CERVICAL BRUCELLOSIS, WITH OR WITHOUT BONE INJURY

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Abstract: Brucellosis is a zoonotic infectious systemic, endemic in Albania, which is accompanied by multi-organ involvement. A common complication is also the vertebral affection. Manifestation in the lower lumbar spine is frequently seen, while cervical involvement is more rare. Diagnose of vertebral involvement is difficult because of non-specific clinical symptoms. We present two cases of cervical brucellosis, with various cervical injuries. Spondylodiscitis with epidural abscess were the cervical manifestation in the first patient, which is very rare and a serious complication and the second patient had epidural involvement without spondylodiscitis. The Rose Bengal, Wright test, ELISA were positive for both patients. The cervical injury was confirmed with MRI exams. After prolonged combination therapy with cervical immobilization, the follow up evaluation demonstrated resolution of the cervical injury. Because vertebral destruction is in the base of this complication, early diagnosis of vertebral brucellosis is important to prevent serious morbidity, if diagnosis and treatment are delayed. Standard Brucella tube agglutination (Wright) test is the primary test and should be performed as a first step in the differential diagnosis of spondylodiscitis. An MRI is recommended for early diagnosis of spinal involvement. Medically treatment of cervical brucellosis has a good prognosis with early diagnosis.

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Introduction

Brucellosis is an endemic zoonosis in Albania. Microorganisms that most often can cause human brucellosis are B. Abortus, B. Melitensis and B. Suius. Brucellosis is more prevalent in rural areas of Albania, being an occupational disease of veterinarians, farmers, hunters. The incidence in our country is high, the reason is connected even with the fact that 60% of people live in rural zones. Spondylodiscitis is a frequent complication, as a result of an Brucella infection. Estimates of the incidence of spondylitis range from 9% to 31% (Colmenero et al., 2008).

While lumbar spine involvement is the most common, cervical involvement is a rare, but more severe complication, accounting for 8.3% of cases of spondylodiscitis in a recent study. Recovery is observed in 60-90% of patients, with specific treatment (Tekkök, 1993).

Spinal pain is a frequent complaint of today. It can be the only manifestation of vertebral brucellosis, making the diagnose difficult and delayed.

The aim of this study is to present the importance that Brucella etiology has for cervical diseases and for the early treatment of local damages.

Material

We present two different cases of cervical brucellosis, with various cervical injuries.

Case 1

A 55- year old male patient, a farmer, presented at our hospital. His symptoms were night sweats, headache, neck pain extending down the back, and myalgias, especially in the right arm. On examination, we revealed neurological deficits as paresis and functional disabilities of the upper extremities, with limited movement of the arm because of pain.

Laboratory results showed an erythrocyte sedimentation rate (ESR) of 39mm/h, a leukocyte count of 14.100/mm^3 with81% neutrophils and 19% lymphomonocytes, a haemoglobin level of 13.3 g/dL and a platelet count of 238 000 /mm3. Liver function tests were normal and lactate dehydrogenase 158 U/L.

Blood urea, creatinine, total bilirubin and electrolyte levels were normal. Blood cultures were negative. There was an increase in C-reactive protein (CRP) to 33.42 mg/L and fibrinogen was 588 mg/dL.

Wright agglutination test was positive (1:640 I.U/ml). Brucella infection was confirmed with another test. ELISA was positive for Brucella IgG antibodies (16.6 U/mL). Cervical magnetic resonance

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imaging (MRI) confirmed the C3-C4 spondylodiscitis and the existence of a minimal inflammatory paravertebral involvement, epidural abscess and medullary compression. (Fig 1).

The patient started treatment with oral doxycycline, 100mg 2 x day, oral rifampicin 600mg daily and intramuscular gentamicin 240 mg × day. The doxycycline and rifampicin regimen continued for a period of 12 weeks, while gentamicin was discontinued after 2 weeks. Cervical immobilization was applied.

Forty days after admission, the patient was discharged free of symptoms with amelioration of the neurological signs. The laboratory tests on his discharge were normal, including ESR and CPR.

Figure 1: C3-C4 spondylodiscitis with the existence of a minimal inflammatory paravertebral involvement, epidural abscess and medullary compression.

Source: Authors (2013)

Case 2

The second case was a female patient 67 –year old, retired, living in village. She presented with a high fever (39-40ºC), that had started 20 days previously and neck pain, without neurological deficits.

Laboratory results showed an ESR of 54 mm/h, a leukocyte count of 9.600 / mm3 with 66% neutrophils and 34% lymphomonocytes, a haemoglobin level of 10.3 g/dL and a platelet count of 281 000 /mm3. Liver function tests were normal and lactate dehydrogenase 280 U/L. Blood urea, creatinine, total bilirubin and electrolyte levels were normal. Blood cultures were negative. There was an increase in C-reactive protein value to 128.8 mg/L.

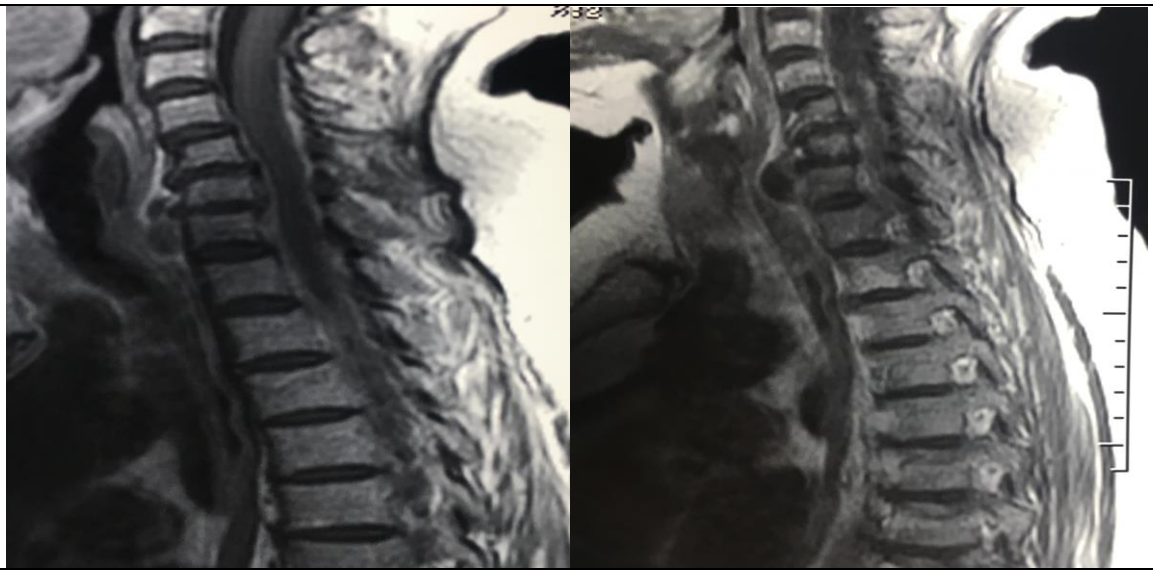
Wright agglutination test was positive (1:1280 I.U/ml). ELISA was positive for Brucella IgG antibodies (13.4 U/mL).

The Cervical MRI confirmed the soft tissue inflammation at the level of C5-C6 vertebrae and epidural inflammatory involvement (Figure 2). The regimen treatment was the same with the first patient. One month after admission, the patient was discharged free of symptoms and the laboratory tests were normal, including erythrocyte sedimentation rate and PCR.

Discussion

Brucellosis is one of the zoonosis with high incidence in Albania. History of brucellosis in Albania begins in 1925 with the diagnosis of several people affected by brucellosis in the south of the country. In 1935, the disease was confirmed also in some goats in the Permet district (Eltari, 1981). However, brucellosis appears to have been endemic to Albania since medieval times. Anthropological analysis of skeletal remains from the ancient Albanian city of Butrint, have confirmed the occurrence of brucellosis in recovered human bone (Motulo et al., 2012).

Figure 2: The soft tissue inflammation at the level of C5-C6 vertebrae with epidural inflammatory involvement



Source: Authors (2015)

During the following forty years, 1960 to 1990 strict measures were taken to limit the infection in cattle and small ruminants in our country. In 1989, the country was declared free of bovine brucellosis and the infection in small ruminants had a very low prevalence in the few flocks in the south of the country (Ilirian et al., 2004). During 1990 -2010, there has been reported a significant increase of the prevalence of brucellosis in Albania, which has influenced a great number of cases and the variety of complications, including those of bones. In Albania, an incidence of 25 per 100.000 inhabitants was reported in 2008 (Mariani et al., 2014). Musculoskeletal involvement typically occurs in men over 40 years of age. The lumbar spine is the most affected, followed by thoracic and cervical spine (Young, 2010, Doganay & Aygen, 2003). The incidence of spinal brucellosis is highly variable (2–54%) (Colmenero et al., 2008, Pourbagher et al., 2006, Mousa et al., 1987, Alp & Doganay, 2008). Involvement of the cervical region is in both our cases. The rate of isolated cervical spondylitis was reported to be 1.2-4% in some studies (Kurtaran et al., 2008, Zormpala et al., 2000).

Muscle, joint and bone pain, and sometimes neurological symptoms (power loss, paraesthesia, paraparesis) can be the manifestation of the musculoskeletal involvement (Young, 2010). Spondylodiscitis is a very insidious disorder, not always accompanied by specific signs. In patients with brucellar cervical spondylodiscitis, back pain has been the presenting complaint (Colmenero et al., 1996). For this reason, in the endemic areas of brucellosis, patients that present with back pain must be examined for Brucella spondylodiscitis also. The back pain was the presenting symptom of our first patient and in the second, it was neck pain.

The diagnosis of spinal brucellosis is the first step to ensure proper treatment. Despite the fact that the diagnosis of cervical brucellosis is not easy, it is very important to initiate specific treatment. Laboratory results alone are not efficient in establishing a correct diagnosis. The standard Brucella tube agglutination (Wright) test is the primary test and should be performed as a first step in the differential diagnosis of spondylodiscitis. Nevertheless, there are seronegative cases, false positive results that have also been reported in the literature (Ulu-Kilic, 2013). In our two cases, the etiologic diagnose was determined by the tube agglutination (Wright) test and serologic test ELISA for Brucella.

The reproduction of Brucella bacteria in cultures taken from appropriate samples is a criterion to diagnose spinal brucellosis. This examination was not performed, because it was refused by both patients. However, serological tests are particularly useful in spinal brucellosis, diagnoses and reducing the need for invasive procedures (Erdem et al., 2015).

MRI remains the gold standard for early diagnosis of spinal involvement. Because bone remodeling can progress slowly, radiographic changes might not be easy to differentiate from those of degenerative disease (al-Shahed et al., 1994). Also, MRI is the method of choice for the assessment of

the disease and follow-up of spinal involvement (Resnick, 1995). When neurologic complications are apparent, the evolution of local spine damages is attended by MRI (Irmak et al., 2004). The diagnose of cervical involvement and its local extension was made by spinal MRI in both cases. This examination was also performed after treatment to follow the effectiveness of it.

Medically treatment of cervical brucellosis has a good prognosis with early diagnosis. No antibiotic combination was proven to be superior, but 14 different regimens were used in the series studied (Pappas et al., 2004). The combination of doxycycline, rifampicin and an aminoglycoside, for two weeks, followed by doxycycline and rifampicin, for 8-10 weeks is the most effective regimen (Madkour et al., 2001). Therapeutic failure and relapse are still reported with this regimen (Ariza, 1985).

The two criteria of anti-brucella treatment efficacy are: first, a normal temperature after the first week of it and second, a normal CRP level within the first month (Ulu-Kilic et al., 2013). Treatment should be continued until the ESR decreases to normal levels and radiological recovery is maintained (Tekkök et al., 1993, Ural et al., 2013). By the end of medical treatment of the two patients, a normalization of ESR and a radiological recovery were evident.

Surgical treatments are performed in cases with neurologic complications, as spinal instability or progressive spinal cord compression (Solera et al., 1999, Samra et al., 1982). None of our patients underwent surgical treatment. Cervical immobilization was applied successfully in our first patient.

Conclusions

Brucellosis is an endemic disease in our country; it is because of that those patients with cervical vertebrae lesions must also be tested for Brucellosis. Early diagnose and treatment will diminish the possibility of serious complications that accompany vertebral involvement and mostly, a total recovery of bone damage. The standard Brucella tube agglutination (Wright) test should be performed as a first step in the etiologic diagnosis of spondylodiscitis. An MRI is recommended for early diagnosis and follow-up of spinal involvement.

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