Original Article

Benign acute childhood myositis in the eastern region of Kingdom of Saudi Arabia; a 5-year experience

Mohammad H. Al-Qahtani, MD*, Abdulelah M. Salih, MD and Abdullah A. Yousef, MD

Department of Pediatrics, King Fahd Hospital of the University, University of Dammam, Al-Khobar, KSA

Received 14 June 2014; revised 8 December 2014; accepted 9 December 2014; Available online 16 January 2015

Abstract

Objective: To assess the epidemiological, clinical and laboratory characteristics, and outcomes of BACM in children diagnosed at a teaching hospital in the eastern province of the KSA.

Methods: A retrospective study of 28 patients admitted to the hospital with the diagnosis of BACM was carried out at King Fahd Hospital of the University, AlKhorab, Kingdom of Saudi Arabia (KSA), from January 2008 to December 2012. Demographics, clinical characteristics, laboratory findings and patients' responses were analyzed.

Results: Thirty episodes were reported over 5 years with incidence of 3.17 per 100,000. The male to female ratio was 6:1. The mean age was 6 years. A seasonal peak during the winter months was observed. The major symptoms were fever, inability to walk in all the patients (100%) and symptoms of upper respiratory tract infections found in 70%. Major clinical findings were the normal deep tendon reflex and calf muscle tenderness in 76.6% and 100% respectively. The prominent laboratory findings were the high creatine phosphokinase (CPK) and leukopenia in 100% and 60%, respectively.

Conclusion: Our findings showed that the clinical and epidemiological features associated with BACM, in the KSA, have the similar pattern of other studies from different regions in the world. A nationwide survey,
BACM is a well-recognized disease first described by Lundberg in 1957 when he reported cases with what is called myalgia cruris epidemica.\(^1\)

It is characterized by severe calf pain and tenderness and inability to walk, preceded by symptoms of upper respiratory tract infection. It mainly affects school age boys and associated with viral agent like influenza virus\(^2\),\(^3\) with high prevalence in winter and spring.\(^4\) Also, there are reported cases associated with mycoplasma and dengue virus.\(^5\),\(^6\)

Serum creatine phosphokinase (CPK) is elevated in the majority of the affected patients.\(^7\) Also there is a decrease in total white blood cells and an increase in the liver enzymes.\(^8\)

Since the inability to walk is the major presenting feature and the most concerning to parents, and since it is a presentation with a list of differential diagnosis including serious diagnoses like Guillain Barre syndrome and acute flaccid paralysis caused by poliomyelitis\(^7\); the pediatricians take this issue seriously and try to exclude the potentially serious or potentially infectious causes at the beginning through appropriate clinical and laboratory assessment tools.

Almost all the cases will resolve spontaneously within days of the onset of the disease including the gradual improvement of the walking ability concurrently with gradual improvement in the laboratory findings.

Although there are many reported cases in the literature from elsewhere, however; there are very few reports about this entity from the Middle East. There are no reported cases or description of the pattern of this disease in Saudi children population.

The aim of this study is to evaluate the clinical and laboratory findings associated with this entity retrospectively in the pediatric age group who were admitted to King Fahd Hospital of the University, eastern province of the Saudi Arabia.

Materials and Methods

The medical records of children admitted to pediatric ward with diagnosis of BACM at King Fahd Hospital of the University; Al- Khobar, Kingdom of Saudi Arabia between January 2008 and December 2012 were reviewed retrospectively. The following variables were retrieved from their files: age, sex, prodromal symptoms, family history of muscle diseases, change in urine color and any physical or neurological abnormalities. Laboratory data like serum creatine kinase (CPK), urine for myoglobin, white blood cell counts (WBCs), Platelet counts, C-reactive protein (CRP), serum aspartate aminotransferase (AST), alanine aminotransferase (ALT), lactate dehydrogenase (LDH), blood urea nitrogen (BUN) and creatinine levels were recorded from the health computer system of the hospital.

The inclusion criteria were as follows: acute onset of symptoms, presence of calf muscle pain or tenderness, gait disturbance and increased serum CPK level. The exclusion criteria were as follows: family history of muscle diseases, presence of neurological abnormalities in the clinical examination and presence of concurrent immune-complex disease.

Statistical analysis: Frequency tables were performed to get the descriptive statistics for the categorical variables, while the continuous data means and percentages were calculated as descriptive values.

Data analysis was done using SPSS version 20.0 for windows (SPSS, Chicago IL, USA).

Results

Total of 29 patients were admitted to the pediatric ward during this period. One patient had been excluded from the study because he was diagnosed with metabolic disorder. Three of the patients had recurrent episodes and there were 2 pairs of siblings affected at same time.

Half of episodes presented during the winter months of December, January and February (n: 15; 50%), while the rest of episodes presented in clusters; Figure 1.

The age of patients ranged from 2 to 11 years with median age of 6 years, 24 of them were boys (85.7%) and 4 were girls (14.3%).

The prodromal symptoms included: fever (100%), cough (70%) and sore throat (20%). Prodromal illness of 2–10 days preceded the onset of pain with median duration of 7 days; Table 1.

All the patients had normal deep tendon reflexes, and the clinical recovery was complete in all patients within one week from the initial presentation.

Laboratory findings showed increased CPK in all the episodes ranging from 297 to 36,852 U/L (normal 75–230 U/L), in the other hand leukopenia was found in 18 episodes (60%) and thrombocytopenia was presented in 4 episodes (13.3%). AST was high in 13 episodes (43.3%) while ALT was high in 6 episodes (20%) and both tests were not done in 15 episodes. CRP was positive in 7 episodes (23.3%) and was not done in 8 episodes, while the lactate dehydrogenase was normal in 16 episodes and was not done in 14 episodes, however; BUN and creatinine were normal in all patients; Table 2.

Urine myoglobin was negative in 23 episodes and was not done in other 7 episodes. No virology study was done for any of the patients.
Benign acute childhood myositis (BACM) is self-limited childhood illness typically affected boys, as shown in this study, 85.7% of the patients were boys which is consistent with other study, the cause of male predominance is unexplained but it could be related to their greater activities or may be caused by genetic predisposition.

There is no Saudi studies to compare our finding with, and there is no established incidence or prevalence of BACM to date in the literature, we found the estimated prevalence of the disease in this study is around 3.17 per 100,000 of pediatric patients less than 12 years of age, however; Buss et al. reported in a retrospective study in Nebraska that influenza-associated myositis incidence estimates per 100,000 of the population less than 18 years old were 2.693 and 0.225 during 2006–2007 and 2003–2004, respectively.

Involvement of sibling and recurrence as shown in our study can raise the possibility of metabolic muscular defects in genetic susceptible patients as hypothesized in another study, although the pathogenesis is unclear yet it is most commonly associated with viral etiology like influenza A and B or other virus, furthermore; Mall S et al. had reported a large outbreak of influenza B-associated benign acute childhood myositis in Germany of 219 cases in winter season of 2007-2008, in addition to that there are reported cases associated with Mycoplasma pneumonia, as well as one case reported with H1N1 which might be related to the winter months season where the clustering of the these viral infections is common, which was also found in this study, most cases we reported were in the period of October to February and another lower peak in the period of May–August, Rajajee S et al. reported 40 cases within 5 months of a year from October to February in India, however; this is in contrary to countries of northern latitudes where the peak occurs in March and April. Mackay MT et al. had collected 38 patients over 20 years in Australia and in a non-published study in a local hospital in the eastern province of the Kingdom of Saudi Arabia they reported 13 cases over 13 years. There was no viral study done for our patients, although it is considered in an ongoing prospective comprehensive study in our hospital.

The reported mean age of BACM is 9.2 years, in our study the mean age was 6 and the range of age of patients was 2–11 years and this is similar to the published reports in the medical literature.

Half of episodes presented during the winter months 50% as clusters and this support the viral etiology hypothesis which is similar to other reported cases, the prodromal period ranged 2–10 days was preceding the pain onset and this is almost similar to previous studies, those prodromal
symptoms were fever (100%), cough (70%) and sore throat (20%) and this percentage is similar to the published studies, although one study only their patients presented with fever and cough in 40.6% and 18.8% respectively.

The clinical presentation was similar in all cases; all patients (100%) presented with calf pain while calf tenderness presented in twenty three episodes (76.7%) and inability to walk in twenty eight episodes (93.3%), these findings are similar to many published studies for all our patients presented with normal neurological examination and got normal reactive deep tendon reflexes which is considered to be an important differentiating feature of BACM from the other serious differential diagnosis like Guillian Barre syndrome, only two studies reported decreased deep tendon reflexes in their patients.

From the laboratory point of view, all our patients (100%) presented with increased in CPK level ranging from 297 to 36,852 U/L (normal 75–230 U/L) on admission indicating the peak activity of the inflammatory process and this result is consistent with all other studies, leukopenia specifically is one of the main laboratory features of BACM, we got 18 patients (58%) presented with leukopenia compared to one study in which they got 27%, thrombocytopenia in the other hand we found it in 4 patients (13%), only one study reported platelet count as normal in their patients, furthermore; our study showed elevated liver enzymes mainly AST in (43.3%) and ALT in (20%) respectively, this result is consistent with the findings of the other reported studies, we did not get an elevation on LDH level in our study patients, finally C-reactive protein (CRP), was positive in 7 episodes (23.3%), this finding is reporting differently from the other studies which founded it to be normal.

Conclusion

The findings of this study have estimated the incidence of benign acute childhood myositis (BACM) in eastern province of the KSA and confirm the clinical and biochemical similarities with various studies from different parts of the world.

Clinical presentation and characteristic laboratory findings of BACM will differentiate it from other serious diagnoses presenting with acute inability to walk in children.

Recommendation

A prospective study at the national level is necessary to determine the accurate incidence and to isolate the actual causative organisms of this disease in the Kingdom of Saudi Arabia.

Authors contributions

All the authors contributed in phrasing the question of the study, designing the study methods. Also they contributing in writing different sections of the article distributed by the corresponding author. At the end each author has reviewed the manuscript and put his own comments and suggestions to improve the writing of the manuscript.

Conflict of interest

The author has no conflict of interests, and the work was not supported or funded by any drug company.

References