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SHORT REPORT

Brucella melitensis infection of ventriculo-peritoneal shunt: A form of neurobrucellosis manifested as gastrointestinal symptoms

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Summary A report of a 9 year-old child with Myelomeningocele who has a ventriculo-peritoneal (VP) shunt presented with gastrointestinal symptoms and peritonitis. The patient had no CNS symptoms but the cerebrospinal fluid was positive for *Brucella melitensis*. We discuss neurobrucellosis in children, its various presentations, complications and challenges in treatment, choice of antibiotics and duration.

Introduction

Brucellosis is an endemic zoonotic disease in Saudi Arabia. It is a disease with protean manifestation that involves almost any body system. Musculoskeletal manifestations in the form of arthralgia and/or arthritis are the most common. However, in endemic areas rare presentations like neurobrucellosis should be diagnosed in any patient with focal or nonspecific neurological manifestations. Rare presentations of neurobrucellosis like brain abscess, cranial nerve neuropathy, Guillian Barre syndrome, and ventriculo-peritoneal shunt infection do occur. Some patients with neurobrucellosis may go unnoticed as they present only non-specific symptoms like headache and depression. This report is about a 9 year-old child with myelomeningocele and ventriculo-peritoneal shunt presented with gastrointestinal but no neurological symptoms, found to have a positive cerebrospinal

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fluid for *Brucella Mellitensis*. Discussions on neurobrucellosis in children, its various presentations, complications, and challenges in treatment, including antibiotic choice and therapy duration are presented herein.

Case report

A 9 year-old child with Myelomeningocele who has a Ventriculo-Peritoneal (VP) shunt inserted at 6 months of age for hydrocephalus was presented with progressive abdominal distension, vomiting, and fever. He had no acute neurological manifestations. Computed tomography (CT) of the abdomen revealed distended bowel loops with no obstruction, multiple mesenteric lymph nodes associated with free fluid and peritoneal enhancement. The findings were suggestive of peritonitis with ileus. There were not enough peritoneal fluid to aspirate but aspiration of the VP shunt reservoir yielded yellowish cerebrospinal fluid (CSF) with white blood cell count 18/μL (88% lymphocytes and 9% polymorphs), protein 780 mg/dL, and glucose 45 mg/dL. The Gram stain was negative but CSF culture yielded growth of *B. melitensis* from two separate taps, 3 days apart. The *B. melitensis* titer in the CSF was only 1:20. Peripheral blood culture was negative in multiple occasions, and the peripheral *B. melitensis* titer was very high at 1:20,480. The patient did not have any neurological symptoms, the VP shunt was functioning, and CT of the brain was read as unchanged from previous one. The patient received intravenous doxycycline, rifampin, ciprofloxacin, and gentamicin for 2 weeks. He was discharged on oral doxycycline, Bactrim, and rifampin following CSF sterility and clinical improvement. Two weeks later he returned with altered mental status (GCS 13/15) and worsening of gastrointestinal symptoms with intestinal obstruction. Abdominal ultrasound revealed a large fluid collection. The VP shunt reservoir tap revealed increased WBC to 126 (84% lymphocyte and 8% polymorphs), sugar of 90 and protein 360 mg/dL with a CSF *B. melitensis* titer of 1:640. The shunt was externalized and external ventricular drainage established. Exploratory laparotomy revealed a peritoneal pseudocyst adherent to the small bowel and multiple small bowel adhesions. Peritoneal fluids culture was negative for *B. melitensis*. Adhesiolysis was done and a longer course of IV antibiotics given (4 weeks). Ventriculopleural shunt was inserted before discharge and the patient received oral doxycycline and rifampin for 10 months in addition to co-trimoxazole for the first 4 months. His condition had improved and *B. melitensis* titre normalized (CSF titer decreased to 1:40 and peripheral titer to 1:5120) after completion of therapy.

Discussion

Brucellosis is an endemic disease in Saudi Arabia. The Ministry of Health reported an annual incidence rate among Saudis at 18.42/100,000 population. However, studies from different regions showed an incidence approaching 138/100,000 population. Sekait reported brucella seroprevalence of 15% in Saudi Arabia [1]. This high incidence is attributed to rearing animals, especially camels, and consumption of their raw milk. It is believed that milk pasteurization remove a lot of its valuable constituents. Brucellosis has varied clinical presentations. Almost any organ system can be involved. The most common form of clinical manifestation is musculoskeletal, i.e., arthritis. Neurobrucellosis is not uncommon in adults but rare in children; 7% and <2% of cases respectively [2].

The clinical presentation of neurobrucellosis is variable with meningitis and meningoencephalitis being the most common. Other presentations of neurobrucellosis include polyradiculopathy, myelitis, hearing loss, visual disturbances, chronic vomiting, coma, and psychiatric disorders mainly depression. Rare CNS manifestations include: brain abscess, hydrocephalus, pseudotumor cerebri, Guillian Barre syndrome, cranial nerve palsy, cerebral venous sinus thromobosis, subdural and epidural collection, and stroke have been reported [3,4]. Therefore, clinical manifestations of neurobrucellosis can range from subtle manifestations like fever and malaise to fortright meningitis. CNS involvement is usually through hematogenous route. However, few case reports showed ascending infection through ventriculo-peritoneal shunt and vice versa where neurobrucellosis could give rise to peritonitis. Locutura et al., reported a male adult who presented signs of peritonitis and was found to have positive ascetic fluid for brucella. He was treated and 4 months later was presented with neurobrucellosis [5].

Anderson and Mortensen reported a 27-year old lady with signs of increased intracranial pressure and found to have hydrocephalus that required VP shunt insertion. Three months later she was admitted with abdominal distension and ascites. *B. melitensis* was isolated from CSF, ascetic fluid, and shunt system [6]. Alexiou et al., reported a 2-year old child who has VP shunt who showed signs of

meningitis and was found to have neurobrucellosis without peritonitis [7].

Another patient with simultaneous VP shunt showed related neurobrucellosis and peritonitis. Although his main complaint was peritonitis, his CSF from the shunt was positive. As it is known that neurobrucellosis may have a very subtle presentation and since this patient was mentally unstable, he might initially had neurobrucellosis that resulted in peritonitis. Primary brucella peritonitis could be the primary source of VP shunt colonization in this patient who showed gastorointestinal symptoms and signs. Peritonitis has been reported in patients with chronic liver disease and ascites, as well as, patients on peritoneal dialysis [8,9]. The presence of extra fluid in peritoneal cavity may allow the growth of brucella in this space.

Abdominal pseudocyst formation and small bowel obstruction complicating such infection is an even rarer occurrence. Diagnosis of neurobrucellosis depends on clinical presentation supported by CSF findings of pleocyotosis, raised protein, low or normal glucose, as well as, positive brucella titre. The patient showed mild pleocytosis with predominance of lymphocytes, which is compat with brucella meningitis. CSF brucella titre is almost always positive in neurobrucellsois. However, in rare occasion it may be negative. This can be explained by a low titre that cannot be detected by the current serology (SAT) test. In cases where SAT is negative and clinical and other laboratory tests are suggestive, use of SAT with Coombs test, ELISA test, and PCR are useful and may yield a positive result. CSF brucella culture is rarely positive occurring in 10—20% of affected patients. The patient had positive culture and raised CSF SAT titre.

Main treatment for shunt infection involves removal of the infected shunt, with establishment of external ventricular drainage (EVD), and antibiotics followed by the placement of a new shunt when CSF sterility is achieved. The patient responded initially to two weeks of intravenous antibiotic therapy without removal of the shunt but he relapsed after two weeks of oral therapy at home. Therefore, shunt was removed and EVD was inserted and he was placed on intravenous therapy for 6 weeks followed by oral therapy for 10 months. Successful antibiotic treatment of *Brucella* VP Shunt infection without shunt removal was proposed [10]. However, others reported neurobrucellosis to occur several months from antibiotic treatment of *Brucella* peritonitis with VP shunt left in situ [5]. For this reason and the complications described in the patient following a temporary improvement, shunt removal should be strongly considered. Intraventricular administration of antibiotics had been used for *Brucella* Shunt infection. The success of antibiotic treatment of neurobrucellosis requires use of a combination of three or four *brucella* effective antibiotics and prolonged duration of therapy. The usual antibiotic regimen should consist of three of the following: rifampicin, doxycycline, trimethoprim-sulfamehtoxazole, ciprofloxacin, in addition to use of either intravenous ceftriaxone or gentamicin, implemented for the first two weeks. Duration of antibiotic therapy was not established, but the consensus is to treat for 4 to 6 months. Full recovery is usually expected with mortality rate less than 1% and morbidity around 20%. The patient completely recovered.

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Competing interests

None declared.

Ethical approval

Not required.

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