Acute Compartment Syndrome- A Rare Complication of Dengue Fever

Authors

Dr Shruthi R*, Dr Adithi Bhandary2, Dr Sudhindra Rao3

1 Junior Resident, Department of General Medicine, K.S. Hedge Medical Academy, Deralakatte, Mangalore
2 Assistant Professor, Department of General Medicine, K.S. Hedge Medical Academy, Deralakatte, Mangalore
3 Professor and unit chief, Department of General Medicine, K.S. Hedge Medical Academy, Deralakatte, Mangalore

*Corresponding author
Dr Shruthi R

Abstract

Background: Dengue fever is a febrile illness caused by infection with one of 4 dengue viruses transmitted by Aedes aegypti. Infection may be asymptomatic or present with a broad range of clinical manifestations including a mild febrile illness to a life-threatening shock syndrome. Incidence of Dengue has increased exponentially with half of world’s population at the risk of infection. We present a case report of 38 year old male who was admitted to our hospital with dengue fever with thrombocytopenia. During his course in hospital he developed swelling over the right forearm and arm which progressed to acute compartment syndrome and emergency fasciotomy was done and his limb was salvaged. This illustrates a rare but an important and potential complication of Dengue fever and also the importance of timing of surgery.

Keywords: Dengue fever, compartment syndrome, fasciotomy, thrombocytopenia.

Introduction

Compartment syndrome is a painful condition caused by the increased intracompartmental pressure within a closed osteofascial compartment. It can present both acutely and as chronic syndrome. Acute compartment syndrome is a surgical emergency, and unless the pressure is relieved quickly, necrosis of the soft tissues and permanent disability may occur. Usually the leg and forearm are the most frequently affected, but it can also involve the arm, hand, foot, and buttock(1). Compartment syndrome can be caused by an increase in tissue volume within a compartment, thereby increasing the pressure, or externally applied pressure compressing a compartment. The commonest cause is trauma, usually after a fracture(2). The treatment is emergency fasciotomy. Here we are reporting a case of an acute compartment syndrome of right upper limb due to intravenous cannula insertion in a patient with dengue haemorrhagic fever.

Case Report

A 38-year-old male admitted to medical ward with the complaints of fever, myalgia, arthralgia, bifrontal throbbing type of headache and nausea for 4 days. On examination, patient was hemodynamically stable. His blood investigations revealed a classic dengue like picture with white blood count of 2,300 c/cumm, platelets of 48,000c/cumm, haemoglobin of 13.9g/dl, hemotocrit of 42.8 and normal coagulation profile. Renal parameters, liver function tests and other investigations were within normal limits. Dengue card test was done and NS1 antigen was positive.
On day 2 of admission he had to be shifted to intensive care unit in view of low platelets (12,000 c/cumm). He did not have any bleeding manifestations in the morning and 4 units of platelets were transfused. In the afternoon patient developed pain and swelling of the right upper limb where an intravenous canula was inserted. On examination, gross swelling of right arm and forearm was noted. There was no redness/local rise of temperature of the limb. Peripheral pulse and circulation were good. Blood investigation showed white blood cells 17,900c/cumm, platelets 35,000c/cumm, haemoglobin 11g/dL, haematocrit 32.8% with normal coagulation profile. A venous doppler was done which showed no evidence of deep vein thrombosis. Emergency Surgery opinion was sought as the swelling increased in size. They noted that the compartment was tense. Subsequently, the pain and swelling worsened after brief period of close observation with the appearance of multiple blebs and bruises at the right cubital fossa (figure 1), a diagnosis of impending compartment syndrome was made and emergency fasciotomy was planned.

In view of severe thrombocytopenia, platelet concentrates and fresh frozen plasma were transfused while preparing patient for surgery. Emergency fasciotomy of forearm and arm with carpal tunnel release was performed under general anaesthesia. Multiple incisions were made over the volar aspect of forearm and lateral aspect of arm releasing the fascia and intermuscular septae’s. Intraoperative findings noted large amount of blood clots. Blood clots were evacuated and haemostasis were secured. Postoperatively, the muscles were healthy with good distal circulation (Figure 2 & 3).

Patient was started on injection piperacillin and tazobactum and continued daily dressing of the wound. Swabs from the fasciotomy site was sent for culture and sensitivity, which showed Acinetobacter baumanni complex and antibiotics was changed to Tigecycline according to culture sensitivity. Patient recovered gradually and was shifted to ward. Once patient recovered from dengue, he was transferred to surgery for secondary closure of the wound.
Discussion

The World Health Organization consider dengue as a major global public health challenge in the tropic and subtropical nations. Dengue viruses are small, enveloped viruses that are members of the family Flaviviridae genus *Flavivirus*. Dengue virus is introduced into the skin when an infected mosquito, most commonly *Aedes aegypti*, takes a blood meal from a susceptible host. Viremia is detectable in humans 6 to 18 hours before the onset of symptoms and ends as the fever resolves. Both innate and adaptive immune responses induced by dengue virus infection are likely to play a role in the clearance of infection.

Figure 3: Post-operative- healthy muscle noted

Plasma leakage, due to an increase in capillary permeability, is a cardinal feature of dengue haemorrhagic fever but is absent in dengue fever. The enhanced capillary permeability appears to be due to endothelial cell dysfunction rather than injury. Endothelial cell activation and injury and activation of coagulation and fibrinolysis have been reported in dengue, particularly in severe infections. Abnormalities that have been described include increased numbers of circulating endothelial cells, elevated levels of von Willebrand factor, tissue factor, tissue plasminogen activator, and platelet activator inhibitor, and an increased fractional catabolic rate of fibrinogen$^{3,4}$. In 1997, the World Health Organization published a classification scheme describing three categories of symptomatic infection: dengue fever, dengue haemorrhagic fever, and dengue shock syndrome. In 2009, the World Health Organization introduced a revised classification scheme consisting of the following categories: dengue without warning signs, dengue with warning signs, and severe dengue$^{5}$. During the early febrile stage (the symptoms of which include fever, malaise, headache, body pains and rash), clinicians cannot predict which patients will progress to severe disease. Later, during defervescence, symptoms such as bleeding, thrombocytopenia of $<100,000$ platelets $\text{mm}^{-3}$, ascites, pleural effusion, haematocrit $>20\%$ and clinical warning signs, such as severe and continuous abdominal pain, restlessness and/or somnolence, persistent vomiting and a sudden reduction in temperature (from fever to subnormal temperature) associated with profuse perspiration, adynamia (loss of strength or vigor) and sometimes fainting, can be indicative of plasma extravasation and the imminence of shock$^{6}$. Acute compartment syndrome commonly develops in traumatized patients with distracting or neurologically inhibiting injuries. Time to diagnosis is the most important prognostic factor for these patients. Insufficient understanding of the natural history and limited evaluation of signs and symptoms primarily account for delays in diagnosis$^{7}$. Compartment syndrome occurs when the interstitial pressure within the compartment exceeds the perfusion pressure at the level of the capillary beds. Elevated intra-compartmental pressure leads to increased pressure at the venous end of the capillary beds causing increased hydrostatic pressure and further increase in intra-compartmental pressure, eventually leading to arteriolar compression. Loss of the perfusion pressure gradient results in the onset of...
ischemia and ultimately leads to cellular anoxia and death\(^{(8)}\).

Immediate management involves the identification and removal of external compressive forces, and releasing casts or dressings down to the skin. The limb should not be elevated and instead kept at the level of the heart so as not to decrease arterial flow any further. If the clinical features of acute compartment syndrome do not improve following simple measures, definitive surgical fasciotomy is required on an emergency basis\(^{(9)}\).

Delay in treatment may lead to tissue hypoperfusion causing muscle necrosis, Volkman’s ischemic contractures and loss of function of the affected extremity. Necrotic muscle is a significant risk factor for bacterial superinfection and may quickly be seeded by bacteria, and lead to sepsis. Necrotic muscle may therefore require repeated debridement and even possible extremity amputation\(^{(10)}\). There are few reported cases of compartment syndrome as result of capillary leakage and hematoma formation in patients with dengue fever and the comparison is highlighted in Table 1.

Table 1: Comparison of five different cases of acute compartment syndrome in patients with dengue fever

|                          | Khoo et al.\(^{11}\) | Bandyopadhyay et al.\(^{12}\) | Kamisan et al.\(^{13}\) | Kishan R S et al.\(^{14}\) | Shruthi R et al. |
|--------------------------|----------------------|-----------------------------|------------------------|---------------------------|-----------------|
| Time of onset            | Day 8 of illness     | Day 3 of illness            | Day 6 of illness       | Day 8 of illness          | Day 3 of illness |
| Gender                   | Female               | Male                        | Male                   | Male                      | Male            |
| History of local injury  | Arterial cannulation | None                        | Venous cannulation     | Venous cannulation        | None            |
| Location                 | Right arm            | Right forearm               | Right arm              | Right arm and forearm     | Right arm and forearm |
| Other leaking sites      | Pleural effusion, ascites | None                        | Pleural effusion       | None                      | Pleural effusion |
| Other bleeding sites     | None                 | Gum bleeding               | None                   | None                      | None            |
| Platelets at the time of onset | 7,000/cumm           | 15,000/cumm                | 42,000/cumm            | 15,000/cumm               | 12,000/cumm     |
| Outcomes                 | Recovered well       | Recovered well              | Ulnar and Radial nerve palsy | Recovered well           | Recovered well |

Conclusion

Acute compartment syndrome is a rare but potentially limb-threatening in patients with dengue fever. We highlight this case as we believe that once the compartment syndrome is clinically detected, even though there is thrombocytopenia, fasciotomy should still be performed as an emergency procedure to salvage the limb. We must be aware of deadly complications of dengue and also aggressive intervention to salvage the limb and to prevent mortality.

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