

Limb-threatening compartment syndrome: A rare complication of dengue fever

Saiful Azlan Bin Kamisan¹, Ren Yi Kow², Ed Simor Khan Mor Japar Khan², Sa'adon Bin Ibrahim¹

Abstract

Dengue fever, a common mosquito-borne disease in Malaysia, has a wide range of clinical presentations. Dengue shock syndrome, a potentially fatal complication of dengue, is associated with derangement of numerous haematologic markers. A compromised coagulation profile, coupled with a traumatic incident may lead to a serious haemorrhagic complication. We present a rare complication of acute compartment syndrome of the right upper limb in a 17-year-old gentleman who was admitted for decompensated dengue shock syndrome after an unsuccessful attempt of venous cannulation. An emergency fasciotomy was performed and the right upper limb was salvaged albeit with ulnar nerve and radial nerve palsy.

Introduction

Dengue fever is common in Malaysia, with 83,849 cases reported in year 2017 alone.¹ Dengue fever is a single stranded RNA virus which is transmitted by mosquitoes, specifically *Aedes aegypti* and *Ae. albopictus*. Dengue fever is an acute febrile illness as defined by the presence of fever and two or more features: rash, headache, arthralgia, leucopenia, retro-orbital pain, a hemorrhagic manifestation with a history of exposure to a dengue endemic area.² Complication of dengue fever can manifest in multiple systems in the human body, including the central nervous system (encephalopathy), gastrointestinal tract (hepatic dysfunction), cardiovascular system (myocarditis), respiratory system (pleural effusion) and coagulation profile (thrombocytopenia). We present a rare complication of acute compartment syndrome of the right upper limb in a patient who was admitted for decompensated dengue shock syndrome.

Case Report

Mr MA, a 17-year-old gentleman, presented to the emergency department with a 4-day history of fever, myalgia, arthralgia, reduced oral intake and epigastric

pain. Upon examination, he seemed lethargic with hypotension, with evidence of plasma leakage. His full blood count showed neutropenia ($3.4 \times 10^9/L$) and thrombocytopenia ($42 \times 10^9/L$), consistent with dengue infection. Dengue serology for non-structural antigen (NS-1), dengue IgM and IgG were all positive. His renal and liver profiles were normal. He was admitted and treated for dengue shock syndrome.

He remained hemodynamically stable while in the ward until day 6 of illness, where he complained of pain and swelling over the right arm after a failure of venous cannulation. The pain was increasing in nature, associated with numbness over the median, ulnar and radial nerve distribution and he had reduced range of motion. The vascular circulation of the right arm remained intact with good Doppler signal for all arteries.

He was diagnosed to have acute compartment syndrome of the right upper limb and an emergency fasciotomy was performed to salvage the right upper limb. A long incision was made over the anterolateral aspect of the forearm and distal aspect of the arm, releasing all the compartments together with carpal tunnel release and cubital tunnel release. Intra-operatively there was hematoma formation over the medial aspect of the cubitus region extending towards the elbow region and along the medial aspect of the arm towards the axillary region. The biceps brachii muscle appeared contuse and swollen.

Post-operatively he was admitted to the intensive care unit as he developed massive plural effusion. He remained stable and subsequently recovered. A secondary suturing and split skin graft was performed for wound closure and he was discharged well. Although the upper limb was salvaged, it was associated with ulnar and radial nerve palsy as evidenced by ulnar clawing and wrist drop of the right hand.

Discussion

Compartment syndrome is defined as a clinical presentation that occurs as the result of increase in tissue

¹Orthopaedic Department, Hospital Sultan Ismail, Johor Bahru, Johor, MALAYSIA

²Orthopaedic Department, International Islamic University Malaysia, Kuantan, Pahang, MALAYSIA.

Address for Correspondence:

Dr Ren Yi Kow, Orthopaedic Department, International Islamic University Malaysia, Kuantan, Pahang, MALAYSIA

E-mail: renyi_kow@hotmail.com

pressure, in a limited fascial space, causing compromise of the circulation and function of the tissue within.^{3,4} Acute compartment syndrome is an orthopaedic emergency as a delay in treatment can result in loss of function or amputation.

A common cause of compartment syndrome is underlying traumatic long bone fractures such as in tibial or forearm fractures, haemorrhage, traumatic crush injuries, or prolonged limb compression.⁴ Young men are more at risk of acute compartment syndrome, due to a relatively larger muscle mass compared with fascial compartment that remains static after growth. Acute compartment syndrome from infectious diseases, particularly as a complication of dengue fever is rare. There are two 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.^{3,4}

In this patient with thrombocytopenia, the haematoma formation resulting from failure of venous cannulation caused an increase in intracompartmental pressure. In patients with thrombocytopenia, the clotting time will be prolonged, hence allowing more time for extravasation of fluid and haematoma formation. The increased intracompartment pressure will induce ischemia when local blood flow cannot meet the metabolic demand in the surrounding tissue.

Early diagnosis is the key to achieve a good treatment outcome of acute compartment syndrome. 'Pain out of proportion to apparent injury' is often the first sign of presentation in patients with acute compartment syndrome. Paresthesia and paralysis are the late signs of acute compartment syndrome, indicating a considerable degree of nerve injury. Clinical examination such as pain upon passive stretch of muscle in the affected compartment (passive stretch test) is an easy and effective test to diagnose acute compartment syndrome.

Acute compartment syndrome requires an emergency fasciotomy to release the intracompartment pressure. Individual muscle belly assessment is required to predict the outcome and careful assessment is required to ensure

each compartment pressure is adequately relieved. Delay in treatment and diagnosis many a times leads to permanent disability. Other complications such as volkmann ischemic contracture, scarring, infection, and amputation have been reported.

Conclusion

Acute compartment syndrome is a rare but potentially limb-threatening in patients with dengue fever. A scrupulous approach must be adopted when performing any vascular cannulation for dengue patients with thrombocytopenia.

REFERENCES

1. iDengue. 2015. <http://idengue.remotesensing.gov.my/idengue/index.php> (Accessed on 14/8/2018).
2. Ministry of Health Malaysia. Management of Dengue Infection in Adults. 2nd ed. 2010.
3. Bandyopadhyay D, Mondal P, Samui S *et al.* Acute compartment syndrome of upper limb as an unusual complication of dengue hemorrhagic fever. *N Am J Med Sci* 2012; 4: 667–8.
4. Khoo CS, Chu GSE, Rosaida MS, Chidambaram SK. Dengue fever with compartment syndrome of the right arm; *J Roy Coll Physicians Edinb* 2016; 46: 241–3.

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

	Khoo <i>et al.</i> ⁴	Bandyopadhyay <i>et al.</i> ³	Kamisan <i>et al.</i>
Time of onset	Day 8 of illness	Day 3 of illness	Day 6 of illness
Gender	Female	Male	Male
History of local injury	Arterial cannulation	None	Venous cannulation
Location	Right arm	Right forearm	Right arm
Other leaking sites	Pleural effusion, ascites	None	Pleural effusion
Other bleeding sites	None	Gum bleeding	None
Outcomes	Recovered well	Recovered well	Ulnar and radial nerve palsy

Figure 1 : Pre-operative swollen arm with bruises around the cubital region.



Figure 2 : Post-fasciotomy wound showing good viability of muscle.

