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ORIGINAL ARTICLE

Acute benign myositis of childhood: Results of a prospective study performed in a pediatric emergency department

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Abstract

Objective: Benign acute childhood myositis is characterized by acute musculoskeletal involvement leading to transient limitations on deambulation followed by a viral illness. Our study objective to evaluate clinical and laboratory features of patients in a pediatric emergency department. **Methods:** We conducted a prospective study in patients with symptoms and laboratory findings compatible with viral myositis in the period of August 2017 to August 2018. **Results:** We assessed 20 patients in the period of twelve months. The mean age was 8,25 years. Of these, 83,3% had infectious symptoms in the week before the musculoskeletal involvement. By the time of the diagnosis, the symptoms were: calf pain, reluctance to walk, gait abnormality, diffuse myalgia and calf weakness. The most relevant laboratory finding was the elevation of CPK (mean 3359,556U/L) level, followed by AST (mean 131U/L) and ALT (mean 64,66U/L) elevation. The mean time for symptom relief was 3 days and in 7 days all exams were normal. **Conclusion:** Though the exact incidence of this condition remains undetermined, the lower extremity pain and the gait abnormality is of concern of both parents and health care providers. We emphasize the importance of knowing this condition to avoid unnecessary exams and the delay in the diagnosis of severe conditions.

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INTRODUCTION

Acute benign childhood myositis, also known as viral myositis, is a self-limited benign disease, characterized by acute musculoskeletal involvement, usually after an infection of the upper airways leading to transient gait limitations¹. Arboviruses represent an important etiology in endemic areas.

The first description of the disease in the literature dates from 1905, when Leichtenstern described pain in the thighs and calves as a complication of an Influenza condition². In 1957, Lunderberg formally recognized the disease among schoolchildren with viral prodromes, who developed severe pain in the calves, making it difficult to walk, and named it epidemic myalgia cruris³.

Benign acute childhood myositis is considered infrequent, and its incidence is not known. The largest series of cases reported in the Brazilian literature includes 35 cases, collected over a nine-year period¹.

The clinical picture is characterized by acute muscle pain in the calves or, more rarely, in the thighs, and sudden difficulty or refusal to walk. In some cases, the patient may still have a bizarre gait. The condition is often preceded by infection of the upper airways, presenting symptoms such as fever, headache, runny nose, cough and sore throat. It occurs mainly in boys of preschool and school age^{1,4}. Creatine phosphokinase (CPK) will be elevated in almost all cases⁴.

The prognosis is excellent, with spontaneous resolution of symptoms between 48 hours and one week, with recurrence being rare.^{1,5}

Among the differential diagnoses are acute myositis caused by fungi, bacteria and protozoa; the myopathies associated with arboviruses, which have an acute onset but a more protracted course; and those with chronic evolution, such as hereditary muscular dystrophy and juvenile dermatomyositis, with a detailed history and a complete physical examination are extremely important^{4,6}. It is necessary to pay attention to urinary changes, mainly in color, since darkened urine may be indicative of rhabdomyolysis.^{1,3}

Despite its transient and self-limited character, viral myositis often leads to the search for emergency services due to parental concerns. It can bring difficulties to the attending physician due to the lack of knowledge about the disease and standardized conducts for the approach and treatment of the disease^{1,7}.

Its early diagnosis improves emergency care, avoids invasive and costly investigations for the health system, in addition to favoring conservative care and reassuring the family.^{1,7}

METHODS

This is an observational and prospective study, which included patients seen between the dates of August/2017 and August/2018.

We established a care protocol for viral myositis cases in order to standardize procedures in the Emergency Department of a Tertiary Care Pediatric Hospital in 2017. After approval by the Ethics Committee and educational measures for resident doctors and supervision, the protocol was implemented in the SUS Emergency Department, and we analyzed the clinical and laboratory data of all contemplated patients.

We invited to participate in the study all the patients who came directly to the Hospital Emergency Room and those referred from healthcare units that presented symptoms and laboratory findings compatible with viral myositis.

We ordered the following laboratory tests according to the protocol implemented in 2017: blood count, erythrocyte sedimentation rate (ESR), C-reactive protein (CRP), glutamic oxalacetic transaminase (TGO), glutamic-pyruvic transaminase (TGP), aldolase, CPK, lactate dehydrogenase (LDH) and urinary tape. Viral serologies and search for respiratory viruses were requested via the Basic Healthcare Unit; however, as a matter of logic, these tests were not feasible and we did not have access to their results.

The patients with microscopic hematuria should be subjected to partial urine collection as the presence of erythrocytes in the urinary strip, without hematuria in the urine, indicates myoglobinuria, suggestive of rhabdomyolysis. In cases suspected of this complication, children should be referred to the hospital and have their kidney function and hydration monitored.

On the other hand, patients without evidence of rhabdomyolysis, were referred for outpatient treatment using non-steroidal anti-inflammatory drugs (NSAIDs), in the case of our service, naproxen. Those responsible were reassured about the benign character of the condition and were advised about alarm signs and possible complications, in which case they should return for immediate reassessment.

As part of the protocol, the patients had clinical and laboratorial reassessment in one week.

Exclusion criteria were defined as: focal neurological findings; previous history of rheumatological, neurological or orthopedic disorders; patients who did not agree to participate in the study; age over 18 years and/or normal muscle enzymes, despite compatible symptoms.

We collected the information using a form with the following request for data: Patient identification; clinical data related to symptoms and duration of symptoms; result of laboratory tests and need for hospitalization. In the reassessment, a form was filled, adding symptom duration and control tests.

We described clinical and laboratory data in absolute numbers and percentages. The laboratory results of the twelve patients analyzed in the two opportunities were paired and analyzed using the t-test. They were considered statistically significant when $p < 0.05$.

The study was approved by the Hospital Ethics Committee (CEP N0 65859217.1.0000.0097). All study participants read and signed the Free and Informed Consent Form and the Agreement Form when applicable.

RESULTS

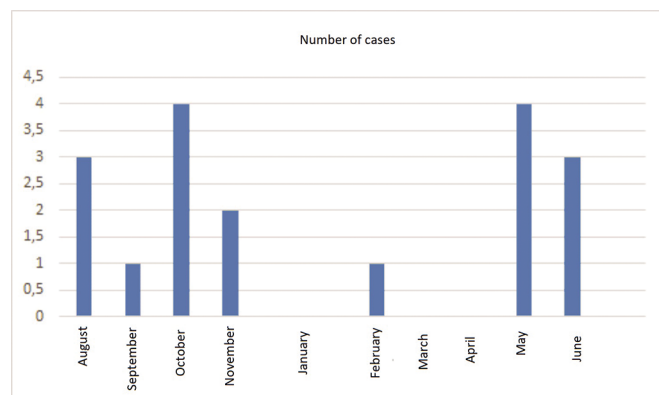
Of twenty patients with clinical and laboratory criteria compatible with the diagnosis of benign acute childhood myositis, two were excluded: one because there was no consent from the legal guardian and another one, who later received the diagnosis of dermatomyositis. Of the 18 patients included in the study, 12 had complete data at the time of the reevaluation, 1 patient had only clinical reevaluation and 5 did not return for reevaluation.

There was a prevalence of males, 11 of the 18 patients analyzed (61.1%). The average age was 8.3 years. The highest incidence of cases was in autumn (38.9%), followed by spring (33.3%) and winter (22.3%) (Graph 1).

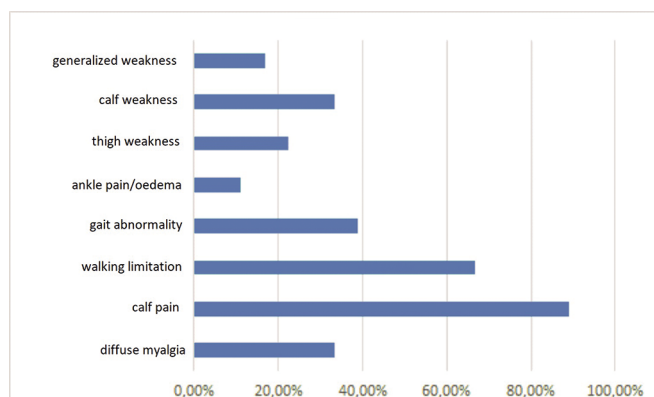
Fifteen patients (83.3%) had clinical changes in the week preceding the musculoskeletal condition, of which fourteen (77.8%) had respiratory symptoms preceding the muscle condition, and one of them, lymphadenitis and fever.

The prevalent clinical manifestations were pain in the calves, 16 of 18 patients (88.9%); gait limitation, 12 out of 18 (66.7%); abnormal gait, 7 out of 18 (38.8%); diffuse myalgia and weakness in the calf, 6 out of 18 (33.3%). Other findings were weakness in the thighs, generalized ankle weakness and pain (Graph 2).

Muscle enzyme analysis showed an average creatine phosphokinase of 3359.556 U/L (minimum, min, 418 U/L and max, maximum, 9964 U/L); mean aldolase of 17.61 U/L (min. 3.8 U/L and max. 29.6 U/L); Mean TGO of 131 U/L (min. 49 U/L and max. 283) and average TGP of 64.66 U/L (min. 25 U/L and max. 303 U/L). As for hematological changes, the results were a mean leukocyte count of 4,522.22 n/mm³, (min. 1,780 n/mm³ and max. 6,650 n/mm³; platelets, mean 188,444 n/mm³ (min. 110,000 n/mm³ and max. 266,000 n/mm³). No patient



Graphic 1. Distribution of cases by months from August 2017 to August 2018.



Graphic 2. Symptomatology presented by patients at diagnosis.

had CRP greater than 40 mg/dL, corroborating a probable viral etiology⁸. The erythrocyte sedimentation rate (ESR) averaged 19mm/h (min. 2mm/h and max. 42mm/h). Table 1 describes the laboratory findings and their reference values.

In our study, we did not find any suggestive case of rhabdomyolysis, which allowed outpatient treatment for all patients.

Symptom resolution occurred between one and six days, with an average of three, from the first evaluation.

At the time of the reassessment, creatine phosphokinase had a mean value of 161.16 U/L, (min. 66 U/L and max. 395 U/L). The leukocyte count had an mean value of 7220 n/mm³, (min 4540 n/mm³ to a maximum of 13820 n/mm³) and platelets, an average of 341,917 n/mm³ (min. 170000 n/mm³ and max. 670000 n/mm³), Mean ESR of 15.7 mm/h (min. 2 mm/h and max. 41 mm/h). The CRP values of the twelve patients remained below 40 mg/ml.

When examining the tests collected at diagnosis and at the time of reevaluation, the normalization of CPK, leukocyte and platelet values were statistically significant with a p value equal to 0.008; 0.004 and 0.0001 respectively. The variations in ESR and CRP were not statistically significant (Table 1).

DISCUSSION

Due to the paucity of information in the Brazilian literature and the lack of standardized approaches for the diagnosis and management of benign acute childhood myositis, patients are often approached with hospitalization or subjected to costly investigations until the diagnosis is established.

In an analysis of 316 patients, Agyeman et al, found a higher incidence in males (2:1), at a mean age of 8.5 years⁹. In a series of 25 Portuguese cases, the ratio found was 4.6 boys to one girl, with an average age of 7 years¹⁰. In the largest Brazilian series of 35 cases over a nine-year period, an incidence of 3.3 boys was found for one girl, with an average age of 7.5 years¹. The findings of the present study are in accordance with the literature, showing a higher prevalence in males (1.57: 1), with ages varying between 4.9 and 15 years.

Table 1. Laboratory parameters in the diagnosis and reassessment of acute viral myositis (Reference values: CPK (45-370 UI/L); Aldolase (5-15 UI/L); TGO (5-60 UI/L); TGP (5-40 UI/L); Leukogram (4000-1000 n/mm³); Platelets (150000-450000 n/mm³); CRP (<5 mg/dL); ESR (0 mm/hour).

Laboratory parameter	Initial value (average $\pm$ DP)	Final value (average $\pm$ DP)	T-test paired
CPK	3359,5 $\pm$ 3111,0	161,1 $\pm$ 95,3	0,008
Leukocyte count	4522,2 $\pm$ 1147,1	7220,0 $\pm$ 2616,0	0,004
Platelet count	188444 $\pm$ 40155	341917 $\pm$ 107451	0,0001
ESR	16,4 $\pm$ 13,3	19,0 $\pm$ 12,9	0,497

In studies carried out in Brazil and Portugal, a higher frequency was found in the months of May, June, July and September, which corresponds to the coldest months of the year, with a distribution similar to that reported by Santos.^{1,10} Despite the disagreements in our data, where the highest incidence was found in autumn (38.9%), followed by spring; 77.8% of the patients in this study had symptoms of airway infections in the week preceding the musculoskeletal condition.^{1,9,10}

The most frequent clinical manifestations at the time of diagnosis were pain and weakness in the calves, similar to those found in the aforementioned studies.^{1,9}

Cardin, Agyeman and Santos,^{1,9,10} also described the increase in CPK and the tendency towards leukopenia and thrombocytopenia found in this study. An interesting aspect of this study was the concomitant increase in transaminases (TGO and TGP) in 72.2% of cases. Despite this data, we emphasize the importance of requesting a complete assessment of muscle enzymes (TGO, TGP, CPK, aldolase and LDH), since not all of them will always be altered.

The results of this study, when paired with the samples, were similar to that of Santos: CPK normalization, elevation of leukocytes and platelets were statistically significant, whereas the values of CRP and ESR were not.

This study helped to create a new protocol, instituted in the emergency department of our Hospital. The PCR collection was not included in the initial screening tests for viral myositis, because it was not statistically significant, its request was at the discretion of the attending physician after evaluating the patient. The erythrocyte sedimentation rate, despite not obtaining statistical significance, was maintained, as it is a relevant finding in patients with autoimmune muscle diseases. The only patient excluded from the study due to an error in diagnosis, had high ESR values and was later defined as dermatomyositis.

Research on respiratory viruses and serologies for Herpes Viruses, Cytomegalovirus and Epstein Barr was added to the new protocol. We emphasize that these exams make up an initial panel to which other serologies can be added

depending on the clinical signs and symptoms and patient origin, such as arboviruses.

In our study, muscle enzyme levels did not show a correlation with clinical severity, prognosis, need for hospitalization or the development of complications, such as rhabdomyolysis.

The treatment of viral myositis is symptomatic, including analgesia using NSAIDs and bedrest. In our study, the NSAID used was naproxen, prescribed at a dose of 20 mg/kg/day. Its choice was based on ease of dosing and proven efficacy in osteoarticular diseases. The use of antivirals has not proven to be beneficial, and thus it is not recommended¹¹.

Despite descriptions of slow recoveries, over a period of up to 30 days, symptom improvement usually occurs in one week, taking an average of three days, similar to what was found in our study^{1,9}.

Among the limitations of the present study, we highlight the small sample, it was held within the limits of a single hospital, but the prospective design promotes an improved description of clinical and epidemiological data. The greater familiarity with the disease enabled us to make a quick and accurate diagnosis, as well as an effective treatment, avoiding burdens on the healthcare system and preventing children from being unnecessarily subjected to invasive procedures such as imaging exams, muscle biopsy and venipuncture puncture.

Finally, we emphasize the importance of knowing this condition and its progress, as well as the need for a careful interview and a complete physical examination, so that conditions that are more serious are not delayed in diagnosis.

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