



## Acute myositis in the paediatric emergency department

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### Abstract

**Introduction:** benign acute childhood myositis (BACM) is a benign, self-limiting disease currently believed to be a sequela of viral infections. The typical clinical presentation is myalgia in the lower extremities accompanied by refusal to walk.

**Patients and methods:** we did a retrospective review of all the cases of BACM managed in our paediatric emergency department over a 25-year period. We collected data on demographic, clinical and laboratory variables through the systematic review of health records. We performed a descriptive statistical analysis, followed by comparison of groups with the Mann-Whitney *U* test. The correlation between quantitative variables was assessed by means of the Spearman rank coefficient.

**Results:** we identified 139 cases of BACM (74.8% in male patients). The highest incidence corresponded to children aged 4 to 8 years and the winter months. Bilateral lower limb myalgia was the most frequent presenting complaint. Fever was reported in 86.3% of cases, accompanied by respiratory symptoms (88.5%); influenza virus infection was confirmed in 37 cases (75.6% serotype B). At diagnosis, the median creatine kinase (CK) level was 1794 U/l, with no significant differences between the sexes ( $p=0.687$ ). All patients had normal renal function, and none required hospitalization.

**Conclusions:** based on its clinical and analytical manifestations, BACM is easy to recognise. Although massive elevation of CK is common, complications are rare, and most patients can be discharged with conservative treatment.

### Key words:

- Benign acute childhood myositis
- Children
- Emergency medicine
- Myositis

## Miositis aguda infantil en urgencias pediátricas

### Resumen

**Introducción:** la miositis aguda benigna de la infancia (MAB) es una enfermedad benigna y autolimitada, considerada en la actualidad como una secuela de enfermedades víricas. Su manifestación clínica típica son las mialgias en las extremidades inferiores acompañadas de rechazo a caminar.

**Pacientes y métodos:** realizamos una revisión retrospectiva de los casos de MAB atendidos en nuestro servicio de urgencias pediátricas durante un periodo de 25 años. Obtuvimos datos demográficos, clínicos y de laboratorio a través de la revisión sistemática de historias clínicas. Se elaboraron estadísticas descriptivas y se efectuaron comparaciones entre grupos a través del test U de Mann-Whitney. Las correlaciones entre variables cuantitativas se realizaron mediante el análisis de rangos de Spearman.

**Resultados:** se identificaron 139 casos de MAB (74,8% varones). La mayor incidencia se produjo en los meses de invierno y en niños de 4-8 años. Las mialgias bilaterales en miembros inferiores fueron el principal motivo de consulta. El 86,3% referían fiebre acompañada de síntomas respiratorios (88,5%), confirmándose en urgencias la infección por virus influenza en 37 casos (75,6% del serotipo B). En el momento del diagnóstico, la mediana de creatininasas (CK) fue 1794 U/l, no existiendo diferencias significativas en función del sexo ( $p = 0,687$ ). La función renal fue normal en todos los pacientes, ninguno necesitó hospitalización.

**Conclusiones:** la MAB es una entidad fácilmente reconocible teniendo en cuenta sus manifestaciones clínicas y analíticas. A pesar de las masivas elevaciones de CK, sus complicaciones son excepcionales y la mayoría de los pacientes pueden ser dados de alta con tratamiento conservador.

### Palabras clave:

- Medicina de urgencia
- Miositis
- Miositis aguda benigna infantil
- Niños

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## INTRODUCTION

Benign acute childhood myositis (BACM) was first described by Ake Lundberg in 1957,<sup>1</sup> who used the term *myalgia cruris epidemica* to refer to the clinical characteristics of 74 children in the context of a flu epidemic, differentiating the particular clinical picture of the myalgias that preceded or overlapped the viral illness.

At present, BACM is considered a sequelae of viral infection and is characterised by myalgia in the lower extremities, weakness, pain in the legs and refusal to walk following the prodromal stage of the infection.

## MATERIAL AND METHODS

We conducted a single centre retrospective observational study in the paediatric emergency department of a tertiary care children's hospital. We included all patients aged 0 to 16 years given a diagnosis of BACM between January 1995 and December 2020. We excluded patients that did not have their levels of creatine kinase (CK) measured in the emergency department and those who received a diagnosis of acute pyomyositis. The study was approved by the research ethics committee of the hospital.

The virological diagnosis of influenza was made by immunofluorescence assay and polymerase chain reaction testing of nasopharyngeal aspirate samples.

We calculated descriptive statistics for the demographic, clinical and laboratory characteristics of the patients. We summarised continuous variables as mean and standard deviation (SD) if they followed a normal distribution, and otherwise as median and interquartile range (IQR). We compared groups with the Mann-Whitney *U* test. We analysed the correlation between quantitative variables with the Spearman rank test. We defined statistical significance as  $p < 0.05$ . The statistical analysis was performed with the software IBM SPSS®.

## RESULTS

We included a total of 139 cases in the period under study. Of this total, 74.8% were male and 25.1% female. The mean age of the patients was  $6.5 \pm 2.1$  years. **Table 1** presents the demographic, clinical and laboratory characteristics of the patients.

The most frequent presenting complaint was bilateral myalgia in the lower extremities (88.5%). Also, 86.3% of patients reported fever associated with respiratory symptoms. Infection by the influenza virus was confirmed in 37 cases, with a predominance of type B (75.6%). The incidence peaked

**Table 1.** Demographic, clinical and laboratory characteristics

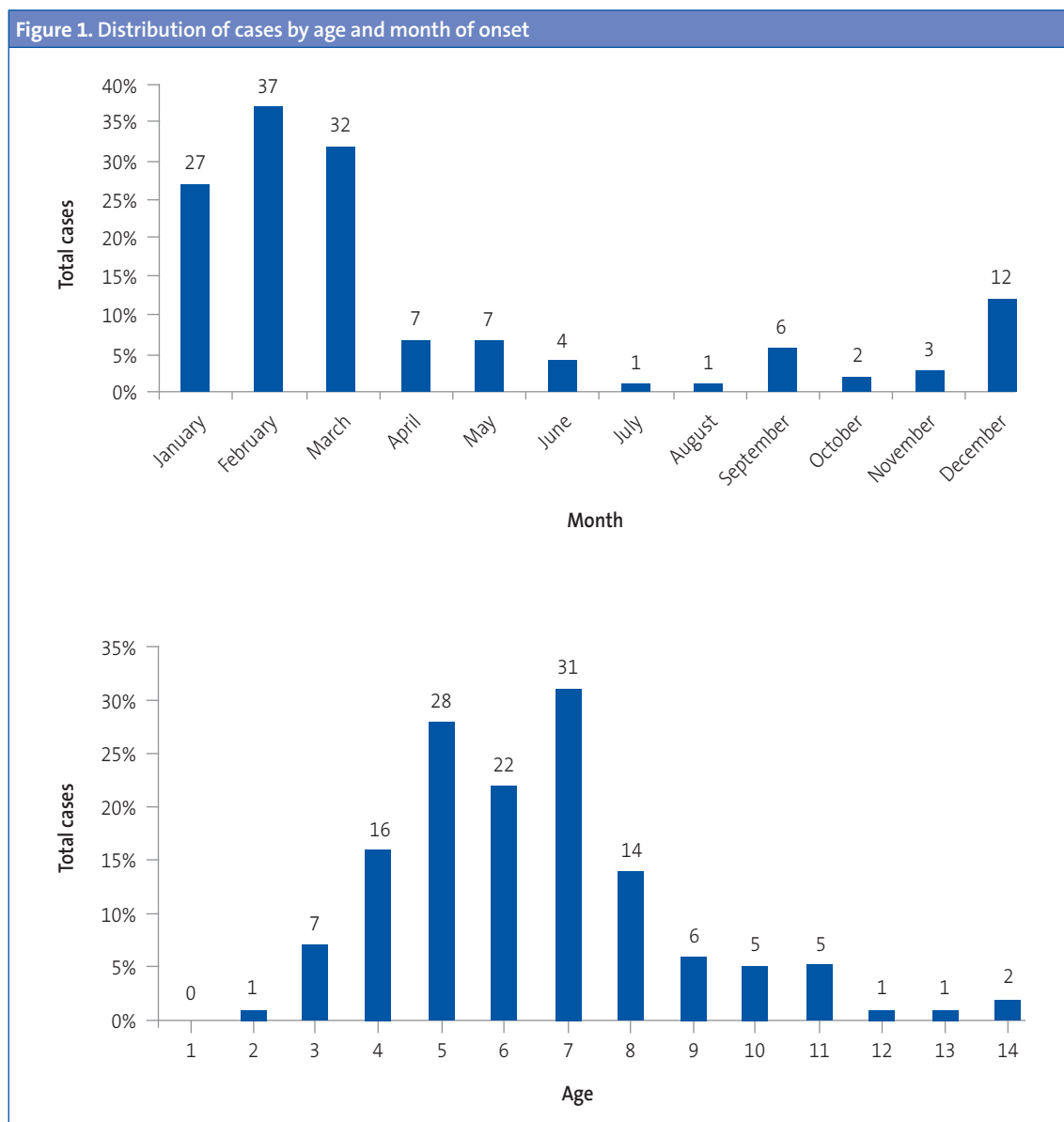
Total episodes	139
Patients with at least 1 episode of BACM	134
• Patients with a single episode of BACM, n (%)	130 (97)
• Patients with recurrent episodes of BACM, n (%)	4 (0)
Sex (male), n (%)	104 (74.8)
Age (years), mean (SD)	6.5 (2.1)
Fever, n (%)	120 (86.3)
Muscle pain, n (%)	135 (97.1)
• Bilateral in lower extremities, n (%)	123 (88.5)
• Unilateral in lower extremities, n (%)	12 (8.6)
Time elapsed from onset of pain	
• ≤24 hours, n (%)	113 (81.3)
• 24-48 hours, n (%)	15 (10.8)
• >48 hours, n (%)	7 (5)
Associated respiratory symptoms, n (%)	123 (88.5)
Laboratory data	
• CK (U/l), median (IQR)	1794 (2798)
• AST (U/l), median (IQR)	94 (81.8)
• ALT (U/l), median (IQR)	37 (32.3)
• Total leukocytes (cells/μl), median (IQR)	4800 (1960)
• Neutrophiles (cells/μl), mean (SD)	2373.6 (1518.5)
• Lymphocytes (cells/μl), mean (SD)	2215.8 (1021.5)
• CRP (mg/l), median (IQR)	4 (3)
Influenza screening in emergency department	
• Type A, n (%)	9 (5.7)
• Type B, n (%)	28 (20.1)

ALT: alanine aminotransferase; AST: aspartate aminotransferase; BACM: benign acute childhood myositis; CK: creatin kinase; CRP: C-reactive protein; IQR: interquartile range; SD: standard deviation

in the winter months and was highest in children aged 4 to 8 years (Fig. 1).

At the time of diagnosis, the median serum CK level was 1794 U/l (range 152-16 813 U/l), with no significant differences based on sex ( $p = 0.687$ ). We found a moderately strong correlation between age and CK levels (Spearman  $r = 0.217$ ;  $p = 0.01$ ). The white blood cell count was normal in 98.5% (normal range:  $4.2\text{-}11.4 \times 10^3/\mu\text{l}$ ). The levels of C-reactive protein were elevated in 25.7% (normal range: 0-5 mg/l) and 96.2% had hypertransaminasaemia (47.1% aspartate aminotransferase [AST] elevation and 52.9% mixed). There was a strong correlation between the serum levels of AST and of CK (Spearman  $r = 0.921$ ;  $p = 0.001$ ). Renal function was normal in all patients, and none of the patients required hospital admission.

Figure 1. Distribution of cases by age and month of onset



## DISCUSSION

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Benign acute childhood myositis is frequently associated with viral respiratory illnesses, chiefly infection by influenza B virus. However, it has also been described in association with other viruses (coxsackie, adenovirus, parainfluenza, respiratory syncytial virus) and bacteria such as *Mycoplasma pneumoniae*.<sup>2</sup> In our case series, the causative pathogen was isolated in 37 patients (28 influenza B virus and 9 influenza A virus).

Abnormal values of CK 20 to 30 times the upper limit of normal confirm the diagnosis in most cases, and usually normalise in a couple of weeks.<sup>3,4</sup> Using sonoelastography, Ekşioğlu *et al.* compared tissue elasticity in children with BACM and healthy controls, demonstrating that this ultrasound technique offers a high sensitivity (80%) and specificity (88%) for the diagnosis of BACM.<sup>5</sup>

In our patients, the median serum CK level at diagnosis was 1794 U/l, similar to the levels reported by other authors (Brisca *et al.*<sup>5</sup> 1413 IU/l, Rosenberg *et al.*<sup>7</sup> 1346 IU/l).

Despite this massive elevation of CK, patients rarely have myoglobinuria and cases presenting with rhabdomyolysis are exceptional.<sup>2,8</sup>

Benign acute childhood myositis is a self-limiting disease with improvement of myalgia in a few days. However, some authors have described cases in which symptoms persisted longer than 1 month.<sup>9,10</sup> Such patients may benefit from rehabilitation, although specific guidelines have not been developed for this approach. Zaldibar-Barinaga *et al.* reported a case in a girl aged 8 years whose symptoms lasted 22 days; the patient improved with rehabilitation consisting of muscle stretches, active and assisted exercises and exercises with a resistance band, correction of abnormal posture, gait rehabilitation with gradually increasing partial weight-bearing and local application of cold to alleviate muscle pain.<sup>11,12</sup>

A study published recently by Turan *et al.*<sup>13</sup> analysed the characteristics of 114 paediatric patients with BACM. Oseltamivir was given to 44 patients (38.6%) with a conformed diagnosis of influenza A/B virus infection. The administered dose varied based on body weight ( $\leq 15$  kg: 30 mg,  $>15$  to 23 kg: 45 mg,  $>23$  to 40 kg: 60 mg and  $> 40$  kg: 75 mg, given twice daily). The authors found that the median time to recovery was shorter in patients treated with oseltamivir (4 days) compared to patients that did not receive it (5 days).

Recurrent episodes of BACM are infrequent—there were only 4 cases in our series—and different viruses may be involved in each episode.<sup>8</sup> In patients with recurrent episodes of BACM associated with influenza, it has been found that the second episode is caused by a different serotype than the one involved in the initial episode. That is, the myositis seems to develop only in response to the very first exposure to a specific virus, which would explain the rarity of cases detected in adults.

## CONCLUSION

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Benign acute childhood myositis is easily recognizable based on its clinical and laboratory characteristics. Despite the massive elevation of CK, complications are rare and most patients can be discharged with conservative management.

## CONFLICTS OF INTEREST

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The authors have no conflicts of interest to disclose in relation to the preparation and publication of this article.

## ABBREVIATIONS

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**ALT:** alanine aminotransferase • **AST:** aspartate aminotransferase • **BACM:** benign acute childhood myositis • **CK:** creatin kinase • **IQR:** interquartile range • **SD:** standard deviation.

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