Case report

Spontaneous splenic rupture in dengue fever with non-fatal outcome in an adult

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Abstract

A 26-year-old male presented with fever for five days and abdominal pain for 24 hours. System examination identified a soft abdomen with diffuse tenderness. CT-abdomen findings were consistent with splenic rupture with intra and peri-splenic hematoma. Laboratory investigations showed a platelet count of 40,000 per mm^3. In due course he developed hypotension and underwent splenectomy. Non-structural protein 1 (NS1) dengue antigen was positive in the admission sample and IgM dengue antibodies were detected in the follow-up sample. Histopathology of the spleen showed normal architecture with no evidence of hyperplasia, cellular infiltrates or haematological malignancy. Splenic rupture is a rare, but potentially fatal complication of dengue fever and severe dengue which should be suspected when a patient presents with abdominal pain and hypotension. Our case highlights the occurrence of splenic rupture in the viremic phase of dengue illness before the development of IgM antibodies.

Key words: dengue; splenic rupture; hypotension;abdominal pain;splenectomy;case report

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Introduction

The spectrum of disease in dengue infection caused by four serotypes (DEN-1 to DEN-4) range from a subclinical or mild illness to a severe form of hemorrhagic fever which may prove fatal [1]. The World Health Organization (WHO) has suggested a revised classification for the illness as dengue (± warning signs) and severe dengue [1]. However, the earlier WHO classification of dengue fever (DF), dengue hemorrhagic fever (DHF), and dengue shock syndrome (DSS) is still widely followed in clinical practice [1]. Prior to 1970 only nine countries had experienced dengue hemorrhagic fever [2]. Today, dengue has spread to over 100 countries; this number includes those countries that were free from the disease 20 years ago [2]. Acute abdominal pain is one of the atypical presentations of dengue fever, and spontaneous splenic rupture is a rare complication of dengue fever. DHF and DSS are prevalent among all age groups in northern India and among children in southern India [2]. Our observation from Chennai (southern India) on 128 DHF patients indicated increased mortality among adults due to the severe form of dengue [3]. However, our report did not identify splenic rupture as a complication [3]. To date, 11 cases of spontaneous splenic rupture in dengue have been reported in the medical literature [4-13]. We report a non-fatal case of spontaneous splenic rupture in an adult patient with dengue fever with no cutaneous or mucosal bleeding tendencies but with features of circulatory collapse due to visceral bleed.

Case report

A 26-year-old male with no pre-existing chronic illness presented to the emergency department of Sri Ramachandra Medical College Hospital, a tertiary care teaching hospital in Chennai (south India), with complaints of fever for five days and abdominal pain during the previous 24 hours. There was no history of bleeding tendencies. His pulse were 120/minute, arterial pressure 100/80 mm Hg, and respiratory rate 34/minute. He appeared febrile and dyspneic, and his tongue was moist; however, there was no pallor, edema, jaundice, lymphadenopathy, rash, petechiae or echymosis. Tournique test was negative. Physical examination identified a soft abdomen with diffuse tenderness over all nine quadrants; no guarding or rigidity was present. Organomegaly could not be properly assessed because of his tender abdomen. Cardiac, pulmonary and nervous system examinations were unremarkable.

He was initially evaluated with a chest X ray, which was normal, followed by ultrasound of the abdomen, which showed a normal-sized liver, normal gall bladder, normal biliary tree, normal pancreas, normal-sized kidneys and pelvic calyceal system, and normal-sized spleen with hypo-echoic regions perpendicular to the anterior margin of the spleen. The ultrasound findings were suggestive of a possible hematoma or abscess in the spleen. A CT-abdomen (GE Health Care, United Kingdom) was requested, which showed an 8 cm × 7.5 cm hypodense lesion extending a few centimetres from the anterior to the medial margin of the spleen through its hilum up to to its lateral margin, causing a wide discontinuity between the upper and lower half of the spleen (Figure 1). These findings were consistent with splenic rupture with intra- and peri-splenic hematoma. Laboratory investigations showed the following: Hb%12.5 gm%; packed cell volume (PCV) 37; total leukocyte count 8,000 per mm^3; platelet count 40,000 per mm^3; blood sugar 120 mg/dl; blood urea nitrogen 28mg/dl; serum creatinine 1.2mg/dl; serum Na 134 meq/L; K 4.2 meq/L; Cl 104 meq/L; and Hco_3 18 meq/L. Liver functions were as follows: total bilirubin 0.9 mg/dl (direct bilirubin-0.2 mg/dl); alanine transaminase 80 IU/L; aspartate transaminase 60 IU/L; alkaline phosphatase 130 IU/L; total protein 7 gm/dl; and albumin 4 gm/dl. International normalized ratio was 1.2 and partial thromboplastin time was 36 seconds (control 28 seconds). Blood smear for malaria was negative. In due course he developed hypotension requiring fluid resuscitation with 3 litres of normal saline, transfusion of 3 units of whole blood and 4 units of platelet concentrate. After hemodynamic stabilization, he underwent splenectomy in view of the hemodynamic status. The surgical procedure was uneventful. There was no fever or bleeding tendencies in the post-operative period. Non-structural protein 1 (NS1) antigen for dengue virus was positive while IgM and IgG antibodies for dengue were negative (Dengue Duo, Standard Diagnostics, Kyonggi-do, Korea) on the sample taken on admission (fifth day of illness), indicating the viremic phase of dengue infection. A repeat sample on the eighth day of illness showed a negative NS1 antigen but with positive IgM antibodies and negative IgG antibodies against dengue. IgM antibodies for leptospirosis (Panbio, Australia) were negative. Blood and urine cultures were sterile. Macroscopic examination of the specimen showed a longitudinal rupture 10 cm in length from the splenic notch to the centre of the hilum, causing dehiscence of the splenic capsule all through the course along with overlying hematomas. Histopathology of the spleen showed normal architecture with no evidence of hyperplasia, cellular infiltrates, or haematological malignancy. Follow-up platelet count initially showed a fall to 34,000 per mm^3 on the eighth day of illness followed by recovery to 1,20,000 per mm^3 on the tenth day of illness. He was discharged on the fourteenth hospital day

Discussion

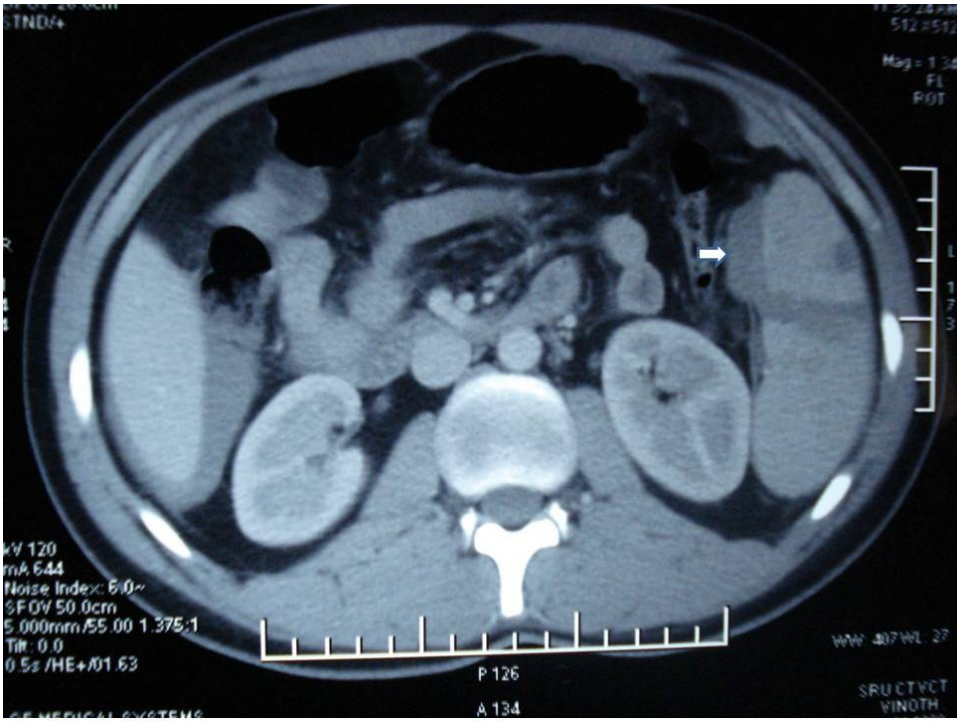
Splenic rupture occurs secondary to abdominal trauma or due to non-traumatic causes [14,15]. Non-traumatic splenic rupture (NTSR) may be either pathologic or spontaneous. The term *pathological splenic rupture* is applied when it occurs in a diseased spleen (with abnormal histology) while spontaneous splenic rupture refers to one occurring in a histologically normal spleen [14]. NTSR can occur in infections, malignancies, and connective tissue disorders [14].

Infections associated with NTSR include infectious mononucleosis, malaria, typhoid, varicella, infective endocarditis, Q-fever, influenza, aspergillosis and dengue [14,16,17].

Splenic rupture associated with dengue can occur in both uncomplicated dengue fever (which lacks features of severe bleeding and plasma leak) and complicated dengue hemorrhagic fever. Though hypothesized to be due to a combination of coagulation factors and severe thrombocytopenia, the mechanism of splenic rupture in dengue is not clear [13]. Of the 11 dengue cases describing splenic rupture, 4 occurred in uncomplicated dengue fever and 7 occurred in DHF of which 8 (out of 11) patients survived. [4-13]. The survival observed in these cases should be attributed to timely diagnosis and management rather than the natural course of splenic rupture, which is expected to be poor if the diagnosis is missed. Unlike the situation in earlier reports, our case highlights the occurrence of splenic rupture in the viremic phase of dengue before the development of antibodies, as illustrated by NS1 dengue antigen detection with no dengue IgM antibodies in the admission sample.

Abdominal pain is the main presenting symptom of splenic rupture in dengue fever. Given that acalculous cholecystitis is a known complication of dengue fever presenting with abdominal pain, which is often treated conservatively, it is possible that clinicians may overlook splenic rupture except when

Figure 1. CT scan of upper abdomen



Arrow showing a 8cm × 7.5 cm hypodense lesion extending from few centimetres anterior to the medial margin of the spleen through its hilum upto to its lateral margin causing a wide discontinuity between the upper and lower half of the spleen

it is specifically sought. Hypotension may be the only clinical clue indicating a possible splenic rupture in a patient with suspected dengue presenting with abdominal pain. This scenario can be complicated with the fact that hypotension is frequently associated with DHF, which may mislead the clinician. It may be prudent to consider that abdominal imaging focussing on splenic rupture is mandatory in a patient with suspected dengue fever presenting with abdominal pain and hypotension, irrespective of the presence of signs of plasma leak. Furthermore, patients presenting with abdominal pain in dengue should undergo careful evaluation since inadvertent surgery may prove fatal [18].

Infectious mononucleosis (IM) is the protean disease affecting the reticuloendothelial system leading to splenic rupture. First described in 1941 by King, the prevalence of splenic rupture in proven infectious mononucleosis is about 0.1 to 0.5% [16]. IM is associated with infiltration of lymphocytes in the splenic pulp, supporting trabeculae, blood vessels, and even the splenic capsule [16], which leads to fragmentation of the splenic architecture facilitating spontaneous rupture.

Splenic rupture is a well-known complication of malaria (more frequent in *Plasmodium vivax* than in *Plasmodium falciparum* infection), with 64 cases reported by the year 1948 [17]. The mechanisms of splenic rupture in malaria include rapid hyperplasia and stretching of the splenic parenchyma and capsule along with occurrence of small infarctions and haemorrhages making the spleen more vulnerable for rupture [17]. Though reports of splenic rupture in typhoid date back to 1898, it is far less common today [19].

Pathologic splenic rupture can occur in malignancies which include haematological conditions such as acute myeloid leukemia, chronic lymphoid leukemia and non-hodgkins lymphoma [14]. Splenic rupture can also occur due to splenic metastasis from choriocarcinoma, malignant melanoma, teratoma, carcinoma of lung, liver, stomach, rectum and urinary bladder [14]. Rheumatoid arthritis, systemic lupus erythematosis, polyarteritis nodosa and Wegener's granulomatosis are connective tissue disorders reported to be associated with splenic rupture [14].

Splenic rupture, either pathological or spontaneous, can be associated with a spectrum of causes. Splenic rupture is a rare, but potentially fatal complication of dengue fever which should be suspected when a patient presents with abdominal pain and hypotension. Dengue and malaria can present with hypotension and they are unique causes

of splenic rupture, which may delay the diagnosis of this complication. Our case highlights the fact that splenic rupture in dengue can occur during the viremic phase of the illness before the development of dengue antibodies.

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