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Case Report

Dengue Hemorrhagic Fever Complicated with Spontaneous Rupture of the Spleen among Patients with Thalassemia and G6PD Deficiency: A Case Report

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Abstract

Dengue infection is endemic in Thailand and Southeast Asia and thalassemia is highly prevalent in this area too. The clinical spectrums of dengue fever range widely, and many complications can occur. Spontaneous splenic rupture is a very rare complication in dengue fever. Here, we report a case of dengue fever complicated with spontaneous splenic rupture in a 22-year-old man with thalassemia presenting acute febrile illness and left upper quadrant pain. Computerized tomography of the upper abdomen with contrast revealed diffuse enlarged size of spleen with focal active contrast extravasation at the spleen surrounded with hematoma, and generalized hemoperitoneum. The patients were treated with exploratory laparotomy and blood transfusion. The outcome depended on early diagnosis and management.

Keywords: Dengue hemorrhagic fever; Hemoglobinopathy; Spontaneous rupture of spleen

1. Introduction

Dengue virus infection is a mosquito-borne disease commonly found in tropic and subtropic regions [1]. The clinical spectrum of dengue fever varies and ranges from asymptomatic infection to severe illness including dengue with warning signs or severe dengue [1]. The many complications of dengue fever include abnormal bleeding, plasma leakage leading to circulatory failure, and other multiple complications such as myocarditis, encephalitis, and liver failure [2]. Spontaneous splenic rupture may be caused by many infections such as infectious mononucleosis, malaria, typhoid, varicella, infective endocarditis, Q-fever, influenza, and aspergillosis [3], but is a very rare complication in dengue fever. Thalassemia is a highly prevalent inherited genetic disease in Thailand and Southeast Asian countries [4]. Despite both dengue fever and thalassemia being common diseases in Thailand, few cases of coincident presentation have been reported [5]. Moreover, spontaneous rupture of the spleen in dengue hemorrhagic fever with thalassemia has never been reported. Thus, we are report a man with thalassemia and G6PD deficiency presenting splenic rupture in the early stages of dengue illness.

2. Case report

A 22-year-old Thai man with pre-existing thalassemia HbH Constant Spring Disease (HbHCS) attended the hospital due to high grade fever one day before admission. Twenty-four hours earlier, he presented fever, myalgia, and anorexia symptoms. Ten hours later, he developed sudden onset of a dull aching pain at the left upper quadrant without referred pain. His pain score was 5 of 10 and he then developed more pain. He denied any history of trauma at his abdomen. The patients did not use any Nonsteroidal Anti-Inflammatory Drugs (NSAIDS) or acetyl salicylic acid. Otherwise his past medical history was unremarkable. His physical examination was remarkable with a temperature of 39.0°C, pulse rate 105/min, BP 144/68 mmHg, respiratory rate 24/min and oxygen saturation 92%. His conjunctivae were pale and exhibited icteric sclerae. The abdomen showed normoactive bowel sound, no abdominal distension, tender at the left upper quadrant but without guarding and rebound tenderness, liver span 14 cm and palpable spleen 2 fingers based below the left costal margin. Other physical examinations were unremarkable. Laboratory studies demonstrated hemoglobin of 9 g/dL with NRC 15%, white blood cell count of 21,000/µL, with 77% neutrophils, 11% lymphocytes, platelets of 159,000/µL and G6PD level 44 iu/100mlRBC (normal range 159 to 297 iu/100mlRBC). His liver test showed total bilirubin 8.25 mg/dL, direct bilirubin 0.36 mg/dL, AST 122 U/L, ALT 25 U/L and ALP 50 U/L. Arterial blood gas at room air (FiO_2 21%) were pH 7.378, pO_2 85.6 mmHg, pCO_2 34.9 mmHg and HCO_3 20.1 mEq/L.

Methemoglobin was suspected due to disproportion of oxygen saturation and oxygen in arterial blood gas. Confirmation test for methemoglobin was done by elevate level of methemoglobin 17% (normal range 0 to 3%). Urgent chest and abdominal radiography revealed no evidence of pulmonary infiltration, pleural effusion, or pneumoperitoneum. An immunochromatographic assay for the dengue NS1 antigen was positive. RT-PCR to detect dengue virus RNA was positive for dengue serotype 1. The diagnosis was dengue fever with hemolytic crisis in a patient with thalassemia and G6PD deficiency. On the next day following intravenous fluid resuscitation, his blood pressure was normal, but hemoglobin fell to 7 g/dL.

Later that day, he developed hypovolemic shock that was characterized by severe hypotension (blood pressure 86/50 mmHg), and severe left upper quadrant abdominal pain. The patient's laboratory results revealed a hemoglobin value of 6.2 g/dL with NRC 41%, white blood cell count of 18,500/μL, with 87% neutrophils, 5% lymphocytes, and platelet count of 111,000/μL. Abdominal ultrasonography was performed showing enlarged size and increased parenchymal echogenicity, and splenomegaly with heterogeneous parenchymal echogenicity. Then the patient was sent for computerized tomography of the upper abdomen with contrast revealing a lobulated contour, diffused enlarged size of spleen (15.8 cm) with calcified capsule, focal active contrast extravasation at the anteroinferior aspect of the spleen surrounded with hematoma, total size about 11.9 × 15.1 × 20.3 cm and associated generalized hemoperitoneum (Figure 1). Exploratory laparotomy was performed at midnight of that day revealing massive hemoperitoneum and ruptured spleen with subcapsular hematoma. Splenectomy was performed and the size of the spleen measured about 20 cm. in length (Figure 2). The pathology of the spleen showed no evidence of malignancy or granuloma. After the operation, the hemodynamics and hemoglobin stabilized and he was then discharged from the hospital after resolving symptoms.

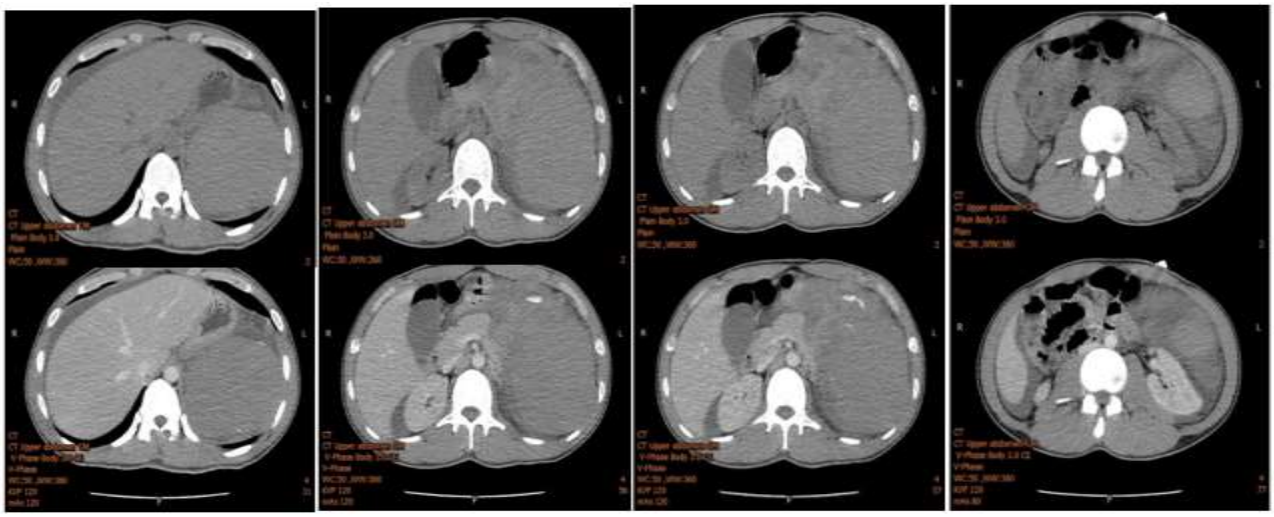

Figure 1: Computerized tomography upper abdomen with contrast: lobulated contour and diffuse enlarged size of spleen, focal active contrast extravasation at anteroinferior aspect of the spleen surrounding with hematoma, and generalized hemoperitoneum.

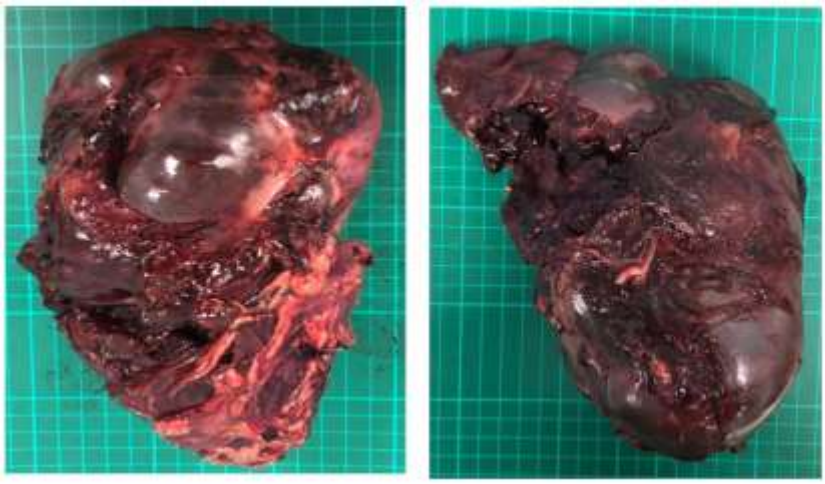

Figure 2: Macroscopic finding: grossly congested spleen with ruptured spleen and subcapsular hematoma.

3. Discussion

Dengue fever is caused by all four serotypes of dengue virus. The past complications of dengue fever primarily affected children, but at present, increasing prevalence has been observed among adults. The pathogenesis of complicated dengue remains not well known. The causes mentioned included thrombocytopenia, abnormal homeostasis, impaired platelet function, disseminated intravascular coagulopathy and increased vascular permeability [6]. Etiology of spontaneous splenic rupture may be caused by lymphoproliferative diseases, connective tissue diseases, aneurysm, and many infectious diseases [3,7-9]. The spontaneous rupture of the spleen comprises a rare complication of dengue fever but can constitute a life-threatening condition. Pathogenesis of splenic ruptured in dengue fever is probably caused by congestion of the spleen and thrombocytopenia [10]. The typical presentation is acute abdominal pain caused by congestion or subcapsular hematomas found in 15% of dengue hemorrhagic fever cases [11]. Shock among patients with dengue includes dengue hemorrhagic fever or dengue shock syndrome that may be misdiagnosed with splenic rupture [11]. Concomitant blood loss is one confounder that may further challenge the diagnostic workup in thalassemia among patients with hemolytic anemia [12]. Early recognition and investigation are important for atypical presentation in the early course of dengue illness. Splenectomy is the treatment of choice for spontaneous splenic rupture with hemoperitoneum for which the prognosis depends on timely diagnosis and management [13-15].

4. Summary

In conclusion, spontaneous splenic rupture can occur and cause shock in the early phases of dengue illness. Patients with hemoglobinopathy may receive a misdiagnosis of hemolytic crisis or blood loss. Patients with dengue and atypical presentation should be aggressively approached. Early diagnosis and management will protect these patients.

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