NEURORADIOLOGY

Kazem Abbassioun MD['], Abbass Amirjamshidi MD², Behrooz Taheri MD.³

Radiologic Changes in Brucella Spondylitis

Experience with 26 Cases.

Background/ Objectives: Brucellosis is an endemic zoonosis in the Middle East and despite all public health efforts it has not yet been eradicated in Iran. We aimed to highlight and categorize the imaging features of Brucella spondylitis.

Material and Method: Twenty six cases of Brucella spondylitis were treated by the authors from 1982 up to 2003. The available imaging studies of all the cases are reviewed and include X-ray films, conventional myelography, computerized tomographic (CTscan) and magnetic resonance imaging (MRI).

Results: There were 21 male and 5 female patients with an age range of 5 to 62 years and the majority (60%) in the 4th and 5th decades of life. Wright hemagglutination tests were positive in all cases. Plain X-ray films typically showed lysis of the end plates with osteophyte formation involving affected vertebrae, followed by narrowing of the interspaces and destruction or collapse of the vertebral bodies in 7 cases. Myelography demonstrated various types of epidural filling defects and obstruction to the flow of contrast material in 10 cases. CT scan, available in 3 cases, showed erosion and cauliflower-like proliferation at the bony edges of the vertebral bodies and end plates. MRI findings varied depending upon the acute or chronic stages of the illness with hypo- or hyper-intense changs on T1 sequences, and primarily hyper-intense changes on T2 sequences in 8 cases.

Conclusion: The findings in this series of patients suggest that imaging findings of Brucella spondylitis are scarcely specific. However when considering the medical history, place of origin of the patients, clinical presentation and laboratory findings, the early diagnosis of the illness may be possible before proceeding to surgical intervention.

Keywords: Brucellosis, Computerized Tomography, MRI, Plain X-ray, Spine.

Introduction

Bdirect skin contact or by ingestion of contaminated milk by-products ^{1, 2}. As an occupation-related disease, males are predominantly affected but females and children are also occasionally involved ^{1.} Brucellosis is uncommon in USA with an annual incidence of 0.07 to 0.1 per 100,000 population.¹⁻⁹ It is also not a common illness in European countries and United Kingdom.¹ It is not so rare in some Mediterranean countries including Israel.⁹ Brucellosis is a common in Iran.

Brucellosis is an intracellular parasite and is gram-negative. Brucella species are often found in reticuloendothelial cells. After an incubation period of 2-3 weeks, the onset of illness is heralded by chills, fever, sweating and physical exhaustion. The majority of patients recover within 3 to 6 months but the chronic illness may remain with varying degrees of disability and different kinds of clinical manifestation. This usually is called the typical 'undulant fever'.^{1,12,13}

Neuropsychiatric manifestations are the most frequent complication of chronic Brucellosis ²² followed by bone and joint involvement in 2 to 85% of patients in reported series.^{8,9} The vertebral column is the most frequently involved site of the musculoskeletal system in up to 50% of cases followed by hip and sacrocoiliac joints.^{4,14-17} This report presents the various imaging findings in one series of patients in suggesting the findings as a suitable preoperative diagnostic tool.

From

1. Professor of Neurosurgery, Tehran University of Medical Sciences, Iran. 2. Associate Professor of Neurosurgery, Sina Hospital, Tehran, Iran 3. Radiologist in Arad Hospital A.Amirjamshidi MD. PO. Box; 19585-678, Tehran, Iran,

Materials and Methods

A retrospective analysis of the results of a series of 26 cases with Brucella spondylitis ranging in age from 5 to 62 years of age, all treated by the authors from 1982 up to 2003 is presented. The study material copmprised files of 26 patients admitted to the departments of neurosurgery in Sina and Arad general hospitals and includes groups of patients treated surgically and/or medically, and discharged with the diagnosis of Brucella spondylitis. The patients are retrospectively reviewed, and the available clinical and imaging studies were reviewed by an independent radiologist and two neurosurgeons (authors) (table 1). The positive findings of all imaging modalities have been categorized according to the specific features of each imaging modality and the more salient radiographic changes in each category have been selected for demonstration as the 'sample cases':

Sample Cases

Case 1) A 41-year-old man presented with lower back pain and intermittent claudication for

approximately 6 months. On physical examination, straight leg raising (SLR) was positive at about 80 degrees, with mild weakness on dorsi-flexion of the toes (EHL). There were no abnormalities noticed as regards the reflexes. The first-hour sedimentation rate (ESR 1hr) was 18 mm, and Wright agglutination test was positive 1/40. Plain X-rays of the lumbosacral spine were prospectively normal, magnetic resonance imaging study (MRI) was demonstrated a triangular T1 hypo-intensity in the posteroinferior aspect of the L4 vertebral body. This corresponded with hypo-intensity on T2W images surrounded by a long-TR hyper-intense rim. The affected part of the vertebral body protruded towards the spinal canal, partly obliterating the intervertebral foramina (fig 1a&b). At operation, this turned out to be a single level narrowing with discolored epidural fat and bulging of the osteophytic bone. The granular fat was removed and discectomy was performed followed by curettage and removal of the osteophytic changes. The histopathological evaluations showed only nonspecific inflammatory changes in the epidural fatty tissue and the adjacent bone.



Figure 1 A

Figure 1 B





Figure 2 B

Figure 2 A

Figure 1 A & B: T1W MRI showing hypointense lesion in the posteroinferior margin of L4 vertebra. In T2W image, the lesion is surrounded by a hyperintense rim and is compressing the dural sac ventrally

Figure 2 A & B : T2W image showing a hyperintense filling defect located ventral to dural sac at L4/L5 level (dark small arrows) and in the horizontal T1W image, the infiltrative lesion located ventro-right laterally to the dural sac(black arrow head)

Case 2) A 48-year-old man was admitted with severe right sciatica of about 3 months' duration, with accompanying weakness on right EHL and hypoesthesia involving right L5 and S1 roots. White blood cell count and the ESR were normal, although the Wright agglutination titer was 1/320. MRI showed a dehydrated L4/5 disc and an epidural lesion filling the right lateral recess at the same level. The lesion was hyper-intense in both T1W and T2W images (fig 2a&b). The vertebral bodies did not show any signal changes. Decompressive laminectomy was performed and a granulomatous epidural lesion was removed in fragments. The degenerated L4/5 disc was also evacuated but did not show any inflammatory changes on histopathological examination; the histopathological evaluation of the specimen was compatible with nonspecific granulomatous inflammatory changes.

Case 3) This 5-year-old girl was admitted for worsening lower back pain of 6 months' duration, and occasional fever. Her neurological examination was normal, with a normal WBC count and ESR of 47 mm. Wright agglutination test was positive in the range of 1/40. Plain films of the lumbosacral spine revealed a widened lumbosacral spinal canal with scalloping of the posterior aspect of the vertebral bodies. Myelography showed severe epidural compression at both the L3-L4 and L5 levels (fig 3 a& b). Decompressive laminectomy was performed; at surgery there was demonstration of grayish red inflamed epidural fat which tightly encircled the dural sac, which was released as much as possible. The vertebral bodies and disc material appeared intact to gross inspection. Histological studies showed a fibro-fatty tissue with large amounts of non-specific chronic inflammatory cells.

Case 4) A 38-year-old man who complained of lower back pain of more than one year's duration with bilateral sciatica was admitted with severe increasing numbness in both legs. All laboratory tests were normal except for Wright agglutination test which was positive with a titer of 1/320. Plain lateral films of the lumbar vertebrae disclosed erosion of the superior plate of the L5 vertebral body in addition to erosion of the inferior plate of the L4 vertebral body, and greater erosive changes demonstrated posteriorly with relative anterior sparing. The myelogram showed ventral compression on the dural sac and a bilaterally truncated L5 root sleeve filling at the L4/5 level (fig 4a&b). Exploration at L4/5 level after interlaminar decompression revealed a degenerated and bulged L4/5 disc which was dark-pinkish and very necrotic when manipulated with surgical forceps. The adjacent vertebral plates were also soft and collapsed adjoining the necrotic disc material. The histopathological examination of the disc material was compatible with nonspecific inflammatory reactions without any visible organism.

Case 5) A 43-year-old farmer was admitted complaining from lower back pain, knee pain, incontinence, and inability to ambulate more than 30 steps. SLR was positive at 45 degrees and femoral stretch test (FST) was also positive. Doriflexors of both feet were weak and knee jerk was not detectable on either side. All the laboratory tests were negative except for an ESR 1hr of 21 and Wright agglutination test which was positive (1/640). On Imaging there was demonstration of L3/4 interspace collapse with posterior spur formation, and a nearly complete myelographic block at L3/4 (fig 5). Decompressive laminectomy was performed at L3 level and a dark purplish, tough and hemorrhagic epidural tissue which was located primarily ventro-laterally to the dural sac was removed. The tissue was a non-specific granulomatous adhesive and heavily cellular tissue on microscopic examination.



Figure 4 A&B: The plain X-ray showing the erosion and raggedness of the L4/L5 interspace (black arrow tip), while the myelogram in lateral view confirms the epidural compression at the same level.



Figure 3 A&B: Myelogram of the 5 years old patient showing sever compression on the dural sac from L2/3 down to the sacrum.

Cases 6-11) There were 6 additional cases presenting with lower back pain and unilateral sciatica ranging in age from 23 to 51, who were initially conservatively managed with rest and antiinflammatory medications. They were referred to our center due to aggravated symptoms and progressive neurological deficits involving one leg. Myelography performed as an urgent diagnostic procedure showed central and/or unilateral epidural filling defects resembling extruded disc fragment in one (fig 6a) or two (fig 6b) adjacent levels. In all cases at exploration, grayish-pink fibro-vascular tissue was demonstrated, compressing or engulfing the demonstrated, compressing or engulfing the symptomatic dural root sleeve. In all cases, this tissue was in continuity with the degenerated and discolored disc space at the same level. On retrospective plain film review, non-specific degenerative changes or erosions and spur formations were visible in all patients (fig 6a&b). The Wright agglutination test was positive in all these cases, even though they became available long after urgent surgical intervention. The histopathological finding was nonspecific inflammatory change either in the epidural fat or degenerated disc tissues.

Case 12) A 42-year-old engineer presented with low back and buttock pain radiating to the groin and



Figure 5: The myelogram showing nearly incomplete blockage of the dye column at L3/L4 level.

down to the knees. On admission, the patient demonstrated a forward-leaning posture with severe tenderness on palpation of the lumbar region. SLR and FST were positive bilaterally with additional severe weakness of dorsiflexors of the feet and toes. intensities in T2W images involving the L3 vertebral body, and a layer of infiltrative tissue which had replaced the epidural fat posterior to the L3 vertebral body. The L5/S1 disc was also focally protruded (fig 7). Decompressive laminectomy at the L3 level was performed, and pinkish gray hemorrhagic areolar tissue which had replaced the epidural fat at this level ,was removed. The vertebral body was porotic and soft on inspection and appeared intact. The specimen was compatible with a nonspecific inflammatory reaction.

Case 13-14) The CT scans of a 54-year-old male patient and a 42-year-old female patient who presented with lower back pain, incontinence and severe bilateral sciatica. Wright hemagglutination test was positive in both cases. Sclerotic changes were circumferentially present involving the edges of the vertebrae, with 'vacuum' phenomenon shown within the discovertebral junctions (fig 8). There were ventral epidural lesions resembling bulging discs in both cases. The lesions were surgically demonstrated to be *inflammatory granulomatous tissue extending to the disc space and into the vertebral body* in both patients.

Case 15) A 45-year-old farmer was admitted with lower back pain, severe right sciatica, and loss of right ankle jerk. MRI showed a hyper-intense and an iso-intense epidural filling defect on T1W and T2W images, respectively. There were T2 hyper-intense changes both in the adjacent L5 postero-inferior vertebral bodies and additionally in the L5/S1 disc. Wright hemagglutination titer was 1/80. The lesion was surgically demonstrated to be a *nonspecific inflammatory granuloma with affection of both the adjacent L5 vertebral body and disc space*.

Case 16) A 54-year-old farmer was admitted with progressive paraparesis of 3 months duration. On plain films, the T12 and L1 vertebral bodies were collapsed. MRI showed destruction and collapse of the same vertebrae with hypo-intense changes in T1W and hyper-intense changes on T2W images, and greater conspicuity of the T12/L1 disc space findings on T2W images. The spinal canal was severely compromised at this level, and decompression was done via lateral extracavitary approach with resection of a *nonspecific granulomatous tissue involving part of each vertebra and the entire T12/L1 disc*, followed by bone fusion using homologous rib graft. Wright hemagglutination titer was 1/640 in this case.



Figure 6A: Myelogram showing epidural filling defect at L4/L5 level indistinguishable from an extruded disc fragment.



Figure 7: MRI showing hypointense changes on L4 vertebral body in T1W images which iturns to be hyperintense in T2W images. The epidural fat located just behind the L4 Vertebra is replaced by and which is isointense in T1W and hypertintense in T2W images. The L5/S1 disc is also dehydrated and hyperbulged.



Figure 6 B : Myelogram showing epidural filling defect both at L4/L5 and L5/S1 levels, hardly distinguishable from hyperbulging



Figure 8: CT scan is horizontal section with saggital reconstruction, showing sclerotic changes around the edge of the vertebral body (cauliflowerer appearance), and the low density areas within the discovertebral junction resembling the vacuum phenomenon. The segm,ental narrowing of canal is evident in saggital view.

Case 17) A 50-year-old man was admitted with severe lower back pain and bilateral sciatica and saddle hypoesthesia of 3 weeks' duration. Wright hemagglutination titer was 1/80 and MRI showed a T1W hyper-intense, T2W hyper-intense epidural filling defect at the L5/S1 level without accompanying appreciable changes in the adjacent vertebrae or disc spaces. The lesion was surgically demonstrated to be *nonspecific granulomatous tissue* compressing S1 roots bilaterally.

Case 18) A 16-year-old patient presented from a rural setting with the diagnosis of lower back pain, leg pain and incontinence of 6 months' duration. MRI showed diffuse hypo-intense changes in the L3 and L4 vertebral bodies in T1W images with continuous extension as a slightly hyper-intense lesion into the spinal canal and secondary severe

canal compromise from the L2 through L5 levels. All the visible lesions were hyper-intense in T2W images. Wright hemagglutination titer was 1/320. The lesion turned out to be a nonspecific inflammatory tissue involving the epidural space, the disc and the adjacent vertebral bodies. There was no abscess collection.

Case 19) A 48-year-old engineer was admitted with severe left shoulder pain. Physical examination revealed mild spastic quadriparesis with mild weakness of the grasp in his left hand. Wright hemagglutination titer was 1/640, and MRI revealed abnormal elongated epidural infiltrative tissue ventral to the cervical dural sac extending from the C3 level, inferiorly, to the C6 level. This epidural material was iso-intense in T1W and hyper-intense in T2W images. The patient was treated with bed

No.	Sex	Age	Presentation	Wright test	Preclinical study	Treatment and Findings
1	F	17	LBP+ IC+ incontinence + High sedR.	1/320	Incomplete block at L2/3 level in myelogram	Laminectomy;Epidural granulation tissue
2	F	5	LBP+ leg pain+ sedR=57 + paralysis	1/40	Wide extradural stricture L3 down to S1	Laminectomy;Epidural granulation tissue
3	М	38	LBP+ sciatalgia + weak Rt EHL + high sedR	1/1280	Incomplete block at L4/5level in myelogram	Laminectomy; normal disc + Epidural grayish granuloma
4	М	50	LBP+ Lt foot drop + sciatalgia + weak Rt EHL	1/80	Sponylosis L4/5 + filling defect at L5/S1 level in myelogram	Extradural granulation tissue
5	М	38	LBP+ sciatalgia + weak Rt EHL	1/320	filling defect at L4/5 level in myelogram+ bone erosion	Epidural granulation tissue + involved disc
6	М	45	LBP+ IC+ sedR=65	1/40	Incomplete block at L4/5level in myelogram	Epidural granulation tissue + involved disc
7	М	24	LBP+ Lt sciatalgia + weak Lt EHL + sedR=33	1/160	filling defect at L5/S1 level in myelogram	Epidural granulation tissue + involved disc
8	М	48	LBP+ Lt sciatalgia + weak EHL + high sedR	1/320	filling defect at L5/S1 level in myelogram	Epidural granulation tissue + normal looking disc
9	F	49	LBP+ IC+ high sedR	1/160	L4/5 block in myelogram	Epidural granulation tissue + involved disc
10	М	51	LBP+ Lt sciatalgia + weak Lt EHL	1/320	filling defect at L5/S1 level in myelogram	Epidural granulation tissue + involved disc
11	М	52	LBP+ Lt sciatalgia + weak Lt EHL	1/40	Sponylosis + tight lateral recesses in myelogram	Adhesions + granulation tissue + involved disc & bone
12	М	62	LBP+ IC+ Lt sciatalgia + weak Lt EHL	1/1280	Bilateral epidural filling defect at L3/4 level	Epidural granulation tissue + involved disc
13	М	26	LBP+ Lt sciatalgia + weak Lt EHL	1/1280	filling defect at L3/4 level in myelogram	Involved & bulged disc and vertebral disc
14	М	43	LBP+ Rt sciatalgia + weak EHL bilaterally	1/640	filling defect at L3/4 level in myelogram	Epidural granulation tissue
15	М	60	LBP+ IC	1/640	L2/3 block in myelogram + ragged vertebral body	Epidural granulation tissue + involved disc & bone
16	F	15	LBP+ IC+ paraparesis + High sedR.	1/640	Lateral recess syndrome + hyperostosis at L4/5	Epidural granulation tissue + involved disc & bone
17	М	54	LBP+ bilateral leg pain + weak EHL bilaterally	1/80	Cauliflower appearance + vacuum phenomenon	Involved & bulged disc and vertebral bone
18	М	16	LBP+ Lt sciatalgia + loss of ankle jerk	1/80	MRI: L5/S1 Epidural infiltration + infarct bone	Epidural granulation tissue
19	F	19	LBP+ Rt sciatalgia + weak EHL	1/1280	filling defect at L4/5 level in myelogram	Epidural granulation tissue
20	М	16	LBP+ Rt sciatalgia + weak EHL+ sedR=32	1/160	L2/3 Epidural infiltration + bone affection in MRI	Epidural granulation tissue+ involved disc
21	М	41	LBP+ Rt sciatalgia + weak EHL	1/320	bone affection at L4/5 level in MRI + epidural filling defect	Epidural granulation tissue + involved bone and disc
22	М	42	LBP+ IC+ paraparesis	1/160	Epidural infiltration at L3/4 + L5/S1 disc in MRI	Epidural granulation tissue + involved disc & bone
23	М	45	LBP+ sciatalgia + loss of ankle jerk	1/80	Epidural infiltration at L5/S1 level in MRI	Epidural granulation tissue
24	М	32	LBP+ IC+ paraparesis	1/640	T12/L1 wedging + epidural infiltration	Epidural granulation tissue + involved disc
25	М	48	LBP+ Rt sciatalgia + weak EHL	1/80	Epidural infiltration at L4/5 level in MRI	Epidural granulation tissue
26	М	42	Neck pain + arm pain + numb feeling in fingers	1/320	Epidural infiltration with disc and bone affection in MRI	Medical therapy leading to gradual improvement of symptoms

Table 1. LBP= low back pain, IC=intermittent claudication, EHL= extensor hallucis longus, Sed R= sedimentation rate.

rest, Cotrimoxasol and Rifampin. There was demonstrated remarkable improvement in the patient's neurological deficits, and the

treatment was continued for an additional 8 weeks. Follow-up images were not available.

Discussion

Apart from the mental and psycho-neurotic manifestations of systemic Brucellosis, osteoarticular disease is the most common physical complication of Brucellosis occurring in 10-85% of patients.^{8,9}. These bone and joint lesions include arthritis, bursitis, sacroiliitis, tenosynovitis, spondylitis and osteomyelitis.² Spondylitis can be considered the prevalent and consequential most form of osteoarticular involvement in adults infected by Brucella species. Spondylitis may become difficult to diagnose; especially in cases complicated bv devastating neurological or vascular complications of the illness.3,5,6,8 The spine is affected mostly in the chronic phase of the illness, presenting as a crippling disease and with positive serum hemagglutination tests which may show a titer less than 1/160. It is almost accepted that if the patient meets at least two of the following criteria, the diagnosis of Brucella spondylitis is considered: 9

1) a Brucella agglutination titer of 1/160 or higher,

2) a positive blood or bone marrow aspirate culture for Brucella species,

3)supportive imagings of the skeletal system (spondylodiscitis / osteomyelitis)

4)presence of non-caseating inflammatory/ granulomatous tissue

5)and we submit for consideration, patients living in areas where, Brucellosis is endemic. In such cases in which there is *chronic* or *partially* treated brucellosis, a lower agglutination titer (less than 1/160) can be considered as a positive titer.

Interestingly enough, the results of this retrospective analysis shows that; 80% (21 out of 26) were male patients all presenting from rural areas and 60%, (15 out of 26) were in the 4th and 5th decades of life. Wright agglutination test was positive in all the cases with varying titers in different types of patients, however, no positive blood or tissue culture for brucellosis could be obtained in the evaluated cases. This is most likely due to either lack of clinical suspicion, or failure of the available laboratories to performing the appropriate culture procedures. All of our surgically treated cases had involvement in the lumbar region. The same anatomic distribution has been mentioned by other authors for Brucella spondylitis along the spinal bony canal.⁸ The main conclusions regarding imaging findings in this series of cases were:

A)In the plain radiographs (7 cases), there were either no signs of spondylodiscitis or only subtle changes including endplate demineralization with or without heterogeneous bone mineralization in the posteroinferior or posterosuperior corners. In the chronic cases, narrowing of the disc space and irregularity of the margins of the vertebral bodies were better visualized (fig. 4a) which may lead ultimately to partial or complete wedging or fusion of the vertebral bodies. These findings are nonspecific for Brucellosis as mentioned by the others in the literature. ^{7,11,12,16,17}

B)Considering that most of the patients became progressively symptomatic neurologically, many of the patients had diagnostic myelograms performed. Myelography was the diagnostic tool of choice in the earliest cases presented in this study (10 cases) and the findings in these cases varied from simple epidural filling defects in one or two adjacent levels (fig. 6a&b), to complete myelographic blocks with a tooth brush appearance. We did not encounter any report emphasizing this particular finding in the literature.

C) Isotope bone scan was available in 3 of our cases which showed nonspecific involvement of the vertebrae.

D)Computed tomography performed in 3 cases yielded positive findings including; flattening of the discs and destruction of the vertebral endplates which was seen on CT but not visible in the plain X-rays, small hypo-dense areas within the disc space (vacuum phenomenon) (fig. 8), intraspinal extension of the inflammatory process with displacement of the dural sac and reactive hyperostotic reaction at the edge of the vertebral bodies (cauliflower appearance).²¹ These findings are also nonspecific for Brucellosis as mentioned by some other authors. 4,9,15,18,19,20

E)MRI (8 cases) demonstrated hypointensity on T1W images and mixed high signal/low signal intensity in T2W images in most of the cases in this series which is somewhat different from those reported in the anecdotal cases previously described in literature.^{3,5,8,9,11,14,20} The anterior aspects of the vertebral bodies and discovertebral junctions were not affected in this series of patients and no paraspinal abscesses were encountered. These findings are also in contrast to the reported characteristics for Brucella spondylitis mentioned elsewhere in the literature.^{5,9,14,20} The otherwise described changes may likely be related to inflammatory processes in different stages of the illness and the ischemic areas developing in the affected zones of the disease. MRI has been able to identify the extension of the inflammation both into the spinal canal and in the paraspinal region more

clearly. Of note, similar changes have been reported in tuberculous and other pyogenic spondylodiscitis cases in the literature.^{5,6}

Conclusion

The findings in this series of patients indicates a low single-study specificity for the various described imaging studies used to help diagnose Brucella spondylodiscitis. The presence of one of those five important criteria (originating from rural areas, positive hemaglutination test, positive culture from peripheral blood or tissue specimens, histopathological examination of the specimens compatible with inflammatory processes, suspicious imaging studies) in a single patient in addition to similar imaging changes presented in this review, can lead to a high level of suspicion for Brucella spondylitis. MRI can be very helpful in obtaining differential diagnosis between spondylodiscitis and other tumoral, traumatic or degenerative spinal pathologies.

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