

Case Report

Colo-renal-duodenal fistula in pediatric patient with acute lymphoblastic leukemia: A rare presentation

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ABSTRACT

Colo-renal fistulas are uncommon presentations in the pediatric population and usually have an underlying insult, which can be vascular, infective, inflammatory, penetrating trauma, tumor, or iatrogenic in postintervention or postoperative procedures. Although few cases of such fistulae are published earlier, there is extreme paucity of literature on colo-renal fistulas in association with acute lymphoblastic leukemia (ALL). A thorough literature search revealed only one similar case report in a 15-year-old patient, to the best of our knowledge. Here, we present another case of a 6-year-old female patient, a known case of T-cell ALL with febrile neutropenia, uncontrolled hypertension, and fever spikes. The patient developed a colo-renal-duodenal fistula subsequent to a vascular and infective insult to the right kidney.

Keywords: Colo-renal-duodenal fistula, Pediatric population, Acute lymphoblastic leukemia, Vascular insult, Infection

INTRODUCTION

Reno-alimentary fistulae are extremely rare, accounting for less than one per cent of all fistulous communications between the intestinal and urinary tracts.^[1] Such abnormal connections are further rare in the pediatric age group. Majority of these fistulae are renocolic followed by the reno-duodenal, the most common being the left renocolic fistula.^[1,2] It can occur either due to long-standing primary renal causes such as nephrolithiasis, pyonephrosis, and xanthogranulomatous pyelonephritis or secondary to iatrogenic insults from minimally invasive/endoscopic procedures and treatments for colon and kidney cancers.^[3] In pediatric patients, these can occur secondary to ischemic insult, infections like tubercular or pyogenic, or an acute or chronic inflammatory process like Crohn's disease at any intestinal segment. The presentation of the patient is highly variable depending on the severity and immune status of the patient. Commonly, these patients present with a history of recurrent urinary tract infections. Other less common clinical findings include abdominal pain, fever, presence of an abdominal mass, pyuria, hematuria, abscess formation, and hematochezia.^[2,4]

The diagnosis of reno-alimentary fistulae is possible with various contrast-based imaging modalities such as barium enema, contrast-enhanced computed tomography (CECT), and antegrade and retrograde pyelography. However, in settings of pre-existing deranged renal functions, where usage of intravenous iodinated contrast agents may prove to be deleterious for the patient, retrograde pyelography comes as rescue imaging gold standard for definitive diagnosis.^[2,5]

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The prognosis of reno-alimentary fistulae depends on disease etiology, duration of the presence of fistula, the general condition of the patient and the degree of renal insufficiency.^[1] Treatment of such cases varies depending on the stage of the disease, localized or advanced disease process and host factors including the nutritional status and other comorbidities.

Management options are mostly surgical; however, conservative management is preferred if the patient has no history of bleeding, intestinal obstruction, renal failure, or sepsis. Kidney preservation should be of utmost importance in all cases where kidney function is not compromised.^[4]

CASE PRESENTATION

A 6-year-old female, known case of acute lymphoblastic leukemia (ALL) on induction therapy with cytarabine presented to our institution with a single episode of fever which was insidious in onset and relieved on medication. The patient was initially clinically diagnosed with necrotizing enterocolitis. Later, the patient complained of right lumbar pain and a history of recurrent urinary tract infections. Baseline investigations reveals hemoglobin - 8.7 g/dL, total leukocyte count - 2700 cells/microliter, absolute neutrophil count - 1430 cells/microliter (febrile neutropenia), platelet count - 34×10^3 cells/microliter and prothrombin time/international normalized ratio - 1.58. Serum galactomannan and Typhi Dot were positive. Renal function tests and liver function tests were normal. Urine routine and microscopic examination revealed the features of infection.

Ultrasound abdomen was done, which suggested a hypoechoic region in the interpolar and the lower pole of the right kidney with a perinephric collection. Subsequently, CECT angiography was done, which showed bulky right kidney with a large hypodense, nonenhancing region in the interpolar and lower pole regions, with the adjacent perinephric collection and thickened fascias [Figure 1]. Hypodense filling defect was seen in the right renal vein and inferior vena cava [Figure 2a]. Furthermore, a fistulous communication between the hepatic flexure and the perinephric collection was observed [Figure 2b and c]. An interval CECT abdomen done after 4 months depicted nonvisualization of the lower pole with heterogeneous enhancement of the interpolar cortex of the right kidney [Figure 3a]. On the delayed phase, contrast opacification of the right renal pelvis with opacification of the hepatic flexure [Figure 3b] and duodenum [Figure 3c] was seen, suggesting autonephrectomy of the lower pole with subsequent reno-colic-duodenal fistula formation. Diethylenetriamine pentaacetate scan also corroborated the computed tomography (CT) findings and showed a fistulous tract between colon and right renal pelvis. The patient was put on broad-spectrum antibiotic cover including cephalosporins

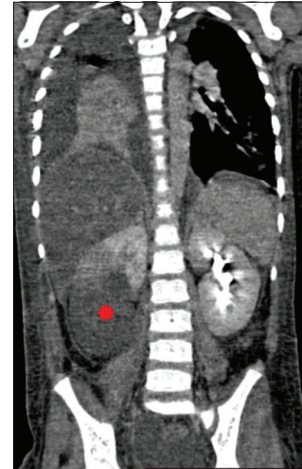


Figure 1: Coronal section of contrast-enhanced computed tomography abdomen shows bulky right kidney with a large hypodense, non-enhancing region in the interpolar and lower pole regions with bilateral pleural effusion (red asterisk).

and vancomycin. Further, he is now planned for fistula repair with diversion colostomy and subsequent primary anastomosis.

DISCUSSION

Colo-renal-duodenal fistula is a rare entity and usually has an underlying inciting event. Renal infections can lead to surrounding inflammation of the viscera (secondary to calculus and diverticulitis), which can progress to form fistulous communication. The malignant causes, such as leukemic deposits or tumors, are less common. It can also occur due to iatrogenic causes like postradiofrequency tumor ablation, percutaneous nephrostomy tube placement, renal biopsy, or during lithotripsy, tumor resection or postradiotherapy.^[1-5] Reviewing the literature, only single case report was found with similar presentation to our case, where a 15-year-old patient with acute lymphoblastic leukemia had recurrent urinary tract infection, subsequently developing colorenal fistula.^[2] Our patient, also a known case of ALL on chemotherapy, which in itself a pro-coagulable and immunosuppressed state, the patient developed necrotizing enterocolitis, renal infarcts and subsequently developed renal infection, followed by perirenal collections, which further complicated to fistulous communication with adjacent viscera - the hepatic flexure of the colon and third part of the duodenum. The preceding event can also be explained as the infection occurring as a prior event followed

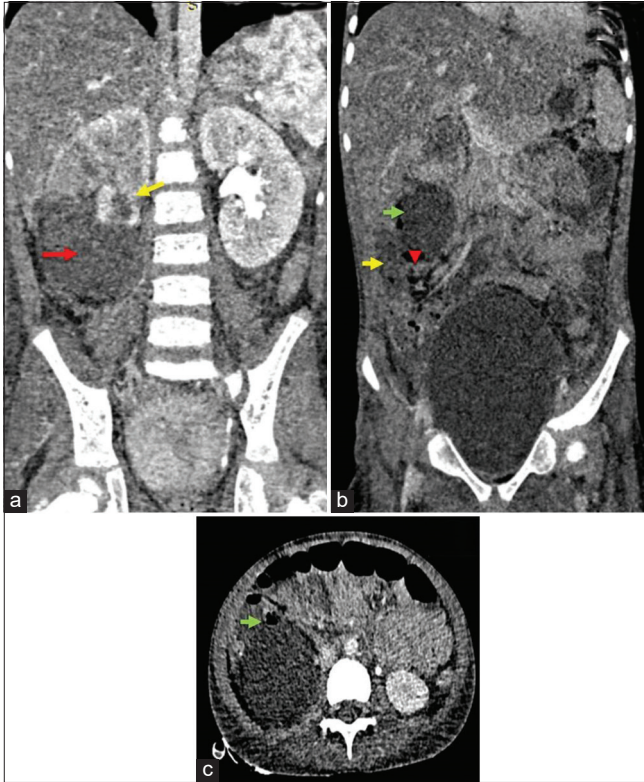


Figure 2: (a) Coronal image of contrast-enhanced computed tomography (CECT) abdomen obtained after 1 month shows bulky right kidney with a large hypodense, non-enhancing region (red arrow) in the interpolar and lower pole regions with adjacent perinephric collection and thickening of the surrounding fascia. Hypodense filling defect is seen in the right renal vein (yellow arrow). (b) Coronal image of CECT abdomen shows communication (red arrow) between hepatic flexure (yellow arrow) and the perirenal collection with air foci within the collection (green arrow). (c) Axial image of CECT abdomen shows communication between hepatic flexure and the perirenal collection with air foci within the collection (green arrow).

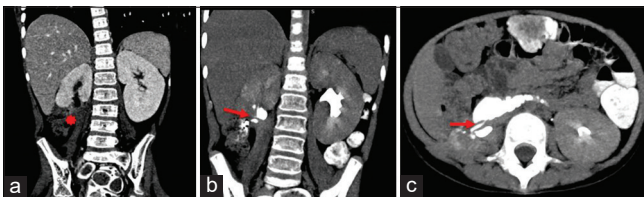


Figure 3: (a) Coronal section of contrast-enhanced computed tomography (CECT) abdomen obtained 4 months later showing nonvisualization of inferior pole of the right kidney-likely auto-nephrectomy (red asterisk) (b) Coronal section of CECT abdomen delayed phase showing abnormal communication between the right renal pelvis and ascending colon (red arrow) (c) Axial section of CECT abdomen showing abnormal fistulous communication between renal pelvis and duodenum (red arrow).

by vascular thrombosis and the subsequent development of a renal infarct. In patients with immunosuppressed states, the

inflammatory cascade is usually less severe, which explains why there was latency in complications in our case. The diagnosis of a reno-alimentary fistula requires both clinical and radiological evidence. Various screening imaging modalities such as ultrasound, intravenous urography, and barium may help in initial diagnosis. However, for confirmation, CECT proves to be highly promising, as in our case, where CT could clinch the diagnosis well, with the demonstration of fistulous communication in the excretory phase, thereby guiding the appropriate management.

CONCLUSION

In children presenting with recurrent enteric or urinary tract infections, imaging is necessary to diagnose the underlying cause and also identify the rare associated complications which proves to be of great help to clinicians for adequate management.

TEACHING POINTS

1. Radiological imaging is warranted in a child with immunocompromised status presenting with recurrent urinary tract or enteric infections.
2. Contrast-enhanced CT abdomen and pelvis play a pivotal role not only in identifying the underlying etiology of recurrent infections but also in depicting the rare complications such as reno-alimentary fistulae, thereby guiding appropriate management.

MCQs

1. In patients with deranged kidney function tests, the investigation of choice for demonstration of reno-alimentary fistulae is-
 - a. Barium enema
 - b. Intravenous urography
 - c. Contrast-enhanced CT abdomen
 - d. Retrograde pyelography

Answer Key: d

2. Which of the following is not a feature of Xanthogranulomatous pyelonephritis?
 - a. Bear paw sign
 - b. Staghorn calculus
 - c. It is a form of acute pyelonephritis
 - d. Increased incidence in diabetes patients

Answer Key: c

3. Jackstone calculus is composed of-
 - a. Calcium oxalate
 - b. Calcium oxalate monohydrate
 - c. Calcium oxalate dihydrate
 - d. Struvite

Answer Key: c

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

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Conflicts of interest

There are no conflicts of interest.

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