

Case Report

Surgical Management of Cervical Spinal Epidural Abscess Caused by *Brucella Melitensis* : Report of Two Cases and Review of the Literature

Mehmet Ali Ekici, M.D.,¹ Zühtü Özbek, M.D.,² Abdülkerim Gökoğlu, M.D.,³ Ahmet Menkü, M.D.³

Department of Neurosurgery,¹ Şevket Yılmaz Training and Research Hospital, Mimar Sinan Ward, Emniyet Avenue, Bursa, Turkey

Department of Neurosurgery,² Yunus Emre State Hospital, Salih Bozok Avenue, Eskişehir, Turkey

Department of Neurosurgery,³ Erciyes University, Medical School, Talas Avenue, Kayseri, Turkey

Spinal epidural abscess, if especially caused by Brucellosis is a very rare disease which is usually a consequence of spondylodiscitis. The spinal column can be affected at any joint; however, the lumbar spine is the most common region, especially at the level of the L4-5 and L5-S1. The frequency of spinal involvement usually seen at the lumbar, thoracic and cervical spine respectively. As an occupational disease in farmers, veterinarians, butchers, laboratory staff and shepherds, brucellosis can also occur by direct contact to animals and infected materials or ingestion of raw cheese, milk or unpasteurized milk products. In this study, we presented two cases with cervical spinal epidural abscess caused by brucella melitensis, which was successfully treated by surgical approach. Initial treatment was combined with antibiotic therapy after the surgery for 3 months.

Key Words : Cervical epidural abscess · Brucella melitensis · Spondylodiscitis · Spinal cord compression · Surgical treatment.

INTRODUCTION

Spinal epidural abscesses (SEA) are rare pathologies and the most frequently cause is *Staphylococcus Aureus*. The incidence of spinal epidural abscesses account approximately 1 in every 10000-hospital admissions²⁹. In addition to *Staphylococcus aureus*, other bacteria or fungi species such as *Candida glabrata*, *Actinobacillus*, *Pseudomonas*, *Mycobacterium tuberculosis*, *Nocardia*, *Pneumococcus*, *Salmonella enteritidis* and *Brucella* species can cause SEA^{6,21,23}. Diabetes mellitus, intravenous drugs, alcohol abuse, and immunosuppression predispose to infections resulting in SEA^{6,21,23,25,31}.

Brucellosis is a systemic, zoonotic disease caused by an aerobic, gram negative and non-encapsulated cocobacillus, which may affect many organ systems^{5,33}. Out of six, four brucella species, *Brucella abortus*, *Brucella suis*, *Brucella canis*, *Brucella melitensis* can cause human diseases. *Brucella melitensis* is the most virulent species. Humans are infected by direct contact with infected animals or materials and ingestion of animal products,

particularly raw milk and cheese^{14,15}. Osteomyelitis usually occurs at the vertebrae especially at the lumbosacral area. Involvement of the epidural space is rare, but the formation of SEA was reported in less than 1.5% of the cases with neurological complications and generally associated with spondylitis^{16,23,28}. The frequency of spinal involvement usually seen at the lumbar spine, thoracic and cervical spine respectively^{1,7,26,30}.

Osteoarticular involvement is the most common complication, which was reported up to 10 to 85% in most series^{1,7,10,19,26,30}. Sacroileitis and arthritis represent the acute form and frequently respond to the standard therapies. It has been reported that the spinal column is generally affected with subacute and chronic forms of brucellosis^{1,22}. Furthermore, spinal brucellosis usually affects elderly patients, whereas sacroileitis and arthritis are usually reported in the first three decades of life^{1,7}. Infection of the epidural space results from hematogenous dissemination or local extension of the infection from adjacent structures, which might be originally infected from hematogenous spread. Approximately half of epidural abscess result from hematogenous

• Received : November 17, 2011 • Revised : March 1, 2012 • Accepted : June 12, 2012

• Address for reprints : Mehmet Ali Ekici, M.D.

Department of Neurosurgery, Şevket Yılmaz Training and Research Hospital, Mimar Sinan Ward, Emniyet Avenue, Bursa 16300, Turkey

Tel : +90-(224) 2955000, Fax : +90-(224) 295 54 97, E-mail : mehmetali.ekici@gmail.com

• This is an Open Access article distributed under the terms of the Creative Commons Attribution Non-Commercial License (<http://creativecommons.org/licenses/by-nc/3.0>) which permits unrestricted non-commercial use, distribution, and reproduction in any medium, provided the original work is properly cited.

spread to the epidural space^{16,23,28}.

CASE REPORT

Case 1

A 61-year-old, male farmer was admitted to the hospital with complaints of weight loss for more than a month, acute severe neck and right shoulder pain of ten days duration. Pain progressively had worsened and became incapacitating before admission. Fever with rigors, fatigue, malaise and profuse night sweating were added to the symptoms. There was no significant predisposing basal illness or factor in history except consumption of fresh goat cheese. Neurological examination revealed 3/5 motor weakness at the shoulder abduction, C4-5 radicular

hypoesthesia, hyperactive deep tendon reflexes and no meningeal irritation signs. The results of the blood tests revealed positive Wright agglutination reaction at a titer of 1/40. White blood cell count (WBC), erythrocyte sedimentation rate (ESR) and C-reactive protein values (CRP) were 8700 mm³, 32 mm/h, 30.7 mg/dL respectively. Blood culture was negative. Magnetic resonance imaging (MRI) studies revealed anteriorly spinal cord compression due to T1-weighted iso-hypointense, T2-weighted hyperintense epidural fluid collection originating from C4-5 disc space. A right anterior cervical approach was performed. A needle was inserted to the C4-5 disc space to confirm by C-arm scopy. Spontaneous drainage of yellowish, purulent material from the disc space (Fig. 1C) was observed after insertion, and culture samples were taken immediately. After achieving the

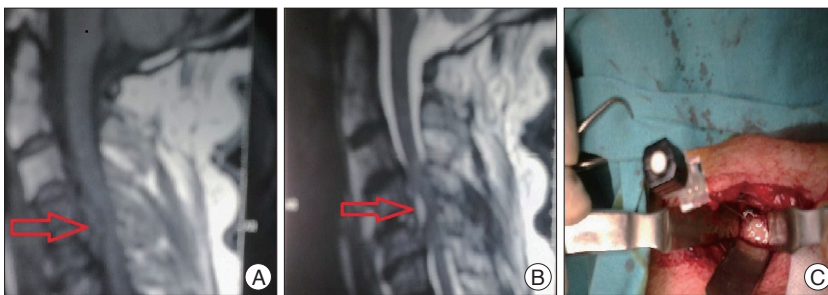


Fig. 1. A : Sagittal T1 weighted magnetic resonance imaging (MRI) of Case1 revealing spinal cord compression at the C4-5 disc level (arrow). B : Sagittal T2 weighted MRI of Case 1 revealing hyperintense epidural lesion and spinal cord compression at C4-5 disc level (arrow). C : The abscess is seen intraoperatively after insertion of needle into C4-5 disc space.

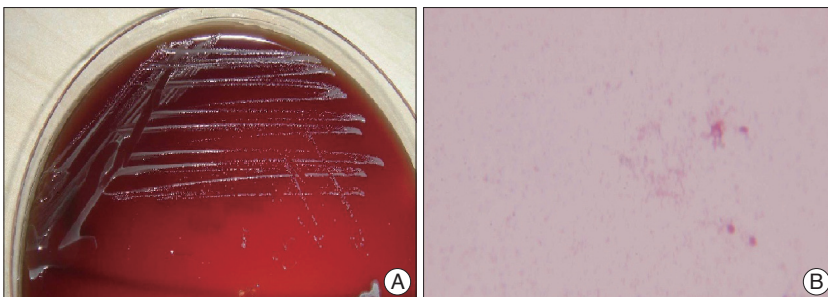


Fig. 2. A : *Brucella melitensis*' colony appearance is also seen with the eosin methylene blue agar. B : Gram negative cocobacillus is being seen at the photomicroscope (H.E×100).

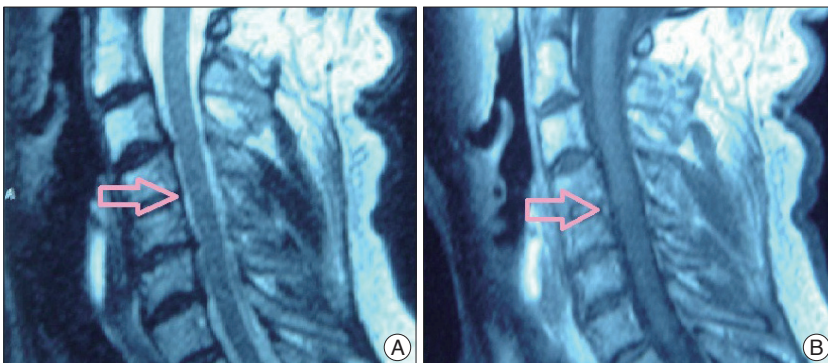


Fig. 3. A : Sagittal T1 weighted magnetic resonance imaging (MRI) of Case 1 demonstrates complete disappearance of epidural abscess at the end of postoperative third month (arrow). B : Sagittal T2 weighted MRI of Case 1 revealing complete recovery at the end of postoperative 3rd month (arrow).

neural decompression due to drainage of abscess, C4-5 simple discectomy was completed without instrumentation because there were no clinical and/or radiological instability or collapse at disk space. A rapid recovery occurred in neurological sign and symptoms of the patient after the operation. Broad spectrum antibiotics consisted from 1 gr IV ceftriaxone BID and 2 g IV ampicilline-sulbactam TID at early postoperative period before isolation of *Brucella melitensis* at eosin methylene blue (EMB) agar, which took 6 days (Fig. 2). Just after achieving the receipt of surgical sample cultures' results, treatment protocol was substituted to a regimen consisting of PO doxycycline 200 mg/day and PO rifampicin 600 mg/day for 3 months. Control MRI was performed at the postoperative 3rd month visit and complete recovery was demonstrated (Fig. 3).

Case 2

A 63-year-old, male farmer was admitted to hospital with severe neck pain and hypoesthesia on his upper extremity lasting for 5-month. Pain was unresponsive to the medical treatment. In the history, he had weight loss, night sweats, fatigue and severe malaise for 5-month. There was no significant predisposing basal illness or factor in history except consumption of fresh dairy products. Neurological examination revealed C3-4 radicular hypoesthesia without motor weakness, hyperactive deep tendon reflexes, no meningeal irritation signs. Wright agglutination reaction tests re-

vealed positive at titer of 1/160 which was. WBC, ESR and CRP values were 7.06 mm³, 12.0 mm/h, 3.78 mg/dL respectively. MRI studies revealed epidural abscess at spinal C3-4 level. T2-weighted non-enhanced sagittal and axial magnetic resonance images revealed C3-4 anterior cervical spinal cord compression due to a hyperintense lesion which was considered as fluid intensity (Fig. 4). Incapacitating severe pain with long duration and unsatisfactory medical treatment led us to perform a right anterior cervical approach. C3-4 anterior micro-discectomy was performed and culture samples were taken immediately during the abscess drainage. Then polyetheretherketone (PEEK) cage was placed in to the C3-4 disk space due to risk of decrease in disk space height. Neurological symptoms and findings of the patient were rapidly recovered after the operation. Postoperatively, triple antibiotics regimen were given, which consisted of 1 g IV ceftriaxone BID, PO doxycycline 200 mg/day and PO rifampicin 600 mg/day until isolation of *Brucella melitensis*. Treatment protocol was substituted to a regimen consisting of PO doxycycline 200 mg/day and PO rifampicin 600 mg/day for 3 months. Control MRI was performed at the postoperative third month visit and complete recovery was demonstrated (Fig. 5).

DISCUSSION

Cervical spinal epidural abscess is an extremely rare condition with brucella species that is responsible for about 0.1% of cases²⁵. Literature research yielded sixteen reports, which are deal with only cervical involvement due to the brucellosis (Table 1). Mortality is seen only in three patients. In a retrospective analysis of 1028 patients, it was revealed that the most frequent involvement was osteoarticular involvement, which was found in 260 cases (25%); followed by spondylitis, which was found in the 32 patients (3.1%)⁵. Patients with spondylitis are more likely to be older and to have longer durations of symptoms before the therapy²⁸.

Brucellar spondylitis is described in two forms as focal and diffuse. The focal disease is seen at the anterior superior portion of an endplate, which is known to have rich blood supply. The diffuse form involves the vertebral body, adjacent disc, and epidural space. Disease spread via the vascular communication and ligaments^{7,16,22,23,28}.

In one of the retrospective multicentre studies with 219 patients, it was reported that, 105 (48%) of these patients had brucellar osteomyelitis and cervical localization was seen in 16 (7.3%)⁹. In another retrospective study on 263 patients, Colmenero et al.¹⁰ reported that 65 patients had osteoarticular complications; and of these patients, 38 (58%) had brucellar spondylitis and cervical localization was seen in 2 (3%). Mortality was seen in only one case secondary to respiratory complications due to the cervical medullar cord compression.

Treatment of brucellosis must control the illness effectively and prevent complications including relapses. Combined medical treatment with a prolonged course is recommended. Cur-

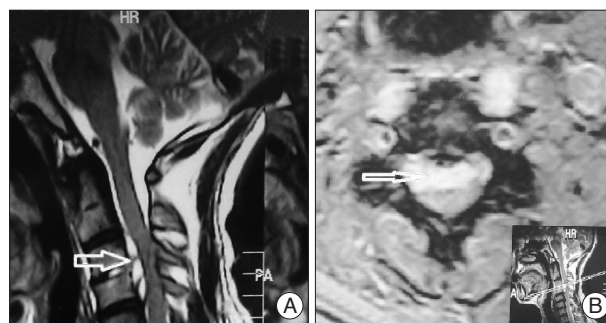


Fig. 4. A : Sagittal T2 weighted magnetic resonance imaging (MRI) of Case 2 revealing hyperintense lesion and spinal cord compression due to epidural abscess at C3-4 level (arrow). B : Axial T2 weighted MRI of Case 2 reveals hyperintense lesion and spinal cord compression due to epidural abscess (arrow).



Fig. 5. Sagittal T2 weighted magnetic resonance imaging of Case 2 demonstrating complete disappearance of epidural abscess at the end of postoperative 3rd month (arrow).

rently, the recommended combinations are doxycycline 200 mg/day, and rifampicin 15-20 mg/kg per day, or doxycycline 200 mg/day plus streptomycin 1g/day i.m. as an initial treatment regimen. The optimal duration of therapy is unknown, but at least 3 to 6 months would be beneficial in the osteoarticular brucellosis⁷. However, there is no standard therapy protocol in chronic brucellosis^{5,33}. First line antimicrobial regimen is doxycycline+streptomycin (200 mg/day/12 weeks and 1 g/day/2-3 weeks, respectively). If there are adverse reactions or contraindications, alternative combination of the antibiotic treatment may be doxycycline+rifampin or co-trimoxazole+rifampin or ciprofloxacin+rifampin or ciprofloxacin+streptomycin¹.

Although, Solera et al.²⁸ reported that initial treatment requires antibiotic therapy, there are a few cases, in which cervical epidural abscess was treated successfully by combined antibiotic regimen without surgery in the presence of spinal cord compression signs. Pina et al.²⁵ reported four cases of cervical spinal epidural abscess due to the brucellosis. Antibiotic therapy was chosen as initial treatment, but three patients underwent surgery due to neurologic deterioration, whereas only one patient had been treated successfully with antibiotics.

Management of cervical epidural abscess due to *Brucella* species remains controversial in terms of antibiotic therapy alone. Ac-

Table 1. Cervical spondylitis and cervical abscess due to the brucellosis, according to literature

Author reference order	Total case (n=41)	Level	Combine antibiotics as a initial treatment	Spinal cord compression	Neurologic deterioration during antibiotics treatment	Surgery	Neurologic sequel	Mortality
Lifeso et al. ¹⁸⁾	6	C1-C7	+	Yes	Yes	Yes	+	No
Mousa et al. ¹⁹⁾	2	C3-C4	+	Yes	Yes	Yes	+	No
		C4-C5	+	Yes	Yes	Yes	+	
Colmenero et al. ¹⁰⁾	2	No level description	+	Yes	Yes	Yes	+	One patients
Solera et al. ²⁸⁾	2	C5-6	+	No	No	No	+	No
		C6-C7	+	No	No	No	+	
Basaranoglu et al. ³⁾	1	C4-5	+	Yes	Yes	Yes	+	No
Pina et al. ²⁴⁾	1	C4-C5	-	Yes	No	Yes	-	No
Pina et al. ²⁵⁾	4	C5-C6	+	Yes	Yes	Yes	+	No
		C4-C5	+	Yes	Yes	Yes	+	
		C5-C6	+	Yes	Yes	Yes	+	
		C6-C7	+	Yes	No	No	-	
Bodur et al. ⁴⁾	2	C3-C4	+	Yes	No	Yes	-	No
		C3-C5	+	Yes	No	Yes	-	
Ates et al. ²⁾	1	C4-C5	-	Yes	No	Yes	-	No
Ugarriza et al. ³²⁾	3	C5-T1	+	No	No	No	-	No
		C4-C5	+	Yes	Yes	Yes	-	
		C5-C6	+	Yes	Yes	Yes	-	
Chelli Bouaziz et al. ⁷⁾	1	C5-6	+	Yes	?	No	?	No
Nas et al. ²⁰⁾	1	C1-C2	+	Yes	No	No	-	No
Colmenero et al. ¹¹⁾	8	No level description	+	Yes	Yes	Yes	+	Two patients
Sengul et al. ²⁷⁾	1	C7-T1	+	Yes	No	No	-	No
Lee et al. ¹⁷⁾	1	C5-6	+	No	No	Yes	-	No
Hantzidis et al. ¹⁴⁾	1	C5-6	-	Yes	No	Yes	+	No

According to the literature, antibiotic therapy with combination of agents is preferred frequently as initial treatment^{3,4,7,10,11,17-20,25,27,28,32)} in contrast some authors suggest surgical approach as an initial treatment^{8,9,12-14,18,30)}. Although neurological sequel is reported frequently, there are only three reports deal with first line treatment option of surgical intervention^{2,14,24)}.

Vertebral osteomyelitis with/without SEA is a serious, potential complication of brucellosis, which often require surgical treatment¹¹⁾. In order to prevent neurologic complications, accurate and early diagnosis is essential²⁵⁾. Although, medical treatment might be enough at early stage of the brucellosis, in older (50-60 years) cases with unsatisfactory respond to medical treatment (antibiotic therapy combined with analgesics), neurologic weakness (tetraplegia or tetraparesis), first motor neuron findings (pathologic reflexes and hyperreflexia on the upper and lower extremities) and long duration of the complaints, it should be kept in mind that disease could be at an advanced stage (subacute or chronic); therefore surgical management should be indicated^{8,9,12-14,18,30)}. Thus, early surgical approach can provide some benefits such as, achieving neural decompression; rapid relief in symptoms and obtaining fresh sample for

microbiologic culture and microscopic analysis for differential diagnosis before deterioration of neurological and clinical parameters^{1,4,11,19,32)}.

Surgical techniques may vary including simple discectomy, aggressive corpectomy plus anterior expandable cage or iliac bone autograft plus anterior plate fixation and simple discectomy plus PEEK Cage fusion.

It might be speculated that first line treatment protocol should be antibiotic therapy with combination of agents, if there are no neurological compression sign and symptoms due to spinal cord compression^{1,8,30)}. Although Case 2 had severe, incapacitating neck and shoulder pain and hyperreflexia without neurological deficit and in contrast Case 1 had neurological deficit, long duration of the complaints and benefits of early spinal cord decompression led us to prefer surgical treatment subsequently combined with antibiotic therapy as initial treatment in both cases.

In conclusion, however, the first line treatment option of the osteoarticular involvement of brucellosis with/without spinal epidural abscess is antibiotic therapy, it should be kept in mind that the first line treatment must be surgical intervention in case of neurological signs and symptoms due to the spinal cord com-

pression, clinical and/or radiological instability and no respond to antibiotic and/or analgesic therapy in order to prevent neurological sequel. Early diagnosis and appropriate early antimicrobial therapy combined with surgery is associated with an excellent prognosis. There are no strict limitations in terms of accurate treatment options; it relies on experience of the surgeon and clinical and/or radiological findings of cases individually.

References

- Alp E, Doganay M : Current therapeutic strategy in spinal brucellosis. *Int J Infect Dis* 12 : 573-577, 2008
- Ates O, Cayli SR, Koçak A, Kutlu R, Onal RE, Tekiner A : Spinal epidural abscess caused by brucellosis. Two case reports. *Neurol Med Chir (Tokyo)* 45 : 66-70, 2005
- Basaranoglu M, Mert A, Tabak F, Kanberoglu K, Aktuglu Y : A case of cervical Brucella spondylitis with paravertebral abscess and neurological deficits. *Scand J Infect Dis* 31 : 214-215, 1999
- Bodur H, Erbay A, Colpan A, Akinci E : Brucellar spondylitis. *Rheumatol Int* 24 : 221-226, 2004
- Buzgan T, Karahocagil MK, Irmak H, Baran AI, Karsen H, Evirgen O, et al. : Clinical manifestations and complications in 1028 cases of brucellosis : a retrospective evaluation and review of the literature. *Int J Infect Dis* 14 : e469-e478, 2010
- Chao D, Nanda A : Spinal epidural abscess : a diagnostic challenge. *Am Fam Physician* 65 : 1341-1346, 2002
- Chelli Bouaziz M, Ladeb ME, Chakroun M, Chaabane S : Spinal brucellosis: a review. *Skeletal Radiol* 37 : 785-790, 2008
- Colmenero JD, Cisneros JM, Orjuela DL, Pachón J, García-Portales R, Rodríguez-Sampedro F, et al. : Clinical course and prognosis of Brucella spondylitis. *Infection* 20 : 38-42, 1992
- Colmenero JD, Jiménez-Mejías ME, Sánchez-Lora FJ, Reguera JM, Palomino-Nicás J, Martos F, et al. : Pyogenic, tuberculous, and brucellar vertebral osteomyelitis : a descriptive and comparative study of 219 cases. *Ann Rheum Dis* 56 : 709-715, 1997
- Colmenero JD, Reguera JM, Fernández-Nebro A, Cabrera-Franquelo F : Osteoarticular complications of brucellosis. *Ann Rheum Dis* 50 : 23-26, 1991
- Colmenero JD, Ruiz-Mesa JD, Plata A, Bermúdez P, Martín-Rico P, Queipo-Ortuño MI, et al. : Clinical findings, therapeutic approach, and outcome of brucellar vertebral osteomyelitis. *Clin Infect Dis* 46 : 426-433, 2008
- Coskun E, Süzer T, Yalçın N, Tahta K : Spinal extradural compression caused by granuloma of brucellosis. *Scand J Infect Dis* 30 : 311-313, 1998
- González-Gay MA, García-Porrúa C, Ibañez D, García-País MJ : Osteoarticular complications of brucellosis in an Atlantic area of Spain. *J Rheumatol* 26 : 141-145, 1999
- Hantzidis P, Papadopoulos A, Kalabakos C, Boursinos L, Dimitriou CG : Brucella cervical spondylitis complicated by spinal cord compression : a case report. *Cases J* 2 : 6698, 2009
- Hasanjani Roushan MR, Mohrez M, Smailnejad Gangi SM, Soleimani Amiri MJ, Hajiahmadi M : Epidemiological features and clinical manifestations in 469 adult patients with brucellosis in Babol, Northern Iran. *Epidemiol Infect* 132 : 1109-1114, 2004
- Izci Y : Lumbosacral spinal epidural abscess caused by Brucella melitensis. *Acta Neurochir (Wien)* 147 : 1207-1209; discussion 1209, 2005
- Lee HJ, Hur JW, Lee JW, Lee SR : Brucellar spondylitis. *J Korean Neurosurg Soc* 44 : 277-279, 2008
- Lifeso RM, Harder E, McCorkell SJ : Spinal brucellosis. *J Bone Joint Surg Br* 67 : 345-351, 1985
- Mousa AM, Bahar RH, Araj GF, Koshy TS, Muhtaseb SA, al-Mudallal DS, et al. : Neurological complications of brucella spondylitis. *Acta Neurol Scand* 81 : 16-23, 1990
- Nas K, Tasdemir N, Cakmak E, Kemaloglu MS, Bukte Y, Geyik MF : Cervical intramedullary granuloma of Brucella : a case report and review of the literature. *Eur Spine J* 16 Suppl 3 : 255-259, 2007
- Nussbaum ES, Rigamonti D, Standiford H, Numaguchi Y, Wolf AL, Robinson WL : Spinal epidural abscess : a report of 40 cases and review. *Surg Neurol* 38 : 225-231, 1992
- Pappas G, Akritidis N, Bosilkovski M, Tsianos E : Brucellosis. *N Engl J Med* 352 : 2325-2336, 2005
- Pérez-Calvo J, Matamala C, Sanjoaquin I, Rodríguez-Benavente A, Ruiz-Laiglesia F, Bueno-Gomez J : Epidural abscess due to acute Brucella melitensis infection. *Arch Intern Med* 154 : 1410-1411, 1994
- Pina MA, Ara JR, Modrego PJ, Juyol MC, Capablo JL : Brucellar spinal epidural abscess. *Eur J Neurol* 6 : 87-89, 1999
- Pina MA, Modrego PJ, Uroz JJ, Cobeta JC, Lerin FJ, Baiges JJ : Brucellar spinal epidural abscess of cervical location : report of four cases. *Eur Neurol* 45 : 249-253, 2001
- Pourbagher A, Pourbagher MA, Savas L, Turunc T, Demiroglu YZ, Erol I, et al. : Epidemiologic, clinical, and imaging findings in brucellosis patients with osteoarticular involvement. *AJR Am J Roentgenol* 187 : 873-880, 2006
- Sengul G, Akar A, Alper F, Uslu H : Nonsurgically treated cervical brucellar epidural abscess causing spinal cord compression. *J Clin Neurosci* 15 : 1411-1414, 2008
- Solera J, Lozano E, Martínez-Alfaro E, Espinosa A, Castillejos ML, Abad L : Brucellar spondylitis : review of 35 cases and literature survey. *Clin Infect Dis* 29 : 1440-1449, 1999
- Tacconi L, Johnston FG, Symon L : Spinal epidural abscess--review of 10 cases. *Acta Neurochir (Wien)* 138 : 520-523, 1996
- Tekkök IH, Berker M, Ozcan OE, Ozgen T, Akalin E : Brucellosis of the spine. *Neurosurgery* 33 : 838-844, 1993
- Turunc T, Demiroglu YZ, Alişkan H, Colakoğlu S, Timurkaynak F, Ozdemir N, et al. : Brucellosis in cases of end-stage renal disease. *Nephrol Dial Transplant* 23 : 2344-2349, 2008
- Ugarriza LF, Porras LF, Lorenzana LM, Rodríguez-Sánchez JA, García-Yagüe LM, Cabezudo JM : Brucellar spinal epidural abscesses. Analysis of eleven cases. *Br J Neurosurg* 19 : 235-240, 2005
- Young EJ : An overview of human brucellosis. *Clin Infect Dis* 21 : 283-289; quiz 290, 1995