Compartment syndrome: A very rare complication of Crimean –
Congo haemorrhagic fever

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ABSTRACT
Background: Crimean-Congo haemorrhagic fever (CCHF) is a worldwide distributed lethal disease. Miscellaneous complications have been reported, however, compartment syndrome is a very rare complication.

Patients: We report two cases of compartment syndrome of the left and right upper limbs following haemorrhage due to Crimean-Congo haemorrhagic fever.

Conclusion: These cases suggest that CCHF should be included in the differential diagnosis of patients with compartment syndrome. Patients are at risk of developing compartment syndromes especially in the forearm. We emphasize the importance of clinical signs in the early diagnosis of this potentially serious complication of viral hemorrhagic fevers.

Keywords: Crimean-Congo haemorrhagic fever, Compartment syndrome, Viral hemorrhagic fever.

INTRODUCTION

Acute compartment syndrome (ACS) is a limb-threatening and life-threatening condition observed when perfusion pressure falls below tissue pressure in a closed anatomic space (1-3). Almost any injury can cause this syndrome (1).

Crimean Congo hemorrhagic fever is a viral disease characterized by fever, thrombocytopenia, myalgia, subcutaneous bleedings and disseminated intravascular coagulation (4). Despite, direct infection of the endothelium and indirect damage by viral factors or virus-mediated host-derived soluble factors that cause endothelial activations and dysfunction, compartment syndrome is a very rare complication in this disease (2,4).

PATIENTS
Case 1:
The first patient was a 30-year-old man who became ill on July 2002, 7 days after he had butchered a goat. He was admitted to the Boo-Ali hospital (Zahedan, Southeast of Iran) because of fever, headache, myalgia, and hemorrhagic rashes. On physical examination, he appeared ill. His temperature was 38.8°C, and he was tachycardic and tachypneic. Pharyngeal mucosa was
erythematous. On abdominal examination, he had epigastric tenderness. A diffuse maculopapular rash and petechiae were seen over the entire body, predominantly, on the chest. On the first day of admission, oral and nasal bleeding occurred. Blood sample showed pancytopenia and platelet count was very low (15,000 platelets/mm³). Serum sample was taken and sent to reference laboratory for further evaluation of CCHF virus by RT-PCR. Treatment was begun with oral ribavirin and intravenous ceftriaxone, along with platelet transfusions and supportive therapy. Three days later, he was better but he complained of pain and burning in right upper limb from the ankle to the wrist. On physical examination, he had pain and it was severe with any movement. Large ecchymoses were seen at venipuncture sites on the volar portion of the right forearm. There was a diffuse ecchymotic rash and swelling in the right arm and also a hematoma within the arm muscles, just above the ankle on sonography. He was referred to neurologist. Electromyography (EMG) findings and clinical manifestations confirmed the diagnosis of acute compartment syndrome. Then, a surgeon was consulted. The result of an IgM enzyme-linked immunosorbent assay (ELISA) for Crimean-Congo hemorrhagic fever virus was positive on the 7th day of admission.

Case 2:
The second patient was a 29-year-old Afghan woman who was referred to Boo-Ali hospital with epistaxis and fever on April 2003. She packed her nose to stop the bleeding without wearing the gloves and taking any other protective measures. She had a high-grade fever (38.8°C) with shivering and severe body aches. She had received anti-malarial drug (chloroquine), antibiotics and strong analgesics four days ago when she had experienced fever and shaking chill. Despite all these medication her temperature and body aches did not improve. Her physician suspected that she may have developed (viral haemorrhagic fever) VHF and advised her to test the platelets count. Initially the platelet count was 30,000/ul, which kept on decreasing and dropped down to 10,000/ul. Then, she tested for CCHF and received oral ribavirin and platelet. On the 4th day of admission, she was afebrile and did not develop any bleeding. But, there was a diffuse ecchymotic rash on the left arm and forearm. On physical examination, she had severe pain and burning sensation in the left forearm that was aggravated by movement. Electromyography and clinical findings suggested compartment syndrome. She referred to neurologic department for further evaluation. The results of both IgM and IgG ELISAs were positive on the 12th day of admission.

DISCUSSION
Acute compartment syndrome (ACS) has been described in association with trauma, crush injury, ischemia, and reperfusion episodes (2-5,7). Non-traumatic ACS occurs most commonly in the lower leg, but has also been reported in the forearm following muscle overuse, undue exertion, and bleeding diathesis (2,3). It requires urgent diagnosis and treatment (2,6).

CCHF virus infection was rarely reported in Iran before 1999 (8,9). Since spring 1999 epidemic of this infection has been occurred in some regions of Iran especially in Sistan and Baluchistan, Southeast of Iran (10,11). Now, Sistan and Baluchistan Province is an endemic region for CCHF and there are a few reports of very rare complications of this disease from Sistan and Baluchistan in the literature (2,12-14). For example, Moghtaderi and his colleagues reported a case of intracerebral haemorrhage due to CCHF in a young man in Zahedan (12). They believe that this is the first patient ever described with parasagittal intracerebral hematomas and gait disorder secondary to CCHF. Meanwhile, he reported the first case of compartment syndrome due to Crimean-Congo hemorrhagic fever in a 45-
year-old man (2). In recent case, compartment syndrome was confirmed by EMG. We reviewed the literature and found a few cases of very rare complications in patients with CCHF that occur due to vascular injury and bleeding in internal organ. Ardalan reported a patients with CCHF who had thrombotic microangiopathy and acute renal failure (13). Ahmati from Kosovo presented a case with CCHF who had severe epigastric tenderness on palpation. Abdominal ultrasonography revealed hepatosplenomegaly and the presence of free liquid in the abdominal cavity, suggestive of hemoperitoneum. In a patient with CCHF during the acute phase of infection, clinical features commonly show a dramatic progression characterised by hemorrhage and bleeding markers are prolonged (7,8,11). Besides direct infection of the endothelium, indirect damage by viral factors or virus-mediated host-derived soluble factors that cause endothelial activations and dysfunction are thought to occur. In the mean time, thrombocytopenia is a risk factor for occurrence of haemorrhage (7,11). Our patients had severe decreased platelet count and repeated measuring of blood pressure was an injury and major cause for haemorrhage within the muscles. Blood sampling is also an important cause of interfascial hemorrhage. Venipuncture sites should be controlled and compressed carefully to prevent continuous bleeding. Reducing blood sampling and controlling venipuncture sites are useful measures for deceasing interfascial hemorrhage in viral hemorrhagic fevers. Progressive increase in muscle compartment pressure may be prevented by early recognition, conservative therapy, and elevation of the affected compartment. Measuring of blood pressure using the involved limb especially with diffuse echymoses could be an inducing factor, especially when venous pressure is less than tissue pressure. Differential diagnosis includes, cellulitis, deep venous thrombosis, gas gangrene and necrotizing fasciitis (1,7).

In conclusion, these cases suggest that VHF including; CCHF infection should be included in the differential diagnosis of patients with compartment syndrome. Patients are at risk of developing compartment syndromes especially in the forearm. We emphasize the importance of clinical signs in the early diagnosis of this potentially serious complication of viral hemorrhagic fevers.

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REFERENCES


