rare, it should be strongly suspected in any patient with unexplainable neck pain and fever, especially when the patient has a predisposing factor for this infectious process. The authors report a case of cervical epidural abscess in a 39-yr-old man with an aorto-duodenal fistula, which complicated the interposition of artificial graft for abdominal aortic aneurysm rupture, which had undertaken 40 months before. Timely detection and intervention rendered him a full neurological recovery. This extremely rare case is presented with a literature review.

Key Words : *Epidural Abscess; Causality; Fistula, Aorto-Duodenal; Transplants; Aortic Aneurysm, Abdominal*

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INTRODUCTION

Spinal epidural abscess (SEA) is not frequently reported. This disease is preferably located in the thoracic and lumbar areas according to the width of the epidural space at each level of the spine (1). And in these levels, abdominal or retroperitoneal pathologic process is a predisposing factor (2, 3). The authors describe a very rare case of cervical SEA and suggest a possible pathogenetic mechanism.

CASE REPORT

A 39-yr-old man presented with a 4-day history of bilateral painful leg swelling, hematemesis, and melena. On admission, he showed pale, dehydrated, and acutely ill appearances with unstable vital signs: blood pressure 92/65 mmHg, temperature 39.5℃, heart rate 112/min, and respiration rate 32/min. The hemoglobin level was 9.3 g/dL, the hematocrit 27.8 %, the white blood cell count 11,100/μL, and the platelet count was 195,000/μL. Blood culture yielded no positive results.

Forty months before, he had been diagnosed with two dissecting aneurysms, one of them had arisen from the abdominal aorta just below the launching of the left renal artery and the other just above the bifurcation of the common iliac artery (Fig. 1). He underwent resection of the aneurysmal segment and interposition of artificial graft. At operation, dirty thrombotic materials had been impacted in the false aneurysmal sacs aside from the true aortic lumen. Histological examination of a surgical specimen showed a marked infiltration of various inflammatory cells with consequent fibrosis and perforations of the vessel wall. Bacteriologic examination of the same specimen did not find any causative organisms, probably due to ample preoperative antibiotic agents coverage.

On the present visit, gastrofiberscopy was performed under the assumption of the aorto-enteric fistula. A huge duodenal ulcer crater and an artificial aortic graft through an aorto-duodenal fistula were seen at the 3rd portion of the duodenum, which located in the retroperitoneal space (Fig. 2). An antiperistaltic extra-anatomic anastomosis between the greater curvature of the lower stomach and the proximal jejunum was undertaken. Bacteriological study revealed polymicrobial organisms, such as Salmonella, Enterococcus, and *Staphylococcus aureus*, suggesting a gastrointestinal origin. Pertinent antibiotics such as ampicillin, amikacin, and vancomycin were used continuously.

Four days after bypass surgery, he complained posterior neck pain and point tenderness. Simple plain cervical film revealed only mild degenerative spondylotic changes at C6-7 level with no evidence of osteomyelitis. We recommended careful observation. From the 9th postoperative day, he suffered voiding difficulty and urinary retention. Bladder dysfunction was suggested by urologic consultation, and urinary catheterization was done. Meanwhile, he developed intermittent fever up to 38.5℃ and the muscle power of his upper extremity decreased progressively, that had been masked by the severe neck pain.

At 16th postoperative day, walking difficulty suddenly

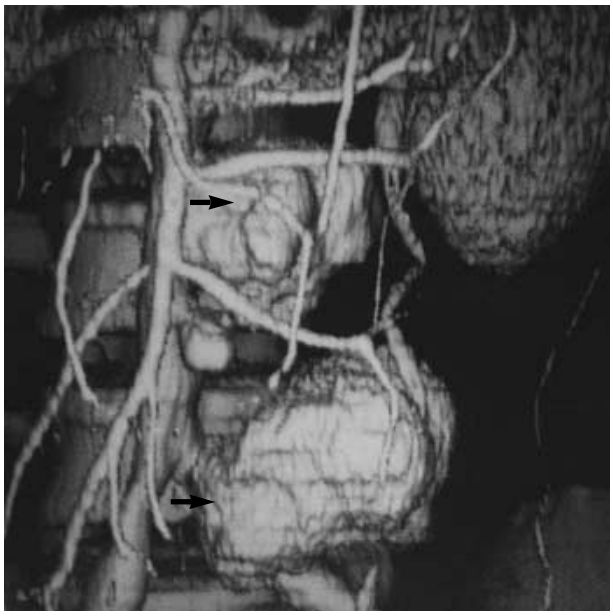

Fig. 1. Three-dimensional abdominal computed tomogram (CT) shows two aneurysmal dilatations of the abdominal aorta, one just below the launching of the left renal artery and the other just above the bifurcation of the common iliac artery (arrows).

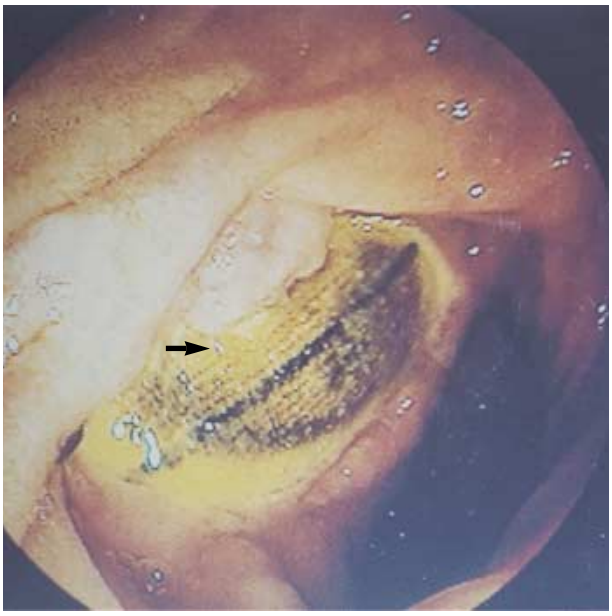

Fig. 2. Gastrofiberscopy demonstrates a large, 3×3 cm sized perforation on the 3rd portion of the duodenum and the artificial graft. Through this fistula was seen the artificial graft which was interposed during the previous operation (arrow).

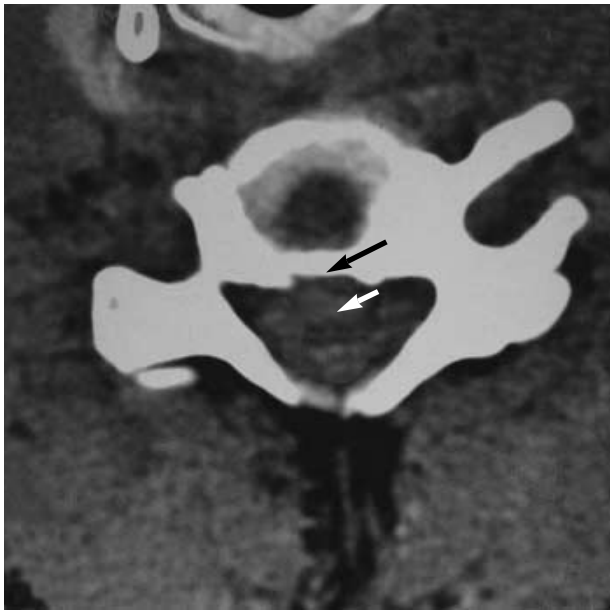

Fig. 3. Cervical axial computed tomogram (CT) discloses resorption of the posterior margin of the upper C6 vertebra body (long arrow) and peripherally-enhanced epidural granulation (short white arrow).

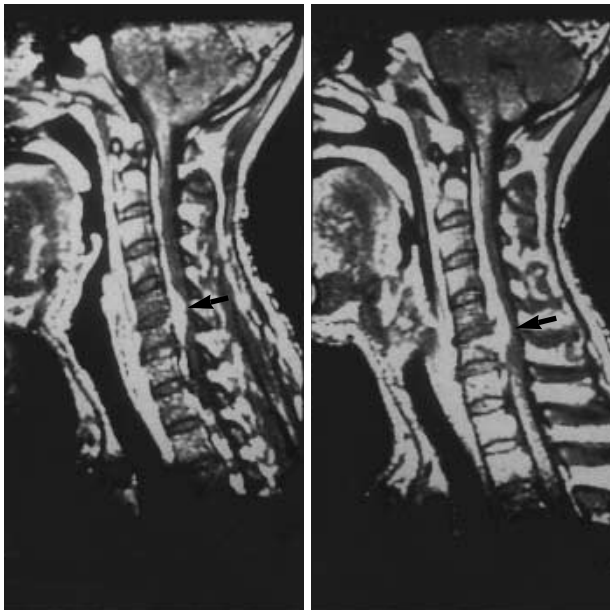

Fig. 4. Cervical sagittal magnetic resonance imaging (MRI) with Gadolinium enhancement shows an enhanced epidural space occupying lesion located anteriorly to the cervical spinal cord, which extends from C1 to C7 with maximum thickness at C5-6 level (arrows).

developed. He was transferred to the neurosurgical department for thorough evaluation. At this time, his neurologic examination revealed a grade II weakness of distal upper extremities bilaterally, especially in hand grasping and digits fanning, a grade III weakness of bilateral lower extremities, and paresthesia below the level of both clavicles. Cervical computed tomogram (CT) revealed resorption of the posterior margin of the upper C6 vertebral body and suspicious peripherally-enhanced epidural granulation (Fig. 3). Subsequent cervical magnetic

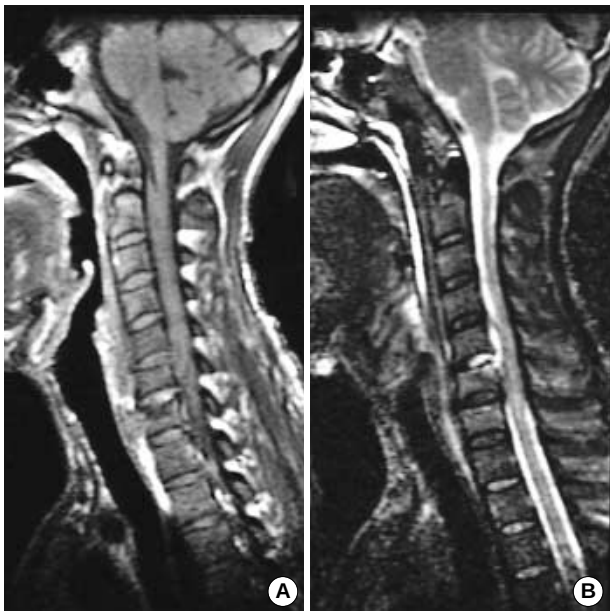

Fig. 5. Cervical T1-weighted sagittal MRI (A) only shows a low signal degenerative change at upper part of C7 vertebral body but T2-weighted sagittal image (B) shows focal high signal intensity lesion anterior to the spinal cord at C5-6 level.

resonance imaging (MRI) study with contrast agent revealed an enhanced epidural space occupying lesion anterior to the cervical spinal cord with maximum thickness at C5-6 level (Fig. 4). T2-weighted image also showed focal high signal intensity lesion anterior to the spinal cord at C5-6 level but T1-weighted image disclosed inconclusive finding (Fig. 5). The patient showed increased deep tendon reflexes and pathologic reflexes at this time point.

An emergent abscess drainage was performed in this patient with general anesthesia. He was placed in prone position and a laminectomy at C5-6 level was undertaken via a posterior approach. At this moment, the posteriorly displaced cord was visible and a thin-walled, yellowish cavity which contains gel-like purulent material was identified on the anterior and lateral aspect of the cord. For fear of undue retraction of the cord and spillage of the contents, gentle aspiration with a 23-gauge needle was done and dissection of abscess wall with surrounding structures was performed. Identification of the posterior longitudinal ligament was followed by meticulous irrigation. Finally a two-way drainage catheter was inserted for both instilling antibiotics and draining necrotic tissue debris. The pathologic report revealed only a diffuse infiltration of lymphoid cells and a focal collection of neutrophils, but no causative organisms were identified in the bacteriologic examination.

Postoperatively, he did not complain a neck pain or abnormal sensation, and was able to ambulate again. Wound irrigation was performed until the 7th postoperative day, when no further exudative fluid egressed. Systemic antibiotics were used for 4 weeks. The patient was discharged with no neurological sequelae. He has been well until the last follow-up.

DISCUSSION

Concerning SEA, clinicians must be aware of the early detection and swift management to improve the outcome of patients harboring this illness (4). Many cases of SEA remain undiagnosed until paraplegia is evident. Paralysis was often overlooked because of excruciating neck pain or of other serious medical problems (5). For this reason, surgeons should seek for potential premorbid conditions responsible for imminent clinical symptoms and signs. In general, the majority of SEA is found posterolaterally in the thoracic or lumbar region, where the spinal canal has the widest dimension (1, 6). Fewer than 20% of these lesions have been reported to occur anteriorly. However, the cervical SEA are typically located in the anterior epidural space due to the lack of a posterior epidural space at this level (1, 7). Although cervical epidural abscess is relatively rare, it should be considered in any febrile patients with unexplained neck pain and weakness, especially when the patient has a predisposing factor for this disease (8).

The pathogenetic mechanisms responsible for the SEA encompass either spread by a hematogenous route from skin, urinary tracts, lung, or dental infections, or by direct extension from a contiguous focus, mainly from vertebral osteomyelitis (1, 2). Infection is thought to reach the vertebrae hematogenously, probably via the arterial route from a remote site, although this culprit site is not always identified. The infection then spreads to the epidural space by direct extension (4). Plain cervical films and CT scans revealed only normal to mild degenerative changes and resorption of vertebrae, but enhanced MRI provided diagnostic clue in our case. These radiological features were comparable to those of another report (7, 9). Under these circumstances, osteomyelitis is thought to be a prerequisite for SEA, however, there may exist routes other than arterial or direct spread. The mechanism of spinal cord compromise and its clinical manifestation is unclear: it may result from a decrease in arterial blood flow, venous thrombosis, or it may be caused by direct compression of the spinal cord (5). Considering the rapidity with which the neurologic symptoms progress, most of the damage is probably related to the thrombosis of the vessels draining from the spinal cord (4).

Staphylococcus aureus has been reported to be the most common infectious organism (2, 10). The polymicrobial nature of the aorto-duodenal fistula swab strongly suggests a gastrointestinal source of infection in general. However, failure to identify a causative microorganism in the operative drainage of cervical SEA in the present case might be due to the 16-day coverage of systemic antibiotic agents before surgical intervention.

In the present case, the artificially grafted area of the abdomi-

nal aorta may rupture into the retroperitoneal space. A concomitant fistulization into the digestive tract may result in an aorto-digestive fistula. On the contrary to the primary aorto-digestive fistula, secondary aorto-digestive fistula (paraprosthesis-enteric fistula) complicates an aortic suture (postanastomotic or postoperative fistulae). Secondary fistulae are more common than primary ones (10). Two main mechanisms of prosthetic rupture are digestive erosion of the prosthesis along the suture line, and a rupture of false aneurysm by organization of a postoperative hematoma; rupture is the major late complication of any aortic repair in the treatment of aneurysm (10). Most of the secondary rupture cases occur into the duodenum, and the association between aortic infection and duodenal rupture is mainly observed in the lumbar region (1, 3). In our case, the secondary aorto-duodenal fistula resulted in anterior cervical epidural abscess rather than lumbar area, and the retroperitoneal venous structures may have played a role in spreading the infection into the remote site, although this was not proven.

Traditional treatment of SEA comprises an initial decompression and surgical debridement followed by a long course of systemic antibiotics administration (6, 11). With regard to the surgical approach to the cervical SEA, Piccolo et al. insisted anterior debridement without fusion in cases where the lesion was located lower than C4 level and spanned for not more than 3 vertebral segments (7). Young et al. performed the anterior debridement and fusion with successful results in patients with cervical osteomyelitis associated with epidural abscess (12). The present authors performed a posterior surgical approach, because the lesion expanded for more than 3 vertebral segments and more importantly, the vertebral bodies seemed to be involved on the very limited area.

Most of the literature supports 4 to 6 weeks of systemic antibiotics, usually antistaphylococcal penicillin or depending on the culture results, followed by oral antibiotics for further 2 to 3 months depending on the extent of osteomyelitis (8). The duration of antibiotics treatment ranged from 4 weeks (in the absence of osteomyelitis) to 12 weeks (for patients with resistant osteomyelitis) (9). Treatment would be conservative, however, if patients had mild symptoms without neurologic involvement or if they were medically unstable and poor surgical candidates (5). In this special setting, the most favorable results are obtainable when antimicrobial treatment was combined with resection of the aorta or the aortic graft and extra-anatomic bypass (3). However, in the sequential occurrence of epidural abscess following fistula repair, abscess drainage combined with antibiotics administration is sufficient to control and eradicate the infection.

Although there were no evidences for causative microbial agents, several clues could be suffice to believe that this cervical SEA was secondary to the paraprosthesis-duodenal fistula, which were temporal occurrence of neck pain, sensory changes, and motor weakness as well as predisposing factor such as aorto-duodenal fistula. Prompt recognition and management warrants successful postoperative results.

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