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Acute Paraplegia Due to *Salmonella brandenburg* Spondylodiscitis: Case Report

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Abstract

The authors present the case of a 48-year-old man admitted for acute onset of paraplegia in a patient suffering from backaches for 1 week. The rapidly progressive motor disturbances had been evolving for approximately 12 hours. The entire spinal MRI showed an epidural mass at T4-T6 associated with extensive lesions of spondylodiscitis and a T7-T8 vertebral body loss of height. A large six-level laminectomy was performed. A tumoral etiology couldn't be entirely excluded intraoperatively so that no fusion has been done at that time. The pathological exam revealed acute inflammatory lesions with no argument in favor of a tumoral process. Bacteriological exam of the pathological specimen and stools cultures were positive for *Salmonella brandenburg*. An episode of gastroenteritis after the ingestion of a pizza has been evoked. The antibiotic medication was prescribed for 12 weeks. Postoperative evolution was favorable with a possible march between bars 6 weeks after. The authors emphasize the pseudo-tumoral presentation in an immunocompetent patient, the lack of complications and the post-ingestion mechanism.

Keywords

Spondylodiscitis, Spinal Epidural Abscess, Paraplegia, *Salmonella brandenburg*, Post-Ingestion, Immunocompetent

1. Introduction

Background. Infections of the spine are still being described in medical literature as spondylitis, discitis, vertebral osteomyelitis, septic arthritis of the facet joints,

spondylodiscitis (SD) and/or spinal epidural abscess (SEA). Terms as pyogenic and hematogenous are often added with claimed intention to point out the non-specific infectious character and, respectively, the most frequently encountered modality of spreading. Also, the term spontaneous is mostly reserved for infections other than post-surgical. The presence or the absence of a spinal epidural abscess could bring out a spinal cord compression which constitutes a neurosurgical emergency. That's why, for more practical reasons, we should classify spinal infections in spondylodiscitis (or vertebral osteomyelitis) with or without spinal epidural abscess.

SD accounts for 2% of bony infections [1]. SEA comprises for 0, 2-2 cases for 10,000 hospital admissions [2].

Age. SEA occurs in patients aged 30 - 60 and two times more frequently in men [2] [3] [4] [5]. Elderly people are more susceptible to developing SD [6] [7].

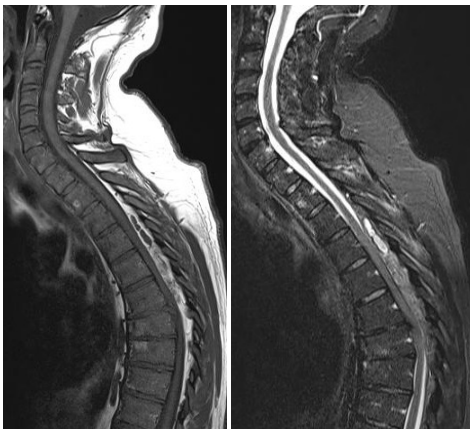
Level. The lumbar spine is affected in 50% of cases with SD [8] [9] [10]. The thoracic level was reported in over 50% of cases with SEA, followed by lumbar, then cervical [8] [11], with a prevalence of midthoracic spine (T6-T8) [12]. SEA appears more frequently in the cervical spine (90%) upon certain authors [13], or lumbar spine in another series [4] but causes a neurological deficit in the thoracic spine (80%) [13]. Over 80% are posterior to the cord [11] [14].

The lumbar spine seems to be more affected by Salmonella infection [15] [16] [17] [18] [19]. Some authors claim that thoracic SD with Salmonella in immunocompetent patients is extremely rare [20].

2. Case Report

A 48-year-old man, with no significant history, has been admitted in the Neurosurgical Emergency Room for acute onset of paraplegia for approximately 12 hours. He had been experiencing progressively increasing backaches for over a week and that morning he awoke noticing he couldn't move his legs. He saw his general doctor and he had been prescribed non-steroidal anti-inflammatory drugs. Examination revealed a 0/5 paraplegia with loss of bladder and bowel function and a sensory level at T8. No fever was noted. Radiographic studies showed an extended T4-T7 area of osteolysis and a T7-T8 vertebral body loss of height. Preliminary laboratory exams showed a white blood cell count (WBC) at 10.600/ml, fibrinogen at 6.9 g/l, platelets at 161.000/ml and TCA ratio at 1.35. Blood cultures remained negative the whole course of hospitalization. MRI showed T6-T7-T8 vertebral body and discal lesions with an epidural mass at T4-T6 with a compressive effect on the spinal cord and bone marrow signal intensity abnormalities (**Figure 1**). He underwent emergent T3-T8 laminectomy and an easily aspirating, greyish/yellowish mass with a purulent liquid at the superior part has been noticed. The relative spinal stability and the tumor-evoking MRI aspect determined us to perform no fusion at that time. The pathological exam revealed an inflammatory, neutrophilic infiltrated adipose tissue with no arguments for a tumoral primitive or metastatic process. Bacteriological specimen exam and stool cultures were, somehow unexpectedly, positive for *Salmo-*

nella brandenburg. The patient's last two-week history revealed an episode of gastroenteritis after the ingestion of a pizza. Antibiotics were prescribed (Rocephin 2 g QD and Tavanic 500 mg QD prior to antibiogram, Tavanic 500 mg QD and Cotrimoxazole BID after obtaining the antibiogram) for 3 months. Immobilization was realized with a thoracolumbar orthosis for 3 months. HIV/AIDS test was negative. Thoracic and abdominal CAT scan showed no evidence of mycotic aortic aneurysm or other infectious foci. Postoperative anemia was due to intra-operative blood loss. Biological exams showed a decrease of C-reactive protein (CRP) from 51.9 mg/l to 5.3 mg/l (normal values < 6 mg/l) at 3 weeks postoperatively. Clinical evolution was favorable, with a 3/5 paraparesis and a caned-march at 6 weeks. He had a urethral sonde at discharge. An MRI performed 4 weeks after the operation showed the resolution of pyogenic lesions (Figure 2). The patient was discharged 7 weeks later for a re-educational hospital.



(a) (b)

Figure 1. (a) Sagittal MRI T1 gado: T6-T7-T8 vertebral body and discal lesions with an epidural mass at T4-T6; (b) Sagittal MRI T2: T6-T7-T8 vertebral body and discal lesions with an epidural mass at T4-T6.

Figure 2. Post operative sagittal dorsal MRI T2: resolution of pyogenic lesions.

3. Discussion

Presentation: Spondylodiscitis associated with a SEA is difficult to recognize disease since the presentation is non-specific [2] [21] [22]. Medical literature reveals high rates of inaccurate first-time diagnosis. A 40-cases retrospective series reports over 25% of misdiagnosed and discharged patients with SEA [23]. For Tang *et al.* the initial accurate diagnostic rate was 11% [24].

Clinical presentation like back pain, fever, local signs and/or neurological impairment associated with an inflammatory syndrome (CRP, WBC, ESR) point out a possible spinal infection [3] [4] [11] [23] [24] [25] [26]. Meningismus may be also associated [19] [21] [27]. Paralysis appears in 34% of cases with SEA [2].

Fever is absent in one-third of the patients [2]. In fact, some authors claim that spondylodiscitis without fever is a diagnostic challenge and they conclude that spondylodiscitis should always be considered when the differential diagnosis is made [28]. Some series of patients with Salmonella spondylodiscitis reported the presence of back pain in 100% of cases and fever in 75% [29].

History of more than 3 months of symptoms is present in 50% of cases [8] [9].

Our patient's presentation was unusual for he had no fever or local signs at the time of admittance and he had been experiencing backaches for only a week. His immunologic condition seemed normal since his medical history was insignificant. His minimal leucocytosis (10.600/ml) and the moderate rise of fibrinogen (6.9 g/l) could be hardly interpreted as specific signs of infection. Also, leucocytosis correlated with lytic lesions on radiographic studies could point out a neoplasia condition. Negative blood cultures added no argument for a spinal infection.

MRI: Plain radiographs may reveal vertebral body collapse and lytic lesions within the vertebral body or posterior spinal elements [4]. MRI is the golden standard nowadays [1] [2] [5] [14] [23] [26] [30], though CT-scan, scintigram and plain X-rays may be helpful [4]. Typical MRI characteristics of SEA, disc and involved vertebral bodies are heterogeneous iso/hypointense on T1-weighted images and hyperintense on T2-weighted images [22]. Inhomogeneous enhancement of disc-vertebral body complex following contrast administration is usually seen on T1-weighted images [1] [4] [22] [31]. Pronounced peripheral enhancement orientates the diagnosis toward an abscess rather than a cystic lesion (old hematoma or serum) [14] [31]. Signal changes of the vertebral body bone marrow can be seen either in infectious or neoplastic diseases of the spine. Differential diagnosis should be made with leptomeningeal metastases [14] [27], epidural hematoma [14], meningitis, acute transverse myelitis, inter somatic herniation [11], multiple myeloma or other hematologic neoplasia, vertebral body collapse followed osteoporosis [9].

Treatment. Treatment can be either conservative (antibiotics and/or immobilisation) or surgical (especially with neurological impairment) [2] [23] [32]. Neurosurgical indications in spinal infections are a diagnostic biopsy, progressive neurologic deficit, spinal instability and failed medical response [12].

Still, no consensus exists in literature concerning indication for surgical treatment for spinal infections. In one series, 90% of patients with SD were treated effectively by non-surgical means [33]. A complete paralysis for more than 3 days seems to be a specific criterion for conservative therapy [34]. SD medical treatment with immobilization and antibiotics would permit 75% pain relief and spontaneously fusion [4]. The use of a thoracolumbosacral orthosis was evoked by certain authors as external stabilization [9]. On the other hand, in a study of 75 patients with SEA, Rigamonti *et al.* conclude that conservative treatment seems to be reserved for selective cases [26].

The main treatment of SEA seems to be early surgery [4] [7] [13] [23]. Primary instrumentation may be considered [9]. There is no consensus among authors for the use of a certain type of surgery or for immediate fusion [35]. In a study of 163 patients, Karadimas *et al.* found that decompression alone was less effective compared with decompression and internal stabilization [33]. The use of metallic implants was reported successful in all 22 patients with spondylodiscitis by Robinson *et al.* [36]. The use of immediate internal fusion, with better materials and broad-spectrum antibiotics, would allow early mobilization, thus avoiding complications such as deep vein thrombosis [7]. Nevertheless, Bostrom *et al.*, in a study of 46 patients with SEA concluded that instrumentation does not seem to be imperative [37]. Nakase *et al.* reported two-stage surgical management of their patients [38]. On a technical note, Greiner-Perth *et al.* present their good results with the use of a less invasive surgical approach of SEA [39].

The use of an anterior or posterior approach is still controversial. Many authors prefer the anterior approach [7], though the posterior approach seems more logical since 80% of SEA are dorsally situated [11] [14] [21]. However, the posterior approach would bear the risk of postoperative instability. The use of intraoperative sonography and a drainage system are also debated [21] [34]. The risk of congestion of irrigation fluid is cited [21].

Some authors cite the postoperative complication rate of 33% [12].

In our case, we chose the posterior approach since the SEA was posterior. Imagistic preoperative and intraoperative criteria of spinal stability determined us to consider fusion as not necessary at that moment. Moreover, we think that an external stabilization should be firstly considered in such cases. Immediate postoperative evolution and pathological and bacteriological exams will dictate an eventual second-time fusion.

Antibiotics: The period of time for antibiotic therapy is between 4 - 6 weeks [19] [28] to 3 months [22] [40] and should comprise a period of 4 - 6 weeks of parenteral administration [12]. Diagnosis should be confirmed by direct exam [6] [10] [27] or blood culture before prescribing antibiotics [4] [9] [28] [40] [41] (nevertheless up to 50% of cultures are negative) [7] [11] [21]. The treatment with Quinolone is considered by a great majority of authors the best for spinal infections with Salmonella [17] [19] [32] [42].

In our case, we used a combination of a quinolone with a sulfonamide (Le-

vofloxacin-Cotrimoxazole) and we considered that a minimum period of three months would be necessary. CRP levels and clinical evolution will be followed and antibiotic therapy will be modified accordingly.

Salmonella brandenburg: The most popular germ is by far *Staphylococcus aureus*, comprising for 40% - 80% of infections [5] [7] [8] [9] [10] [11] [14] [26], followed by other Gram-positive cocci (*Streptococcus*) [3] [26]. Gram-negative organisms, such as *Escherichia coli*, *Pseudomonas*, *Proteus*, *Diplococcus pneumoniae*, *Serratia*, *Enterobacter*, and *Salmonella* seem to have an increasing incidence, especially due to IV drug use [4]. Also, spinal infections with anaerobic bacteria (Bacteroides, Peptostreptococcus), fungi and parasites have been cited [10].

The most common *Salmonella* serotypes were: S choleraesuis C1 [29] [43], S typhimurium and S enteriditis [16] [17] [19] [42], S paratyphi B [44], S typhi [3] [18] [32] [45].

In a study of 151 patients, Ben Taarit *et al.* found that Salmonella was the third cause of pyogenic SD, following Staphylococcus and Brucella [6], whereas Maiuri *et al.* reported Salmonella as the fourth cause of SD in a study of 65 patients [1].

In a similar case report, Abdullah *et al.* present a thoracic SEA caused by Salmonella Typhi in a patient suffering from diabetes mellitus, with a skin infection. Favorable outcome was obtained after early surgery and antibiotic therapy (Ceftriaxone and Ciprofloxacin) [45].

Perras *et al.* report the case of a 59-year-old man who developed *Salmonella enteritidis* SD 4 weeks after bronchopneumonia and was treated with antibiotics for 3 months (Ciprofloxacin) [46].

Liu *et al.* present the case of a 60-year-old woman who developed a thoracic SD with *Salmonella choleraesuis*, treated successfully with antibiotics (ceftriaxone) after percutaneous transpedicular biopsy, in spite of a 4/5 graded lower limbs weakness at admittance [47].

Özturk *et al.* present the case of a 53-year-old woman under immunosuppressive treatment who developed a thoracic Salmonella typhimurium SEA 2 month after laparoscopic cholecystectomy. Treatment consisted of anterior and posterior fusion after decompression and antibiotic therapy (Ciprofloxacin) for 6 weeks followed by complete recovery 15 months later [16].

We can conclude that the SEA with Salmonella seems to be more frequent in aged and/or immunosuppressed patients [48] [49].

Spreading. The most common is hematogenous spreading from various foci: skin, parenteral, urinary tract, endocarditis, respiratory or upper digestive systems [35], the arterial spread being more frequent than venous transmission [9]. The latter is implied in genitourinary or gastrointestinal prior infections, posterior elements of the spine being mostly implicated [10]. Direct extension, post-operative and trauma are other mechanisms of transmission, though no source can be identified in up to 50% [11]. Pathogenesis of the neurologic deficits comprises a mechanical factor (compression of a SEA, the collapse of the

vertebral body), an ischemic factor (decreased arterial flow, venous stasis due to local thrombosis) and a toxic factor (inflammatory products) [22], the first being considered the most important [21].

Postingestion: The prior digestive signs seem non-specific for the infection with Salmonella. In fact, in certain series of spondylodiscitis with Salmonella, gastrointestinal symptoms were completely absent [29]. Further, the post-ingestion mechanism appears to be very rarely encountered in literature. Our patient's gastroenteritis and positive stool cultures for Salmonella point out ingestion as the probable mechanism of infection. Consequently, venous spreading through Batson's valveless epidural venous plexus seems to be more probable.

Immunologic condition, Co-morbidities. A large consensus among authors states that spinal infections are associated with chronic diseases in up to 65%. Conditions like diabetes mellitus, IV drug abuse, advanced age, cancer, alcoholism, liver cirrhosis, HIV/AIDS, chronic renal failure, repeated urinary tract infections, medical acts will compromise immunity and will favor the development of infection [4] [5] [7] [9] [14]. Many authors found a frequent association of SD with aortic aneurysms [29] [50] [51], sickle-cell disease [11] [26] [44] [52] or systemic leptospirosis [49]. SEA—mycotic aortic aneurysm association seems to affect especially aged, immuno-depressed patients, being highly lethal [50] [53], whereas SD—sickle-cell disease association appears more frequent in the pediatric population [54]. Some authors state that routine echocardiography should be implied in infections with Staphylococcus or Streptococcus [40].

In a review of 44 patients of Salmonella SD, Santos and Sapico found that fever was present in 87%, blood cultures were positive in 48% of cases and mycotic aortic aneurysms were seen in the over 50-year-old group [51]. In a series of 8 patients with Salmonella SD, 2 had concomitant mycotic aortic aneurysm [29]. Among the germs that provoke SD, Salmonella is the most common cause of mycotic aortic aneurysm [53]. Among *Salmonella serotypes*, *S. typhi* and *S. choleraesuis* are the most common species associated with mycotic aortic aneurysms [50]. Chen SH *et al.* report 3 cases of *S. choleraesuis* and 1 case of *S. enteritidis* SD associated with mycotic aortic aneurysm [53].

No complications were described in our case probably due to the patient's age and prior good condition. A thoracolumbar CT-scan revealed no aortic aneurysmal lesion. The early surgical decompression (within 18 hours) and the targeted antibiotics therapy permitted a favorable evolution.

Outcome. Favorable in spondylodiscitis with variable reports concerning pain-free rate (25% - 100%) [3] [11]. Outcome may be fatal in approximately 6% - 20% of the patients with SEA [2] [4] [7] [11] [12] [25] [37]. Morbidity rates are very high in patients with previous neurological deficits (even for those operating within 12 hours of onset) and depend on early diagnosis [22] [23] [25] [43]. In a study of 101 patients with spinal infection, 23% of patients recovered completely after surgical treatment [13]. In another study of 24 cases, 8 from 13 patients with neurological deficits never recovered [7]. In a study of 27 cases of

SEA, the 3 primary paraplegic patients never improved. The authors concluded that thoracic localization has the poorest outcome [21]. However, complete recovery after paraplegia remains possible [7] [22].

Our patient's short follow-up doesn't allow us to formulate definitive conclusions concerning his neurological evolution. Two months after the intervention his motor, sensorial and bowel disturbances improved (3/5 paraparesis, walk with walking sticks and an external anal sphincter control) but bladder dysfunctions remained unchanged.

4. Conclusion

Spinal epidural abscess (SEA) is an uncommon disease. Symptoms involve a classic triad of fever, backache and neurological deficits but all the symptoms are rarely seen at the first contact. The treatment of SEA involves emergent laminectomy, drainage of the abscess and antibiotic therapy. Prompt diagnosis and early surgical intervention, are associated with a favorable outcome.

Conflicts of Interest

The authors declare no conflicts of interest regarding the publication of this paper.

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