CASE REPORT

MYALGIA CRURIS EPIDEMICA: AN UNUSUAL PRESENTATION OF DENGUE FEVER

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Abstract. We describe a 5-year-old girl who had sudden onset difficulty in walking after 3 days of febrile illness. In the emergency department her creatine kinase level was elevated but urine myoglobin was normal. She was diagnosed as having benign acute childhood myositis. Because of poor oral intake and dehydration, she was admitted to the pediatric ward. The next day she had a petechial rash over the antecubital fossa, and dengue IgM back was positive. She was treated conservatively and recovered uneventfully. Despite dengue fever being endemic in Malaysia, this is the first case report of myositis following dengue infection in Malaysia.

INTRODUCTION

In 1957, Lundberg published the first report on "Myalgia Cruris Epidemica" (Hussin *et al*, 2003). This syndrome of muscle pain is characterized by sudden onset calf pain after a period of rest and refusal to walk following viral illness (Rennie *et al*, 2005). It has been suggested that calf pain causes the weakness instead of true inability to generate power. The majority of children have an elevated creatine kinase (CK). Leukopenia, thrombocytopenia and elevated serum glutamic oxaloacetic transaminase (SGOT) are less consistent laboratory findings. Neurological examination has been reported as normal (Rennie *et al,* 2005).

"Myalgia Cruris Epidemica" predominantly affects school age children and typically occurs in boys (Lundberg, 1957). Myositis and rhabdomyolysis associated with viral infection is a well-described entity. This syndrome is also known as benign acute childhood myositis (BACM) and is self limited. Normally patients with BACM will be discharged with medications and proper instructions. However, "Myalgia Cruris Epidemica" is a rare manifestation of dengue infection, of which hemorrhagic shock is one of the life threatening complications. We report a preschool girl who presented to the emergency department with muscle pain and difficulty in walking who was initially diagnosed as having benign acute childhood myositis, but later she was confirmed to having dengue infection.

CASE REPORT

A 5-year-old girl was brought to the emergency department because of sudden onset difficulty in walking. She was apparently well until 3 days previoulsy when she developed a high-grade fever, but had no chills or rigors. The fever decreased temporarily after taking paracetamol syrup. At the same time she had lethargy and loss of appetite. She denied any respiratory tract symptoms, headache, retro-

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orbital pain or joint pain. On the morning prior to admission, she developed bilateral calf pain which was dull and aching in nature. The pain was aggravated by movement and relieved by rest. There was no swelling or discoloration of the calf muscles. Late afternoon, after she woke up from her nap, she developed progressive muscle weakness of the lower extremities associated with difficulty in walking. She denied difficulty in breathing, double vision, vomiting, diarrhea, or urinary symptoms. Her family and medical history were unremarkable.

On presentation, she appeared weak, but alert. Her pulse was 90 beats/minute and regular rhythm, her blood pressure was 90/60 mmHg, her respiratory rate was 26 breaths/minute, and she was febrile (38.5ºC). Her hydration status was normal. She had no rashes, pallor or jaundice. Cardiopulmonary and neurological examinations were unremarkable. Both her calves were warm and tender on active stretching. There was no discoloration or swelling of the calf muscles, fasciculations, myoclonus, or muscular atrophy observed. Examination of gait revealed an unsteady, broad-based gait. The remainder of the physical examination was normal. The presentation of fever with inflammation of the calf muscles made acute viral myositis the most likely diagnosis.

Her electrocardiogram showed no abnormalities. Her complete blood count showed white blood cells of 3,100/mm^3 and platelets of 92,000/mm^3. Her CK was markedly elevated (1,400 IU/l), however, urine for myoglobinuria was negative. Serum biochemistries were normal. These findings are consistent with benign acute childhood myositis. She was admitted to the pediatric ward for further evaluation and treatment.

In the ward, she was treated conservatively with intravenous fluid and paracetamol 250 mg every 4 hours. The next day the managing team noted a petechial rash over the right antecubital fossa. A Pan Bio Rapid test (immunochromatography method) for dengue was sent. Her dengue serology for IgM was positive while the IgG was negative. During hospitalization the fever subsided, her platelet count increased and she began to walk again. She was discharged on the 4th day of hospitalization after two sets platelet counts were more than 100,000/mm^3 and the creatine kinase level was normal (45 IU/l).

DISCUSSION

Dengue is the most common arthropod borne arboviral infection in the world today. In Malaysia, the disease is endemically transmitted by both *Aedes aegypti* and *Aedes albopictus* (Wallace *et al*, 1980; Rebecca, 1992).

It is estimated that there are at least 100 million cases of dengue fever (DF) annually, and 500,000 cases of dengue hemorrhagic fever (DHF) require hospitalization. In Malaysia, it has become a major public health problem (Hussin *et al*, 2003).

The clinical features of dengue fever vary according to age of the patient. Infants and young children usually have a febrile illness with a maculopapular rash and older children have a mild febrile illness or the classic incapacitating disease, with abrupt onset, high fever, severe headache, pain behind the eyes, muscle pains, joint pains and rash (WHO, 2002). Some authors have reported atypical presentations of dengue virus infection, such as mononeuropathies, encephalitis, cardiomyopathy or rhabdomyolysis (Solomon *et al*, 2000; Gibbon and Vaughn, 2002).

According to the WHO (Mackay *et al*, 1999), classic dengue fever is considered as a diagnosis if the patient presents with an acute febrile illness and two or more of the following: headaches, retro-orbital pain, myalgia, athralgia, rash, hemorrhagic manifestations and leukopenia. Those patients need to

be notified to the local health authorities even before serology is known for preventive measures.

In this case, our patient presented with a typical viral syndrome but the absence of other features of dengue fever. Therefore, the initial diagnosis made for her illness was benign acute childhood myositis secondary to viral infection.

Many viruses may cause a single episode of myositis and are unlikely to be recurrent. The most commonly reported infection is influenza B virus. Other viruses include influenza A, parainfluenza viruses 2 and 3, echoviruses 6 and 9, coxsakievirus, rotavirus, measles, mumps, rubella, adenoviruses and viruses of the herpes group (Rebecca, 1992; Rennie *et al,* 2005).

It is important for emergency department physicians to distinguish benign acute childhood myositis from other more severe illnesses. If patients present with classical signs of BACM, the physicians should feel comfortable discharging them with medications but have them follow up. However, emergency department physicians need to look for features not associated with BACM, because some potentially life threatening diseases may mimic BACM, such as dengue fever and Guillain Barre Syndrome. A thorough clinical history and physical examination, especially neurological examination, and blood investigations, such as full blood count, and biochemistry investigations, particularly CK, urine myoglobin, and viral serology, should be performed in order to rule out the above matter. Failing to differentiate between BACM and possible life threatening events may lead to inappropriate management and jeopardize the patient's life.

A series of patients with BACM (Rajajee *et al*, 2005) was reported for Kanchi Kamakoti CHILDS Trust Hospital, Chennai, India. Serological tests were positive for primary dengue infection (Elisa PAN BIO) in 8 (25%) and secondary infection in 12 (30%). According to few study, there were two possible mechanisms for this: direct invasion of the muscle fibers and release of myotoxic cytokines by the dengue virus, or myotoxic cytokines, particularly tumor necrosis factor (TNF), injured the affected muscle. (Greco *et al*, 1977; Pratt *et al*, 1995; Seibold *et al*, 1998; Gagnon *et al*, 2002). However, we could not explain why it mainly affects the calf muscles.

Our patient developed petechial rashes on the 5th day of the febrile episode, which turned out to be dengue virus infection. Being a country that is endemic for dengue infection, we recommend reviewing classic BACM patients in the outpatient department, following up for at least a week. If dengue fever can be ruled out, it is advisable to review the patient after a couple of weeks to ensure complete resolution of symptoms.

We found little local medical literature regarding this unusual presentation of dengue infection. In a study by Horvath *et al* (1999), they reviewed 100 hospitalized dengue patients in North Queensland from 1997 to 1999. They found musculoskeletal pain, headache, gastrointestinal symptoms, and nausea were common, 99, 95, 95 and 92%, respectively. Creatine kinase levels were measured in 15 patients; 6 patients had elevated plasma creatine kinase levels (NR <200 U/l). They concluded that the high prevalence of elevated CK levels in the small group tested suggests that myositis was common in this outbreak of dengue fever. This is no great surprise since musculo-skeletal symptoms are common. Therefore, we postulate that myalgia cruris epidemica in dengue infection is under-reported. In highlighting this condition, we hope that more CK levels, urine myoglobin and viral studies will be performed in future cases.

In conclusion, by reporting this case we hope to raise awareness of unusual presentations of dengue (myalgia cruris epidemica)

among healthcare providers as the incidence of dengue infection increases. Emergency department physicians need to differentiate myalgia cruris epidemica of self limiting classical BACM from myalgia cruris epidemica of dengue and Guillain Barre Syndrome, as the latter is a potentially life threatening condition.

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