

Spondylodiscitis in two children: case reports and review of the literature

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ABSTRACT:

- Spondylodiscitis has a higher incidence in children under 3 years of age than other age groups. The onset may be insidious and clinical signs can be mild and unspecific, as well as the laboratory tests, often within the normal range. Hypomobility and pain are the main symptoms, and spine MRI is the gold standard for the diagnosis. The duration and type of treatment are controversial. Treatment with antimicrobial therapy, together with rest and immobilization, has given good results in specific cases, leading to a progressive recovery. The authors report on two patients, in whom no direct infectious pathogen was identified. In these patients, an infectious process following a bacterial or viral infection has been hypothesized, but an autoimmune cause could not be excluded. In both cases, the children had a good outcome.
- **Key words:** Discitis, Spondylodiscitis, Vertebral osteomyelitis, Intervertebral disc, Musculoskeletal infections, MRI.

INTRODUCTION

Spondylodiscitis is a primary infection or autoimmune inflammation of the intervertebral disc space and vertebral endplate^{1,2}. It is an uncommon disorder and the number of affected individuals is difficult to estimate^{3,4}. The disorder affects all ages from infancy to adulthood, with relatively higher incidence in toddlers and young children due to the increased blood supply to the endplate, that occurs at this age and which tends to undergo progressive involution during subsequent ages⁴. Various pathogenic factors have been identified as the etiologic agents of this disorder, including bacteria, fungi, and occasionally parasites^{4,5}. It is estimated that *Staphylococcus aureus* causes 30-80% of spondylodiscitis cases¹, and other reported infective agents are *Brucella melitensis*, *Mycobacterium tuberculosis*, and *Kingella kingae*⁶. The infection may spread endogenously through the arterial venous network to reach the disc and the vertebral body. Less commonly, the infectious factors may result from direct inoculation, as a result of surgical intervention or other spinal procedures, or by contiguous spread from adjacent infections. Predisposing factors in the pathogenesis of this disorder include cardiovascular and renal diseases, and immunodeficiency².

We report on two patients affected by spondylodiscitis, who had a benign course with a rapid clinical resolution of the symptoms. At two- and three-year follow-up, complete recovery was confirmed.

CASE REPORTS

Case 1

A two-year old girl was born at term after an uncomplicated pregnancy and a normal delivery. The family history was unremarkable. She was admitted to the Orthopedic Clinic, University of Catania, Italy for a diagnostic work up due to a four-day history of irritability and back pain. The parents stated that a week before admission the girl had an episode of fever lasted for two days and was treated with paracetamol. Physical examination was normal, there were no dysmorphic signs or congenital birthmarks. The body temperature was 36.4°C, lower lumbosacral pain was elicited by palpation and movements. Cranial nerves were not impaired and plantar reflexes were normal. The complete blood count was as follows: red blood cells 4,280,000/mm³, white cells 10,200/mm³ with 58% neu-

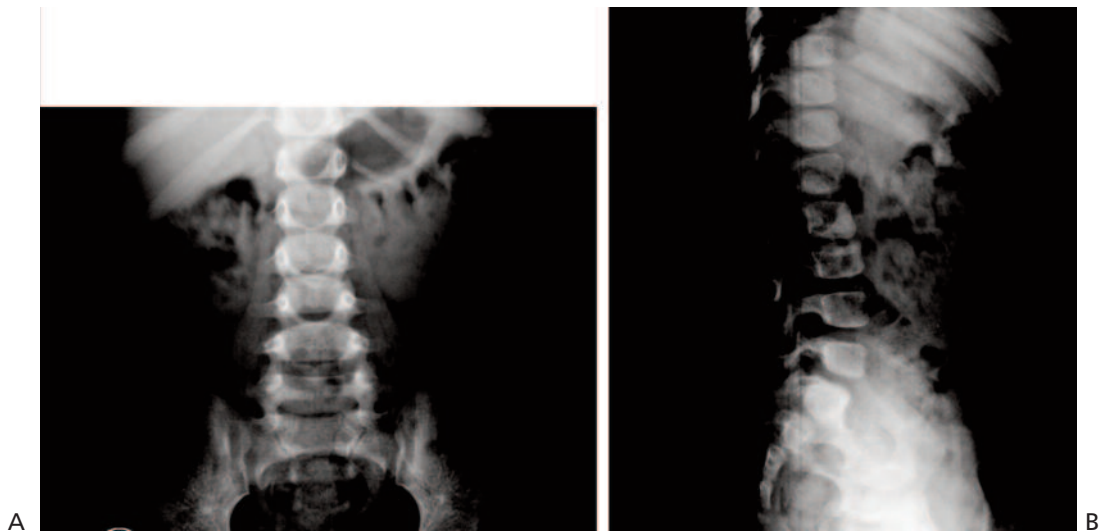


Figure 1. X-rays of a two-year-old girl showing L2-L3 narrowing space. *A*, postero-anterior (PA). *B*, Lateral view.

trophils, 44% lymphocytes, 4% eosinophils and 4% monocytes. ESR of 15 mm/h, the other laboratory tests were normal; serum antibodies, anti-Herpes Simplex Virus, *Mycoplasma pneumoniae*, *Borrelia burgdorferi*, Enterovirus, Adenovirus, and Parvovirus B19, and blood cultures were negative. Cerebrospinal fluid analysis was negative with no albumin-cytological dissociation, and 8-10 leucocytes. Ophthalmologic examination and fundus oculi were normal, as were ECG, echocardiogram, and video-EEG registration, when awake and during sleep. Conventional thoracic and lumbar X-rays showed a reduction of disc space with erosions of adjacent vertebral endplates at the level of L2-L3 (Figure 1A-B). Brain MRI was normal, while the spinal MRI before and after gadolinium enhancement showed a reduction of hyper-intensity on T2-

weighted images at the L2-L3 level, with vertebral bodies hyper-intensity on T1-weighted images, suggestive of bone marrow edema (Figure 2). Treatment with intravenous amoxicillin-clavulanate and cefuroxime at the standard dosage was started. Treatment was continued for two weeks and then was switched to oral one for another three weeks. At the same time, body plaster cast was introduced and maintained for two months, followed by a brace. At two-year follow-up, results of clinical and neurological examination confirmed complete recovery. X-rays taken after 1 year showed a slight segmental reduction of lordosis at the level of L2-L3, with no functional significance.

Case 2

A 3-year-old girl was the second child of healthy, unrelated parents. The girl was born after 38 weeks of gestation following a normal pregnancy and normal delivery. Birth weight, height, and head circumference were within the normal range. The girl was referred to the Orthopedic Clinic University of Catania, Italy for consultation for difficulty in walking and painful back. The parents stated that the clinical course had progressed rapidly. A week before the admission the girl had episodes of fever that lasted for three days. At admission, physical examination was normal. Cranial nerves examination was normal, she walked only if supported, and the pressure on her back caused pain and crying. Laboratory analysis showed 8,000 white blood cells/mm³, of neutrophils 63%, ESR 12 mm/h, and C reactive protein (CRP) 0.5 mg/L (n.v. 0-5 mg/L). The remaining laboratory parameters were normal, including immunoglobulins. Serum and CSF antibodies, Anti-Herpes Simplex Virus, Varicella Zoster Virus, Epstein-Barr Virus, *Mycoplasma pneumoniae*, *Borrelia burgdorferi*, Enterovirus, Adenovirus, and Parvovirus and blood cultures were all negative. The CSF was also negative both for chemical and intrathecal oligoclonal bands. Ophthalmologic findings with fundus oculi, ECG, and echocardiogram ultrasound examination were normal, as was the EEG performed while awake and during sleep. Brain MRI was normal, while spine MRI showed signs of hyper-in-



Figure 2. Sagittal MRI showed spondylodiscitis localized at the L2-L3 level.

tensity at L4-L5 and the adjacent vertebral body on T2-weighted images. Treatment with intravenous cefuroxime was started for a period of two weeks, followed by oral administration for three weeks. A body plaster cast was used. At three months follow-up, a new spinal MRI showed a favorable evolution. Neurological examination was normal. Follow-up at three years showed complete resolution.

DISCUSSION

Two female patients presented with lumbosacral pain. At admission, both patients did not have fever and laboratory findings were negative for specific infective agents. Treatment with antibiotics and body plaster cast was efficacious, leading to complete resolution and prompt recovery in few months.

Spondylodiscitis is a very insidious disorder, not always accompanied by specific signs (i.e., blood analysis or CSF evaluation). In both patients no laboratory signs of infection were found. Spine MRI remains the gold standard to obtain a diagnosis. Brown et al⁷ reported a series of 11 consecutive cases and found that the most common presenting signs were refusal to walk, with limping and hip or leg pain reported in 63% of patients. Neither fever nor abdominal pain were observed in these patients⁷ and some laboratory tests including ESR, CRP, and white cell count were normal or only slightly elevated. Blood cultures and CSF were mainly negative and thus unable to provide sufficient etiologic information. Spiegel et al⁴ reported on the results of a biological analysis of the disc space that was performed in three patients before they received antibiotics treatment; two biopsies contained only inflammatory cells and in the third no infections were found. Moreover, the biopsy performed in the paravertebral inflammatory masses was sterile. A spine MRI with gadolinium enhancement is diagnostic, showing an enhancement within the disc and adjacent vertebral body (i.e., often associated with a paraspinal inflammatory mass). The area of the spine most frequently involved is the lumbar one, particularly in the region L5-S1, although the thoracic region and cervical spine are also sometimes affected⁸. According to Chandrasenan et al⁹, the most frequent causes of infection are *Staphylococcus aureus*, *Streptococcus epidermidis* and *Streptococcus pneumoniae*. Two patients with a spondylodiscitis that was linked to *Kingella kingae* infection were reported by Ceroni et al⁶.

In the present cases, it is not possible to rule out an autoimmune process following a previous bacterial or viral infection. This pathogenic hypothesis may explain the negativity of the serological investigations in our patients and in several other reported patients.

The treatment of spondylodiscitis is still under debate. The use of antibiotics is recommended in cases of clear infection. In a report by Ring et al¹⁰, treatment with intravenous antibiotics resulted in a significant reduction in the duration of symptoms compared with those who had no antibiotics or oral treatment.

Antibiotic treatment certainly plays an important role in cases with clear infective factors, but may be useful also in cases with no evident infections to eliminate po-

tential residual pathogens. Bracing has been advised in conjunction with antibiotics or alone. Brown et al⁷ suggested bracing in children who are symptomatic but who otherwise exhibit no evident signs of infections. However, these authors reported a complete recovery in all patients except one, who continued to have mild back pain with premature ankylosis of the affected spine. At follow-up (which ranged from 12 to 38 months), almost half of their patients showed evidence of early degenerative change or ankylosis at radiologic examination. Persistent radiological abnormalities and asymptomatic restriction of spinal movements in a group of patients were reported by Kayser et al¹¹ in a longer follow-up of 23 years.

CONCLUSIONS

Spondylodiscitis is an uncommon disorder but its diagnosis should not be neglected, since the signs are often insidious; irritability, refusal to walk, and spinal pain are the most common ones. A possible etiological role of the autoimmune process should be considered when there are no signs of direct infectious origin. In any case, treatment with antibiotics should be carried out in the silent forms, since the disorder may result in progressive and relevant consequences. In our patients, the course of the disorder was benign and follow-up at two and three years respectively, did not show any sign of persistent spine impairment. A longer follow-up may give useful information for these patients.

CONFLICT OF INTERESTS:

The Authors declare that they have no conflict of interests.

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