

## Acute Compartment Syndrome of Upper Limb as an Unusual Complication of Dengue Hemorrhagic Fever

Dear Editor,

Dengue fever with its hemorrhagic complications is a common viral illness in India and other south Asian countries. We present a case of dengue hemorrhagic fever associated with capillary leak syndrome presenting as a compartment syndrome involving the right upper limb in the background of iron deficiency, which we believe to be the first such presentation of dengue in medical literature.

Our patient, a 40-year-old man, presented with high grade continuous fever, retro-orbital headache, and generalized body ache for 5 days along with scanty gum bleeding for 2 days. He also had red spots and swelling of the right forearm for 2 days, the swelling being gradual in onset, painful, reddish in appearance, and rapidly progressing to restrict movements of the right upper limb. There was no history of trauma, insect bites, rash, drug reaction, or past history suggestive of any coagulation disorder.

On examination, the patient was febrile, conscious, oriented, had mild pallor, and active gum bleeding in the oral mucosa. There was a warm, tense, tender, erythematous swelling involving the entire right forearm, wrist, hand, and all fingers, with hypoaesthesia involving all sensory modalities but without any ulceration or discharge. Both active and passive movements at all joints involving the right hand and wrist were severely painfully restricted and resisted. Laboratory tests revealed hemoglobin (Hb) 7.2 g/dL, platelets 15,000/ $\mu$ L and Hematocrit 58%, and erythrocyte sedimentation rate (ESR) of 36 mm in first hour. Abnormal cells or parasites were absent on the peripheral smear. Biochemical tests including liver function test (LFT), kidney function test (KFT), electrolytes and urine routine, and microscopic examination (RE/ME) were all normal. Malarial antigen tests were repeatedly negative. Dengue IgG and IgM were strongly positive and an initial diagnosis of dengue hemorrhagic fever was entertained. Disseminated intravascular coagulation (DIC) was ruled out with

a normal serum level of fibrin degradation products (8.2  $\mu$ g/dL). Platelet transfusion support, fluids, and paracetamol were instituted. Chest X-ray, X-rays of the right arm, forearm, and hand were all normal. Ultrasonic (USG) Doppler of the right forearm documented gross soft tissue swelling with no compromise of the venous or arterial blood flow. Orthopedic consultation concurred with our diagnosis of right forearm compartment syndrome with Volkmann's Ischemic contracture (VIC) grade III.

Despite low platelets, high risk surgical decompression of the right forearm was performed after 2 days with blood component support, and extensive muscle swelling discoloration was demonstrated during operation. Large clots were removed from the anterior compartment and the muscles began to regain their natural color immediately following fasciotomy. Post-operative recovery was uneventful over the next 12 days [Figure 1], without any fever, secondary infection, or swelling on any other site of the body. Hb and platelet counts subsequently rose to 8.6 g/dL and 1,10,000/ $\mu$ L, respectively. Secondary closure of right forearm fasciotomy was done at 2 week follow-up.

As documented in the literature, the cardinal feature of dengue hemorrhagic fever (DHF) results from a transient increase in vascular permeability that causes fluid leakage from the intravascular to the interstitial compartment, leading to hemoconcentration, and circulatory collapse, which contribute to mortality and significant morbidity.<sup>[1]</sup> Capillary leak syndrome is seen in various situations, sepsis and dengue being the major infectious causes.<sup>[2]</sup> The mechanisms of capillary leak



**Figure 1:** Photograph of the right forearm 3 days after emergency fasciotomy. The edematous muscles of the anterior compartment of the right forearm are visible

may involve increased hydrostatic pressure, decreased oncotic pressure, or increased permeability of the capillary endothelium.<sup>[3]</sup> Among the other infectious causes of capillary leak leading to compartment syndrome, Brucellosis, Hantavirus infection, and sepsis have been mentioned in literature.<sup>[4]</sup>

Idiopathic systemic capillary leak syndrome (SCLS) or Clarkson's disease is another important cause of capillary leakage, where there are recurrent episodes of hypovolemic shock, edema, hemoconcentration, and hypoalbuminemia. Episodes of systemic capillary hyperpermeability due to various cytokines have been implicated in the pathogenesis of this disorder.<sup>[5,6]</sup> The most common bleeding manifestations of dengue include petechiae, gum bleeding, GI bleeding and hemoptysis although rarer sites are often involved.<sup>[7]</sup> Even after thorough clinical and radiological evaluation (chest skiagram and abdominal ultrasound), we did not find evidence of capillary leakage at the common sites like pleural cavity, pericardial cavity, and peritoneum. Workers from South India have shown that bleeding in dengue might lead to abdominal compartment syndrome in rare cases.<sup>[8]</sup> The solitary involvement of the right forearm in this scenario is indeed uncommon, and adds to the clinical interest of this case.

In conclusion, internists must be aware of these different forms of dengue complications, where urgent and aggressive interventions may enable physicians to salvage limbs and prevent mortality. We report this case to underline the importance of internal bleeding and capillary leak in dengue, as also to record a clinical scenario as yet unreported in the world literature.

**Dipanjana Bandyopadhyay,  
Pradip Mondal, Samiran Samui,  
Saptarshi Bishnu, Sukanta Manna**

*Department of Medicine, MCH Building, Medical College,  
Kolkata, India. E-mail: drsamiransamui@gmail.com*

## References

1. Wills BA, Oragui EE, Dung NM, Loan HT, Chau NV, Farrar JJ, *et al.* Size and charge characteristics of the protein leak in dengue shock syndrome. *J Infect Dis* 2004;190:810-8.
2. Dhir V, Arya V, Malav IC, Suryanarayanan BS, Gupta R, Dey AB. Idiopathic systemic capillary leak syndrome (SCLS): Case report and systematic review of cases reported in the last 16 years. *Intern Med* 2007;46:899-904.
3. Erkurt MA, Sari I, Gül HC, Coskun O, Eyigün CP, Beyan C. The first documented case of brucellosis manifested with pancytopenia and capillary leak syndrome. *Intern Med* 2008;47:863-5.
4. Chang B, Crowley M, Campen M, Koster F. Hantavirus cardiopulmonary syndrome. *F Semin Respir Crit Care Med* 2007;28:193-200.
5. Takabatake T. Systemic capillary leak syndrome. *Intern Med* 2002;41:909-10.
6. Cicardi M, Gardinali M, Bisiani G, Rosti A, Allavena P, Agostoni A. The systemic capillary leak syndrome: Appearance of interleukin-2-receptor-positive cells during attacks. *Ann Intern Med* 1990;113:475-7.
7. Said SM, Elsaed KM, Alyan Z. Benign acute myositis in association with acute dengue viruses' infections. *Egypt J Neurol Psychiatr Neurosurg* 2008;45:193-200.
8. Kamath SR, Ranjit S. Clinical features, complications and atypical manifestations of children with severe forms of dengue hemorrhagic fever in South India. *Indian J Pediatr* 2006;73:889-95.

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