Case Report

Pure Brucellar Discitis Mimicking Lumbar Disc Herniation: A Case Report and Review of The Literature

Murat Şakir EKŞİ1, Yaşar BAYRİ1, Ali ÖZEN1, Adnan DAĞÇINAR1, Deniz KONYA2

1Marmara University Medical School, Neurosurgery, Istanbul, Türkiye 2Bahcesehir University Medical School, Neurosurgery, Istanbul, Türkiye

Summary

Brucellosis is a zoonotic infection, endemic in the Middle East, the Mediterranean region, Central and South America. Osteoarticular involvement is the most common form of the disease process with spinal complications in third place in this group. We presented a 36-year-old male patient with signs and symptoms of lumbar disc herniation. On lumbar magnetic resonance imaging, a right-sided L5-S1 disc extrusion was detected and the patient was operated. Frozen material pointed out an inflammatory process with Brucella melitensis in disc specimen culture. He was put on rifampicin and doxycycline chemotherapies. After 6 months of follow-up; his clinical, laboratory, and radiological findings became normal. Medical treatment was stopped with ongoing routine outpatient follow-ups. Discitis without spondylitis in Brucella infection is very rare. There have been 2 case reports in the literature. Also, Brucella disc infection should be in differential diagnosis of lumbar disc herniation clinic in endemic parts of the world.

Key words: Lumbar disc herniation, brucellar discitis, spine, MRI

INTRODUCTION

Brucellosis is a multisystem infection present worldwide but endemic in the Middle East, the Mediterranean region, Central and South America(3,9). It is still a public health problem with a disability weight of 0.150 for chronic, localized; 0.190 for acute diseases(4).

Brucellosis is a zoonosis of aerobic, non-encapsulated, non-motile, non-hemolytic,
gram-negative intracellular coccobacillus\(^{(2,6,9)}\). It is metabolically inactive outside host cell\(^{(2)}\). It was first described by Bruce (Malta) in 1887\(^{(1)}\). Its prevalence in Turkey is 20.32/100000\(^{(7)}\). It affects male gender a little more than females (55%) and neurological manifestations are present in 4% of patients\(^{(4)}\). Brucella has 4 species infecting humans: B. abortus (cattle), B. suis (swine), B. melitensis (goat), B. canis (dog)\(^{(2-3)}\). B. melitensis is the most common pathogen in spinal infections and the most virulent type in human brucellosis\(^{(2-3,5)}\). Humans get infected through contaminated non-pasteurized milk and dairy products or direct contact with infected animals\(^{(3,5-6,9)}\).

Spinal brucellosis confines vertebral body and spreads to intervertebral disc space. Discitis without spondylitis is a very rare manifestation\(^{(9)}\). In this case report, we present a 36-year-old young man with brucellar discitis having low back pain and right sciatica presenting as lumbar disc herniation clinic.

CASE PRESENTATION

A 36-year-old male patient applied to our clinic with low back pain and right sciatica. He was an officer in a government institution. He had had the pain for 2 months before seeking medical aid. He had no history of consumption of unpasteurized milk or dairy products. He had type 2 diabetes mellitus and hypertension on past medical history.

On admission, the patient had hypoesthesia on lateral side, 4\(^{th}\) and 5\(^{th}\) fingers of right foot. There was 1/5 motor weakness of right gastrocnemius and posterior tibialis muscles. Achilles deep tendon reflex was hypoactive on the right side. Also, straight leg raising test was positive at 45 degrees from horizontal plane on the right leg. There was no tenderness over the lumbar spinal processes. His body temperature was 36.8°C. Routine pre-operative laboratory work-up results were within normal range (white blood cell count: 6000/uL). On his lumbar MR examination, a right sided disc herniation was observed at L5-S1 level. It was hyperintense to both spinal cord and intervertebral disc material, on T1- and T2-weighted images (Fig. 1). After diagnosis of lumbar disc herniation, right L5 hemilaminectomy, right L5-S1 microdiscectomy, right S1 foraminotomy were performed.

Extruded disc material was excised. Macroscopically, it was bloody mucoid in nature. A piece of disc material was sent to frozen and the other piece to microbiology laboratory for culture. On frozen, a granulation tissue with polymorphonuclear leukocytes was observed. On further immunohistochemical studies, CD3+, CD20+ lymphoid cells and immunoreactivity in kappa and lambda plasma cells were detected.

After the operation, the patient was immediately consulted to infectious diseases department. Blood erythrocyte sedimentation rate (ESR) and C-reactive protein (CRP) were 32 mm/h and 7.8 mg/L, respectively. Moxifloxacin was proposed as initial chemotherapy. On post-op 6\(^{th}\) hour, the patient was mobilized, he spontaneously urinated. His pain had been relieved by the surgery. On post-op 1\(^{st}\) day, he had a spot high fever of 38.1°C. A blood sample was taken for culture. A week after I.V. antibiotic therapy, he was well and afebrile. After re-consultation to infectious diseases department, his medical therapy was turned to oral form and he was discharged from hospital.

The patient was routinely followed by our outpatient clinic with infectious diseases department. No organism was cultured in the blood sample. But, disc specimen culture became positive for B. melitensis. Also, serum agglutination test was positive at 1/320 titer. Moxifloxacin was exchanged with a combination therapy of rifampicin 2 x 600 mg and doxycycline 2 x 100 mg. A control MR examination was performed 1 month after operation. The posterior part of L5-S1 intervertebral disc and an area
immediately surrounding right S1 root were enhanced homogenously with I.V. contrast material compatible with discitis. His follow-up continued for 6 months with the same regimen. In some controls, he reported that he had undulant fever, night sweats. After 6 months, a new lumbar MR scan showed only granulation tissue. Discitis had regressed. On laboratory tests, his last ESR and CRP were 11 mm/h and <3.36 mg/L, respectively. Wright test was still positive at 1/160 titer. But, both inflammatory markers and radiological appearance of the operated site were compatible with total cure. Chemotherapy was stopped with ongoing clinical follow-ups.

Figure 1: On lumbar MRI, there is right-sided extruded disc material at L5-S1 level, hyperintense to spinal cord on T1- and T2-weighted slides (A-C). The spinal column is intact without any degenerative changes on plain radiographs (D, E).

DISCUSSION
Spinal infections include a wide variety of disease entities like spondylitis, discitis, spondylodiscitis, meningitis, myelitis, pyogenic facet arthropathy, epidural abscess. Immunosuppression, diabetes mellitus, chronic renal failure, old age, alcohol abuse are predisposing factors\(^5,\)\(^8\). Our patient had type 2 diabetes mellitus. Brucellosis is one of the important and common sources of spinal infections in endemic parts of the world.

Brucellosis is a systemic granulomatous infection and may present with undulant fever (rises in the afternoon, falls during the night), malaise, anorexia, fatigue, sweats, chills, headache, weight loss, nausea, vomiting, abdominal pain, hepatosplenomegaly, lymphadenopathy, neurological signs, other signs and symptoms related with skeletal system\(^2\)-\(^4,\)\(^6\). Spinal involvement is the third most common skeletal disease process\(^6\). Spinal Brucellosis mainly affects male gender (1.5 : 1) and middle ages between 5th and 7th decades of life. Brucellar spondylodiscitis presents with non-specific signs and symptoms\(^6,\)\(^8\). The most common presenting symptom is back pain. But, it is the most common symptom of all infectious spondylodiscitis cases\(^8\).

There are two different theories about the origin of infection in spine. The first theory states that Brucella has an affinity for embryonic tissues such as intervertebral
disc, as it is a notochordal derivative\(^{(1)}\). The second opinion insists that infection starts from anterior superior end plate in the spinal column. Because the infection spreads via blood system and the blood supply is very rich in this site. But rarely it may emerge from the inferior end plate\(^{(3,5,9)}\). It encloses all vertebral body and spreads to the neighbor intervertebral disc space and the other intact vertebral body\(^{(9)}\). So, the process is called as spondylodiscitis. Spinal cord or nerve root compression signs and symptoms due to brucellosis have been reported in the literature\(^{(5-6,9)}\). Rate of neurological deficit is 25.8% in Brucella spondylodiscitis patients\(^{(8)}\). Paraspinal collections are present in 10-20% of the spinal cases\(^{(5-6)}\). Even in the cases presenting with only disc herniation symptoms, spondylitis or spondylodiscitis is detected\(^{(5)}\). Discitis without spondylitis in Brucellosis is very rare and there have been 2 case reports in the literature (Table 1)\(^{(5,9)}\). All patients (including our case) were male and had discitis at L5-S1 level. Two were right-sided and the other was left-sided.

**Table 1**: Features of the reported cases of pure Brucellar discitis in the literature

<table>
<thead>
<tr>
<th>Author(s)</th>
<th>Patient age/sex</th>
<th>Infection location</th>
<th>Presentation</th>
<th>Treatment</th>
<th>Follow-up time</th>
</tr>
</thead>
<tbody>
<tr>
<td>Demirci(^{(5)})</td>
<td>30 year/male</td>
<td>L5-S1</td>
<td>Low back and left leg pain</td>
<td>L5-S1 discectomy; after discitis diagnosis doxycycline (200 mg/daily) and streptomycin (1 g/daily) for 21 days, rifampicin (600 mg/daily) and doxycycline (200 mg/daily) for 3 months</td>
<td>4 months</td>
</tr>
<tr>
<td>Yılmaz et al.(^{(9)})</td>
<td>50 year/male</td>
<td>L5-S1</td>
<td>Low back and right leg pain</td>
<td>L5-S1 discectomy; after discitis diagnosis doxycycline and rifampicin</td>
<td>3 months</td>
</tr>
<tr>
<td>Present case</td>
<td>36 year/male</td>
<td>L5-S1</td>
<td>Low back and right leg pain</td>
<td>L5-S1 discectomy; after discitis diagnosis doxycycline and rifampicin (200 mg/daily) and rifampicin (1200 mg/daily)</td>
<td>6 months</td>
</tr>
</tbody>
</table>
Brucella discitis could not be diagnosed before the surgery in our case; because clinical or radiological signs and symptoms were not related with an infection. The patient had only low back and radiating right leg pain resembling lumbar disc herniation clinic without any fever, fatigue, malaise or any other non-specific constitutional symptoms. On MR imaging, right sided L5-S1 disc extrusion over S1 root was observed. Herniated disc material was hyperintense to the L5-S1 disc, much more resembling extruded disc material containing water in it. The L5-S1 intervertebral disc space was hypointense meaning as disc degeneration. There was no other pathological signal change in any vertebral bodies or other disc spaces (Fig. 1A-C). On lumbar plain radiography, there was no abnormal change dealing with infection (Fig. 1D, E). So, post-contrast MR scanning protocol was not performed before the surgery. Even though, if it had been applied, it would not have given us more information than T2-weighted scans. Only, epidural extensions and meningeal spread could be better visualized, if any of them had been present.

Non-specific erosions in consecutive vertebral bodies (brucellar epiphysitis) and narrowed disc space are detected in usual spinal brucellosis cases. In acute stage, infected disc spaces and vertebral bodies appear more hypo- to isointense on T1- and hyperintense on T2-weighted MR images. On chronic stage, signal intensity varies. Beside MR, being the method of choice in imaging the disease, computed tomography and bone scintigraphy (sensitivity between 69% and 91%) can be used as trustful imaging modalities. Some radiological properties are important to differentiate the disease from a more common entity: spinal tuberculosis. In tuberculosis; paraspinal involvement is markedly more, making out gibbous deformities, and posterior vertebral involvement is also important.

Although posterior vertebral elements are usually protected in Brucellosis, facet joint arthritis may occur. But, radiological changes usually appear 3 months after clinical presentation of spinal signs and symptoms. We suspected an infection, due to mucoid structure of extruded disc material observed in the surgery.

Diagnosis is confirmed with positive blood culture (56.2% of cases), specimen culture or if not possible (especially in the chronic stage of the disease), with positive blood serology tests such as standard tube agglutination (Wright test), Rose-Bengal test. IgG agglutination titers above 1:80 is indicative for active infection. High ESR (>20 mm/h) and CRP (>6 mg/L) are not pathognomic but useful for diagnosis of infection and follow-up of the disease process. In our patient, serum agglutination test was positive at 1/320 titer. At the end of the medical therapy, even it became lesser, still it was higher than normal (1/160).

Infected disc material enlarges because of inflammatory process in the nucleus pulposus. After the pressure inside the disc passes over the extrinsic pressure, disc material extrudes mimicking a disc herniation. Discitis without spondylitis in this case might have proposed an early phase of Brucella infection such as Yilmaz et al. mentioned spondylitis had evolved in their pure discitis case after 10 days of Brucella diagnosis. As mentioned before, because of the acute stage of the infection, other radiological changes of spondylitis probably had not evolved in time to diagnose in our case.

In medical therapy, prolonged antibiotic usage is mandatory because Brucella species are obligatory intracellular organisms. Tetracycline, trimethoprim-sulphamethoxazole, rifampicin, aminoglycoside and quinolone are useful and combination treatments are more reliable. Surgery is conserved for cases with mass effect causing neurological
deficit or spinal instability\textsuperscript{(3,8)}. Relapse rate is 40% in the literature\textsuperscript{(6)}. Prognosis is related with early diagnosis and specific treatment directed at the causative agent\textsuperscript{(8)}.

There are some other diseases in the differential diagnosis of this case such as disc herniation, metastasis, bacterial or tuberculosis spondylitis. Metastasis was out of scope because of age of the patient and absence of any signs and symptoms pertinent to a primary disease. Pyogenic spondylodiscitis presents more acute than Brucellosis\textsuperscript{(3)}. Patient history and radiological findings were inconsistent with a tuberculosis infection. It was really hard to differentiate the infection from lumbar disc herniation. There was neither spondylitis appearance on MR scans nor other paraspinal inflammatory process.

CONCLUSION

This case showed us, there is not a rule that spondylitis should be present in every spinal brucellosis case. A high level of suspicion should be conducted in every case of lumbar disc herniation in endemic parts of the world. In such a case, serological tests should be performed before the surgery; so medical therapy without surgery can be chosen if neurological deficits have not evolved already.

Correspondence to:
Murat Şakir Eksi
E-mail: muratsakireksi@gmail.com

Received by: 01 August 2013
Revised by: 12 August 2013
Accepted: 27 August 2013

REFERENCES


The Online Journal of Neurological Sciences (Turkish) 1984-2013
This e-journal is run by Ege University Faculty of Medicine, Dept. of Neurological Surgery, Bornova, Izmir-35100TR as part of the Ege Neurological Surgery World Wide Web service.
Comments and feedback:
E-mail: editor@jns.dergisi.org
URL: http://www.jns.dergisi.org
Journal of Neurological Sciences (Turkish) Abbr: J. Neurol. Sci.[Turk]
ISSNe 1302-1664