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Benign Acute Childhood Myositis - A Great Mimicker

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Abstract: ***Benign acute childhood myositis is a transient, self-limiting inflammation of muscles, particularly gastrocnemius and soleus following a viral respiratory infection in children; as the name suggests its benign in nature; symptoms usually resolve within 5-7 days. There is characteristically bilateral calf muscle pain which leads to gait abnormalities in the children with near normal neurological examination. Occasionally it can lead to serious complications like rhabdomyolysis and renal failure; hence crucial monitoring of the affected children is mandatory. Here we are presenting two such cases managed successfully in our centre.***

Keywords: myositis, influenza, post-viral, CPK, rhabdomyolysis, children

1. Introduction

BACM is a rare transient self-limiting condition that occurs in children during the convalescent phase of a viral respiratory illness(1). This is characterized by acute onset of calf pain that leads to difficulty in walking. We report two such cases of BACM – 7-year-old male child and 5-year-old female child with acute onset severe bilateral calf pain following viral pharyngitis. Adequate supportive management helped both the patients recover completely in about 5 days.

BACM predominantly affects school-aged children, with male predominance and characteristically follows a viral infection. It is characterized by acute onset muscle pain, with the kids refusing to walk. Serum CPK (Creatinine Phospho Kinase) levels are typically elevated in them. This pseudo-paralysis can be confusing to the clinician who may go for extensive workup(2).

This illness lasts for a short period, usually one week. This kind of illness has to be promptly detected to avoid serious complications(3). Children with BACM do not usually require hospitalization, aggressive medical intervention or long-term follow-up.

2. Case Report

In this case report, we present two cases of BACM, one in a seven-year-old male child and other in a five-year-old female child in a span of one month, admitted in our hospital and successfully treated with conservative measures.

Case 1:
7-year-old boy with high grade fever for 4 days, presented with dehydration to our paediatric ER. He has had calf muscle pain since that day morning. Family history and personal history were not suggestive of neuromuscular disorders. The child was vaccinated up to the age of 5 years as per the national immunization schedule followed in India. Previous history was not correlating with any remarkable illness. There was no history of trauma or vigorous exercise reported. There were no similar such episodes in the past.

On admission, the child was alert, pyretic, his vitals were stable. No evidence of trauma to the spine and lower limbs. Neurological examination revealed normal muscle power and tone. Tendon reflexes and sensations were normal. The gait was of tip-toe walking. Other neurological findings were normal and suggestive of non-neurological illness. Calf muscles on both sides were soft on palpation and there was no evidence of inflammatory changes. There were complaints of sharp pain in calf muscles on passive stretching at the knees and ankles as well. This sharp pain was present also on gentle palpation of the calves.

Test	D1	D2	Follow up
TC	2600/mm^3	3700/mm^3	4800/mm^3
Platelet	1,34,000/mm^3	1,35,000/mm^3	1,67,000/mm^3
BUN	15.18 mg/dl	12.99 mg/dl	-
Serum creatinine	0.58 mg/dl	0.61 mg/dl	-
CRP	0.3 mg/dl (negative)	-	-
Serum CPK	5100 IU/L (Normal 0-197 IU/L)	-	120 IU/L
Serum AST(SGOT)	144.5 IU/L	-	36 IU/L
Serum ALT(SGPT)	56.9 IU/L	-	14.2 IU/L
Urine analysis	Normal	Normal	-

Urine analysis was normal, it was absent for myoglobulin, blood and protein. Dengue and Leptospira PCR were negative. Throat swab was negative for influenza A & B. Since his oral intake was low, he was maintained on intravenous fluids after correction of dehydration. Intravenous paracetamol was given at the dose of 10mg/kg/day thrice daily to alleviate pain and fever. His urine output was well maintained throughout the hospital stay. On the 4th day of hospitalization, his calf pain started resolving and a normal gait was regained.

The boy was discharged on ninth day of illness with an advice to review after a week to recheck his clinical findings and serum CPK levels, which turned out to be in normal range. On follow-up, the child didn't have any neurological complications.

Case 2:
A five-year-old girl presented to us on 5th day of illness with history of refusal to walk since morning. The child had fever and throat pain for the past 4 days with pain in both calves and limping on walking for the past several hours. This is the first time, the child is having such complaints and as the previous case, there was no family history or personal history suggestive of neuromuscular disorder.

Neurological examination of the child was completely normal with calf muscles tender to touch. Other system examination was normal. Blood investigations ruled out severe infections. Urine analysis was normal.

She was treated with oral ibuprofen for pain and intravenous crystalloids for hydration. Since her calf muscle tenderness worsened the next day, she was re-examined; her neurological examination was completely normal other than tip toe walking. Her serum CPK levels showed a decline and there were negative reports in dengue and leptospirosis. Nasopharyngeal swab was not done for viral panel due to financial constraints. Child was closely monitored for any worsening of symptoms. The next day, the child showed significant improvement and started to walk without much pain.

On 4th day of admission child was taking feeds well, maintaining urine output and walking without difficulty. Hence the child was discharged and upon review after one week her serum CPK levels returned to normal and neurological examination was completely normal.

Test	D1	D2	Follow up
TC	5800/mm3	-	-
Platelet	1,83,000/mm3	-	-
BUN	14.18 mg/dl	11.77 mg/dl	-
Serum creatinine	0.55 mg/dl	0.56 mg/dl	-
CRP	0.26 mg/dl (negative)	-	-
Serum CPK	3430 IU/L (Normal 0-197 IU/L)	2376 IU/L	131 IU/L
Serum AST(SGOT)	142.6 IU/L	-	28 IU/L
Serum ALT(SGPT)	38 IU/L	-	19.8 IU/L
Urine analysis	Normal	Normal	-

3. Discussion

Benign Acute Childhood Myositis is a self-limiting process which can involve any muscles, but the gastrocnemius and soleus muscles frequently. The aetiology of BACM is typically viral and the most frequent viruses involved are influenza A& B. It affects preschool and school aged children of a median age of 6-9 years, with a male predominance, though it has been reported in few adolescents during the 2009 influenza pandemic.(3) In a study conducted by Costa Azevedo et al. in Portugal, out of 174 cases of elevated CPK levels, 100 cases corresponded to BACM compatible clinical presentation. There was male predominance (77%) with a median age of 6 years. Adults are rarely affected.(4) (5)

Infective myositis can also be caused by coxsackie, parainfluenza, adenovirus, enterovirus, human T-cell leukemia-lymphoma virus, hepatitis B and C, and even SARS-coronavirus. (6)

BACM was first formally described in a series of case reports by Lundberg, who studied 74 patients in Sweden with an illness named 'myalgia curis epidemica'.(7) The prodromal symptoms are usually fever, cough and rhinorrhoea. The pain is worse after a period of rest, usually in the early morning.

In a Portugal study published in 2022, the incidence of BACM was about 2.6 cases per 1,00,000 children under 18 years during epidemic seasons and 0.23 cases during non-epidemic seasons. BACM typically manifests with acute myositis and increased serum CK levels following a viral infection. A family history of neuromuscular disorders, myoglobinuria, trauma, rash, oedema and muscle weakness are not typically associated with BACM. This is common during winter and early spring. During the 2019/2020 flu season, influenza B was the most prevalent virus and it is known to be the most myotropic one. (4)

Despite the clinical features and course of this condition in children clearly described, reports showed that only 24% of 165 participating physicians were able to recognize BACM. Furthermore, the exact current incidence, prevalence and pathogenesis of BACM are not well known.(8)

Although most cases of BACM are children, it's incidence has also been described in adolescents. This could simply be explained by the increased tropism of viruses for immature muscle cells. Furthermore, each virus could act as a trigger in genetically predisposed children and in few patients with undiagnosed metabolic diseases.

Regarding the management of BACM, it doesn't need any invasive tests or medical therapy. Still the sudden onset of illness may be mistaken for severe neurological illness like Guillain Barre Syndrome, transverse myelitis or other autoimmune conditions.(9) This might lead to unnecessary investigations like electromyography, CSF analysis. Otherwise, daily thorough clinical examination, mainly complete neurological examination and urine dipstick to detect myoglobinuria are sufficient to detect complications promptly and to rule out severe illness. Serum CPK levels should be measured early and monitoring of CPK levels in case of worsening of clinical condition is of due importance.

Though this condition follows a viral upper respiratory infection, anti-viral drugs are usually not indicated since when myositis develops, the child is already in early convalescent phase of illness.(10) Also tests for BACM-related viruses are usually not indicated routinely because of the time interval between infection and seroconversion.

In the Neurology article published in 1999 in children with BACM, two characteristic gaits were noted; toe-walking and a wide-based stiff legged gait. Muscle tenderness was isolated to the gastrocnemius soleus muscles in 82% of episodes. (11)

The Italian study suggests few red flags in patients with suspected BACM upon which they should be admitted, evaluated further and managed conservatively. (12) Those are

- Age less than 2 years

- Family history positive for neuromuscular diseases
- Recurrent episode of BACM
- Abnormal neurological examination or poor general condition.
- Dark coloured urine.
- Abnormal renal function tests
- Serum CPK more than 10 times the normal

The most common haematological findings were leukopenia and thrombocytopenia.(13) Once child has improved clinically, follow up is to be done after a week to check for serum CPK levels and if the weakness persists, electromyography or nerve conduction study are needed as per the clinical condition. Hospitalization is advised when there are red flags in a case of suspected BACM.

The exact mechanism of the virus causing myositis remains undetermined, but some theories have been suggested, such as direct invasion of the muscle cells by the virus, myotoxic cytokines released in response to viral infection and immunologic processes induced by the viral infection. It is known that the infection causes necrosis and degenerative changes to the muscle fibre, which results in elevation of CPK levels.(7)

Myoglobinuria is rare and if it occurs, the patient should be admitted to monitor renal function periodically and dialysis is done if there is development of rhabdomyolysis. (14) (15) This serious complication seems to be more common in girls than boys. The prognosis is usually excellent and there are no functional sequelae. Complete clinical and biochemical recovery can be expected in 1-2 weeks. (2)

4. Conclusion

Despite the benign nature, BACM can be confusing to physicians who are not familiar with this entity, leading to unnecessary extensive workups. This occurs following a viral URI, most commonly influenza B in young children, predominantly boys. The actual mechanism remains unclear whether the myositis is due to direct viral invasion or by immune-mediated mechanisms. Leukopenia and thrombocytopenia have been described in many cases; it could be a finding related to the preceding viral illness. Conservative management with adequate hydration and analgesics for pain relief along with crucial monitoring for progression to severe complications such as myoglobinuria and renal failure due to rhabdomyolysis are the important measures in management of BACM cases.

Conflicts of interest
None

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