CASE REPORT

A rare case of Dengue Fever associated intestinal perforation in a Pediatric patient

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Abstract:
Dengue fever is a common viral disease presenting with a flu-like illness and bleeding manifestations. Although abdominal pain is a common presenting symptom of Dengue fever, gastrointestinal perforations are extremely rare. To date, only ten cases of gastrointestinal perforation have been reported in association with Dengue fever.

We report a case of ileal perforation in a young patient who was diagnosed with Dengue fever. The disease was diagnosed with an x-ray abdomen and was treated with surgical exploration.

The perforation was brought out as an ileostomy. Although rare, perforation peritonitis is highly fatal if left untreated. This must be considered early in the differentials of a Dengue-infected patient with signs of peritonitis. More studies are required to see into the patho-physiology of this association.

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Introduction:
Dengue is a viral disease with a reported incidence of up to 96 million cases per year worldwide.[1] The common symptoms are flu-like illness and severe presentations include dengue hemorrhagic fever and, potentially fatal dengue shock syndrome.[2] Gastrointestinal manifestations include abdominal pain, hepatitis, a-calculous cholecystitis, and acute pancreatitis.

Although abdominal pain is a common symptom of dengue fever, the disease is very rarely associated with bowel perforation.[2,3] To date, seven studies have reported a total of only ten cases of this association.[3-8] The pathogenesis for this association is not known, but it has been suggested that the dengue virus, either directly or by the release of endotoxins, may cause mucosal damage, mucosal ischemia, and subsequently perforation.[9]

Depending upon the site and size of the perforation, these cases can present with severe abdominal pain, nausea, vomiting, fever, gastroenteritis, sepsis or shock.[10] Management includes emergency resuscitation, detailed work-up to find the cause of the perforation, and a definitive surgical repair including primary repair, omentopexy, and/or stoma formation.[7] The prognosis of intestinal perforations is very poor with high mortality if left untreated and early diagnosis can be life saving.[11]

Here we present one such rare case of ileal perforation in association with dengue fever that was successfully treated surgically.

Case Presentation:
A 14-year-old male patient presented to the surgical emergency with severe abdominal pain for 8-hours. He had a history of fever and vomiting for 4-days for which he was taken to a physician.

He was diagnosed with gastroenteritis and was

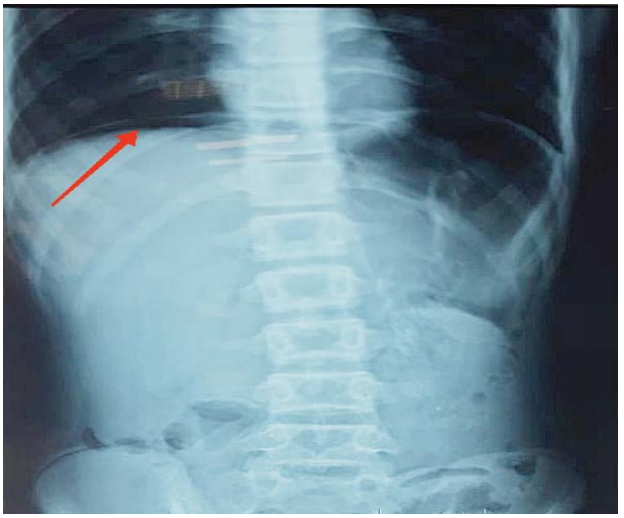

Figure-1: Plain X ray abdomen erect Postero-anterior view showing radiolucent shadow under the right hemidiaphragm (red arrow), suggestive of pneumo-peritoneum

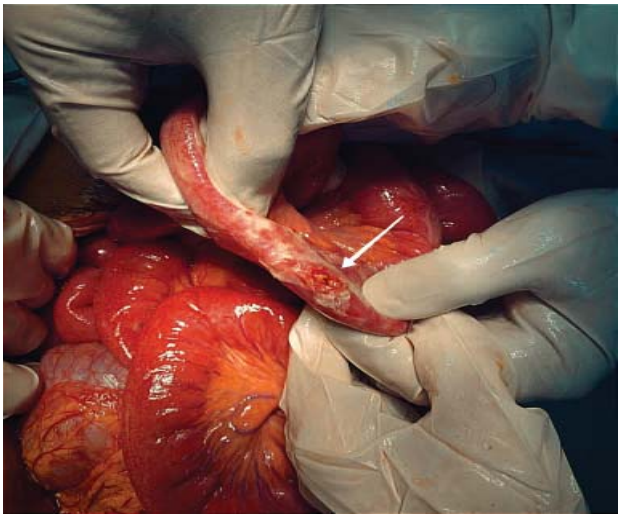

Figure 2: Intra-operative image showing a single perforation (white arrow) in the wall of the ileum one and a half feet proximal to the ileocecal junction.

Table 1: Literature review of all the cases of dengue fever associated bowel perforation reported to date

S.No	Primary Author (year of publication)	Country	Age, (Gender)	Site of perforation	Diagnosis	Treatment	Outcome
1	Kumar P(2016)	India	10 years,(F)	Ileum	X-ray	Primary repair	Improved
2	Kumar P(2016)	India	7 years,(M)	Ileum	X-ray	Primary repair	Improved
3	Jain AKC(2014)	India	64 years,(F)	Jejunum	Exploratory Laparotomy	Resection and stoma	LAMA due to financial burden
4	Desai G(2014)	India	-	Appendix	Ultrasound	Exploratory laparotomy	Pleural effusion, SSI
5	Desai G(2014)	India	-	Appendix	CT scan with contrast	Appendicectomy	improved
6	Desai G(2014)	India	-	Appendix	Ultrasound	Exploratory laparotomy	Pleural effusion, SSI
7	Mandhane N (2015)	India	17 years, (M)	Stomach	-	Exploratory laparotomy	Improved
8	Jibril H	Pakistan	37 years (M)	Duodenum	X-ray	Graham patch omentopexy	Improved
9	Pillai M(2019)	India	12 years,(F)	Stomach	X-ray	Graham's omentopexy	-
10	Ng CY(2019)	Malayasia	39 years,(M)	Stomach	CT scan	Primary repair	Improved
11	Haseeb A (Current case)	Pakistan	14 years,(M)	Ileum	X-ray	Ileostomy	Improved

advised oral antipyretic, oral fluids, and bed rest.

Unfortunately, the patient's condition deteriorated, and he was brought to a tertiary care hospital. On examination, he had a sick and toxic look, with a blood pressure of 80/60mmHg, Pulse rate of 126 beats/min, Oxygen saturation of 94%, and respiratory rate of 26 breaths/min.

He had a tense, distended, and tender abdomen with generalized rigidity. There was a tympanitic percussion note with absent bowel sounds on auscultation. The rest of the examination was unremarkable. There was no history of NSAID use, peptic ulcer disease, tuberculosis, enteric fever, or other inflammatory diseases. Past medical and family history was unremarkable. The patient was resuscitated with intravenous fluids, antibiotics, analgesics, and antiemetics in the emergency department.

The patient's blood workup revealed a mild leu-

kocytopenia (3500x103/uL) thrombocytopenia (45000x103/uL), and positive dengue-NS1 serology. This confirmed a diagnosis of dengue fever with peritonitis. A plain erect X-ray abdomen revealed free gas under the right hemidiaphragm, suggestive of a hollow viscus perforation (Figure 1).

After informed consent, the patient underwent an emergency exploratory laparotomy. Upon exploration, we noted dilated gut loops, 50 ml purulent fluid, and a single perforation in the ileum (2x2 cm in size) about one and a half feet proximal to the ileocecal junction (Figure2). The rest of the bowel was found disease free. The abdominal cavity was washed thoroughly. A biopsy was taken from the margins of the perforation, and the perforation was brought out as a loop ileostomy. After hemostasis, the sheath was closed with the skin left open. The post-anesthesia recovery and postoperative course were unremarkable. Both his fever and abdominal pain resolved soon afterward, and he was discharged with medications on the 3rd post-operative day.

On a two-week follow-up, the patient was found healthy. His histopathology report was unremarkable except for acute inflammatory cells, excluding other causes of intestinal perforation like tuberculosis, enteric fever, neoplasia, and inflammatory bowel disease, and the perforation peritonitis was associated with dengue fever itself. The patient remained stable at the one-monthly follow-up.

Discussion:

Intestinal perforation is an extremely rare complication associated with dengue fever. After an extensive literature search on PubMed and Google Scholar, we found only ten reported cases of bowel perforation in association with Dengue fever.[3-8] Only three of these cases belonged to the pediatric age group (Table 1). Kumar et al reported the first two cases of dengue fever-associated bowel perforation in children.[3] Both presented with sudden-onset severe abdominal pain along with flu-like symptoms. Our patient had a similar presentation and was diagnosed with dengue fever and perforation peritonitis.

Although acute abdominal pain is a common manifestation of Dengue fever, only a very few cases develop perforation and peritonitis.[2] This makes it a diagnostic challenge to diagnose the disease early. However, due to the life-threatening nature of perforation peritonitis, it should be considered among the top differential diagnosis in a suspected dengue fever patient with signs of peritonitis.

The diagnosis of a visceral perforation is usually made with a combination of clinical presentation of peritonitis along with imaging. Ultrasound abdomen and Erect abdominal x-ray might show intraperitoneal free gas. A computed tomography (CT) scan abdomen is more sensitive in detecting even a small amount of pneumoperitoneum.[12] In cases of severe peritonitis with sepsis, and/or when the above diagnostic modalities are equivocal or unavailable, an exploratory laparotomy should be undertaken. The perforation in our case was diagnosed with an x-ray abdomen and confirmed upon exploration. The association with dengue fever was established by exclusion when all the other etiologies were negative and dengue serology was positive.

The site of perforation varies from the stomach to the duodenum, jejunum, ileum, and appendix. In children, including in our case, ileal perforations are more common.[3]

Abdominal pain in dengue fever is treated medically. However, abdominal pain refractory to medical treatment along with signs of peritonism and/or sepsis should suggest bowel perforation and might need urgent surgical exploration. Depending upon the site, size, the number of perforations, and associated findings, the surgical treatment can range from primary repair to omentopexy, bowel segment resection, and even stoma formation[7] as shown in table-1. With a single ileal perforation but inflamed surrounding tissue in our case, we brought out the perforation as a loop ileostomy.

The pathogenesis is not entirely understood. One study suggests mucosal damage, both from

direct viral infection and cytokines mediated, as the basis for the perforations.[9] More research is required to delineate the underlying pathophysiology of this disease.

Conclusion:
Dengue fever-associated intestinal perforation is an extremely rare but life-threatening association. It is important to consider this association in a suspected dengue fever patient who presents with severe acute abdominal pain. Early treatment can be life saving. Further study is required in to the pathophysiology of this association.

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Role and contribution of authors:
Abdul Haseeb, collected the data, references and did the initial writeup.

Almas, collected the data and helped in introduction writing.

Waseem Khan, collected the references and helped in discussion writing.

Shandana Khan, collected the data, and helped in case report discussion.

Qaidar Ali Zai, collected the data, references and helped in compiling the result.

Muhammad Jawad Zahid, critically review the article and made final changes.

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