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Benign childhood acute myositis: clinical and laboratory findings of 15 cases

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Summary

Aim: In this study, clinical and laboratory findings of 15 cases with benign acute childhood myositis are presented to review pathognomonic findings of the disease.

Material and Method: Fifteen typical cases with benign acute childhood myositis referred to our Pediatric Neurology Clinic because of inability to walk from 15th of January to 15th of March 2011 were enrolled into this study. Eighty percent of the cases were male and the mean age was 6.3 years. Guillian-Barre's syndrome was the most common initial diagnosis (47% of the cases).

Results: The prodromal period lasted for 2- 10 days and common symptoms included sore throat, fever and cough. When they were referred to our clinic, the patients complained of pain in both calves and inability to walk. The symptoms resolved rapidly and spontaneously in 3 days. Leucopenia was found in 87% of the patients, thrombocytopenia was found in 47% of the patients and mildly high alanine transaminase was found in 87% of the patients. Serum creatinine kinase level ranged from 580 to 8250 U/L and returned to normal within one week.

Conclusions: In patients whose clinical and laboratory findings are suggestive of benign acute childhood myositis, it is important to consider the diagnosis early in terms of preventing unnecessary laboratory evaluations and different therapeutic approaches. *(Turk Arch Ped 2012; 47: 55-8)*

Key words: Childhood, CPK, gait abnormalities, Guillian-Barré syndrome, myositis

Introduction

Benign acute childhood myositis (BACM) is a benign picture which is usually characterized by sudden bilateral calve pain and inability to walk following viral infections in school-aged children and which resolves spontaneously (1-6). It was described by Lundberg (1) in 1957 for the first time in 74 pediatric cases. It occurs in boys more frequently. The typical laboratory finding is high serum creatinine phosphokinase (CPK) level (2,3). Differential diagnosis with Guillain-Barre syndrome (GBS) which leads to difficulty to walk and with more severe diseases which proceed with myoglobinuria is important (3). Detailed knowledge of the characteristics of benign acute childhood myositis will prevent unnecessary tests and treatment. In this study, the clinical and laboratory findings of 15 cases with BACM were presented to review the significant criteria of the diagnosis of BACM.

Material and Method

A total of 15 BACM cases who were referred to our Pediatric Neurology Clinic with different prediagnoses between January the 15th and March the 15th 2011 from different pediatric clinics where they presented with the complaint of inability to walk were included in our study. The prediagnoses at presentation included GBS (46.6%) and transverse myelitis (13.3%). In 40% of the patients no prediagnosis was stated. All patients were referred on the first day when the complaint of inability to walk started.

The mutual features of the patients:

1. Presence of sudden bilateral calve pain, inability to walk or gait disturbance
2. Upper respiratory infection before and/or during the attack
3. Moderate increase in serum CPK level
4. No marked pathology on neurological and muscle examination
5. Improvement of the clinical picture in 48 hours

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None of the patients had trauma, familial muscle disease, history of dark urine, arthritis, skin eruption, prolonged fever or systemic disease finding including nephritis.

On the initial evaluation, complete blood count, serum CPK, aspartate aminotransferase (AST), alanine aminotransferase (ALT), complete urine analysis, C-reactive protein (CRP) and renal function tests were ordered urgently. Neurologic examination was performed every day on the first three days and repeated at the first week. Electromyogram (EMG) was performed in a total of four patients. Complete blood count, CPK, ALT, AST were repeated on the 3rd day and at the first week. To elucidate the etiology toxoplasma, M. Pneumonia, rubella, adenovirus, parvovirus, Ebstein Barr virus, herpes simplex virus and hepatit markers were ordered.

Results

12 (80%) of the 15 patients included in the study were male and three (20%) were female. The age ranged between 3 and 10 years and the mean age was 6.3±2.1 years. In the prodromal period, all patients had sore throat (100%), 14 patients (93.3%) had fever, 14 patients (93.3%) had cough, three patients (20%) had vomiting, one patient (6.6%) had otitis and one patient (6.6%) had diarrhea (Table 1). In three patients (20%), fever was continuing at presentation to our clinic. 14 of our subjects (93.3%) could not stand and walk when they presented to our clinic. One subject could only achieve the position of standing on fingertip, but could not walk. Although pain and stiffness was observed in the muscles of the calves in all patients, their muscle strengths were perfect. No abnormal finding was found on neurological examination in any of the patients except for decrease in deep tendon reflexes in one patient (6.6%). Leucopenia was found in 13 of the subjects (86.6%), thrombocytopenia was found in 7 (46.6%). CRP was negative in all patients. Serum CPK values ranged between 580 and 8250 U/L (mean 2 970±2 190) (Table 2). EMG results were found to be normal in a total of four patients including the patient whose deep tendon reflexes were decreased. Improvement in signs and symptoms during the clinical course started in 24-48 hours in all subjects and was completed in 3 days at the latest. The patients achieved firstly the position of standing on fingertip. Afterwards, walk on fingertip started in 11 subjects (73.3%) and broad based walking started in four subjects (26.6%). Serum CPK levels started to decrease after the third day and reached normal values in all patients at the end of the first week. In the tests performed to find the etiology, human parvovirus B 19 was found in only one patient.

Discussion

Benign acute childhood myositis is a condition which is characterized by sudden calf pain, inability to walk or difficulty in walking occuring after a prodromal period of 1-5 days following a viral upper respiratory disease and which resolves spontaneously (3). Previous studies reported the prodromal period to range between 2 and 31 days (1-5,10). In our subjects, the prodromal period was 2-10 days. Sudden bilateral calf pain which is reported to be the typical clinical finding was

Table 1. Characteristics of the prodromal period of the patients

Patient Number	Age /Gender	Fever	Sore throat	Cough	Vomiting	Otitis	Diarrhea	Prodromal period (days)
1	8/M	-	+	+	-	-	-	7
2	4/M	+	+	+	-	-	-	3
3	7/M	+	+	+	-	-	-	3
4	3/F	+	+	-	-	-	-	10
5	6/M	+	+	+	-	-	-	3
6	8/M	+	+	+	-	-	-	7
7	10/M	+	+	+	+	-	+	3
8	3/F	+	+	+	-	-	-	7
9	6/M	+	+	+	-	-	-	2
10	6/M	+	+	+	-	+	-	2
11	6/M	+	+	+	+	-	-	2
12	10/M	+	+	+	-	-	-	7
13	6/F	+	+	+	+	-	-	3
14	6/M	+	+	+	-	-	-	5
15	5/M	+	+	+	-	-	-	3

M: male; F: female.

Table 2. Initial laboratory findings of the patients

Patients number	CK (U/L)	ALT (U/L)	Leukocytes (/mm^3)	Thrombocytes (/mm^3)
1	3070	108	3 600	176 000
2	787	67	3 800	220 000
3	4040	205	2 600	255 000
4	804	41	7 700	424 000
5	3172	109	3 000	200 000
6	5002	322	2 400	80 000
7	4640	166	3 180	144 000
8	580	61	3 800	109 000
9	1512	78	2 600	149 000
10	680	26	4 000	268 000
11	3265	291	2 900	188 000
12	8250	229	3 100	138 000
13	2294	108	3 000	141 000
14	5227	78	3 200	145 000
15	1237	65	3 500	191 000

present in all of our patients (1-3). This pain started after getting out of the bed (after a long rest) as described (7-9). Our patients had complaints including inability to step on the feet, inability to stand and inability to walk. Therefore, all families presented to pediatric clinics (frequently to the emergency department) with a high degree of axiety with their children on their laps. The most common prediagnosis in the patients who were referred to our Pediatric Neurology Clinic from other clinics was GBS (in 46.6% of our subjects).

In a study which evaluated 32 subjects with benign acute childhood myositis, 78% of the subjects were reported to be referred by a family practitioner or pediatrician with different diagnoses other than BACM (3). These diagnoses included GBS (37%), transverse myelitis (18.8%), meningitis (12.5%) and postinfectious cerebellitis (9.4%) (3). Rajajee et al.(10) reported that BACM subjects were referred to clinics most frequently with a diagnosis of GBS. However, in GBS, bilateral muscle weakness progressing from bottom to top and pain are observed, decreased deep tendon reflexes are found on physical examination and serum CPK level is within normal limits. There are many reasons which may lead to muscle pain in children. During the course of viral infections, widespread myalgia can be observed, but BACM is differentiated with typical bilateral gastrocnemius muscle involvement. In muscular distrophies which are considered in differential diagnosis because of high serum CPK level, unclear pain and lack of improvement in clinical findings during the course of the disease are significant clues. Growing pains should be considered in the differential diagnosis though it does not cause difficulty in walking (11). Dermatomyositis which may cause a similar picture in children is characterized with progressive bilateral proximal muscle weakness, increase in CPK level, EMG changes, extraordinary muscle biopsy findings and typical cutaneous findings (12,13). Pyomyositis is a purulent infection which primarily involves skeletal muscle and it should be considered in the differential diagnosis, since high CPK levels have been reported in cases with pyomyositis (14,15).

The fact that no pathology was found in 14 of our subjects on neurological and muscle examination was a significant finding in terms of the diagnosis of BACM. It has been reported that gastrocnemius, soleus and hamstring muscles may be affected and rigitidy in these muscles may be observed on examination (10). In this context, Mackay et al. (2) defined two types of gait disturbances including widebased walking and walking on fingertip. Zafeiriou et al.(3) reported that muscle strength was full in all their patients with BACM and deep tendon reflexes were decreased in 15.6% (3). While muscle strength was full in all of our subjects, deep tendon reflexes were found to be decreased in only one of our subjects (6.6%). Walking on fingertip was observed in 73.3% of our patients who started to walk and wide based walking was observed in 26.6%. In all of our subjects, clinical improvement started in 24 hours at the latest and complete improvement occured in 3 days.

The most typical and diagnositc finding for benign acute childhood myositis is high serum CPK level (2,3). Moderately high (558- 6800) CRP is being reported (3). In cases with higher levels, it is recommended to pay attention in terms of rhabdomyolysis and renal failure and to monitor the color of urine (16). CPK levels in our subjects ranged between 582 and 8250 U/L and started to decrease on the third day. In all subjects, CPK levels were observed to have returned to normal values. In addition, other laboratory findings included transient increase in ALT/AST, leucopenia and thrombocytopenia (2,3,10,17,18). Rajajee et al.(10) reported leucopenia with a rate of 27.5%, thrombocytopenia with a rate of 40% and increased ALT, AST levels with a rate of 70%. In 86.6% of our subjects, ALT levels returned to normal or near-normal values in one week. Negative CRP, transient leucopenia with a rate of 86.6% and thrombocytopenia with a rate of 46.6% supported viral etiology.

The etiopathogenesis in benign acute childhood myositis is controversial. The most recent theories may be summarized as immune processes triggered by viral agents or demage caused by invasion of viral particles in the muscle tissue (2). The second theory was supported by investigators who showed that viral particles were found in muscle biopsies in patients with BACM (25). However, nonspecific degenerative changes and muscle necrosis are usually observed in muscle biopsies (10). In the series reported in the literature, it is reported that BACM is associated with influenza epidemic (especially influenza B) and other agents (adenovirus, parainfluenza, rotavirus, Coxsackie, Dengue virus, herpes simplex virus, Ebstein Barr virus, M. Pneumonia etc.) (2-5,10,19-24). BACM is frequently observed epidemically and most cases (60%) occur in winter months (2,3). Although detailed viral etiology was not

investigated in this study, human parvovirus B19 was found in one of our subjects. Although we could not determine etiologic agents, we can state that viral agents are involved in our cases who presented consecutively in a two-month period in the winter considering the prodromal symptoms including fever, sore throat and cough in addition to the above mentioned laboratory findings.

In cases of BACM, EMG is frequently normal or patchy myopathic changes are found (1,5,6). In our four subjects in whom EMG was performed, the result were found to be normal.

While the mean age in our subjects was 6,3 years, it was found to be 5,3-9,2 years in previous series (4,10). Although this was related to genetic predisposition or the fact that boys are more restless, the cause is not known exactly (2,10).

Conclusively, in cases of sudden bilateral leg pain and inability to walk following an upper respiratory infection especially in winter months, BACM should be considered as a prediagnosis, if neurological and muscle examination is normal and CKP is found to be moderately high. Making a diagnosis of BACM by observing a rapid improvement in clinical findings in follow-up is important in terms of avoiding unnecessary tests and the differentiation of BACM which resolves spontaneously and other conditions which can show a more severe course and require different therapeutical options.

Conflict of interest: None declared.

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