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Case report

Severe neurobrucellosis in a young infant

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1. Introduction

Brucellosis is the most common zoonotic infection, which occurs through direct contact with infected animals or the consumption of infected animal products, especially milk. Brucellosis displays broad clinical polymorphism and almost every organ can be affected during the infection. Neurobrucellosis is rather rare in pediatric patients, comprising 3–10% of all cases reported in the literature. Neurobrucellosis is generally in the form of meningoencephalitis [1]. Brucellar brain abscess is extremely rare in children with brucellosis [2–4]. We describe herein a case of multiple brain abscesses due to brucellosis possibly transmitted by breast milk that occurred in a 4-month-old infant.

2. Case report

A 4-month-old, previously healthy exclusively breast-fed female infant was admitted to our clinic with a 10-day history of fever. She had one attack of generalized tonic–clonic convulsions and altered sensorium of several days in duration. Physical examination on admission revealed a temperature of 38 °C. The patient was conscious but irritable with bulging fontanelle. Isocoric pupils with direct and indirect light reflexes (+) were detected during neurological examination. There was no loss of strength in the bilateral upper and lower extremities. There was no rash or lymphadenopathy. The liver was 2 cm, and the spleen was 1 cm below the costal margins. There were no focal neurodeficits. Deep tendon reflexes were increased. There was no history of contact with tuberculosis. Brucella has been diagnosed in her father eleven months ago, and he has been successfully treated. Her mother has exhibited fever, back pain and fatigue, 30 days after delivery. The mother's blood has been found positive for brucella seroagglutination titer 1:320 at the second month after delivery. Coombs antibrucella test titer has been found 1:640. She has been treated with injectable streptomycin 1 g for the first 21 days together with oral tetracycline (2 g) in four divided doses for 45 days. Breast milk culture could be done after her treatment, and the organism was not isolated from culture of the breast milk. She has sustained breastfeeding after her diagnosis of brucellosis. She has not gived raw's milk or any formula to the patient.

In the patient's laboratory investigation, hemoglobin was 10 g/dl, white blood cell count was 6500/mm^3 with a 70% lymphocyte ratio, and platelet count was 120,000/mm^3. The peripheral blood smear for malarial parasites and the Gruber–Widal test were negative. Serum glucose, electrolytes and liver function tests were normal. His serum immunoglobulin levels and clotting tests were within normal limits.

No abnormality was found by chest X-ray. The Wright agglutination test was positive with a ratio of 1:320 in blood. Brain magnetic resonance imaging (MRI) showed multiple brain abscesses characterized by a lesion with ring enhancement surrounded by edema in both hemispheres and a communicating hydrocephalus causing dilation of the 3rd and lateral ventricles

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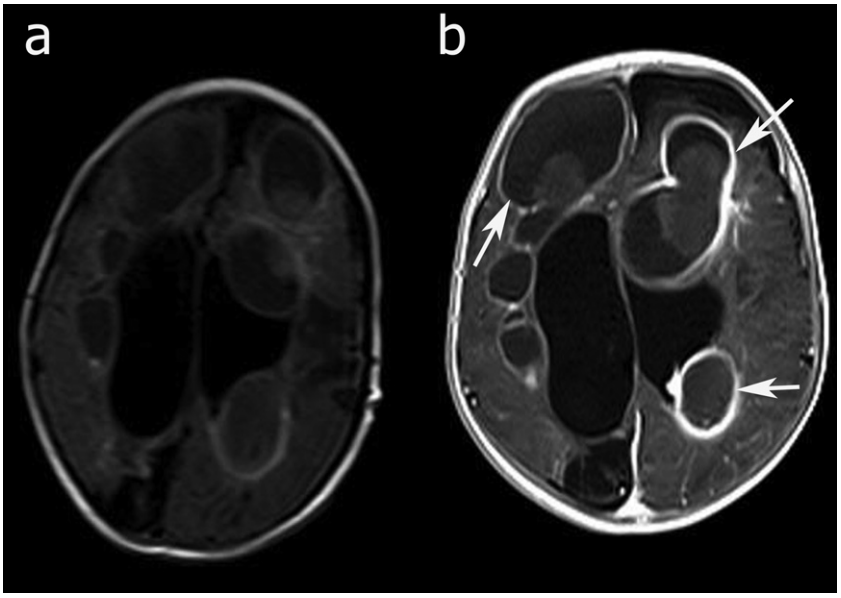

Fig. 1. Magnetic resonance (MR) images T1WI before (a) and after (b) gadolinium administration showing multiple brain abscesses characterized by ring enhancement hypointense lesions in both hemispheres.

(Fig. 1). Diffusion-weighted image (a) demonstrating hyperintense (restricted diffusion) lesions. ADC map (b) demonstrating a predominantly hypointense lesions (Fig. 2).

Examination of the cerebrospinal fluid (CSF) samples disclosed a white blood cell count of 75 cells/mm^3 with 65% lymphocytesd 35% polymorphs, a glucose level of 55 mg/dl and a protein concentration of 360 mg/dl except for *Brucella melitensis* was isolated from CSF. No recovery was found in terms of bacteria, mycobacteria and fungi in cerebrospinal fluid. Blood samples yielded no growth.

After the diagnosis of *Brucellar meningitis*, brucellar brain abscesses and hydrocephalus, external ventricular drainage was performed, and ventriculoperitoneal shunt was performed 10 days after the insertion of external ventricular drainage. Treatment was started with injectable gentamycin (5 mg/kg/day, 7 days), oral trimethoprim–sulphamethoxazole (10 mg/kg/day, 4 months) and rifampicin (20 mg/kg/day, 4 months). Prednisone was given for 3 weeks. She became afebrile on the 10th day, and her irritability gradually disappeared. Repeated CSF examination revealed no leukocytes, glucose 75 mg/dl, and protein 120 mg/dl at 4 weeks after diagnosis. The CSF was sterile at this time. The blood brucella agglutination titer was 1:160 at two months after the diagnosis. The brain MRI at two months after the initial brain MRI revealed sustained brain abscesses. For this reason, long-term antibiotic treatment was planed. The patient represented increased generalized hypertony and poor head control at this time. Baclofen (0.5 mg/kg/day) was added treatment regimen. Physical therapy was initiated. A new ventriculoperitoneal shunt was inserted for shunt obstruction at six months. The patient's serum electrolytes, renal and hepatic functions remained within the normal range. She still receives combined antibiotic regimens mentioned above.

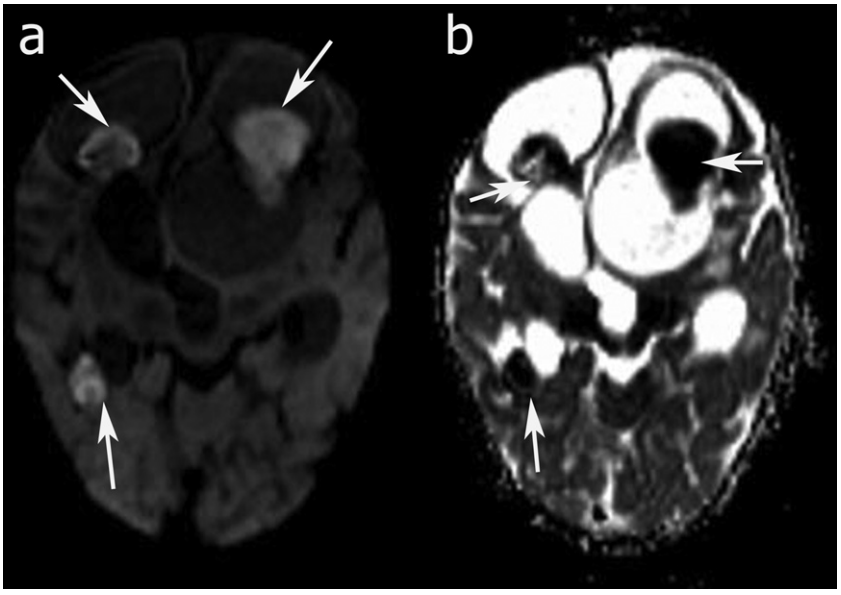

Fig. 2. Diffusion-weighted image (a) demonstrating hyperintense (restricted diffusion) lesions. ADC map (b) demonstrating a predominantly hypointense lesions.

3. Discussion

The principal clinical presentation of neurobrucellosis in children is displayed as acute meningitis or meningoencephalitis [1,2]. Usually, because *Brucellar meningitis* may not be included in the CSF assessment for differential diagnosis, diagnosis of the condition can be difficult. Neurobrucellosis is a severe form of the disease and it may rarely be complicated by the formation of multiple abscesses in brain parenchyma. There are several reports of brain abscess caused by *Brucella* spp. in children [3,4]. To our best knowledge, brucellar brain abscess has not previously been reported during infancy. Our case presented here is the youngest case with brucellar brain abscess concomitant with *Brucellar meningitis*. Brucellosis in humans is usually associated with the consumption of unpasteurized milk and soft cheeses made from the milk of infected animals, primarily goats, and with the occupational exposure of laboratory workers, veterinarians and slaughterhouse workers. Transmission from human to human, through sexual contact or from mother to child, is rare but possible. An infected mother may transmit the infection to the infant via breast milk [5].

In our case, it seems that the mother had brucella with clinical and laboratory findings and was likely bacteremic postnatally, at which point brucella was transmitted to the infant via her mother's breast milk. Because the patient was exclusively breastfed and was not exposed to raw animal milk.

The suspicion of neurobrucellosis should always be entertained in patients, living in or coming from endemic areas, who present with obscure, confusing and mixed constitutional or neurologic symptoms for which no obvious explanation is readily apparent. Our patient presented with fever, vomiting, altered sensorium and generalized tonic–clonic seizure with a bulging fontanelle. Analysis of the CSF revealed elevated protein, reduced glucose and lymphocyte pleocytosis. A definitive diagnosis was made of neurobrucellosis, as established by recovery of *Brucella melitensis* from CSF in our case. The radiologic imaging representing typical contrast enhancement and diffusion-weighted magnetic resonance imaging findings combined with positive serologic and bacteriologic evidence confirmed brucellar brain abscess.

There are few guidelines for the appropriate duration of neurobrucellosis treatment. Although the standard management of intracranial abcess is antibiotherapy combined with or without surgical drainage, we could not perform surgical drainage because of the multiplicity of abcesses in our case. Improvement with antibiotherapy as we have seen on serial imagings did not necessitate re-assessment for a surgical management. Cotrimoxazole and rifampicin can cross the blood–brain barrier and have synergistic effects. A complicated form of the disease, such as neurobrucellosis, should be treated for 4 months or longer duration [1,2]. The treatment period should be individualized according to clinical presentation and the normalization of CSF profiles. There are some neurobrucellosis patients who were given treatment as long as 17 months [1]. Long-term antibiotic treatment is required in our case with severe neurobrucellosis.

4. Conclusion

In summary, although neurobrucellosis is very rare in infants, it is considered in the differential diagnosis of a central nervous system infection, especially in an endemic region.

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Ethical approval

Not needed.

Conflict of interest

No conflict of interest.

Contributors

Calik M. proposed the study and wrote the first draft. Iscan A. helped in the writing. All authors contributed to the design and interpretation of the study. Calik M. is the guarantor.

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