Original Research Article

A prospective study to describe the clinical and laboratory features of benign acute childhood myositis

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Abstract

Background: Benign acute childhood myositis (BACM) is a rare transient clinical condition that mainly affects preschool and school age children, preceded by a viral illness, with an excellent prognosis and no functional sequelae. **Objective:** To describe the clinical and laboratory features of BACM. **Study design**: Retrospective cross sectional study. **Study setting**: Department of Pediatrics, Gadag Institute of Medical Sciences. **Methodology:** Nineteen children of benign acute childhood myositis were seen during September 2017- August 2018. **Inclusion criteria:** Children presenting with preceding history of fever, cough, cold, headache/myalgia followed by acute onset of pain in both legs and difficulty in walking and unable to bear weight. **Exclusion criteria:** Children with neuromuscular disorders, children with altered sensorium, recent intramuscular injection, history of vigorous exercise and history of trauma. **Results**: Out of nineteen children, the mean age of presentation was 7.5 years, predominantly affecting males 68%(n=13) with male: female ratio of 2.1:1.Leucopenia was seen in 32%(n=6) with neutropenia in 26%(n=5) with reactive lymphocytes in all patients(100%). Creatinine phosphokinase was elevated in all children with mean of 1892U/L. Dengue serology was negative in all patients. Mean duration of time of clinical recovery from the onset of pain was 3.32days(3.32+0.48 days) with mean duration of hospital stay of 4.5days.There was no significant association noted between CPK levels and duration of clinical recovery(p=0.087). The outcome of therapy was mainly supportive with complete clinicalrecovery. **Conclusion:** BACM is a benign entity with a characteristic clinical presentation that can be managed with supportive treatment, avoiding unnecessary diagnostic investigations.
Key Word: Myositis; Benign; Creatinine phosphokinase; Dengue serology.

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INTRODUCTION

Benign acute childhood myositis (BACM) is a transient and rare inflammatory condition. Benign acute childhood myositis (BACM) is a well-recognized disease, first described in 1957 by Lundberg .[1]It occurs mainly in school and pre-school aged children, predominantly affecting males and occurs during the acute convalescent phase of viral illnesses, more prevalent in the late winter and early spring .is characterized by calf tenderness and the sudden onset of difficulty in walking after a viral illness, sometimes associated with influenza epidemics in the wintertime. BACM has also been seen in the adolescents age group .[2]This can be substantiated by the explaination that the increased tropism of viruses for immature muscle cells.[3]Further every virus can act as a trigger in genetically predisposed children and in few patients with undiagnosed metabolic diseases.[3,4] Benign acute childhood myositis does not require any invasive tests or medical therapy. Nevertheless, the onset may be mistaken for very severe neurological illness such as Guillain–Barrè syndrome or chronic autoimmune diseases. As a consequence unnecessary invasive tests,

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such as radiography, echocardiography, electromyography and magnetic resonance are still performed.[2,3,5] The daily medical examination by the pediatrician followed by urine examination for the presence of myoglobin can be used a tool for detecting complications and the progress of the disease . The measurement of Creatinine Kinase is usefull in the final diagnosis of the condition and it also helps out in ruling out degenerative disease. Once Symptoms have resolved the patient does not require any follow up and should be educated to monitor the color of urine and the presence of swollen legs as a indictor for immediate medical care. BACM typically manifests with acute myositis and increased CK level, following a viral infection. A family history of neuromuscular disorders, myoglobinuria, trauma, chronic progression, rash, edema, muscle weakness or neurological diseases are not typically associated to BACM; in these cases further investigation is required. This study focus to characterize BACMs clinical picture, laboratory features to provide greater awareness to its benign course and helping to differentiate it from more serious disorders

OBJECTIVE

To describe the clinical and laboratory features of BACM

MATERIALS AND METHODS

A cross sectional study was conducted by the Department of Pediatrics, GadagInstittue of Medical Sciences from September 2017 to August 2018. A total of Nineteen children of benign acute childhood myositis (BACM) were seen the study period.

Inclusion criteria: Children presenting with preceding history of fever, cough, cold, headache/myalgia followed by acute onset of pain in both legs and difficulty in walking and unable to bear weight.

Exclusion criteria: Children with neuromuscular disorders, children with altered sensorium, recent intramuscular injection, history of vigorous exercise and history of trauma.

Physical examination showed calf tenderness of both legs on palpation and on dorsiflexion, with no swelling or erythema None of the children had rashes. Complete neurological examination was done in all cases. All children had normal muscle power and tone with preserved tendon reflexes. Laboratory investigations were sent including complete blood count(CBC), differential count, peripheral blood smear(PS), creatinine phosphokinase (CPK), C-reactive protein(CRP), Erythrocyte sedimentation rate(ESR), blood urea, serum creatinine, urine for myoglobin, protein and red blood cells, aspartate transferase (AST), alanine transferase (ALT) and Dengue serology. Fourteen children were admitted and treated with adequate bed rest, hydration and symptomatic treatment. Five children were treated on outpatient basis with close follow up. Complete clinical recovery was seen in all children.

RESULTS

From September 2016-January 2017, 19 patients with BACM were seen with maximum number of case seen during September (47%). [**Table1].** The mean age of presentation was 7.5years (3.8-12years), predominantly affecting males 68% (n=13) with male : female ratio of 2.1:1**[Table 2]**Preceding symptoms were fever (100%), followed by cold/ rhinorrhoea (66%), cough (49%) and headache (32%). Children presented with acute onset of pain in both the legs (100%) with unable to bear weight (74%) and difficulty in walking (26%).Onset of pain in relation to fever was 3.52days. On physical examination, muscle tenderness on palpation and stretching was noted in all children (100%). Provisional diagnosis of viral myositis was made and laboratory investigations were sent. Leucopenia was seen in 32% (n=6), with neutropenia in 26% (n=5) and thrombocytopenia in 32%(n=6) children. CRP was raised in 15% (n=3) and ESR in 37% (n=7) patients. CPK was elevated in all children with a mean of 1892U/L (249-9080U/L). All children had raised AST (100%) with a less increased ALT in 47% (n=9). To rule out rhabdomyolysis urine dipstick and urinalysis was done in all children which was normal. All children also had normal blood urea, serum creatinine and serum electrolytes. Dengue serology was negative in all children. However, due to non-availability of specific probes, virus characterization could not be done. Fourteen children (74%) were admitted in view of functional impairment and to monitor for complications. Higher CPK levels were not associated with acute renal failure, progression to rhabdomyolysis or other complications. Daily urine dipstick test was done along with CPK values. Complete clinical recovery was seen in all children with symptomatic treatment. Mean duration of hospitalization was 4.5days. Children treated on outpatient basis(26%) were also closely followed up till complete clinical recovery with an advice to review if noted to have dark colored(red to brown) urine. Follow up CPK values were also done. Fischer's exact test showed no significant association between CPK levels and duration of clinical recovery (p=0.087). All children recovered with a mean duration of 3.32 days and CPK values were normalized in all children with a mean duration of 4.94 days. (table 3)

Table 1: Distribution of Case based on the month

Month	No of Cases	Percentage
September	9	47.4
October	1	5.2
November	2	10.5
December	4	21.1
January	3	15.8
Total	19	100

Table 2: Gender wise distribution of Cases

Gender	No of Cases	Percentage
Male	13	68.4
Female	6	31.6

Table 3: Creatinine phosphokinase levels and clinical recovery

Case no.	Age (Years)	Sex (Male/ Female)	CPK (U/L) Day0/Admission (normal range 20-200U/l)	Duration for normal CPK levels (days)	Duration for clinical recovery (days)
1	7	M	1118	5	3
2	9	M	3537	4	3
3	7	M	1849	4	3
4	5	M	1274	4	3
5	7	M	305	4	3
6	10	M	980	5	3
7	3.8	F	249	5	4
8	5	M	337	5	4
9	12	F	1478	4	3
10	10	M	5426	7	4
11	9	F	4492	4	3
12	12	M	9080	7	4
13	5.6	F	514	5	3
14	4.8	M	2588	4	3
15	6	M	2265	7	3
16	7.6	F	1128	5	4
17	4.9	M	899	5	4
18	5.8	M	1465	5	3
19	11	F	969	5	3

DISCUSSION

BACM is an acute self limiting condition frequently affecting the gastrocnemius and soleus muscles. This was first described in a series of case reports by Lundberg in 1957, who studied 74 patients in Sweden with severe calf myalgia as “myalgia crurisepidemica” by following an upper respiratory infection [1]. Middleton *et al*.[6] in 1970 were the first to provide laboratory evidence in support of an association with influenza virus infection and showed elevated creatinine phosphokinase. Association with other viruses such as coxsackie, adenovirus, parainfluenza, respiratory syncytial virus, among others, and bacteria such as Mycoplasma pneumoniae has also been described .[7,8] In our case series we noted that all children belong to school age, predominantly affecting boys.[9,10] All children in our study presented with classic clinical picture, with predominance of bilateral calf pain and refusal to walk and to bear weight and with a febrile respiratory prodrome. The age of presentation also coincided with that described in the literature.[4,9,10] The symptoms of BACM are alarming and can cause concern and confusion both in parents and health professionals and these children are prone to extensive investigations because of their clinical presentation. Children present to their doctors because of refusal to bear weight [11]. Differential diagnosis for spectrum of diseases that present with claudication and/or muscle pain include infectious, muscular and neurological diseases such as acute myositis associated with other infectious diseases (eg dengue), toxic myoglobinuria, rhabdomyolysis, Guillain- Barre syndrome, transverse myelitis, muscular dystrophies, polymyositis, juvenile dermatomyositis, trichinosis , osteomyelitis, arthritis and deep vein thrombosis, among others [12,13]. Therefore these children

may be subjected to repeated blood taking, lumbar puncture, sedation risk of neuroimaging, electromyography and nerve conduction tests [11,13]. The author groups from Greece [11] and Switzerland [13] noted that with careful history, physical examination and neurological examination, a clinical diagnosis of BACM could be made and the investigations aforementioned may not be necessary. Clinical observation with follow up is recommended until the functional status of the patient returned to normal. The most striking laboratory result is the marked elevation of the CPK muscle enzyme (20-30 times higher than normal values) which typically normalizes within a week to couple of weeks [7,8.]Even in cases where CPK has been massively elevated it is seldom associated with myoglobinuria and significant rhabdomyolysis [14]. A bedside urine dipstick test may be done to exclude myoglobinuria. As mentioned in our study, CPK values had no implication on the severity, duration of clinical recovery (p=0.087) which was similar to study done by Sham *et al.*[15] Other findings are leucocytopenia with moderate neutropenia, thrombocytopenia and a mild and transient elevation of transaminases [16,17] . Inflammatory markers are generally normal, although ESR and CRP may be slightly increased. This coincides with the laboratory parameters observed in our patient group. Dengue virus was reported to be associated with 50% of the cases reported in a case series by Sarala Rajajee *et al.*[17] but none of our cases were found associated with this virus. Management of BACM is symptomatic .The use of antivirals in the case of viral infection is of little benefit, since in most cases the acute respiratory infection is already in resolution [12]. The process is self-limited with complete recovery between the third and tenth days, complications are infrequent and do not leave functional sequelae which was similarly observed in our study.[16,17]

CONCLUSION

By presenting this clinical series, we fulfill the objective of emphasizing that BACM is a benign, self-limited entity with an excellent prognosis, with a characteristic clinicalpresentation and that in most cases it can be managed on an outpatient basis and avoid invasive studies and unnecessary hospitalizations.

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