J Med Sci 2022;42(6):285-288
DOI: 10.4103/jmedsci.jmedsci_210_21

CASE REPORT

Polymicrobial Spontaneous Pyogenic Spondylodiscitis: A Very Rare Entity!

Vishal Mangal[1], Jayaraj Hasvi[1], Subir Dey[2], Anil Shankar Menon[1]

[1]*Department of Internal Medicine, Armed Forces Medical College,* [2]*Department of Neurosurgery, Command Hospital Southern Command, Pune, Maharashtra, India*

Spontaneous pyogenic spondylodiscitis (SPS) is an uncommon disease. The most common causative organism of SPS is *Staphylococcus aureus*, followed by Gram-negative bacilli in 4%–30% of the cases. Polymicrobial SPS is very rare, with only a few cases reported in the literature. We report a case of polymicrobial SPS in a 43-year-old male who was recently diagnosed with autoimmune hepatitis with cirrhosis of the liver and presented with a 1-month history of progressive low backache with radiation to both the legs. Initially, he was managed as a case of Pott's spine with antitubercular therapy on the basis of immunocompromised status with positive ascitic fluid *Mycobacterium tuberculosis* polymerase chain reaction. However, due to lack of clinical and radiological response after 6 weeks, he underwent computed tomography-guided biopsy of the affected region. The culture from the biopsy specimen yielded *Klebsiella oxytoca* and *Proteus vulgaris*. He was managed with culture-sensitive antimicrobials. To the best of our knowledge, this is the first case of polymicrobial SPS caused by *K. oxytoca, P. vulgaris,* and *M. tuberculosis*.

Key words: *Klebsiella oxytoca, Proteus vulgaris, Mycobacterium tuberculosis* spondylodiscitis, polymicrobial, case report

INTRODUCTION

Spontaneous pyogenic spondylodiscitis (SPS) is defined as the infection of the intervertebral disc and/or adjacent vertebra without any prior spinal instrumentation or procedure. It is an uncommon disease with an estimated incidence of 4 cases per million population per year in developed countries.[1] The most common causative organism of SPS is *Staphylococcus aureus*, followed by Gram-negative bacilli in 4%–30% of the cases.[2] Polymicrobial SPS is very rare, with only a few cases reported in the literature.[3,4] In the majority of these cases, tuberculosis was associated with a pyogenic organism. We could find only one case of SPS caused by *Klebsiella oxytoca* in a 51-year-old male with a history of intravenous heroin addiction.[5] Similarly, *Proteus* species have been rarely associated with SPS.[6] The SPS can also be caused by anaerobic organisms such as *Parvimonas micra* and *Fusobacterium nucleatum.* Establishing the microbiological diagnosis with conventional culture methods is difficult, and the newer methods such as Bactec blood culture system and matrix-assisted laser desorption ionization-time of flight mass spectrometry can help to solve this problem.[7] The co-infection by *K. oxytoca*, *Proteus vulgaris*, and *Mycobacterium tuberculosis* in a case of polymicrobial SPS has never been reported in the literature, and we report the first such case with a favorable outcome.

CASE REPORT

A 43-year-old male, recently diagnosed as a case of autoimmune hepatitis and diabetes mellitus on oral prednisolone 40 mg once a day for the last 8 weeks and oral antidiabetic drugs, now presented with complaints of low backache of 1-month duration along with radiation to both lower limbs. He denied any history of fever, fall, trauma, sphincter incontinence, or neurologic claudication. The general physical examination and neurological examination were unremarkable; however, he had Grade 2 ascites. Straight leg raise was bilaterally restricted to 60°. The spasm of the paraspinal muscles was present along with restriction of spinal movements, and he had an antalgic gait. Local tenderness was present at the level of the lower back. The baseline laboratory parameters are depicted in

Received: June 21, 2021; Revised: September 06, 2021; Accepted: September 09, 2021; Published: November 25, 2021
Corresponding Author: Dr. Vishal Mangal,
Department of Internal Medicine, India-Tajikistan Friendly Hospital, Qurghon Teppa, Bokhtar, Tajikistan - 735140.
E-mail: vishal_rimshi@rediffmail.com

How to cite this article: Mangal V, Hasvi J, Dey S, Menon AS. Polymicrobial spontaneous pyogenic spondylodiscitis: A very rare entity! J Med Sci 2022;42:285-8.

Table 1. His ascitic fluid *M. tuberculosis* polymerase chain reaction (MTB-PCR) was positive. He underwent magnetic resonance imaging (MRI) of the lumbosacral (L-S) spine which suggested infective spondylodiscitis [Figure 1]. With the background of glucocorticoid use, positive ascitic fluid MTB-PCR, and an MRI suggestive of infective etiology, the provisional diagnosis of Pott's spine was made and he was empirically started on antitubercular therapy (ATT) with tablet isoniazid 300 mg, tablet rifampicin 600 mg, tablet levofloxacin 750 mg, and tablet ethambutol 1200 mg once daily. The spinal immobilization was achieved using lumbar-sacral orthosis. The patient tolerated ATT well with mild improvement in his symptoms. Ten days later, the patient had worsening of low backache. On examination, his left ankle-deep tendon reflex was absent however the rest of the neurological examination was within normal limits. He underwent an urgent MRI of L-S spine which showed extensive destruction of the L5-S1 intervertebral disc [Figure 2]. The patient was managed conservatively with analgesics, and ATT was continued. The patient had mild improvement in his symptoms over the period of the next 6 weeks. A repeat MRI was done after 6 weeks which showed an increase in marrow edema of the fifth lumbar (L5) and first sacral (S1) vertebral bodies [Figure 3]. Because of increased inflammatory changes on imaging despite 6 weeks of ATT, the diagnosis of Pott's spine was reviewed. The patient was subjected to computed tomography (CT)-guided biopsy from the affected vertebra and disc. The culture of the CT-guided biopsy revealed *K. oxytoca* and *P. vulgaris*; however, no *M. tuberculosis* was detected on cartridge-based nucleic acid amplification test of the biopsy specimen. The diagnosis was revised to polymicrobial SPS, and the patient was started on culture-sensitive antibiotics injection cefoperazone plus sulbactam 2 g intravenous twice a day, injection amikacin 750 mg intravenous once a day along with tablet trimethoprim plus sulfamethoxazole 160 mg/800 mg twice daily and the ATT was stopped. His symptoms improved remarkably following 2 weeks of therapy. The injectable antibiotics were continued for a total duration of 6 weeks. He underwent a repeat MRI of the L-S spine which revealed progression of disease. At this stage, he was again started on ATT, considering that he already had ascitic fluid MTB-PCR positive with spondylodiscitis. His final diagnosis was spontaneous polymicrobial spondylodiscitis caused by *K. oxytoca* and *P. vulgaris* in the background of tuberculous spondylodiscitis. He underwent repeat MRI L-S spine after 8 weeks of ATT which suggested a 30% reduction in the soft-tissue component along with reduction of bone marrow edema. Informed written consent was taken from the patient for submission of this case report.

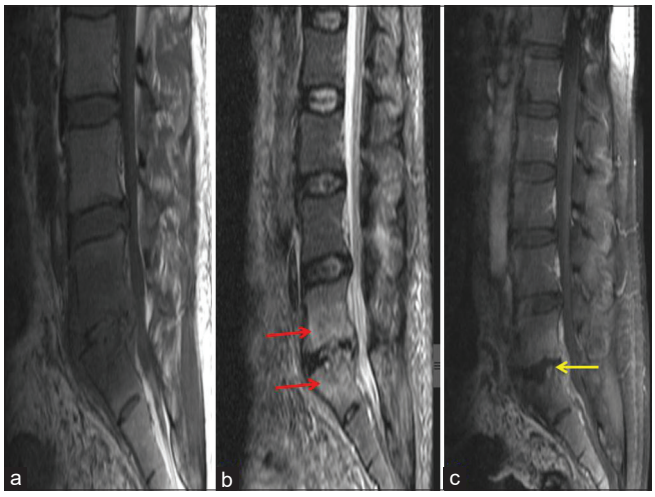

Figure 1: Baseline magnetic resonance imaging: (a) T1 sagittal, (b) T2 sagittal, (c) postcontrast images: The yellow arrows show mild hyperintense signal in the intervertebral disc between L5 and S1. The red arrows show marrow edema in L5 and S1 vertebral bodies. There is no enhancement of the intervertebral disc

DISCUSSION

Polymicrobial SPS is a rare disease with an indolent course. The low backache is present in 90% of the patients. The majority of the patients have paravertebral muscle tenderness and spasm, and restriction of spinal movement. Our patient also presented with low backache with pain radiating to the legs along with spasm of paraspinal muscles and marked limitation of spinal movements.

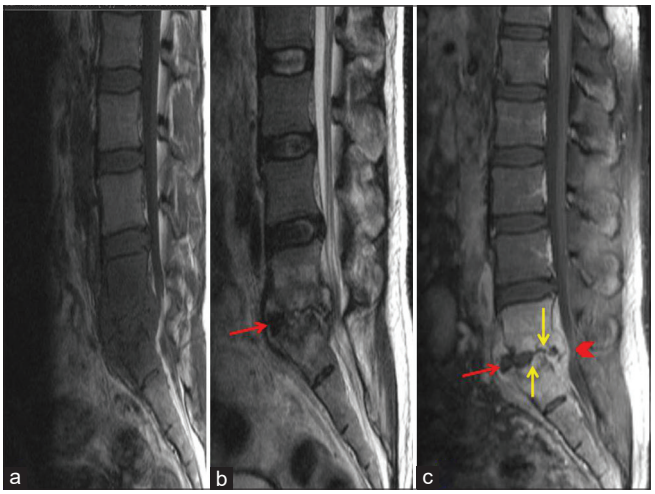

Figure 2: Magnetic resonance imaging after 10 days of baseline magnetic resonance imaging: (a) T1 sagittal, (b) T2 sagittal, (c) postcontrast images: Show extensive destruction of L5-S1 intervertebral disc with reduced disc height. The yellow arrows show an increase in the irregularity of the subjacent end plates with postcontrast enhancement. The red arrows show the anterior epidural soft-tissue component measuring 9.5 mm × 50 mm (anteroposterior × craniocaudal) indenting the cauda equina is seen (the red arrowhead)

Table 1: Laboratory parameters at different time points

Laboratory parameter	Baseline	After 6 weeks of ATT	Reference range
Hemoglobin (g/dL)	8.6	9.3	12-16
Total leucocyte count (cells/microL)	6300	2300	4000-11,000
Platelets (cells/microL)	102	114	150,000-450,000
Total serum bilirubin (mg/dL)	0.8	0.3	0.3-1
Serum aspartate aminotransferase (U/L)	104	25	10-40
Serum alanine aminotransferase (U/L)	48	29	10-40
Blood urea (mg/dL)	25	23	8-20
Serum creatinine (mg/dL)	0.82	0.75	0.50-1.10
Serum C-reactive protein (g/L)	Positive	Positive	1.00-10.00
Erythrocyte sedimentation rate (Westergren)	34	26	0-15 mm/Hr
Ascitic fluid MTB-PCR	Positive		
Blood culture	Negative		

PCR=Polymerase chain reaction; ATT: Antitubercular therapy; MTB=*Mycobacterium tuberculosis*

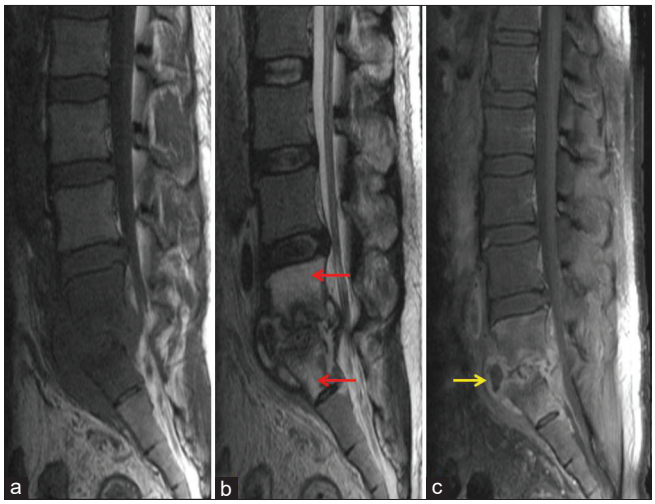

Figure 3: Magnetic resonance imaging after 8 weeks of baseline magnetic resonance imaging: (a) T1 sagittal, (b) T2 sagittal, (c) postcontrast images: The red arrows show an increase in marrow edema of L5 and S1 vertebral bodies as compared to previous scans [Figures 1 and 2]. A mild increase in sclerosis of L5 subjacent to L5-S1 disc is noted. The yellow arrow shows a minimal increase in the prevertebral collection; however, no significant change in the soft-tissue component as compared to previous scans [Figures 1 and 2]

Neurological complications such as cord or nerve root compression, meningitis, and cauda equina syndrome can be seen in 12% of the patients.[8] An epidural abscess is a dreaded complication marked by worsening back pain along with new-onset weakness. In our patient, pain worsened after 10 days of starting ATT and his left ankle reflex became absent. We also considered the possibility of nerve root compression or epidural abscess. An urgent MRI LS spine was done which ruled out the epidural abscess, however, compression of the cauda equina was noted.

The organisms mainly associated with polymicrobial SPS are *S. aureus,* coagulase-negative staphylococci, *Enterococcus faecalis, Escherichia coli,* and *Klebsiella pneumoniae.*[6] Recently, a prospective study enrolling 98 patients of pyogenic spondylodiscitis reported that only 12/98 patients had a polymicrobial infection. In these 12 patients, the most common organisms were coagulase-negative *Staphylococcus*, *Staphylococcus*, and *K. pneumoniae*. None of the patients had *K. oxytoca* or *P. vulgaris.* [9]

The most common risk factors which predispose to SPS can be classified as (a) patient-related: immunocompromised states such as active malignancy, glucocorticoid therapy for autoimmune diseases (daily dose of more than 10 mg of prednisolone and cumulative dose of more than 700 mg of prednisolone increase the risk of infections by 1.6 times),[10] diabetes mellitus, intravenous drug use, increased age, and harmful use of alcohol and cirrhosis of the liver; (b) concomitant infections: urinary tract infections, gastrointestinal tract infections, infectious endocarditis, postoperative infections, and sepsis; and (c) iatrogenic: spinal surgeries such as laminectomy, discectomy, vertebroplasty, spinal fixation, and diagnostic and therapeutic lumbar puncture. [2,11] Our patient had diabetes mellitus and cirrhosis of the liver, and he was on glucocorticoid therapy as risk factors.

The treatment of SPS depends on the accurate microbiological diagnosis, presence of neurological complications, and spinal deformity or instability. In our patient, the exact microbiological diagnosis was established and he was started on culture-directed antimicrobial therapy. The exact duration of antimicrobial therapy in SPS has not been established. Some studies advocate that 6 weeks of antibiotic therapy is optimum; however, others have demonstrated that

the chance of vertebral osteomyelitis relapse is less following 12 weeks of therapy.[12]

Surgical treatment is required for drainage of abscess, sequestrectomy, and subsequent mechanical stabilization of the spine. Surgery can be done by less invasive techniques such as endoscopic spinal surgery or by the classical open approach. The indications for surgery are limited to (1) poor response to medical therapy; (2) epidural abscess; (3) spinal instability or deformity due to bone destruction; and (4) imaging evidence of nerve roots, spinal cord, or dura mater compression. Epidural abscess and nerve or cord compression are indications for immediate surgical decompression.

Our patient had a 50% improvement in his functional status and pain after 6 weeks of therapy. However, the imaging suggested disease progression, hence he was again started on ATT with the background of positive ascitic fluid MTB-PCR and spondylodiscitis. Thus, we hypothesized that our patient had primarily tuberculous spondylodiscitis which was complicated by secondary culture-positive *K. oxytoca* and *P. vulgaris* infection. We plan to continue ATT for a total of 9 months.

CONCLUSION

Tuberculous spondylodiscitis is the most common occurrence in the background of immunocompromised state; however, there should be a high index of suspicion for the secondary bacterial infection in case of failure of radiological and clinical improvement. Early diagnosis of SPS by CT-guided biopsy and prompt treatment with culture-sensitive antimicrobial therapy can improve the outcomes and prevent the patient from lifelong morbidity arising out of the complications.

Acknowledgment

We are grateful to the Department of Radiology, Armed Forces Medical College, for their continuous support in the diagnosis of this patient.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given his consent for his images and other clinical information to be reported in the journal. The patient understands that name and initials will not be published and due efforts will be made to conceal his identity, but anonymity cannot be guaranteed.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

REFERENCES

1. Fantoni M, Trecarichi EM, Rossi B, Mazzotta V, Di Giacomo G, Nasto LA, *et al.* Epidemiological and clinical features of pyogenic spondylodiscitis. Eur Rev Med Pharmacol Sci 2012;16 Suppl 2:2-7.
2. Skaf GS, Domloj NT, Fehlings MG, Bouclaous CH, Sabbagh AS, Kanafani ZA, *et al.* Pyogenic spondylodiscitis: An overview. J Infect Public Health 2010;3:5-16.
3. Goehrs L, Baudry E, Bornand A. Spondylodiscitis due to *Mycobacterium tuberculosis* and *Streptococcus agalactiae*. Med Mal Infect 2014;44:486-7.
4. Boudokhane M, Mahmoud I, Saidane O, Mahmoud M, Ben Tekaya A, Tekaya R, *et al.* Double bacterial pyogenic spondylodiscitis: A diagnostic challenge. J Clin Rheumatol 2021;27:e69-70.
5. Sabio JM, López-Gómez M, Jiménez-Alonso J. Spontaneous spondylodiscitis caused by *Klebsiella oxytoca*. Ann Rheum Dis 2002;61:758-9.
6. Smits JP, Peltenburg HG, Mooi-Kokenberg EA, Koster T. Proteus mirabilis spondylodiscitis complicating a urinary tract infection. Scand J Infect Dis 2006;38:575-6.
7. Cleaver LM, Palanivel S, Mack D, Warren S. A case of polymicrobial anaerobic spondylodiscitis due to *Parvimonas micra* and *Fusobacterium nucleatum*. JMM Case Rep 2017;4:e005092.
8. Wisneski RJ. Infectious disease of the spine. Diagnostic and treatment considerations. Orthop Clin North Am 1991;22:491-501.
9. Widdrington JD, Emmerson I, Cullinan M, Narayanan M, Klejnow E, Watson A, *et al.* Pyogenic spondylodiscitis: Risk factors for adverse clinical outcome in routine clinical practice. Med Sci (Basel) 2018;6:E96.
10. Stuck AE, Minder CE, Frey FJ. Risk of infectious complications in patients taking glucocorticosteroids. Rev Infect Dis 1989;11:954-63.
11. Friedman JA, Maher CO, Quast LM, McClelland RL, Ebersold MJ. Spontaneous disc space infections in adults. Surg Neurol 2002;57:81-6.
12. Grados F, Lescure FX, Senneville E, Flipo RM, Schmit JL, Fardellone P. Suggestions for managing pyogenic (non-tuberculous) discitis in adults. Joint Bone Spine 2007;74:133-9.