

**CASE REPORT**

# A rare presentation of benign acute childhood myositis

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**Key Clinical Message**

Benign acute childhood myositis is a self-limiting muscle disorder characterized by calf pain with an isolated finding of elevated serum creatine kinase, being preceded by an influenza-like illness. The classic clinical and laboratory features may allow for a correct diagnosis. This report describes some accompanying symptoms which are not usually perceived. An incorrect diagnosis could lead to unnecessary treatments.

**KEYWORDS**

calf pain, creatine kinase, influenza, myositis, self-limiting disease

## 1 | INTRODUCTION

Benign acute childhood myositis (BACM) is a rare muscle disorder predominantly affects school-age children, with an incidence of 2.6 cases per 100 000 children under 18 years old in epidemic times.<sup>1</sup> The disease is usually occurring at the early convalescent phase of a viral illness, particularly influenza B infection.<sup>2</sup> Many theories regarding the origin of the disease have been postulated so far. It is unclear whether this is due to direct infection of the muscle by the virus, myotoxic cytokines or other immunological processes, as the viral agent has not been consistently demonstrated in biopsy.<sup>3</sup> Clinically, it is characterized by a sudden reluctance to walk due to severe bilateral calf pain and by increased creatine kinase (CK) level. Histopathologic examination of muscle reveals degeneration and necrosis, with overall little inflammatory infiltrates. The diagnosis can be made by a febrile prodrome, otherwise normal exam, positive CK level and viral studies.<sup>4,5</sup> Typically, the illness is self-limiting and patients recover spontaneously in about a week.<sup>2</sup> Only symptomatic therapy is required during the acute illness and antivirals are unlikely to be beneficial.<sup>6</sup> However, nonclassic presentation of BACM would trigger a more urgent and extensive investigation of serious differentials.

## 2 | CASE PRESENTATION

An 11-year-old girl with a history of recurrent episodes of calf pain and difficult walking for the last 3 months sought chiropractic care for pain relief. In late January 2018, the child had suffered from fever (38.5°C), headache, coryza, cough, and diarrhea. A rapid flu testing at a district hospital led to a diagnosis of influenza B. She was then given oseltamivir (Tamiflu®, an antiviral drug), ibuprofen (a nonsteroidal anti-inflammatory drug, NSAID), chlorpheniramine (an antihistamine), bromhexine (an expectorant), and diphenhydramine compound linctus (an antitussive) for 5 days.

Eleven days later, on recovery from the flu symptoms, the child was brought to the emergency department because of nuchal pain, calf pain, and difficulty walking. She was then hospitalized (8/Feb- 22/Feb) in order to allow medical observation and exclude severe disorders. Increased creatine kinase 244 IU/L (CK range: 42-186) and phosphate 1.78 mmol/L (pediatric range: 0.7-1.43) were the alterations seen in laboratory tests. Serum calcium of 2.37 mmol/L (range: 2.15-2.55), other routine blood parameters and inflammatory markers were within normal limits. Blood cultures were sterile, and PCR for mycoplasma pneumoniae was negative. Chest X-ray and brain and lumbar MRI were observed normal. There was no family history of neuromuscular disorder. With a

provisional diagnosis of benign acute childhood myositis, the patient was given acetaminophen (an analgesic), ibuprofen (a NSAID), magnesium trisilicate (an antacid), and IV fluids. Serum CK had returned to 71 IU/L, her myalgia had resolved, and she was discharged from hospital.

After returning home, the child had fainting, slipped and fell sporadically and had involuntary twitching of arms and legs while sleeping. Within the next few days, she progressed to catastrophic manifestations of diplopia, occipital headache, and excruciating calf pain, which led to another hospital stay (28/Feb- 5/Mar) for further neuropsychological investigation. Pain intensity rating was 4-5/10 for headache and 8/10 for calf myalgia on Numeric Rating Scale (NRS). Muscle weakness was subtle in the upper limbs (5-/5) and mild in the legs (4+/5), according to the Medical Research Council (MRC) scale. No common external signs were seen other than occasional involuntary twitching for a minute or two noticed in bilateral brachioradialis muscles and the anterior thighs. Findings of electroencephalography/nerve conduction test were normal. Psychiatric, neurological, and ophthalmic investigations were generally unremarkable, with the exception of suspected attention deficit disorder by a psychoeducational assessment. The patient returned home after having an exclusion of more serious illness and her gait improved. She had been placed on ibuprofen (a NSAID), famotidine (an acid control tablet), and topical application of anti-inflammatory emollients with hydrocortisone acetate cream, heparinoid cream, and calamine cream which provided little relief of symptoms.

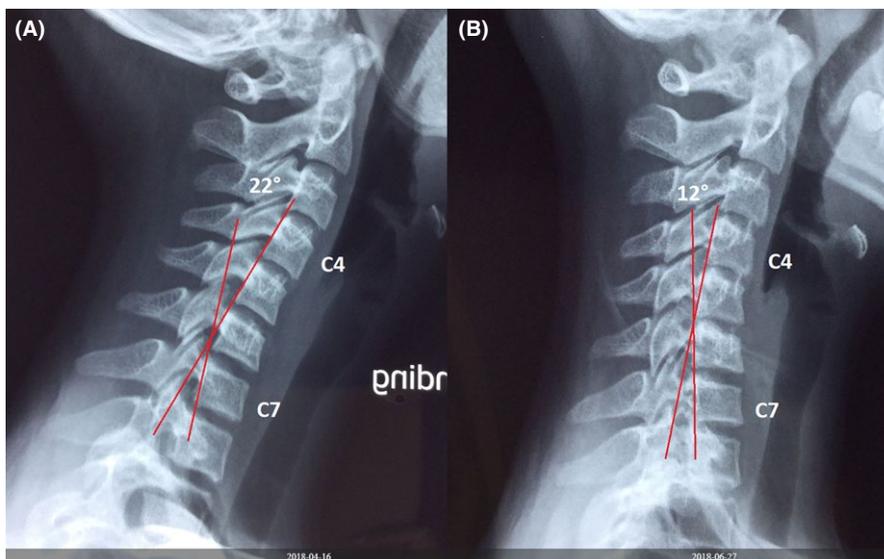
On April 13th, the child was brought to our clinic in a wheelchair by her mother for a holistic approach to pain management. Examination showed restricted cervical extension and bilateral rotation. Her upper arms, thighs, and calves were tender on palpation and on stretching. Muscle power was symmetrical in upper and lower limbs. The child also described vague paresthesias in the extremities which

were inconsistent with dermatomal manner, more likely resulting from distorted sensory perceptions. Cervical radiographs demonstrated reversal of normal lordosis in the lower C-spine (Figure 1A). Prevertebral space measurements: nasopharyngeal space, 3 mm; retropharyngeal space, 3 mm; and retrotracheal space, 10 mm.

Chiropractic regimen was in an attempt to release cervical restriction and stretch musculature. Treatment program included ultrasound therapy over the upper trapezius muscles, and adjustment covering of the cervical and the upper thoracic spine. The patient was treated with five sessions for the first week. She reported substantial pain relief shortly after initiation of adjustment and continued to receive chiropractic treatment thrice a week for 1 month, followed by twice-weekly until a 2-month assessment. After 25 treatment sessions, the painful symptoms and muscle twitching had disappeared. She returned to school and fully participated in all activities. A follow-up assessment demonstrated a reduction in cervical kyphosis ( $22^\circ$  vs  $12^\circ$ ; Figure 1B). The patient remained virtually symptom-free.

### 3 | DISCUSSION

Benign acute childhood myositis (BACM) was first described in 1957 by Lundberg in 74 Sweden children under the name of myalgia cruris epidemica.<sup>7</sup> Cases tended to cluster and were consistent with a viral etiology. Isolated cases or small clusters of BACM reported from different regions of the world had similar cardinal features (a flu-like prodrome, bilateral calf pain, increased CK levels, and spontaneous remission). Sporadically, a diverse pattern of acute viral myositis had also been observed, in which the trapezius muscle rather than the calves was involved. The outbreaks had been recorded under the name of myalgia nuchae epidemica.<sup>8,9</sup> A



**FIGURE 1** Cervical kyphosis before and after chiropractic adjustment. A, Sagittal radiographs demonstrate a reversal in the cervical curve over C4 to C7 levels. B, Some correction of the cervical curvature is noted. Stress angle returns from  $22^\circ$  to  $12^\circ$  after spinal adjustment

study in 72 Swedish cases (collected in between 1946 and 1951) revealed that clinical features of myalgia nuchae epidemica also included an antecedent flu-like illness, severe muscle pain affecting an isolated musculature, and spontaneous remission without lasting consequences.<sup>10</sup> The cardinal features of myalgia nuchae epidemica are quite identical to those of classic BACM, but the affected muscles and predominant adult population are dissimilar.<sup>10</sup>

It should also be remembered that our patient had severe nuchal pain as well. Sagittal radiographs also demonstrated a reversed curvature of her lower cervical spine (Figure 1A). Posture problems in the neck, in general speaking, are often related to the trapezius. The trapezius muscle is a postural and movement muscle, used to tilt and turn the head and neck, raise the shoulders and twist the arms. The superior part of the trapezius arises from the spinous processes of C7 vertebra, the external protuberance of the occipital bone and the nuchal ligament. The tightened trapezius could pull the spine taut, straightening the cervical curvature, and even causing the perception of reverse cervical lordosis. As reported in BACM with calf pain, often an abnormal gait is noticed, as a result, the nuchal pain can have the same consequences on nuchal posture. It is reasonable to speculate that the trapezius muscle had been involved concurrently.

The case presented is unique in that our patient with BACM experienced relapses in subsequent 3 months rather than a transient process.<sup>2,6</sup> Furthermore, the symptom of classic BACM is isolated to the gastrocnemius and soleus complex. Associated pain or tenderness in the upper extremities or trunk has been rarely reported.<sup>11</sup> A concomitant involvement of the trapezius muscle (dual musculature involvement) has not been described. The diverse manifestations could be explained by an increased tropism of viruses for muscle cells,<sup>4</sup> different virus subtypes,<sup>12</sup> or preexisting conditions affecting individual susceptibility.<sup>13</sup>

The chiropractic approach performed seems reasonable as an option to treat the musculoskeletal pain in this case. Since the typical course of BACM is self-limiting without lasting consequences, it remains possible that the symptomatic remission was due to spontaneous healing rather than to chiropractic therapy. Several findings suggest, however, that our patient appeared to benefit from incorporating chiropractic above that expected by natural evolution alone. First, the patient reported substantial pain relief shortly after treatment initiation. Moreover, symptom improvement observed during the treatment sessions was notably larger than that seen during the period of medication preceding it. Finally, clinical improvement was not restricted to pain relief and was also significant for restoration of cervical curvature (Figure 1).

The mechanisms responsible for the therapeutic effects of spinal adjustment remain unclear. Likely, muscle stretching by spinal adjustment may help reduce the spinal nerve tension, release its pinched nerve, and subsequently alleviate

neurological symptoms.<sup>14</sup> This presentation is limited by its retrospective nature specifically reliability of self-reporting and medical records. We were unable to prove the efficacy of chiropractic alone only by a single case. Second, the diagnosis of BACM is made principally based on the medical history and clinical symptoms.<sup>4,5</sup> When the patient has clinical pictures resembling the cardinal features of typical cases, the diagnosis is made. Whereas patient's symptoms to a varying extent differ from the typical features, the diagnostic gray zone can therefore be wide, as seen in this case. More such cases being reported would shed more light on various aspects of BACM. It has also been emphasized that children with a rapidly worsening condition, no symptomatic resolution after a few days, chronic course and atypical presentation, should be evaluated in order to rule out neurological illness, metabolic diseases, LPIN1 gene mutations, or immune-mediated syndromes with similar characteristics. In such cases, identification of autoantibodies or muscle biopsy should be considered. Furthermore, an incorrect diagnosis may lead to treatments that do no good and perhaps do harm. For instance, the use of heparinoid cream and hydrocortisone cream is not justified in this patient.

## 4 | CONCLUSION

In conclusion, symptoms of BCAM are alarming and can cause confusion both in parents and physicians. This report puts forth the need to recognize such diverse scenarios, not only from a challenging diagnostic standpoint, but also help avoid unnecessary workup and treatment.

## INFORMED CONSENT

Written-informed consent was obtained from the parent of the patient for publication of this case report and any accompanying images.

## CONFLICT OF INTEREST

The authors have no conflicts of interest to declare.

## AUTHOR CONTRIBUTION

EC: managed patient and wrote the manuscript as main author. AY: critically reviewed and helped organize the manuscript. All authors: read and approved the final manuscript.

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