Non Contiguous Multilevel Spondylitis: an exceptional presentation of spinal brucellosis

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La spondylodiscite multi-étagée non contiguë: Une présentation exceptionnelle de la brucellose rachidienne

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RÉSUMÉ

But : Décrire les signes radiologiques de la spondylodiscite brucellienne multifocale et en discuter les difficultés diagnostiques

Observation : Les auteurs rapportent une observation de spondylodiscite brucellienne multi-étagée intéressant de façon concomitante le rachis dorsal et lombaire

La brucellose est une zoonose relativement fréquente dans les pays méditerranéens et du moyen orient. Il s'agit d'une infection systémique, causée par une bactérie intra-cellulaire faculltative de l'espèce brucella qui peut intéresser plusieurs tissus et organes. Le rachis est le siège le plus fréquent de la brucellose ostéoarticulaire. Cependant, la brucellose rachidienne multifocale est très rare et seuls 10 cas ont été rapportés dans la littérature.

Conclusion : Bien qu'il s'agisse d'une forme exceptionnelle, la spondylodiscite brucellienne multi-étagée mérite d'être connue afin d'éviter des erreurs diagnostiques.

SUMMARY

Aim: The aim of this study was to describe the imaging features of multilevel brucellar spondylitis and discuss the diagnostic challenges

Case: The authors report describes one case of noncontiguous synchronous multifocal involvement of thoracic and lumbar spine.

Results: Brucellosis is a zoonosis of worldwide distribution, relatively frequent in Mediterranean countries and in the Middle East that can involve many organs and tissues.

The spine is the most common site of musculoskeletal involvement, but multilevel involvement is uncommon and only ten cases were reported in literature.

Conclusion: Although it is an exceptional form, multifocal brucellar spondylitis is worth to be known to avoid diagnostic mistakes

Mots-clés

Brucellose, Infection, Rachis, Radiographies, TDM, IRM

Key-words

Brucella, Infection, Spine,Radiographs , CT. MRI

التهاب الفقرات و الأقراص متعدد الدرجات توضع استثنائي لداء البروسيلات في النخاع

الباحثون : شلي بوعزيز. م - بوقمرة . إ - كفال . ض - كشيري . م

الهدف من هذه الدراسة هو استعراض العلامات التصويرية لالتهاب الفقرات و الأقراص بسبب داء البروسيلات و مناقشة الصعوبات التشخيصية .تشتمل دراستنا على حالة واحدة لهذا المرضى الذي يتمثل في خمج مجموعي تسببه بكتيريا من عائلة البروسيلا ويعتبر التعدد الموضعي لداء البروسيلات في النخاع نادر جدا ولم يذكر في المقالات الطبية إلا 10 مرات و رغم ندارة هذا المرض فإنه يجب علينا التعرف عليه لتفادي الأخطاء التشخيصية.

الكلمات الأساسية : داء البروسيلات - خمج - نخاع - تصوير - تصوير بالرنين المغناطيسي.

Brucellosis, also called Malt fever, sweat-painful fever, ondulary fever, melitococcis or Mediterranean fever is a zoonosis of worldwide distribution, relatively frequent in Mediterranean countries and in the Middle East. It is a systemic infection, caused by facultative intra-cellular bacteria of the genus Brucella. [1]. Four species are responsible for brucellosis in humans: Brucella abortus, Brucella suis, Brucella canis and Brucella melitensis, the latter being the most virulent and invasive [2, 3]. Brucella organisms are shed in the excreta (urine, stool, milk and products of conception) of infected animals. Virtually all infections occur as a result of direct or indirect exposure to animals; the main modes of transmission are ingestion of unpasteurized milk and milk products. Human-to-human transmission is unusual [4]. The wide spectrum of clinical involvement and the non-specific signs and symptoms interfere with an early diagnosis of this disease [5].

CASE REPORT

A 66-year-old man, without any history of exposure to dairy animals or ingestion of raw dairy products, was referred to our hospital with a 3-month history of low back pain and productive cough. He also reported low-grade fever, night sweats and a weight loss of approximately 4 kg during the last 3 months. On physical examination, he was afebrile and his vital signs were normal. Spine mobility was not restricted. There was no splenomegaly, hepatomegaly, or lymphadenopathy. The PPI skin reaction was negative.

Laboratory workup revealed elevated erythrocyte sedimentation rate (ESR=58 mm on the first hour), C-reactive protein (CRP=148 mg/l) and a cholestasis syndrome with elevated gamma GT and Alcaline phosphatasis rates.Total blood count was otherwise normal.

The abdominal ultrasound (US) examination was normal. Plain

Figure 1 : a and b: anteroposterior and Lateral radiographs of the thoracic spine show a narrowing of the T9-T10 disc space. c and d: anteroposterior and lateral radiographs of the lumbar spine show lateral narrowing of the L2-L3 disc space associated with well-defined erosions (arrow) and sclerosis of vertebral endplates

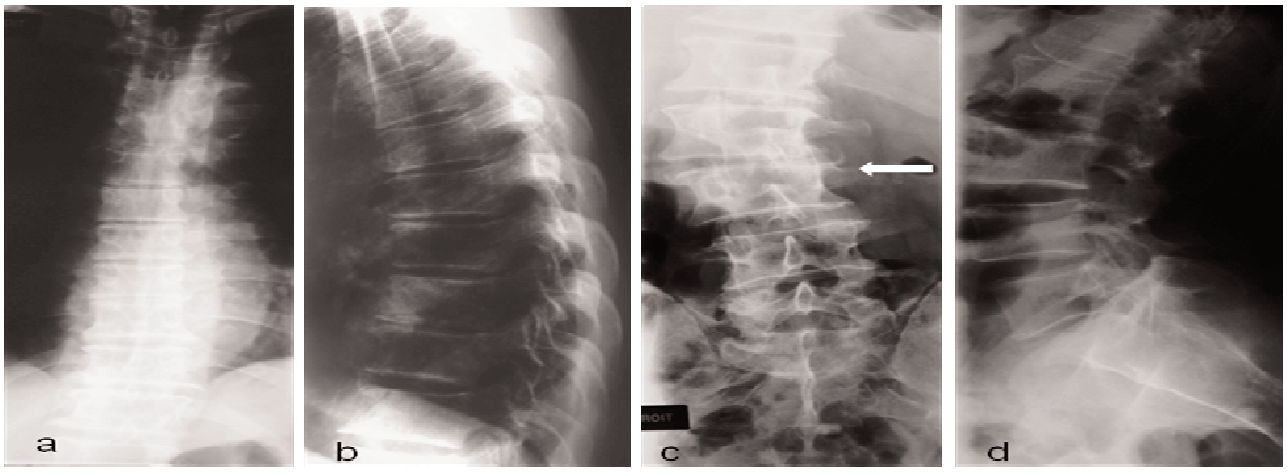
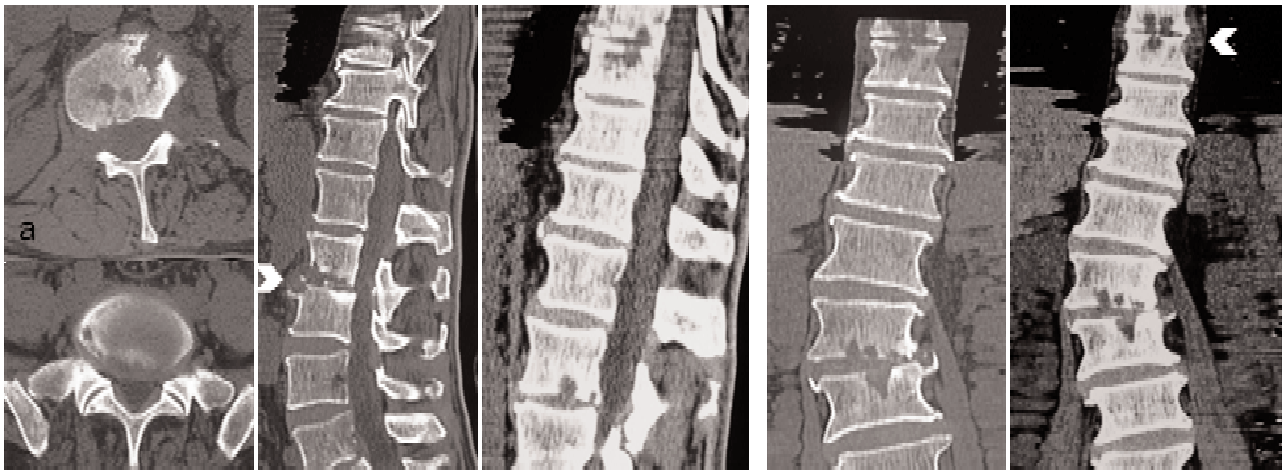


Figure 2 : (a, b, c, d, e and f): transversal, sagittal and coronal CT images in bone and soft tissue algorithm show a narrowing of T9-T10 and L2-L3 disc, erosions of the adjacent endplates with presence of disc vacuum (arrow) , new bone formation of the L2-L3 space with a "parrot beak appearance" and moderately swollen paravertebral soft tissues (arrowhead).



radiographs of the spine where performed, showing a narrowing of T9-T10, L2-L3 and L5-S1 disc spaces (Figure 1).
Chest X-ray completed with thoracic CT didn't detect pulmonary lesions but showed a narrowing of T9-T10 and L2-L3 disc, erosion of the L2 anterior endplates with presence of disc vacuum and new bone formation (Fig2) The paravertebral soft tissues were moderately swollen which was unusual for degenerative disease.
MRI of the whole spine revealed abnormal signal intensity of the discs and adjacent vertabrae at the descriped levels (Figure 3). Paraspinal and epidural abcesses were detected at L2-L3 and L5-S1 levels on T2 Weighted and T1 weighted images after Gadolinium administration (Figure 4). Brucella agglutination test turned positive (240 IU/ml Wright) establishing the diagnosis of brucellar spondylodiscitis.
Patient received an 8-week, 2-drug combination treatment (Rifamycin 900 mg/day and Rovamycine 200 mg/day p.o.) associated to spine immobilization with a favourable outcome.

DISCUSSION

Any part of the body may be affected by active brucellosis but osteoarticular involvement remains the most common location.

Figure 3 : MRI of the dorsal spine
A, Sagittal spin-echo T1-weighted image shows decreased signal intensity T9–T10 vertebral bodies.
B, Sagittal STIR-weighted image shows increased signal intensity in affected vertebrae with irregular vertebral body endplates and narrowing of intervertebral disk spaces.

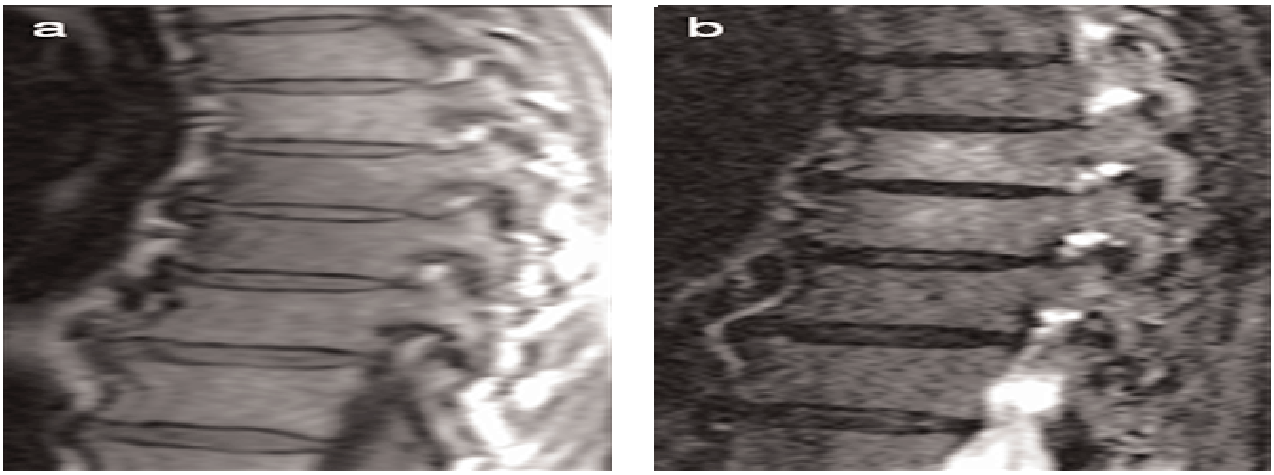
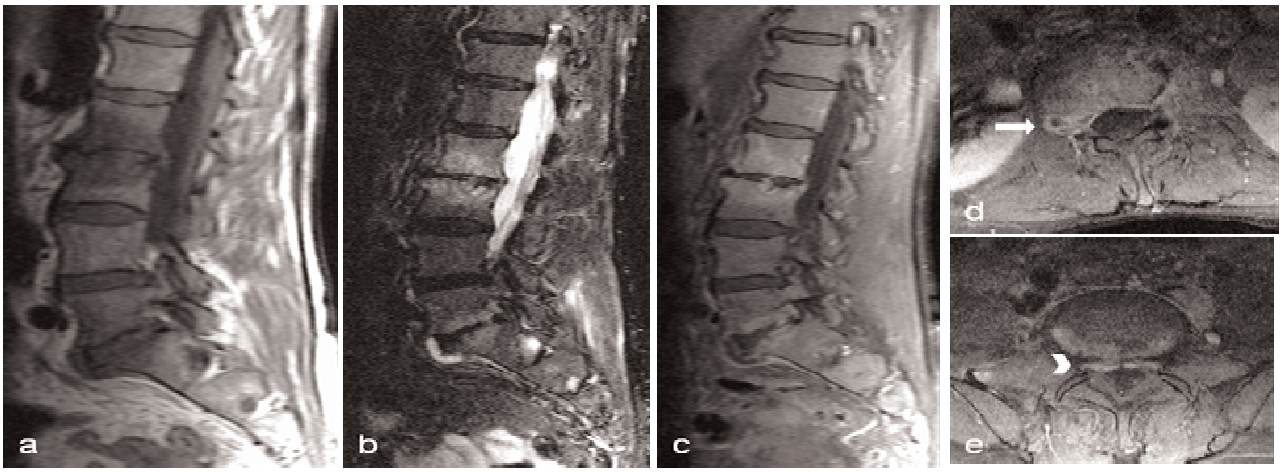


Figure 4 : Images of the lumbar spine from an MR examination show bone edema of the L2, L3, L5 and S1 endplates and paravertebral and epidural abscess in L2-L3 level (arrow) and epidural thickening in L5-S1 level (arrowhead)
A, Sagittal spin-echo T1-weighted.
B, Sagittal turbo spin-echo T2-weighted
C, Sagittal gadolinium-enhanced T1-weighted with fat saturation
D and F, transverse gadolinium-enhanced T1-weighted with fat saturation



The reported frequency of osteoarticular brucellar involvement varies between 10% and 70%. This variation is related to the difference of species pathogenicity and to differences in diagnostic criteria [6]. The spectrum of musculoskeletal manifestations of brucellosis includes spondylitis, sacroiliitis, arthritis, osteomyelitis, tenosynovitis and bursitis. Rarely, multifocal involvement of the skeleton may be observed [7].
The type of skeletal involvement depends on the patient's age [3]. The sacroiliac joints and knee arthritis predominate in children and young adults whereas the spine remains the most common site of involvement in elderly patients [8]. Brucellar spondylitis represents 6% to 58% of osteoarticular localizations [1]. It typically occurs in men over 40 years of age. Most affected is the lumbar spine (60%), followed by thoracic (19%) and cervical spine (12%). More than one level is affected in 3% to 14% of the cases [8, 1]. Multi-level involvement is an exceptional form of brucellar spondylitis and to our knowledge only 18 similar cases have already been reported in the world literature [6, 8, 9, 10, 11, 12].
MRI is the imaging method of choice for the diagnosis and follow-up of brucellar spondylitis. MRI has high sensitivity for detecting the disease in the early stages and provides excellent definition of paravertebral and epidural extension. It also allows the detection of otherwise unsuspected additional non-contiguous spinal foci. In acute brucellar infections, MRI shows low to- intermediate signal intensity on T1-weighted images of the intervertebral disc and low signal intensity in the adjacent vertebral bodies. The signals in these areas usually become hyperintense on T2-weighted MRI sequences, with either a homogeneous or heterogeneous pattern. However, the disc signal may remain low on T2 weighted images in brucellosis. The intravenous administration of Gadolinium allows better definition of the spinal inflammatory lesions and a more complete assessment of soft tissue involvement and epidural extent. These features are best shown when fat-suppression techniques are applied to the contrast enhanced images. Paravertebral abscesses are observed in approximately 30% of cases and are typically characterised by well-defined margins. In the chronic stages, the MRI pattern of the discs and vertebral bodies may vary. However, vertebral bodies usually show heterogeneous signal intensity [1].
Multilevel brucellar spindylodiscitis can be detected with bone scintigraphy as multilevel increased uptake of involved areas [13]. MRI also allows this diagnosis as it can image the whole spine. MRI offers a better specificity than bone scintigraphy and has the advantage to assess very precisely disco-vertebral, soft tissue and epidural involvement.
The major diagnostic problem of multilevel spinal brucellosis is the differential diagnosis with degenerative disc disease and tuberculous spondylitis.
Distinctive criteria between spinal brucellosis and degenerative disc disease include well-defined erosions and sclerosis of vertebral endplates on radiographs and CT and high signal strip surrounding low signal of vertebral endplates on MRI T1-weighted images, these features being suggestive on degenerative disc disease (Table 1) [14]. Discal vacuum phenomenom, although rare, can be seen in brucellar spondylitis. Inflammatory modifications of the vertebral endplates in degenerative disc disease can mimic infection on MRI but in these cases, the disc usually show a low T2 signal intensity and soft-tissue and epidural abcesses are absent.
When clinical and radiographic presentations are typical, the distinction between tuberculous and brucellar spondylitis is rather easy. Brucellar spondylitis most commonly affects the lower lumbar spine, whereas tuberculous spondylitis is most commonly located in the midthoracic spine. The combination of severe vertebral collapse, large paraspinal abscesses extending beyond the area of vertebral disk involvement, and gibbus deformity are typical of tuberculosis.
An intact vertebral architecture, despite diffuse high signal intensity with T2-weighted MR sequence or abnormal uptake with bone scintigraphy, the presence of end plate defects, moderate paraspinal soft tissue abcesses; and the presence of disk gas and peri-lesional bone formation with osteophyte formation at the anterior vertebral endplate (parrot's beak) are typical of brucellosis [13, 15].
However, important bone destruction or large or calcified paraspinal soft tissue collections may be observed in brucellar spondylitis, constituting the so-called brucellar pseudo-Pott's disease [16]. One more feature of brucellosis mimicking tuberculosis in our case is the multilevel non contiguous spinal involvement.

BACTERIOLOGICAL AND SEROLOGICAL CHARACTERISTICS

Haematological and biochemical testing yield no specific findings to suggest the diagnosis of focal brucellosis. Complete blood counts most frequently reveal leucopenia; pancytopenia can be observed in as many as 20% of patients [17]. The levels of C reactive protein (CRP) may be elevated [17]. Isolation of the organism in culture is the conclusive diagnostic procedure. Brucella spp. are cultured in standard biphasic (solid and liquid) mode or with the Castaneda bottle, which incorporates both solid and liquid media in the same container. The use of automated blood culture systems has shortened the detection time of Brucella. [18]. Standard culturing techniques require prolonged incubation periods (up to 30 days) under special conditions. If the diagnosis is suspected clinically, one should be careful to notify laboratory personnel of the concern for brucellosis so that proper specimen processing occurs. Pus culture gives positive findings in 10% to 20% of cases. Sensitivity of blood cultures ranges from 17% to 85%, depending on the strain involved [19, 2]. Brucella melitensis and Brucella suis are more likely to have detectable bacteraemia. Failure to culture the organism may result from inadequate microbiological techniques or from the administration of antimicrobial agents [17]. Sensitivity decreases for all strains as the disease duration progresses. The serum tube agglutination test (Wright test) is rarely positive. In rare instances, positive blood culture results and negative serology findings have been reported [18]. Results are considered positive if antibrucella titres ≥1/160 are obtained in standard tube agglutination tests. In general, serological tests that provide immediate information, such as the Rose Bengal test or card test are desirable. This test is almost as sensitive as

the standard tube agglutination test [19]. Enzyme-linked immunoassay, indirect immunofluorescence and counter-immuno-electrophoresis are more sensitive than agglutination assay.

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