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CASE REPORT

Positive Brucella serological findings with incidental arteriovenous malformation: A case report

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Abstract

Brucellosis is one of the most important zoonosis. It causes significant economic and medical problems in countries throughout the world, including Iran. According to the available literature, brucellosis rarely causes intracranial space-occupying lesions. In this case study, we reported an intracranial mass in a 52-year-old male who were affected with brucellosis. The patient was referred to Vali-e-Asr hospital with right hemiparesis and transient speech impairment. The symptoms had begun sub-acutely and had gradually progressed. His past medical history was completely normal. Moreover, he had not consumed raw meat, had not been exposed to animal bite and slaughter within the recent weeks. In the initial assessment, the results of Wright agglutination and Coombs Wright tests level were 1/640 (a Wright's titer of less than 1/80 is considered normal in Iran). The serum level of 2-Mercaptoethanol was 1/320 that was higher than the normal value. Brain computer tomography (CT) scan and magnetic resonance imaging (MRI) were performed due to the inconsistency of neurologic findings with the usual presentations of brucellosis. MRI and CT scan findings showed a 35×35×34 mm heterogeneous mass lesion containing multiple vascular components located in the left paraventricular region along with mild peripheral edema and mass effect. Due to the size and the location of the lesion and the neurosurgery consultation, surgery was avoided and follow-up suggested. Treatment of brucellosis was initiated and completed according to the national protocol with rifampin and doxycycline. Follow up were performed in 6, 12, and 24 months after completion of the treatment. The growth of brain lesion was also controlled and the serologic test results of brucellosis were obtained normal.

Key Words: Adult, Arteriovenous malformations, Brucellosis

Introduction

Brucellosis is a common bacterial zoonosis that is transmitted to human through contact with the body fluids of infected animals such as sheep, goat, cattle, and pigs (1, 2). This endemic disease in

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many regions of Iran could also be transmitted to humans through the use of raw meat, non-pasteurized dairy products, exposure to aerosols, skin ulcers, and direct contact with genital mucous membranes (1, 3, 4). Half a million new cases of human brucellosis are discovered annually having numerous negative effects on individuals' health and economic indicators, especially in developing countries (2).

International tourism and migrations play important roles in the global expansion of this disease. Neurobrucellosis occurs in 5-10% of cases of brucellosis, and its most common manifestation is meningoencephalitis (5, 6). Arteriovenous malformations (AVM) are complex vascular lesions that are seen in both male and female patients with similar symptoms. The prevalence of this lesion ranges from 15 to 18 cases per 100,000 populations (7). In these lesions, arterial blood flow enters the venous system directly. The absence of the capillary system leads to the direct transfer of arterial pressure into the venous system; as a result, it increases blood flow and causes dilatation and tortuosity in the venous system (7, 8).

These lesions may also impair the cerebral hemodynamic, including the reversal of the venous blood flow, venous hypertension, and decreased blood flow to the region surrounding the AVM. There is a probability regarding the congenital origin of these lesions; however, the majority of these lesions are formed during the fetal period and are not hereditary (7).

The clinical presentations of these patients are similar to those of cases with other intracranial space-occupying lesions which are caused by pressure on the affected hemisphere and adjacent cranial nerves.

Edema of the surrounding area also plays a role in creating the clinical picture. The clinical presentations include decreased level of consciousness, increased intracranial pressure, neurologic deficits, and intracranial hemorrhage due to the rupture of the AVM. Intracranial hemorrhage is the most serious and common presentation of these lesions (7, 9). These malformations also cause seizure and focal neurologic deficits as a consequence of affecting the adjacent cortex. In the present study, a rare case of brucellosis presented with AVM is reported.

Cases

A 52-year-old male referred to Vali-E-Asr hospital with right hemiparesis and transient speech impairment. The symptoms had begun sub-acutely and had gradually progressed. His past medical history was completely normal and had no history of diabetes, hypertension, cancer, tumors, transfusion, hospitalization, surgery. In addition, he did not use any drug. He had not consumed raw meat, been exposed to animal bite and slaughter within the recent weeks.

In the physical examination , the right facial nerve was found impaired, the gate was disturbed and was deviant to the left, the right plantar reflex was extensor, and the left plantar reflex was flexor. The patient was alert and could recognize the place, time and people. No fever and hypothermia was reported. Blood pressure, heart and respiratory rates were within the normal ranges. Other clinical and neurological examination findings revealed no abnormalities. Furthermore, the complete blood cell count, blood glucose, urea, creatinine, calcium, phosphorus, thyroid profile, liver enzymes, lipid profile, procalcitonin, alkaline phosphatase, serum sodium, potassium levels, prothrombin time, partial thrombin time, international normalized ratio, and urinalysis were within the normal ranges. Troponin, C-reactive protein, erythrocyte sedimentation rate, and rapid plasma reagin test results were negative. The results of Wright agglutination and Coombs Wright were 1/640 (a Wright's titer of less than 1/80 is considered normal in Iran). The serum level of 2-Mercaptoethanol was 1/320 that was higher than the normal value.

According to the findings derived from MRI, there was a 35×35×34 mm heterogeneous mass lesion in the left paraventricular region. It contained multiple vascular components along with mild peripheral edema and mass effect. Central and mild peripheral enhancement of the lesion was noted after contrast injection. These findings are in favor of vascular tumors including AVM or cavernous hemangioma (Figure 1).

Brain magnetic resonance arteriography with and without enhancement showed a vascular lesion with multiple internal microvascular regions in the left side, which was in favor of AVM or cavernous hemangioma (Figure 1).

Figure 2 demonstrates a hypervascular mass in the left suprasellar region, suggesting an AVM. The arterial blood supply of the lesion was from the posterior left cerebellar artery and the posterior left communicating branch. The venous drainage of the lesion was into the rectus vein. A few arteries from the left cerebral artery were also supplying the lesion.

According to the results of clinical examinations, brain imaging, and laboratory tests, the patient was diagnosed with brucellosis and treatment was initiated. He was treated with 1-gram intravenous

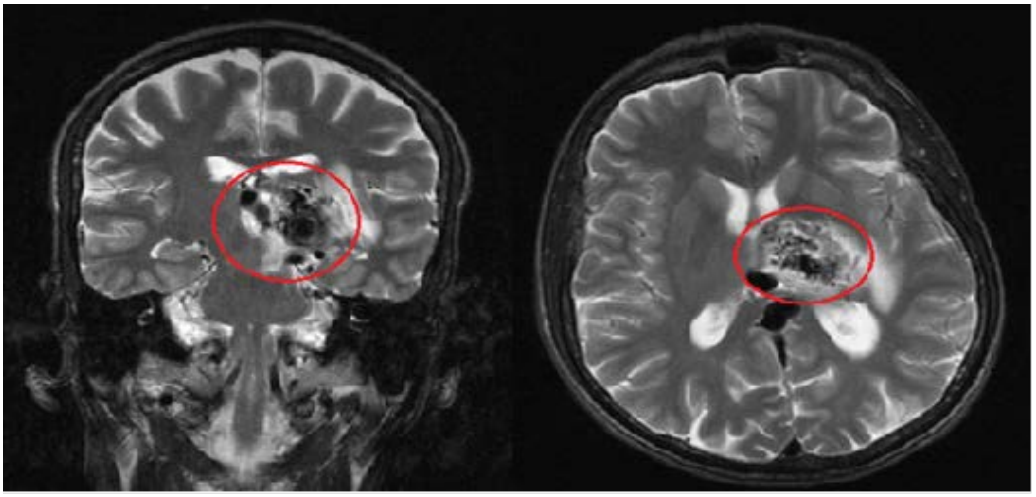

Figure1: Brain MRI showing a large hypervascular mass lesion in the left paraventricular region

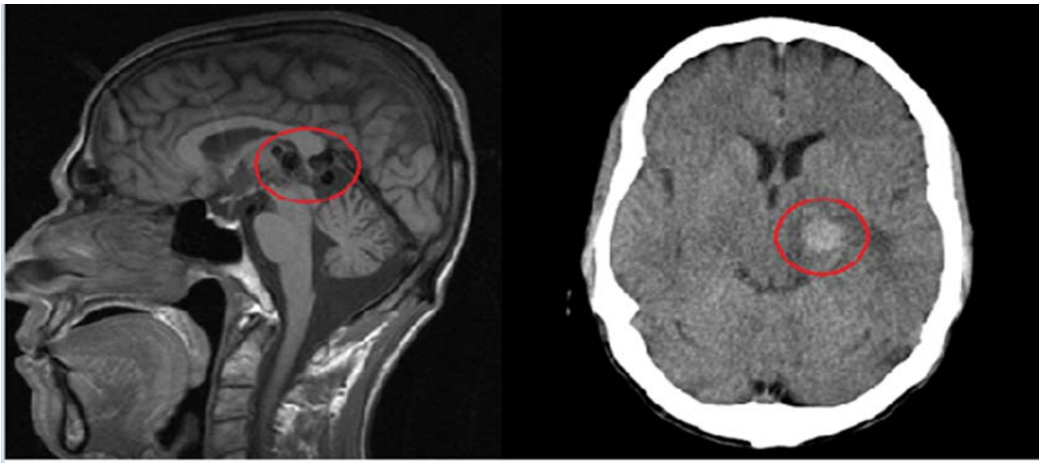

Figure2: Contrast brain CT-angiography showing a space-occupying lesion in the left suprasellar region, suggesting an AVM

ceftriaxone every 12 hours. The treatment was continued after discharge with 600 mg rifampin once daily and 100 mg doxycycline every 12 hours.

Due to the size and location of the lesion and the neurosurgery consultation, surgery was avoided and the patient follow-up was suggested.

Therefore, follow-up visits were performed within 6, 12, and 24 months after the completion of the treatment. The serologic tests of brucellosis were normal; however, no clinical and radiological changes were observed in findings.

Discussion

Brucellosis is a systemic disease that can affect almost all organs of the body; however, the exact mechanism of its transmission to the nervous system is still unknown (1). The diagnosis may also be delayed due to the absence of specific symptoms and signs, (2). The central and peripheral nervous system can be affected by neurobrucellosis (6). As a result, bacteremia and the involvement of the meninges can occur followed by the development of polyradiculoneuropathy, and meningitis or meningoencephalitis.

Some of the rare manifestations of neurobrucellosis include intracranial lesions, brain abscesses, and AVM (2) which can be due to the direct bacterial invasion or immune-mediated destruction of the nerve tissue affected by its endotoxin (1). The common clinical findings of neurobrucellosis include back pain, areflexia, paraparesis, cranial nerves damage, myelitis, and meningovascular injuries such as strokes, neuropathy, and depression (1). This study was a rare case of brucellosis with incidental AVM.

In a study performed by Riasi et al., the

main clinical manifestations of arteriovenous malformations of neonates were congestive cardiac failure (54%), seizure (31%), brain hemorrhage (15%), and hemorrhagic stroke and hydrocephaly (35%) (8). Grillner et al. conducted a study on the presence of a gene called RASA1 that was associated with an increased risk of vascular malformations (9).

In a study carried out by Altekin et al., a patient affected with brucellosis was reported to have aortic valve endocarditis and cerebral aneurysm due to the direct invasion of brucella. As a result, brucellosis can cause neurologic complications through cerebral aneurysms and ensuing hemorrhage (10).Tufan et al. described brain white matter involvement and inflammation in a patient affected with neurobrucellosis leading to brain mass (11). Fincham et al. showed that anemia associated with brucellosis could also be correlated with brain damage (12).

Conclusions

Brucellosis is not considered as the main cause of cerebral AVM in this patient and this can probably be a coincidence. These lesions have probably a genetic origin, and this is why they are persistent after the completion of antibiotic therapy.

Conflict of Interest

The authors declare that there is no conflict of interest.

References

1. Sari I, Altuntas F, Hacioglu S, Kocyigit I, Sevinc A, Sacar S, et al. A multicenter retrospective study defining the clinical and hematological manifestations of brucellosis and pancytopenia in a large series: hematological malignancies, the unusual cause of pancytopenia in patients with brucellosis. Am J Hematol. 2008; 83(4):334-9. PMID: 18069671 DOI: 10.1002/ajh.21098
2. 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. 2010; 14(6):e469-78. PMID: 19910232 DOI: 10.1016/j.ijid.2009.06.031
3. Solera J. Update on brucellosis: therapeutic challenges. Int J Antimicrob Agents. 2010; 36:S18-20. PMID: 20692127 DOI: 10.1016/j.ijantimicag.2010.06.015
4. Guven T, Ugurlu K, Ergonul O, Celikbas AK, Gok SE, Comoglu S, et al. Neurobrucellosis: clinical and diagnostic features. Clin Infect Dis. 2013; 56(10):1407-12. PMID: 23446629 DOI: 10.1093/cid/cit072
5. Tuncel D, Uçmak H, Gokce M, Utku U. Neurobrucellosis. Eur J General Med. 2008; 5(4):245-8.
6. Galinska EM, Zagórski J. Brucellosis in humans-etiology, diagnostics, clinical forms. Ann Agric Environ Med. 2013; 20(2):233-8. PMID: 23772567
7. Tabasizadeh H, Salehi F, Moayyed ME, Moayyed ME. A newborn infant with congenital cerebral arteriovenous malformation, and congestive heart failure: case report. Tehran Univ Med J. 2017; 75(3):235-9.
8. Riasi HR, Salehi F, Sanati AR. Fluctuating neurologic symptoms in a patient with brain lesion. J Med Cases. 2017; 8(2):67-9. DOI: https://doi.org/10.14740/jmc2706w
9. Grillner P, Söderman M, Holmin S, Rodesch G. A spectrum of intracranial vascular high-flow arteriovenous shunts in RASA1 mutations. Childs Nerv Syst. 2016; 32(1):213. PMID: 26552382 DOI: 10.1007/s00381-015-2952-7
10. Altekin RE, Karakas MS, Yanikoglu A, Ozbek SC, Akdemir B, Demirtas H, et al. Aortic valve endocarditis and cerebral mycotic aneurysm due to brucellosis. J Cardiol Cases. 2011; 4(3):e179-82. DOI: 10.1016/j.jccase.2011.07.008
11. Tufan K, Aydemir F, Sarica FB, Kursun E, Kardes Ö, Cekinmez M, et al. An extremely rare case of cervical intramedullary granuloma due to Brucella accompanied by Chiari Type-1 malformation. Asian J Neurosurg. 2014; 9(3):173-6. PMID: 25685211 DOI: 10.4103/1793-5482.142740
12. Fincham RW, Sahs AL, Joynt RJ. Protean manifestations of nervous system brucellosis: case histories of a wide variety of clinical forms. JAMA. 1963; 184(4):269-75. PMID: 13945207