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Diaphragmatic Paralysis during Mediterranean Spotted Fever (MSF)

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Background

Rickettsia conorii is epidemic during summer in Algeria. Uncommon manifestations of MSF, including neurological manifestations such as mononeuropathy, encephalopathy, transverse myelitis, polyradiculopathy and Guillain-Barre syndrome have also been recognized in the past.

Case Presentation

A 34 year-old man from Oran, was admitted with a seven day history of high fever and a generalized rash. He had a history of animal contact with the household dogs of the family. He had a left basithoracic pain. He had no cough on admission and did not complain of breathlessness.

He denied any recent travel to Malaria endemic regions. He had no previous co-morbidities of significance. His physical examination revealed generalized rash and a black escharr on his scrotum **(Figure 1)** on admission and a body temperature of 39.8°C. His full blood count revealed a total white blood cell count of 5700/mm^3 with normal differentials, hemoglobin 14.4 g/dl and with a platelet count of 120.000/mm^3 (**Table 1**). The Chest X-ray on admission showed an elevated left hemi-diaphragm .MSF was confirmed with a positive MSF IgM antibodies. He was managed with intravenous fluids and doxycyclin. He did not complain of paroxysmal nocturnal dyspnoea. He had no wheezing, or palpitations. Physical examination at this point revealed normal vital parameters with decreased breath sounds of the left lung base with a stony dull percussion note.

Ultrasound scan chest did not reveal any pleural effusion but noted reduced diaphragmatic movements with respiration on the affected side. ECG and doppler echocardiography did not show abnormalities. A thoracoabdominopelvian computed tomography did not reveal any lung parenchymal or mediastinal abnormalities. An oesogastroduodenal transit did not reveal diverticulitis or hiatal hernia. His lung function test revealed a sitting forced vital capacity of 2.11 L and supine 1.4 L (difference of 34%). He subsequently underwent nerve conduction studies of the phrenic nerves which revealed a decreased conduction amplitude on the left side suggestive of a demyelinating neuropathy, but was not suggestive of Guillain-Barre syndrome. There was no evidence of a neuromuscular junction disorder as suggested by normal electromyography. He was managed and recovered within a few days of hospitalization and was discharged home.

Discussion

Neurological involvement has been reported in 28% of patients with Mediterranean spotted fever and constitutes a negative

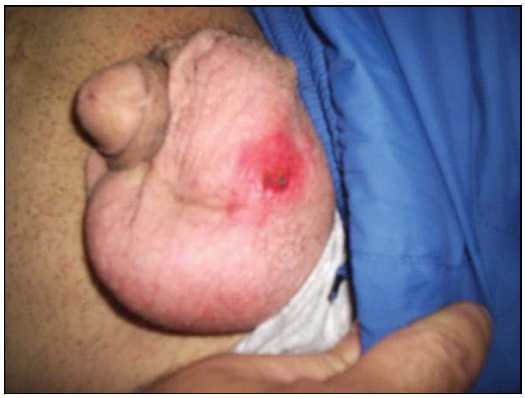

Table 1. Results of the laboratory tests.

	Results	Normal	units
WBC	5700	5.2-12.4	$x10^3$/ml
RBC	5020000	4.7-6.1	$x10^6$/ml
HGB	14.4	14-18	g/dl
HTC	50.4	42-52	%
platlets	120 000	(130-400)	$x10^3$/ml
neutrophils	70	(40-74)	%
lymphocytes	19	(19-48)	%
monocytes	7	(3.4-9))	%
eosinophils	2.5	(0-7)	%
basophils	1.5	(0.0-1.5)	%
Triglycerids	1.10	0.4-1.5	g/l
cholesterol	1.71	1.30-2.00	g/l
Alanine transferase	84	0-45	U/l
Aspartate aminotransferase	34	0-45	U/l
glucose	0.99	0.7-1.10	g/l
creatinine	9	6-13	mg/l

prognostic factor for the outcome of the disease [1,2].Unusual neurological manifestations of the disease such as encephalitis [3], Guillain-Barré syndrome [4],facial palsy [5] were reported.

Our patient had confirmed MSF. The indirect immunofluorescence test is generally considered to be a sensitive and specific test for confirming the diagnosis of R conorii infection. Titre above 1/150 for IgM antibodies is suggestive of acute infection. In a second sample, the diagnosis was confirmed by a fourfold increase of the specific antibody, four weeks after the onset of the disease.

In this report we presented a case of MSF with left diaphragmatic paralysis, as confirmed by nerve conduction study and lung function tests. A myopathy or a neuromuscular junction pathology were excluded by normal electromyography. Phrenic nerve compression by mass lesions was excluded by a normal thoracoabdominopelvian CT and oesogastroduodenal transit.

The presence of f waves on nerve conduction study made Guillain-Barre syndrome an unlikely cause although there was evidence of demyelination.

Post viral phrenic neuropathy has previously been documented following Polio-virus infection [6], Herpes Zoster infection [7], dengue [8], and Human Immunodeficiency virus infection [9].

Conclusions

With Mediterranean spotted fever, more unusual complications are to be expected. In such light isolated phrenic nerve palsy causing diaphragmatic paralysis should be considered a recognized complication of this disease. In conclusion, the outcome of such cases associated with rickettsioses seems excellent.

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