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A Case of Refractory Salmonella Spondylodiscitis in an Immunocompetent Patient Treated Via an Extracavitary Approach, Corpectomy and Placement of Expandable Cage

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Abstract: Problem statement: Salmonella spondylodiscitis is a rare condition that is more prevalent in patients with sickle cell disease or immunosuppression. However, it can also be found in immunocompetent patients. It usually responds well to proper antibiotics. However, surgery is rarely indicated when the infection is refractory to antibiotics, the spine is destabilized or there is an epidural component that is compressing the thecal sac and causing neurological deficit. We report the case of a 60 year-old African American woman with borderline diabetes mellitus controlled with diet, who presented with Salmonella osteomyelitis of the thoracic spine that was associated with an epidural abscess. **Approach:** The patient presented to an outside hospital with diarrhea, fever and chills. This occurred after eating a chicken sandwich. Stool and blood cultures were positive for Salmonella. Initially she was placed on triple antibiotics (ceftriaxone, vancomycin and levofloxacin) and was later switched to levofloxacin alone given that the bacteria were sensitive to this drug. Several weeks later, she presented with severe back pain. Imaging studies showed a significant lytic lesion in her thoracic spine at T11-12 with an epidural component. Given the instability of the lesion and the failure of medical treatment a decision was made for surgical intervention. **Results:** She underwent an extracavitary approach for partial T11-T12 corpectomies and fusion with instrumentation with continuous administration of the proper antibiotics. The patient was then discharged to an acute patient rehabilitation center. Postoperatively, she was able to ambulate with full strength in her extremitiesand her back pain had resolved. **Conclusion/Recommendations:** To the best of the authors' knowledge this is the first reported case of Salmonella spondylodiscitis in an immunocompetent patient treated with a corpectomy via an extracavitary approach with the use of an expandable cage and posterior instrumentation.

Key words: Salmonella, spondylodiscitis, epidural abscess, corpectomy, extracavitary approach, expandable cage

INTRODUCTION

Despite the fact that salmonella spondylodiscitis accounts for less than 0.5% of all bone infections, it causes significant difficulties in management and can be associated with increased morbidity and mortality for it usually eludes timely diagnosis (Ozturk *et al*., 2006; Korovessis *et al*., 2008). Although it is usually managed with intravenous antibiotics, surgical intervention is indicated in cases of neurological compromise, spinal deformity and concomitant epidural abscess (Arnold *et al*., 1997). The usual need for a corpectomy during the operation makes the stable reconstruction of the anterior column of the spine imperative. The use of titanium mesh cages and most recently expandable cages through an anterior approach has revolutionaries this process (Korovessis *et al*., 2008; Hee *et al*., 2002; Kuklo *et al*., 2006; Liljenqvist *et al*., 2003; Fang *et al*., 1994; Altman *et al*., 1996; Bhat *et al*., 1999; Auguste *et al*., 2006; Arts and Peul, 2008). In an effort to minimize the morbidity associated with the anterior approach, we report to the best of our knowledge the first case of vertebral osteomyelitis to be treated with an extracavitary approach. In fact this appears to be the first case of salmonella spondylodiscitis in an immunocompetent patient in which an expandable cage was used.

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MATERIALS AND METHODS

Presentation: This 60 year-old African-American woman with a history of childhood rickets, borderline Diabetes Mellitus (DM) type 2and arterial hypertension developed food poisoning characterized by severe diarrhea after eating a chicken sandwich. She was admitted to an outside hospital with dehydration, fever and chills. Stool and blood cultures were positive for salmonella. She was initially started on triple antibiotics (vancomycin, ceftriaxoneand levofloxacin). She was then kept on levofloxacin alone since the bacteria were sensitive to this antibiotic. The patient did well and was discharged home on levofloxacin to return to the outside hospital several weeks later with fever, chills, severe back pain and inability to walk. On further questioning, she revealed that she had recently fallen from a standing position. Her physical exam revealed that she was febrile (T 102°F). Her strength in her lower extremities was full. However, she was unable to ambulate due to pain. Her sensory examination was intact to light touch and pinprick. Her spine was tender to palpation at the thoracolumbar junction. Computed Tomography (CT) and Magnetic Resonance (MR) imaging of the thoracolumbar spine revealed a destructive lesion at T11 and T12 causing a kyphotic deformity with a retropulsed fragment into the spinal canal Fig. 1 and an epidural abscess causing spinal cord compression Fig. 2 and 3 Imaging studies of the rest of her spine did not reveal any other sites of infection. Laboratory findings included a White Blood Cell (WBC) count of 13,200 mm^{-3}, C-Reactive Protein (CRP) of 22.3 mg dL^{-1} and Erythrocyte Sedimentation Rate (ESR) of 82 mm h^{-1}. Treatment options were discussed with the patient and given the existence of an epidural abscess and the severity of the lytic lesion at T11/T12 with an inability to ambulate, she was offered surgery.

Operation: After the patient was intubated uneventfully she underwent an extracavitary approach that has been described by other authors (Snell *et al*., 2006; Shen *et al*., 2008; Sciubba *et al*., 2007), T11-12 corpectomies, drainage of the epidural abscess, placement of expandable cage (Synex®, Synthes, West Chester, PA) and T9-L3 posterior instrumentation and correction of the kyphotic deformity Fig. 4. Intraoperatively, purulent discharge was encountered in the affected vertebral bodies and samples were sent for culture. Intraoperative monitoring using Somatosensory Evoked Potentials (SSEPs) and Motor Evoked Potentials (MEPs) remained at baseline throughout the procedure. Two drains were placed; the muscles and fascia were closed in layers and the skin with staples. There were no intraoperative complications. Her postoperative examination revealed full strength in all her extremities.

Fig. 1: Preoperative sagittal Computed Tomography (CT) image of the thoracic spine revealing a destructive lesion at T11 and T12 causing a kyphotic deformity with a retropulsed fragment into the spinal canal

Fig. 2: Preoperative sagittal T2 weighted Magnetic Resonance Imaging (MRI) of the thoracic spine revealing a destructive lesion at T11 and T12 causing a kyphotic deformity and an epidural abscess causing spinal cord compression

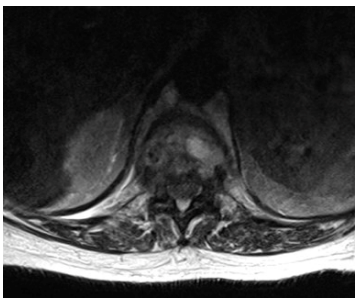

Fig. 3: Preoperative axial T2 weighted Magnetic Resonance Imaging (MRI) at the level of the T12 vertebra revealing a destructive lesion of the T12 vertebral body and an epidural abscess causing spinal cord compression

Fig. 4: Postoperative sagittal CT of the thoracic spine demonstrating the position of the construct

Post-operative course: Postoperatively, she was transferred to the neurosurgical Intensive Care Unit (ICU) overnight and was consequently transferred to the regular floor. The infectious disease service was consulted and she was kept on levofloxacin. Postoperatively, her motor strength remained normal in all her extremities and her sensory examinations remained intact to light touch and pinprick. Her pain was well controlled. On postoperative day 4, she was transferred to an acute rehabilitation center to follow a course of physical and occupational therapy in order to gain mobility and ambulatory independence. She also continued on levofloxacin orally (750 mg daily) for an additional 6 weeks post-operatively. Further workup was performed in an effort to reveal any immunologic compromise, infection with HIV and sickle cell anemia or sickle cell trait, which was negative. On follow up visits, she was able to ambulate without any assistance and her back pain had resolved except from some stiffness. Likewise, her infection had resolved and laboratory values had normalized two months postoperatively (WBC was 6,160 mm^{-3}, CRP was 0.5 mg dL^{-1} and ESR was 30 mm h^{-1}).

RESULTS AND DISCUSSION

Salmonella is a non-spore-forming gram-negative bacillus of the family Enterobacteraciae, which can be easily cultured on simple media (Osebold, 2008). In most cases humans ingest the organism from contaminated food or water and their small bowel becomes its habitat (Osebold, 2008). Salmonella can be either quiescent in an asymptomatic carrier state or manifest as gastroenteritis, typhoid fever, or bacteremia (Cobos *et al.*, 1993; Gupta *et al.*, 2004). Infections with this bacterium represent a rare cause of osteomyelitis since the advent of antibiotics, accounting for approximately<% of all cases (Cobos *et al.*, 1993; Gupta *et al.*, 2004). These cases are thought to occur secondary to hematogenous spread after an episode of bacteremia (Ozturk *et al*., 2006; Cobos *et al.*, 1993; Gupta *et al.*, 2004) as is the case with our patient. Most incidences of salmonella osteomyelitis are caused by *S. typhimurium* and *S. enteritidis*, whereas the typhi serotypes have been rarely reported (Cobos *et al.*, 1993).

Salmonella osteomyelitis is usually seen in patients that are immunologically compromised (Osebold, 2008). It occurs more often in people suffering from sickle cell disease or sickle cell traitand other hemoglobinopathies, but also in conditions that may suppress the immune system such as chronic alcoholism, hematologic malignancies, Systemic Lupus Erythematosus (SLE), Diabetes Mellitus (DM), solid tumors, long term therapy with steroids and possibly Acquired Immunodeficiency Syndrome (AIDS) (Cobos *et al.*, 1993; Gupta *et al*., 2004; Chambers *et al*., 2000; Vichinsky and Lubin, 1980; Wu *et al*., 2004). In osteomyelitis patients without sickle-cell anemia the infection is attributed to Salmonella in only 0.5% of the cases, one forth of which involves the spinal column (Ozturk *et al*., 2006). The most common spinal region involved in salmonellosis is the lumbar area, followed by the thoracic region that was the one affected in our patient (Santos and Sapico, 1998). The major clinical manifestations and laboratory findings include fever, back pain, leukocytosis and elevated ESR and CRP (Ozturk *et al*., 2006; Wu *et al*., 2004; Santos and Sapico, 1998; Carragee *et al*., 1997).

Various authors have reported the rare occurrence of salmonella osteomyelitis involving the spine in immunocompetent patients (Ozturk *et al*., 2006; Santos and Sapico, 1998; Carvell and MacLarnon, 1981; Dolan *et al*., 1987; Hunt *et al*., 1996; Hunt, 1965; Le, 1982; Miller *et al*., 1988; Mnaymneh, 1977; O'Keeffe *et al*., 1978; Ortiz-Neu *et al*., 1978; Sapico and Montgomerie, 1979; Schweitzer *et al*., 1971; Govender *et al*., 1999; Cottalorda *et al*., 1997; Abdullah *et al*., 2008; Acharya and Bhatnagar, 2004; Akagi *et al*., 1998). From these groups a few have reported surgical intervention (Ozturk *et al*., 2006; Wu *et al*., 2004; Hunt *et al*., 1996; Le, 1982; Mnaymneh, 1977; Abdullah *et al*., 2008; Acharya and Bhatnagar, 2004; Akagi *et al*., 1998) for the treatment of the infection (Table 1). None of these case reports though, provides a possible explanation for this incidence. Salmonella infection is more prone to disseminate in areas of inflammation or trauma (Osebold, 2008), probably because of the higher vascularity and the local inflammatory activity rendering the capillaries of the affected tissue more permeable. We speculate that the cause of our patient's osteomyelitis was her mild spinal trauma secondary to the fall she sustained.

Table 1: Reports of immunocompetent patients treated with surgery for salmonella spondylodiscitis

Author	Year	Procedure	Indication	Outcome
Mnaymneh	1977	Anterior drainage of paravertebral abscess and vertebral interbody fusion with bone graft	T8-9 spondylodiscitis, spinal cord compression	Mild lower extremity weakness 6 months postoperatively consequently lost at follow up
		Anterior drainage of paravertebral abscess and vertebral interbody fusion with bone graft	L2-3 spondylodiscitis, spinal cord compression	Neurologically intact
Le	1982	Anterior debridement and vertebral interbody fusion with bone graft	L1-2 spondylodiscitis, back pain refractory to antibiotic treatment	Neurologically intact
Hunt *et al*.	1996	Posterior debridement	Sacroiliac osteomyelitis	Neurologically intact
Santos and Sapico	1998	L4 hemilanectomy, L5 laminectomy, nerve root decompression and debridement	L5-S1 spondylodiscitis and spondylolesthesis, epidural abscess, cauda equina syndrome	Neurologically intact, mild back pain
Akagi *et al*.	1998	Anterior debridement and vertebral interbody fusion with bone graft	C5-7 spondylodiscitis, epidural abscess, spinal cord compression, tetraplegia	Ambulatory with crutches 6 months postoperatively
Acharya and Bhatnagar	2004	Anterior debridement and anterior fixation with bone graft	L1-2 spondylodiscitis and L1-2 paravertebral collection	Ambulating on custom spinal orthoses
Ozturk *et al*.	2006	Anterior debridement, T8-10 corpectomy with titanium mesh cage, T6-12 posterior instrumentation and fusion	T9 spondylodiscitis, spinal cord compression	Neurologic status at Frankel E
Abdullah *et al*.	2008	T3-4 costotransversectomy and epidural abscess drainage	Epidural abscess	Not mentioned

Salmonella spodylodiscitis is usually treated with the appropriate antibiotic therapy. Cases of medically controlled Salmonella spodylodiscitis and paravertebral abscess have been reported (Arnold *et al*., 1997). Surgical intervention is necessary however, when epidural abscesses, neurologic impairment or instability are present (Arnold *et al*., 1997; Chang, 2005; Matsui *et al*., 1998; Suchomel *et al*., 2003). Other indications include intractable pain, failed medical therapy and the need to establish tissue diagnosis (Arnold *et al*., 1997; Chang, 2005; Matsui *et al*., 1998; Suchomel *et al*., 2003). There is much evidence suggesting that better outcomes can be achieved with surgery in comparison to medical treatment (Quinones-Hinojosa *et al*., 2004) in patients with intractable pain.

There has been a long lasting debate on whether instrumentation in spondylodiscitis patients increases their risk of persistent infection by providing a scaffold for biofilm formation and thus reducing antibiotic effectiveness (Korovessis *et al*., 2008). Titanium mesh cages though appear to be a safe choice since titanium has been shown to be less prone to bacterial colonization than stainless steel (Chang and Merritt, 1991). There is currently strong evidence in the literature that titanium mesh cages provide better biomechanical results than simple bone grafts in spondylodiscitis patients, without increasing the risk of complications and especially the risk of persistent infection (Ozturk *et al*., 2006; Korovessis *et al*., 2008; Kuklo *et al*., 2006; Liljenqvist *et al*., 2003; Fang *et al*., 1994; Altman *et al*., 1996; Bhat *et al*., 1999). Hee *et al*. (2002) was the first group to support the superiority of titanium cages in the treatment of spondylodiscitis patients. They have shown a statistically significant improvement in sagittal alignment in patients receiving cages or posterior instrumentation only in comparison to those treated with simple bone grafts. Additionally, those who received cages had greater correction of their coronal alignment than did the patients without cage ($p = 0.0006$).

Various groups have extended these promising results with titanium mesh cages to the use of the newer expandable cages in patients with the same characteristics (Auguste *et al*., 2006; Arts and Peul, 2008). However, this is the first report of the use of an expandable cage after corpectomy in Salmonella spondylodiscitis in an immunocompetent patient. To minimize the surgical burden of our patient we used for both procedures a single extracavitary approach that has been previously described (Snell *et al*., 2006; Shen *et al*., 2008; Sciubba *et al*., 2007) was the first group that showed excellent results with the use of the extracavitary approach in patients with metastatic tumors of the lumbar and thoracic spine. Sciubba *et al*. (2007) have expanded the use of this technique to include patients with spinal trauma and prior vertebral osteomyelitis. These patients however, were operated for correction of deformity several years after their infection has been medically treated with success. On the other hand, our patient is the first one reported who has been operated with this technique during the active phase of a salmonella infection, a factor that did not negatively affect her recovery. Despite the limited

surgical exposure, our patient did not develop any intra or postoperative complications, she remained neurologically intact and her infection resolved promptly. It is felt (Sciubba *et al*., 2007) that this technique imparts a lower morbidity associated with a lone posterior approach, while conferring the benefits of anterior spine decompression and active intraoperative kyphosis correction via an expandable cage.

CONCLUSION

In summary, we report the first case of salmonella spondylodiscitis in an immunocompetent patient, treated with an expandable cage for corpectomy reconstruction and posterior instrumentation through an extracavitary approach. The surgical exposure allowed for removal of the infected bone, drainage of the epidural abscess and stabilization of the spine. We therefore demonstrate that this procedure is an excellent option for the treatment of patients with infectious processes of the spine that needs surgical drainage and deformity correction.

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