—Case Reports—

Fungal Thoracic Spondylodiskitis in an Immunocompetent 14-year-old Girl

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Abstract

Fungal spondylodiskitis (inflammation of intervertebral disk tissue and adjacent vertebrae) is rare, particularly in immunocompetent patients. Here, we report a case of fungal and bacterial thoracic spondylodiskitis in a 14-year-old girl with abdominal and back pain. The spondylodiskitis was diagnosed on the basis of the presence of β-D glucan and the unusual clinical course, although cultures for fungus were negative. We conclude spondylodiskitis must be considered in cases of abdominal pain without clear etiology and in cases of fungal infection with unexplainable findings after standard treatment for bacterial infection, even when fungal cultures are negative.

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Key words: spondylodiskitis, fungal infection, childhood

Introduction

Spondylodiskitis (inflammation of intervertebral disk tissue and adjacent vertebrae) is being diagnosed with increasing frequency as a result of advances in magnetic resonance imaging (MRI) technology[1]. However, fungal spondylodiskitis in childhood is rare, particularly in immunocompetent patients. Here, we report a case of fungal and bacterial thoracic spondylodiskitis in a 14-year-old girl.

Case Report

A 14-year-old girl was admitted to our hospital with abdominal and back pain. Her history did not suggest immunodeficiency. Two months before admission, the patient experienced abdominal and back pain that resolved spontaneously. Four days before admission, she experienced abdominal and back pain again; antibiotics and nonsteroidal anti-inflammatory drugs were prescribed, but the pain increased. On admission, body temperature was 38.5℃, and physical examination revealed severe tenderness on the central back at the L3–4 level rather than in the abdomen. However, no redness or swelling was observed on the abdomen and back. Laboratory examination revealed the following: white blood cells, 7,820/μL (neutrophils, 68.4%; lymphocytes, 24.8%; monocytes, 4.6%; eosinophils, 1.8%; and basophils, 0.4%); red blood cells, 434×10^4/μL; hemoglobin, 12.9 g/dL; platelets, 26.5×10^4/μL; aspartate aminotransferase, 10 IU/mL; alanine aminotransferase, 18 IU/mL; C-reactive protein (CRP), 5.45 mg/dL; and erythrocyte sedimentation rate (ESR), 82 mm/h. Plain X-ray films of the spine showed no abnormalities (**Fig. 1a**). However, MRI

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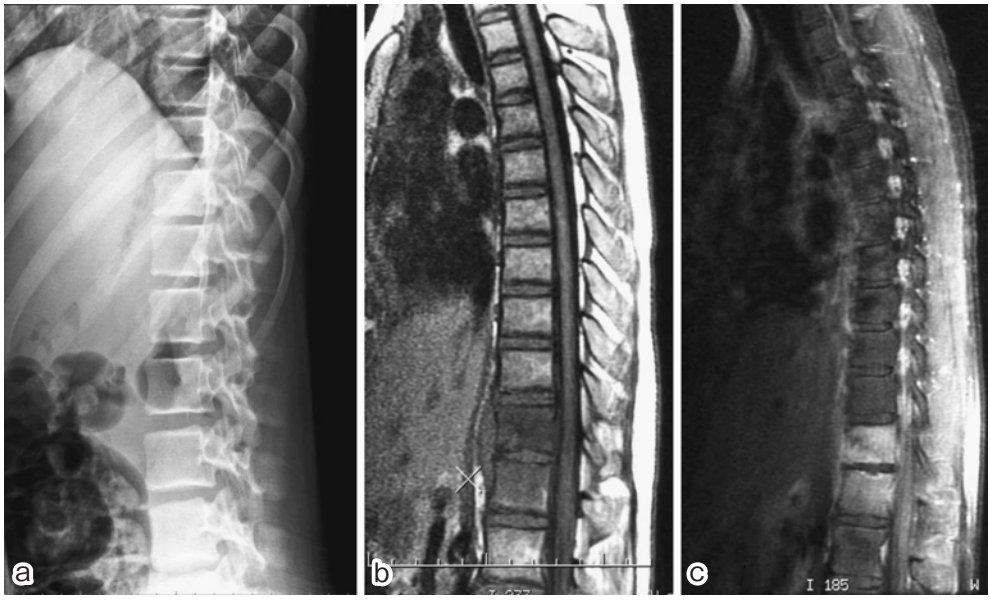

Fig. 1 Radiographic findings of the spine (**a**, plain X-ray film of the spine; **b**, T1-weighted MRI; **c**, T2-weighted MRI with gadolinium enhancement) on admission: Low-intensity area on T1-weighted images and high-intensity area on gadolinium-enhanced T2-weighted images can be seen. Plain X-ray film showed no significant changes in the same area.

showed low intensity on T1-weighed images (**Fig. 1 b**) and high intensity on T2-weighed images, with gadolinium enhancement at Th11–12 (**Fig. 1c**). Although blood, pharyngeal, urine, and stool cultures were all negative, bacterial spondylodiskitis was diagnosed, and antibiotics (cefazolin sodium) were administered intravenously. Subsequently, the back pain and the high-grade fever resolved, and CRP levels decreased, but a low-grade fever and the elevated ESR remained (**Table 1**, 75 mm/h 14 days after admission). In addition, the β-D-glucan level was 83.9 pg/mL (normal range, <20) on day 14 after admission. The purified protein derivative reaction for tuberculosis was negative. We then intravenously administered 400 mg of fluconazole, because we believed that the spondylodiskitis was also caused by fungal infection, and applied an insulated thorax brace to prevent compression fractures of vertebrae and deformity. As a result, by 24 days after admission the CRP and β-D-glucan levels and ESR had decreased to 0.05 mg/dL, 27.3 pg/mL, and 36 mm/h, respectively, and the patient was discharged on day 27. The patient continued to receive oral fluconazole (200 mg) (**Table 1**).

Two years after disease onset, the affected vertebrae had increased in intensity on T1- and T2-weighed MRI, thus suggesting an increased percentage of adipose tissue, and the ESR and levels of β-D-glucan and CRP were normal. Fluconazole was discontinued, and the brace was removed. Three years after disease onset, the patient had no abnormalities, deformities, or limitations of movement.

Discussion

Fungal spondylodiskitis is a rare disorder in childhood, particularly in immunocompetent patients[2,3], although the incidence of invasive fungal infection has been steadily increasing because of such factors as aggressive chemotherapy for cancer, bone marrow and organ transplantation, acquired immunodeficiency syndrome, and advanced critical care[4]. Lucio et al have reported that 3 of 27 cases of pyogenic spondylodiskitis were due to fungal organisms (2 due to *Candida* species and 1 due to Blastomyces dermatitidis)[1]. Here, we have reported on a 14-year-old girl who had fungal and bacterial spondylodiskitis with no immunodeficiency.

We diagnosed fungal spondylodiskitis and treated the patient for it on the basis of the presence of β-D-glucan and prolonged ESR elevation, without positive culture colonies. β-D-glucans are part of the outer cell wall in most pathogenic fungi (except for *Zygomycetes* and *Cryptococcus* species), and a multicenter clinical study has demonstrated the utility of the β-D-glucan test in screening for deep mycosis, with a sensitivity of 90% and a specificity of 100%[5]. Establishing a histopathological diagnosis and obtaining culture specimens from protected

Table 1 Clinical course of the patient

days after admission	1	5	10	14	24	55	85	115	128	168	203	233	273	302	362	480	720
ESR (mm/hr)*	82			75	36	24	15	14	19	14	20	15	12	21	10	17	11
β-D-glucan (pg/mL)*				83.9	27.3		13.4			7.8	15.7	16.9	13.1	10.5	13.7	8.5	7.8
CRP (mg/dL)*	5.45	3.11	1.45	0.33	0.05					>0.05	0.39	>0.05	0.07	0.06	0.05>	0.05>	0.05>
Fever (>37.5℃)																	
Back pain																	
Status	admission					outpatient service											
Therapy	cefazolin sodium				fluconazole										discontinued		

*Normal values: ESR, >10 mm/hr; β-D-glucan, >20 pg/mL; and CRP, >0.3 mg/dL.

anatomical sites, such as the thoracic vertebrae in our case, are often not possible in critically ill patients or young children. Because blood cultures are positive in only 50% of cases of invasive candidiasis and in 10% of cases of invasive aspergillosis[6] and because accurate diagnosis and early treatment prevent serious complications and deformities, we believe that the diagnosis of fungal infection in the present case was appropriate.

Fluconazole is the drug of choice for fungal spondylodiscitis because it can be orally administered and is better tolerated and less likely to cause nephrotoxicity than is amphotericin B[7]. Although there is no consensus regarding the optimal duration of treatment, treatment can be continued for 1 to 14 months, depending on the patient's underlying immune status[7]; in the present case treatment was continued until the CRP level and ESR had normalized[8,9], the symptoms had resolved, and the resolution of inflammatory changes had been confirmed with imaging studies[10].

In conclusion, spondylodiskitis should be considered in cases of abdominal pain without clear etiology and in cases of fungal infection with unexplainable findings after standard treatment for bacterial infection, even when fungal cultures are negative.

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