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Recurrent Life-Threatening Lower Gastrointestinal Bleeding Secondary to Abdominal Tuberculosis in a Case of Moderate Hemophilia A

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Abstract

Background: Gastrointestinal tuberculosis can have a plethora of presentations, commonly abdominal discomfort, altered bowel habits, weight loss, fever, or mild rectal bleeds. Massive and recurrent life-threatening Lower Gastrointestinal Bleeding (LGI) is an unusual presentation in mild-moderate hemophilia cases. Our case is a moderate hemophilia patient with a significant recurrent GIT bleeding where ileocecal tuberculosis was identified later as the culprit.

Clinical Description: We report an uncommon case of recurrent episodes of life-threatening LGI bleeding in a patient with moderate hemophilia A. The child was given factor VIII replacement therapy with each episode of bleeding. The radionuclide scintigraphy revealed the ileal origin of the bleed and contrast CT abdomen showed features suggestive of ileocaecal tuberculosis which was confirmed later microbiologically.

Management and Outcome: The patient showed significant improvement after the use of anti-tubercular drugs and achieved complete remission.

Conclusion: An alternate diagnosis concurrent to the non-severe form of hemophilia should always be sought as a possibility in case of severe spontaneous and recurrent LGI bleeding.

Key Words: Hemophilia A, Intestinal Tuberculosis, Massive Gastrointestinal Bleeding.

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1- INTRODUCTION

Tuberculosis (TB) is known as a grand masquerader with variable clinical presentations. Ileo-caecal tuberculosis is one of the most common surgical emergencies due to obstruction (adhesion or stricture formation) and perforation. However, Massive Lower Gastrointestinal (LGI) bleeding in abdominal tuberculosis is a rare manifestation (1). Few cases of GI TB have reported severe hematochezia in patients with other comorbidities and additional risk factors, i.e. tubercular endarteritis of gut vessels, and fistulas (2). Severe hemophilia (fVIII<1%) can manifest as spontaneous and life-threatening GI bleeding but is rarely seen in mild-moderate cases (3). In the present manuscript, we report an unusual case of moderate hemophilia A, who developed recurrent episodes of severe LGI bleed and was treated spuriously with factor replacement therapy on multiple occasions, and was later diagnosed with intestinal tuberculosis. The patient achieved complete remission after initiating anti-tubercular drugs.

2- CLINICAL DESCRIPTION

An 11-year-old boy from Rewari, state of Rajasthan/India who was a known case of moderate hemophilia A (factor level 4.8 %) diagnosed at age 4 years, and presented to the emergency room with severe pallor and shock. History revealed passage of rectal blood clots over the past 2 days. There was no associated fever, jaundice, pain abdomen, altered bowel movement, or bleeding from any other sites. There have been similar 2 such episodes of severe hematochezia over the past 1.5 months where blood transfusions were required in each episode. On clinical examination, he was found to be in a hemodynamically unstable state with poor perfusion and altered sensorium. Vitals showed HR: 150/min, RR: 34/min, BP: 70/50 mmHg, cold peripheries, and low volume thready pulse; he looked severely pale and drowsy (E3V3V4). General physical examination showed average built habitus with no much-cutaneous or musculoskeletal bleeding manifestations. Cardiac examination showed tachycardia with gallop rhythm. On abdominal examination there was no tenderness, no distension, no lump, and no signs of free fluids. The child was resuscitated with Normal Saline fluid boluses; and subsequently packed RBC was given in two aliquots (15 ml/kg each).

2-1. Investigation and Management

Investigation revealed severe anemia, (Hb was 3.2 gm/dL, Hct-12%, TLC-15400 cells/cumm, Platelets count- 3.8 lac/microL), Blood Urea- 22 gm/dL, Creatinine: 0.4, Serum Sodium- 145 meq/L, Serum Potassium-4.5 meq/L, serum lactate- 4.5, Prothrombin time- 14 seconds, INR-0.9 and aPTT- 45 seconds. Peripheral blood smear showed NCNC anemia with increased reticulocytes. Factor VII -5.4 %, the mixing study (Bethesda assay) was not suggestive of the presence of inhibitors. Rectal examination showed hematochezia with the passage of clots, there were no polyps or ulcers. After initial resuscitation, a sigmoidoscopy was done which revealed normal findings. The child was administered factor VIII, 50 units/kg to achieve 100% serum factor levels; and bleeding stopped after 24 hours of factor replacement.

The history revealed that the child had had 2 subsequent emergency room visits to the same hospital in the past 1.5 months, with similar complaints of massive LGI bleeding requiring blood transfusions and factor VIII administrations, and had been discharged on oral hematinics. This recurrent and spontaneous hematochezia in the case of moderate hemophilia warranted a detailed investigation to rule out other potential organic causes.

99^{m} technetium RBC scintigraphy done to trace the origin of the bleed, revealed

active GI bleeding from the ileocaecal junction and caecum. Contrast CT of the abdomen showed features of abdominal tuberculosis; irregularly enhancing circumferential thickening involving terminal ileum associated with fat stranding and necrotic abdominal lymphadenopathy, hepatosplenomegaly with splenic granuloma (Fig. 1A & B). CECT chest revealed multiple centrilobular nodules with tree-in-bud necrotic mediastinal lymphadenopathy (Fig. 2A & 2B). The gastric aspirate was negative but the stool sample tested positive by TB rapid diagnostic test (NAAT). The child was started on Anti-Tubercular drugs (ATT) which showed significant clinical improvement and didn't bleed again over the next 6 months of follow-up.

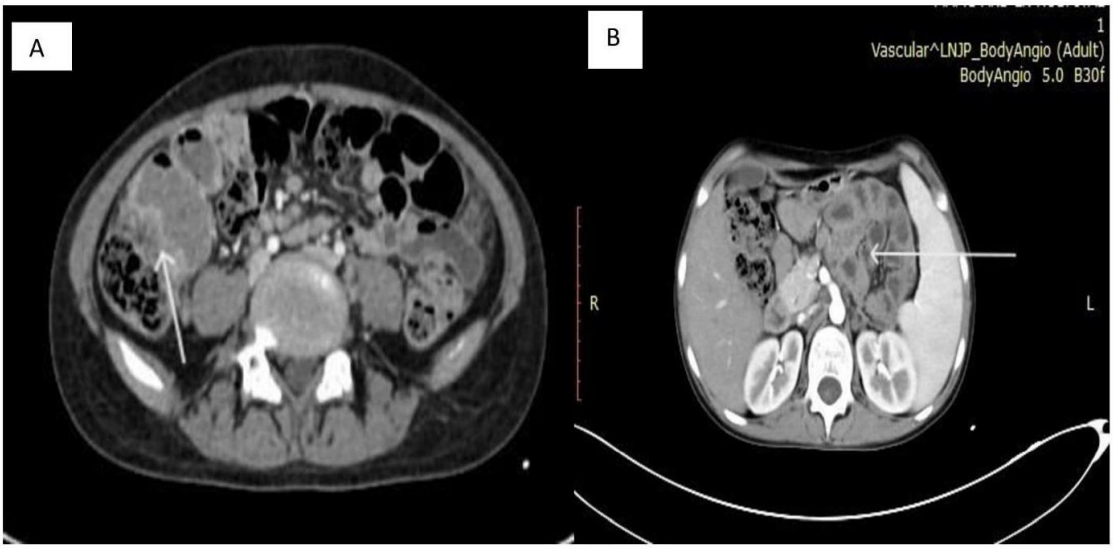

Fig. 1: Contrast CT of the abdomen; features of abdominal tuberculosis are shown, irregularly enhancing circumferential thickening involving terminal ileum associated with fat stranding and necrotic abdominal lymphadenopathy, hepatosplenomegaly with splenic granuloma.

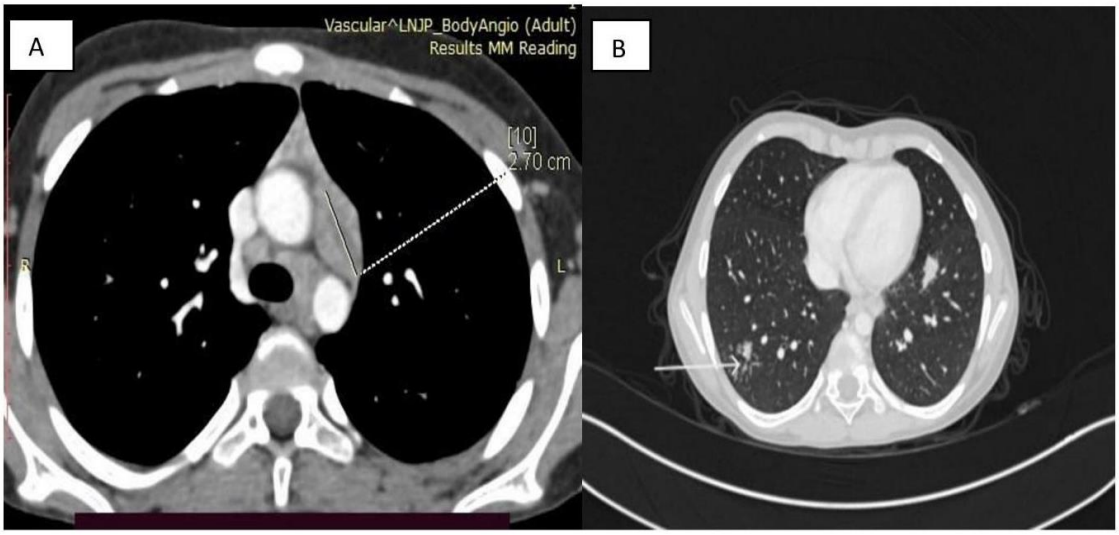

Fig. 2: CECT chest: multiple centrilobular nodules were revealed with tree-in-bud necrotic mediastinal lymphadenopathy

3- DISCUSSION

Abdominal tuberculosis has a myriad of presentations and mimics a variety of diseases like Crohn's disease, Ulcerative colitis, and intestinal malignancy. Usually, the symptoms are vague with abdominal discomfort, change in bowel habits, weight loss, fever, and other constitutional symptoms (4). Gastrointestinal bleeding is frequently minor and massive bleeding is a rare finding seen usually in diverticulosis and angiodysplasia (5). The common presentations of abdominal tuberculosis in the pediatric population described are adhesive peritonitis followed by nodal disease and less frequent intestinal perforation (6). There are few case reports available where GI TB has led to severe bleeding due to the presence of other risk factors like anticoagulant medicines, immunosuppressant therapy, TB aortitis, or vascular fistulas due to mycotic aneurysm (7, 8).

The severity of hemophilia symptoms is often related to the level of factor activity, which helps classify the condition further. Individuals with greater than normal (>5%) factor activity (classified as mild hemophilia) typically experience bleeding primarily, after significant injuries or surgical procedures. Spontaneous bleeding episodes are rare in mild hemophilia, and the diagnosis is often made incidentally or during routine pre-surgical lab tests. In cases where factor activity ranges from 1% to 5% of normal (moderate hemophilia), bleeding tends to occur following trauma, injuries, dental work, or surgery. Moderate hemophilia may also involve recurrent joint bleeding in up to 25% of cases, and the diagnosis can be delayed (9). Our patient was moderate hemophilia A with factor VIII levels of 4.8%. The sudden onset of recurrent episodes of severe melena in a moderate hemophilia patient compelled us to investigate other associated causes for melena. In our patient, we started with 99mTc RBC scintigraphy, which was useful in locating the bleeding site and planning further relevant investigations. ^{99m}Tc RBC scintigraphy is more sensitive than angiography for detecting bleeding sites and can detect sites with a minimum bleeding rate as low as 0.1 ml/min (10). It helped us reach the diagnosis of intestinal TB with the use of a CT scan and target biopsy from the site of the bleeding.

4- CONCLUSION

It is exceedingly uncommon for children with mild to moderate hemophilia to experience massive gastrointestinal bleeding. Also, in Abdominal tuberculosis, LGI bleeding has not been mentioned as a frequent symptom among children. Hence, having a high index of suspicion to rule out other organic lesions in the gut or other concurrent illnesses is of paramount importance and would prove potentially life-saving.

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