Benign acute childhood myositis secondary to parainfluenza A virus

Introduction

Benign acute childhood myositis is an uncommon, acute, self-limiting muscle disorder that mainly affects school-aged boys (Neocleous et al, 2012). The condition typically follows a viral illness and is characterized by sudden onset of leg pain and tenderness over the calf muscles after rest, and refusal or inability to walk. The majority of affected children have significantly elevated blood levels of creatine phosphokinase. Benign acute childhood myositis has an excellent prognosis and generally requires only supportive management. This article presents the case of a 5-year-old boy with painful legs and elevated creatine phosphokinase level following an upper respiratory tract infection.

Discussion

Children often complain of muscle pain in association with a viral illness. Lundberg (1957) first published the report entitled 'myalgia cruris epidemica' describing a newly identified syndrome of muscle pain, predominantly affecting the calf muscles, and occurring mainly in school-aged children. The precise pathophysiology of the disorder remains unclear, although an immune-mediated inflammatory process initiated by viral infection has been hypothesized (Crum-Cianflone, 2010). Various descriptions have been given to the disorder, and despite some debate about the actual muscle pathology present, the term myositis seems to be

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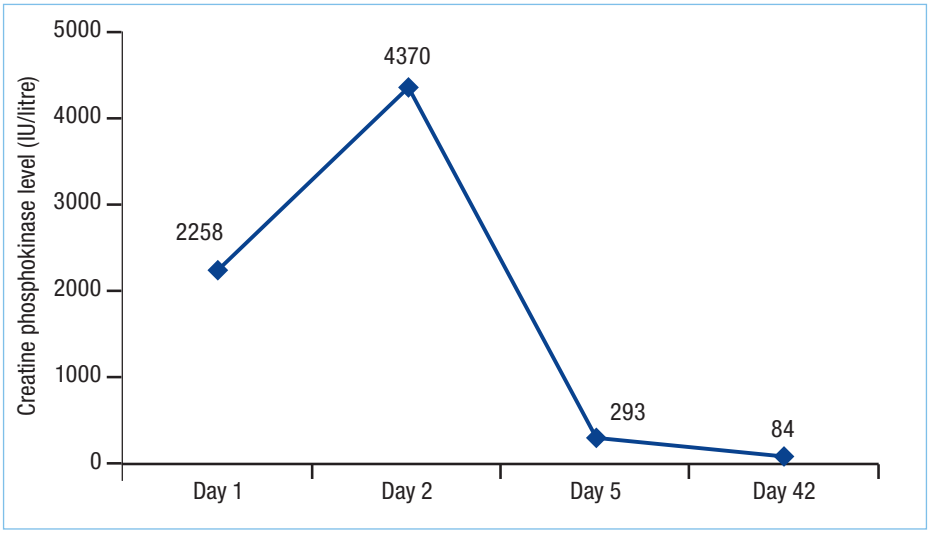
Figure 1. Serial creatine phosphokinase levels during the acute illness and after.



CASE REPORT

A previously fit and healthy 5-year-old boy presented with a 24-hour history of excruciating pain in his legs and refusal to weight bear. He had had a febrile illness for 3 days previously although his fever had resolved at presentation. There was no history of trauma. He was reported to be drinking well and passing urine normally. The only history of note was that he had returned from a 3-week visit to Pakistan a week earlier.

His vital observations were within normal limits and he appeared to be systemically well. Clinical examination revealed exquisite tenderness to palpation over both his calf muscles, but the area showed no redness or warmth. The pain was exacerbated by an attempt to gently dorsiflex his ankle joints. Joint swelling and tenderness were absent. Tendon reflexes were normal. A provisional diagnosis of myositis was made. Other potential serious pathologies such as leukaemia, rhabdomyolysis and compartment syndrome were considered as diagnostic possibilities. He was admitted for bed rest and further investigations, with strict recording of fluid intake and urine output.

Blood investigations revealed normal renal and liver function functions, full blood count (including blood film), haemoglobin, erythrocyte sedimentation rate, and C-reactive protein, lactate dehydrogenase, calcium and phosphate levels. In light of his recent travel history, thick and thin films for malaria screening and antigen test for Plasmodium were sent, and both were negative. Arbovirus serology (to rule out dengue fever) could not be obtained for logistical reasons. Mycoplasma serology was negative.

The only abnormal blood result was an extremely high creatine phosphokinase level of 2258 IU/litre (range 25–190 IU/litre) which increased further over the next 24 hours (*Figure 1*). He was managed conservatively with intravenous fluids (needed because he was vomiting) and analgesics. He was closely monitored for urine output and for myoglobinuria (risk of rhabdomyolysis); gentle mobilization of the lower limbs in bed was encouraged.

A viral throat swab which became available 72 hours later confirmed the presence of parainfluenza A virus, thus confirming the diagnosis of benign acute childhood myositis secondary to viral aetiology. The pain settled over the next 3–4 days and he started to gradually weight bear on his legs. He was discharged home after 4 days with advice to return if symptoms worsened or dark-coloured urine was noted. At a clinic review 6 weeks later, he had returned to normal health with complete resolution of symptoms and normalization of his creatine phosphokinase levels.

generally used. This reflects the finding that there must be at least an element of muscle inflammation to account for the release of creatine phosphokinase. Viruses may affect the musculature directly by invasion or indirectly via immune mechanisms (Crum-Cianflone, 2010). Some authors believe that its occurrence primarily in boys may reflect a genetic predisposition or a previously unknown metabolic defect (Crum-Cianflone, 2010).

Viruses commonly associated with benign acute childhood myositis include influenza A and B viruses (Ruff and Secrist, 1982; Mall et al, 2011; Neocleous et al, 2012). Other organisms which cause benign acute childhood myositis include parainfluenza virus, dengue virus, toxoplasma, *Mycoplasma*, cytomegalovirus, adenovirus and swine flu virus (H1N1). In a Portuguese study of 25 children with benign acute childhood myositis, organisms were isolated in nine cases (Santos et al, 2014).

Benign acute childhood myositis has a predilection for males. The Portuguese study of 25 cases of benign acute childhood myositis noted a 4.6:1 male:female ratio; the median age was 7 years (range 4–10 years), and 23 patients (82%) were aged >5 years (Santos et al, 2014). The onset of pain tends to occur 'first thing' in the morning. Neurological examination is usually normal, and it is suggested that mild weakness is the result of calf muscle pain rather than true inability of muscle to generate power (Neocleous et al, 2012). In a Greek study with 32 children (18 males), 30 patients had calf tenderness, especially pronounced while walking, 19 had profound gait abnormalities with wide-based gait, and calf muscles were the only muscle group affected in 29 patients; tendon reflexes were normal in 27 patients while the remaining five cases had decreased tendon reflexes (Zafeiriou et al, 2000).

Most children with benign acute childhood myositis have a raised level of creatine phosphokinase. In the Portuguese study creatine phosphokinase levels at admission ranged between 785 and 26 863 IU/litre (median 4181 IU/litre) (Santos et al, 2014). A similar trend was seen in the Greek study where creatine phosphokinase levels at admission varied between 558 and 6800 IU/litre (median 2850 IU/litre) (Zafeiriou et al, 2000). Other less consistent laboratory findings are leucopenia, thrombocytopenia, raised erythrocyte sedimentation rate and raised serum aspartate transaminase levels (Zafeiriou et al, 2000; Santos et al, 2014).

In many cases although creatine phosphokinase levels are massively elevated at presentation, rhabdomyolysis and myoglobinuria usually do not occur (Santos et al, 2014). The condition is managed by pain relief, rest and gentle remobilization. Patients can usually be discharged if they have typical clinical presentation, normal renal function, and follow up is arranged to ensure complete resolution (Santos et al, 2014). Further investigations are not indicated unless there is clinical suspicion of more serious disorders. Rhabdomyolysis, acute renal failure and extensive compartment syndrome should be considered in the differential diagnosis of benign acute childhood myositis, especially in a child presenting with a decreased level of consciousness, severe dehydration, and severe bilateral thigh and calf pain and swelling, or those with a pre-existing neuromuscular condition during or after a viral illness (Vrsalovic et al, 2007; Ebbeson et al, 2009).

Muscle studies have been performed relatively infrequently in view of the short duration of symptoms and the good prognosis in cases of benign acute childhood myositis. When electromyography has been performed the findings have either been normal or have detected brief motor units (<3 ms) of low amplitude (<300 µV) found in a patchy distribution in several upper and lower extremity muscles (Ruff and Secrist, 1982). When muscles biopsies have been taken most have been reported as normal, or occasionally found to demonstrate moderate muscle necrosis and regenerating muscle fibres with interstitial oedema and foci of polymorphonuclear and mononuclear cell infiltrates (Ruff and Secrist, 1982).

Conclusions

Benign acute childhood myositis is a transient, self-limiting condition which requires no specific treatment. Correct diagnosis is made by recognizing the characteristic clinical and laboratory findings associated with this condition. As the differential diagnosis includes more severe pathological conditions which must be excluded, prompt accurate diagnosis can prevent undertaking unnecessary diagnostic investigations and causing anxiety both to parents and children. Benign acute childhood myositis should be considered among the main causes of sudden onset gait abnormality or refusal to walk, with calf tenderness, in young children. **BJHM**

LEARNING POINTS

- Benign acute childhood myositis is an acute, self-limiting muscle disorder especially affecting school-aged boys.
- Benign acute childhood myositis usually occurs after an episode of viral infection, influenza viruses being the commonest pathogen detected.
- Benign acute childhood myositis should be considered as important differential diagnoses of sudden onset gait abnormality or refusal to walk, with calf muscle tenderness.
- Raised creatine phosphokinase levels are often the only abnormal result detected following blood investigations.
- The condition is managed by pain relief, rest and gentle remobilization.

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