INTRODUCTION

Spinal epidural abscess (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 admissions[29]. In addition to Staphylococcus aureus, other bacteria or fungi species such as Candida glabrata, Actinobacillus, Pseudomonas, Mycobacterium tuberculosis, Nocardia, Pneumocococcus, 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[5,19]. 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 vertebras 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[4,5,13]. The frequency of spinal involvement usually seen at the lumbar, thoracic and cervical spine respectively[^21,26,30] .

Osteoarticular involvement is the most common complication, which was reported up to 10 to 85% in most series[^7,10,14,26,30]. 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[^22]. Furthermore, spinal brucellosis usually affects elderly patients, whereas sacroileitis and arthritis are usually reported in the first three decades of life[^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.
spread to the epidural space.\(^{26,23,28}\)

**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 mm\(^3\), 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 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 cases. 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%)%. Patients with spondylitis are more likely to be older and to have longer durations of symptoms before the therapy.

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. 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%)%. In another retrospective study on 263 patients, Colmenero et al. 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. Currently, the recommended combinations are doxycycline 200 mg/day, and rifampicin 15-20 mg/kg per day, or doxycycline 200 mg/day plus streptomycine 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 brucellosis. However, there is no standard therapy protocol in chronic brucellosis. First line antimicrobial regimen is doxycycline+streptomycine (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+streptomycine.

Although, Solera et al. 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

| Autor reference order | Total case (n=41) | Level | Combine antibiotics as a initial treatment | Spinal cord compression | Neurologic deteriation during antibiotics treatment | Surgery | Neurologic sequel | Mortality |
|-----------------------|------------------|-------|-------------------------------------------|------------------------|----------------------------------------------------|---------|-----------------|-----------|
| Lifeso et al. 28       | 6                | C3-C7 | +                                         | Yes                    | Yes                                                | No      | No              | No        |
| Mousa et al. 29        | 2                | C3-C4 | +                                         | Yes                    | Yes                                                | Yes     | No              | No        |
| C4-C5                 | +                | Yes   | Yes                                       | Yes                    | Yes                                                | +       | No              | No        |
| Colmenero et al. 30    | 2                | No level description                      | +                                         | Yes                    | Yes                                                | Yes     | No              | One patients |
| Solera et al. 28       | 2                | C5-C6 | +                                         | No                     | No                                                 | No      | No              | No        |
| C6-C7                 | +                | No    | No                                        | No                     | No                                                 | +       | No              | No        |
| Basaranoglu et al. 31  | 1                | C4-5  | +                                         | Yes                    | Yes                                                | Yes     | No              | No        |
| Pina et al. 11         | 1                | C4-C5 | -                                         | Yes                    | No                                                 | Yes     | -               | No        |
| Pina et al. 32         | 4                | C5-C6 | +                                         | Yes                    | Yes                                                | Yes     | +               | No        |
| C4-C5                 | +                | Yes   | Yes                                       | Yes                    | Yes                                                | +       | No              | No        |
| C5-C6                 | +                | Yes   | Yes                                       | Yes                    | Yes                                                | +       | No              | No        |
| C6-C7                 | +                | Yes   | No                                        | No                     | No                                                 | -       | No              | No        |
| Bodur et al. 4         | 2                | C3-C4 | +                                         | Yes                    | No                                                 | Yes     | -               | No        |
| C3-C5                 | +                | Yes   | No                                        | No                     | Yes                                                | -       | No              | No        |
| 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                    | Yes                                                | -       | No              | No        |
| C5-C6                 | +                | Yes   | Yes                                       | Yes                    | Yes                                                | -       | No              | No        |
| Chelli Bouaziz et al. 7| 1                | C5-6  | +                                         | Yes                    | ?                                                  | No      | ?               | No        |
| Nas et al. 30          | 1                | C1-C2 | +                                         | Yes                    | No                                                 | No      | -               | No        |
| Colmenero et al. 13    | 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. 12    | 1                | C5-6  | -                                         | Yes                    | No                                                 | Yes     | +               | No        |

According to the literature, antibiotic therapy with combination of agents is preferred frequently as initial treatment\(^4\,^{17,28,29,30}\) in contrast some authors suggest surgical approach as an initial treatment\(^8,^{12,14,18,30}\). Although neurological sequel is reported frequently, there are only three reports deal with first line treatment option of surgical intervention\(^2,^{14,18}\).

Vertebral osteomyelitis with/without SEA is a serious, potential complication of brucellosis, which often require surgical treatment\(^1\). In order to prevent neurologic complications, accurate and early diagnosis is essential\(^30\). 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 tetraparasia), 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\(^8,^{12,14,18,30}\). Thus, early surgical approach can be 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\(^2,^{14,18,30}\).

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\(^8,^{12,14,30}\). 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 ostearticular 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
1. Alp E, Doganay M: Current therapeutic strategy in spinal brucellosis. Int J Infect Dis 12: 573-577, 2008
2. Ates O, Cayli SR, Koçak A, Kutlu R, Oral RE, Tekiner A: Spinal epidural abscess caused by brucellosis. Two case reports. Neurol Med Chir (Tokyo) 45 : 66-70, 2005
3. 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
4. Bodur H, Erbay A, Colpan A, Akinci E: Brucellar spondylitis. Rheumatol Int 24 : 221-226, 2004
5. Buzgan T, Karahocagil MK, Irmak H, Baran AI, Karsen H, Evrigen 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
6. Chao D, Nanda A: Spinal epidural abscess: a diagnostic challenge. Am Fam Physician 65 : 1341-1346, 2002
7. Chelli Bouaziz M, Ladeb MF, Chakroun M, Chaabane S: Spinal brucellosis. Clin Infect Dis 21 : 235-240, 2005
8. Colmenero JD, Gineros JM, Orjuela DL, Pachón J, García-Portales R, Rodríguez-Sampedro F, et al.: Clinical course and prognosis of Brucella melitensis infection. Arch Intern Med 154 : 1410-1411, 1994
9. 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
10. Colmenero JD, Reguera JM, Fernández-Nebro A, Cabrera-Franquelo F: Osteoarticular complications of brucellosis. Ann Rheum Dis 50 : 23-26, 1991
11. Colmenero JD, Ruiz-Mesa JD, Plata A, Bermódez P, Martín-Rico P, Queipo-Ortuño MJ, et al.: Clinical findings, therapeutic approach, and outcome of brucellar vertebral osteomyelitis. Clin Infect Dis 46 : 426-433, 2008
12. Coskun E, Sizer T, Yalcin N, Tahta K: Spinal extradural compression caused by granuloma of brucellosis. Scand J Infect Dis 30 : 311-313, 1998
13. 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
14. Hantziatis 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
15. Hasanjani Roushan MR, Mohrez M, Smailnejad Gangi SM, Soleimani Amiri MJ, Hajiahamdi M: Epidemiological features and clinical manifestations in 469 adult patients with brucellosis in Babol, Northern Iran. Epidemiol Infect 132 : 1109-1114, 2004
16. Izi Y: Lumbosacral spinal epidural abscess caused by Brucella melitensis. Acta Neurochir (Wien) 147 : 1207-1209; discussion 1209, 2005
17. Lee HJ, Hur JW, Lee JW, Lee SR: Brucellar spondylitis. J Korean Neurosurg Soc 44 : 277-279, 2008
18. Lifeso RM, Harder E, McCorkell SJ: Spinal brucellosis. J Bone Joint Surg Br 67 : 345-351, 1985
19. Mousa AM, Bahar RH, Araj GF, Koshy TS, Muhtaseb SA, al-Mudallal DS, et al.: Neurological complications of brucella spondylitis. Acta Neurochir Scand 81 : 16-23, 1990
20. Nas K, Tasdemir N, Cakmak E, Kemaloglu MS, Bakte 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
21. Nuusbaum 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
22. Pappas G, Akritidis N, Bosilkovski M, Tsianos E: Brucellosis. N Engl J Med 352 : 2325-2336, 2005
23. Pérez-Calvo J, Matamala C, Sanjoaquin I, Rodriguez-Benavente A, Ruiz-Laiglesia F, Bueno-Gomez J: Epidural abscess due to acute Brucella melitensis infection. Infection 20 : 38-42, 1992
24. Pina MA, Ara JR, Modrego PJ, Iyul MC, Capablo JL: Brucellar spinal epidural abscess. Eur J Neurol 6 : 87-89, 1999
25. Pina MA, Modrego PJ, Uroz JJ, Cobeta JC, Lerin FJ, Baiges J: Brucellar spinal epidural abscess of cervical location: report of four cases. Eur Neurol 45 : 249-253, 2001
26. Pourbagher A, Pourbagher MA, Savas L, Turunç 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
27. Sengül G, Akar A, Alper F, Uslu H: Nonsurgically treated cervical brucellar epidural abscess causing spinal cord compression. J Clin Neurosci 15 : 1411-1414, 2008
28. Solera J, Lozano E, Martínez-Alfaro E, Espinosa A, Castillejos ML, Abad I: Brucellar spondylitis: review of 35 cases and literature survey. Clin Infect Dis 29 : 1440-1449, 1999
29. Tacconi L, Johnston FG, Symon L: Spinal epidural abscess--review of 10 cases. Acta Neurochir (Wien) 138 : 520-523, 1996
30. Tekkök IH, Berker M, Ozcan OE, Ozgen T, Akalin E: Brucellosis of the spine. Neurosurgery 33 : 838-844, 1993
31. Turunç T, Demiroglu YZ, Alışkan H, Colakoğlu S, Timurkaynak F, Özdemir N, et al.: Brucellosis in cases of end-stage renal disease. Nephrol Dial Transplant 23 : 2344-2349, 2008
32. Ugarriza LF, Porras LF, Lorenzana LM, Rodrigues-Sánchez JA, García-Yagüe LM, Cabezudo JM: Brucellar spinal epidural abscesses. Analysis of eleven cases. Br J Neurosurg 19 : 233-240, 2005
33. Young EJ: An overview of human brucellosis. Clin Infect Dis 21 : 283-289; quiz 290, 1995