A Case of Neurobrucellosis with Hydrocephaly

The Editor,

Sir,

A seven-year old girl presented with fever, vomiting and headache. Her speech was incoherent and she had generalized tonic-clonic convulsions twice in approximately five minutes. She was lethargic. Body temperature was 35.5 °C. Petechial lesions were observed in the periumbilical area. Nuchal rigidity and Brudzinski signs were positive. There were bilateral papilloedema at fundus examination. In the complete blood count, white blood cell was 11 200/mm³, haemoglobin 12 g/dL and platelet count 244 000/mm³. Hepatic and renal function tests were normal. Prothrombin time was 17.1 seconds and partial thromboplastin time 32.0 seconds. C-reactive protein was 3.19 mg/dL and erythrocyte sedimentation rate 20 mm/h. Protein was 129 mg/dL, glucose 22.5 mg/dL (simultaneously, blood glucose was 96 mg/dL) lymphocytes 30/mm³ and neutrophil count 10/mm³ at cerebrospinal fluid (CSF) examination. Phenytoin and ampicillin-sulbactam were administered. Wright agglutination test for brucellosis was positive 1/1280 titration in blood and CSF. Rifampicin, streptomycin and trimethoprim-sulfamethoxazole were started for neurobrucellosis treatment. Brucellosis was positive in CSF and blood cultures but negative in bone marrow culture. Hydrocephaly was diagnosed on cranial computed tomography. Dilatation in the third and lateral ventricles on magnetic resonance imaging, contrast agent in the basal cisternas and contrast in meningeal areas were observed. External shunt drainage was applied for hydrocephaly (Figure) and the hydrocephaly improved. The patient was discharged, with antibiotic therapy scheduled for another three months. No symptoms have been observed on outpatient follow-up.

Approximately 18 000 new brucellosis cases are diagnosed in Turkey annually (1). Acute or chronic meningitis, encephalitis, multifocal white matter disease, subdural abscesses, subdural empyema, intracranial hypertension, mycotic aneurysms, papilloedema, cranial neuropathies, psychosis, parkinsonism, radiculopathies and paravertebral abscesses have been reported among neurobrucellosis manifestations (2). The diagnosis of neurobrucellosis requires: clinical features of the illness compatible with a known neurobrucellosis syndrome, typical CSF changes (pleocytosis, elevated protein concentration), positive results of either blood or bone marrow or CSF culture or appropriate serological tests (eg agglutination test titres of > 1:160 in blood or any positive titre in CSF), clinical improvement after starting an appropriate treatment and the clinical findings not explained by another disease (3). Radiological imaging is non-specific in neurobrucellosis (4). The first case of neurobrucellosis associated with hydrocephalus in the literature was reported in a 38-year old farmer. His neurological examination revealed dysmetria, dysdiadochokinesia, ataxia and bilateral sensorineural hearing loss (5). The pathophysiological mechanisms of hydrocephalus in paediatric neurobrucellosis may be due to blocked CSF reuptake by arachnoid villi via granulomatous inflammation within the CSF (5).

Our patient’s family job is animal husbandry and farming. Her family has made cheese without boiling the milk. Lethargy, convulsions, senseless speeches, papilloedema and meningitis/meningoencephalitis was observed in the index patient. No cells were seen on CSF microscopy. Brucellosis agglutination was positive in CSF at 1/1280. Brucellosis is endemic to our region. A result is considered positive if p ≥ 1/160 in the Wright agglutination test. Brucellosis-associated hydrocephaly has rarely been reported in the literature. Neurobrucellosis should be considered when there are findings of meningeal irritation in endemic brucellosis infection areas such as Turkey.

C Yilmaz¹, A Kaya², AS Güven², N Yilmaz³, H Çaksen⁴

From: ¹Department of Paediatric Neurology, Yuzuncu Yil University, School of Medicine, Van, Turkey, ²Department of Paediatrics, Yuzuncu Yil University, School of Medicine, Van, Turkey, ³Department of Neurosurgery, Yuzuncu Yil University, School of Medicine, Van, Turkey and ⁴Department of Paediatric Neurology, Necmettin Erbakan University, Meram Medical Faculty, Meram, Konya, Turkey.

Correspondence: Dr A Kaya, Yuzuncu Yil University, School of Medicine, Department of Pediatrics, Van, Turkey. E-mail: avnikaya@gmail.com
REFERENCES