Burkholderia Pickettii Spondylitis

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Abstract

Study design: Case report describing *Burkholderia pickettii* spondylitis in a healthy adult.

Objectives: To describe this very rare form of spondylitis and to discuss some of the difficulties in the diagnosis of *Burkholderia pickettii* spondylitis.

Setting: Department of Orthopaedic Surgery, Nayoro City General Hospital, Japan.

Methods: A 48-year-old woman presented with a complaint of severe back pain radiating from the right side of her chest. Plain radiographs of the spine showed osteolytic destruction of the right side of the T10 vertebral body at T10 level, with an involvement of the pedicle. Magnetic resonance image of the spine showed a low signal intensity from the T10 vertebral body on a T1-weighted image and an increased signal intensity on T2-weighted sequence image. These lesions were enhanced when a contrast medium was used. The patient underwent open biopsy and specimens were collected through the right pedicle.

Results: Diagnosis was established on the basis of direct identification of the microorganism. Histological findings were consistent with examination of *Burkholderia Pickettii* spondylitis. Chemotherapy (intravenous cefepime and *per os* minocycline) resulted in complete cure.

Conclusion: *B. Pickettii* is widely distributed in aqueous sources in nature and has not previously been considered to be an aggressive pathogen towards humans. This case report will help to improve our understanding of the ecology and virulent pathogenicity of this organism. A biopsy is an essential and reliable method for the early etiologic diagnosis, which will lead to prevent the development of more severe complications such as spinal cord compression.

Key words; Spondylitis, *Burkholderia Pickettii*

Introduction

Burkholderia Pickettii is a nonfermentative gram-negative bacillus isolated commonly from environmental sources and only rarely from clinical samples.[1] The species has very rarely been identified as a human pathogen and has been associated with pseudobacteremias or in asymptomatic colonization of patients.[2,3] The cases in which infection has been reported generally involve simple bacteremia, related to contamination of manufactured products.[4-6] The authors describe a rare case of *B. Pickettii* spondylitis in a healthy adult.

Case report

A 48-year-old previously healthy woman was referred to our hospital in March 2003 with a complaint of severe back pain radiating from the right side of her chest. The pain had lasted for more than five weeks. Her past history was unremarkable, except that a total hysterectomy had been performed for hysteromyoma five years previously. On admission, the patient's temperature was 37.2 ^{o}C, her pulse rate was 66 beats / min, and her blood pressure was 110/70 mm Hg. No skin lesions were observed. Physical examination revealed tenderness on the spinous processes of the thoraco-lumbar spine. No neurological other deficits were identified. The peripheral white blood cell count was 5600 /mm^3, and her C-reactive protein level was 2.8 mg/dL. Other laboratory data including tumor markers were within normal limits. Cultures of blood and urine yielded negative results.

Plain radiographs of the spine showed destruction of the right-side vertebral body at T10 level, with an involvement of the pedicle (Figure1). An axial image of the computed tomography scan at T10 level showed an osteolytic area on the right side of the vertebra and pedicle (Figure2). Magnetic resonance image (MRI) of the spine showed a low signal intensity in the T10 vertebral body on a T1-weighted image (Figure3A) and a hyper-intense signal on T2-weighted sequence image. These lesions were enhanced with a contrast medium(Figure3B). A bone scan showed increased

uptake in the T10 vertebra and ^{67}Gallium scan showed pathological uptake in the same lesion.

The patient underwent open biopsy and specimens were collected through the right-side pedicle. Histopathological studies showed multiple foci of polymorphonuclear cell infiltration. No malignant lesion was identified. All cultures of samples obtained during biopsy yielded *Burkholderia Pickettii*. The isolate was sensitive to cefotaxime, cefepime, minocycline, and ciprofloxacin. The patient was treated with intravenous cefepime (1g / day) for 2 weeks, followed by *per os* administration of minocycline (100mg / day) for 3 months. The patient's condition slowly improved and her back pain subsided. At 18-month follow- up, the patient was free of symptoms. All laboratory data were within normal limits. Plain radiographs did not show further collapse of the vertebral body. MRI at T10 showed the same intensity as other vertebral bodies.

Discussion

B. Pickettii is classified in the rRNA homology group II.[7] It is widely distributed in aqueous sources in nature and has not previously been considered to be an aggressive pathogen towards humans.[2,3] Cases in which *B. Pickettii* infection has been reported, generally involve simple bacteremia related to contamination of hospital pharmacy sources.[4-6] Many cases were associated with a direct inoculation of the blood or another site of infection. Maki et al [4] reported an outbreak of nosocomial bacteremias caused by *B. Pickettii* in surgical patients and the linked this to the first traces in a contaminated narcotic. The epidemic was caused by fentanyl that became contaminated in the Central Pharmacy of their hospital. The contamination was introduced during convert removal of fentanyl from predrawn syringes and replacement by an equal volume of contaminated distilled water. Antiseptics such as chlorhexidine and ethacridine lactate have also been reported to be contaminated with *B. Pickettii.*[8,9] In addition to the bacteremia, the organism has been reported to cause endocarditis and meningitis.[10,11] These previous reports described that most of the *B. Pickettii* infection was associated with an immunocompromised state such as haemodialysis or cancer therapy.[10,11]

Spondylitis caused by *B. Pickettii* is extremely rare. Most of the reported cases of spondylitis or discitis are caused by more-virulent organisms.

Staphylococcus aureus is the more common pathogen, followed by gram-negative bacteria and *Streptococcus* species.[12] To our knowledge, only one case of *B. Pickettii* spondylodiskitis has been reported. Wertheim et al [7] described a case of spondylodiskitis in a patient with chronic renal failure. They emphasized that *B. Pickettii* might be a more virulent and invasive pathogen than previously recognized. As to the portal or mechanism of *B. Pickettii* entry, they suspected that the patient might have been infected during a preceding period of hospitalization, by introduction of *B. Pickettii* through an intravenous catheter or during cardiac catheterization. Moreover, their patient's situation was complicated by underlying diabetes, liver disease, and uremia and by administration of corticosteroids, all of which increased the risk of infection as previously described.[10,11]

In the present case, however, the patient was a previously healthy, ordinary housewife. Pyogenic spondylodiskitis is uncommon in healthy people. We believed that *B. Pickettii* was unlikely to be a contaminant because the specimens were taken using a careful, sterile technique. *B. Pickettii* was only isolated in pure culture from an intraoperative direct inspection of the bone, and despite extensive investigation, no alternative etiologic agent was identified. No cases of *B. Pickettii* infection were uncovered in our hospital. The route of entry for the organism remains unclear. Since blood cultures are negative in more than two-fifths of

spondylodiskitis, vertebra or disc-space aspiration biopsy offers an essential and reliable method for the rapid etiologic diagnosis, like in this case.[12]

Although clinical findings in the present case were unremarkable compared with other cases of pyogenic spondylitis, radiological findings showed some distinctive features. Plain radiographs showed that the bony destruction had spread over the pedicle. These images also demonstrated that the structure of the vertebral endplates were relatively well maintained. MRI at the infected vertebra exhibited a diffuse, homogeneous intensity area. This lesion was enhanced when contrast medium was used. Intensity changes in the intervertebral discs and vertebral endplates were not observed. Intravertebral abscesses were not found. All these findings are similar to metastases to the spine and can help aid the diagnosis of *B. Pickettii* spondylitis.

In summary, we report the first case of spondylitis due to *B. Pickettii* in a previously healthy adult, which will help us as physicians to improve our understanding of the ecology and virulent pathogenicity of this organism.

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Figure legend

Figure1

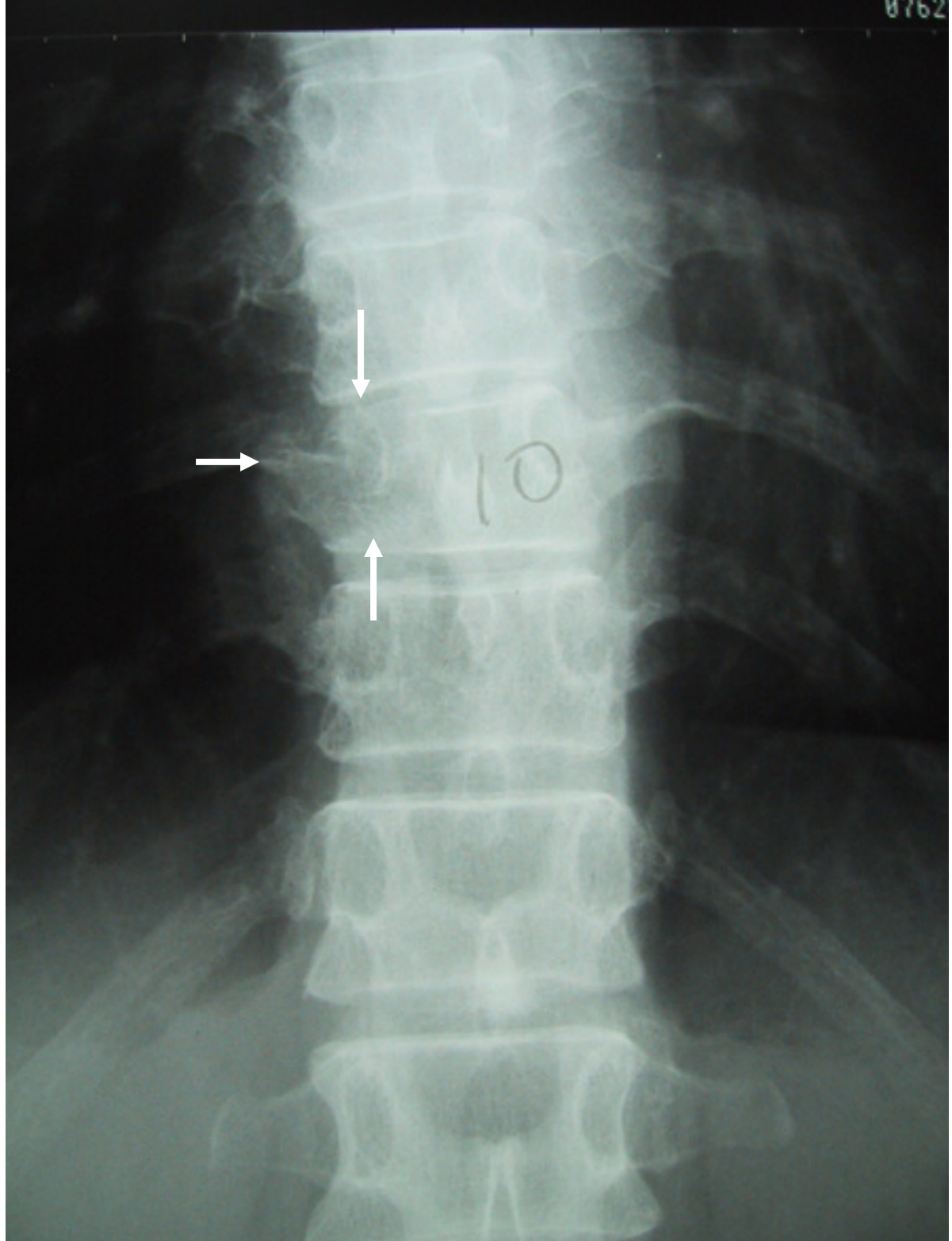
Plain radiographs of the spine showed destruction of the right-side vertebral body at T10 level, with an involvement of the pedicle.

Figure2

An axial image of the computed tomography scan at T10 level showed an osteolytic area on the right side of the vertebra and pedicle.

Figure3

MRI at T10 level demonstrated an osteolytic area on the right side of the vertebra and pedicle, which exhibited a diffuse, homogeneous low intensity on a T1-weighted image (Figure3A). These lesions were enhanced with a contrast medium (Figure3B).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