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Inflammation and infection

Reno-appendiceal fistula in autosomal dominant polycystic kidney disease

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ABSTRACT

We present a very rare Case of a 53-year-old female with autosomal dominant polycystic kidney disease (ADPKD) who was incidentally found to have a reno-appendiceal fistula while undergoing open bilateral nephrectomy. The mid-portion of the appendix was fistulized to a cyst in the lower pole of the right kidney. The etiology was likely due to chronic inflammation. An appendectomy was performed along with the planned right nephrectomy to ensure complete removal of the fistulous tract.

Introduction

The development of a fistulous communication between the kidney and intestinal tract is a rare event that has been cited in the literature a little over 100 times with the ascending and descending colon being most commonly implicated. The etiology may be secondary to trauma, iatrogenic, or spontaneous due to underlying renal or intestinal tract pathology.[1] To the best of the authors' knowledge, there has never been a report of a reno-appendiceal fistula in a patient with ADPKD. We present a Case of a patient with ADPKD incidentally diagnosed with a reno-appendiceal fistula while undergoing an open bilateral nephrectomy.

Case presentation

A 53-year-old female with a past medical history of ADPKD, on hemodialysis for 10 years, presented for a scheduled open bilateral nephrectomy, cholecystectomy, and ventral hernia repair. She had previous abdominal surgery for diverticular disease, including colostomy creation and take-down, which was complicated by a ventral hernia. The patient also had a history of an infected left renal cyst requiring drainage by interventional radiology, but no history of procedures on the right kidney. She was, however, evaluated in the emergency room two years prior to presentation for right-sided abdominal pain and fever, and was found to have an infected cyst in the lower pole of the right kidney. Imaging at that time showed inflammatory fat stranding anterior to the right kidney. She was treated with antibiotics and the infection resolved.

Pre-operative evaluation with a computed tomography (CT) scan of the abdomen and pelvis with intravenous contrast showed large bilateral polycystic kidneys, intrahepatic biliary ductal dilation, and a normal-appearing appendix that was adjacent to the lower pole of the right kidney (Fig. 1). Contrast enhancement was limited by the patient's end stage renal disease and a fistulous tract could not be appreciated. At the time of presentation, the patient was afebrile and hemodynamically stable, without leukocytosis. She did not report any abdominal pain and had no history of lower urinary tract symptoms or recurrent urinary tract infections (UTIs).

After informed consent was obtained, the patient was taken to the operating room. During the procedure, a large intestinal phlegmon was found adhered to the lower pole of the right kidney. With further dissection of the cecum, it became evident that the mid-portion of the appendix had fistulized to one of the renal cysts. An appendectomy was performed to allow for complete removal of the fistulous tract along with the right kidney (Figs. 2 and 3). The remainder of the surgery was uneventful and there were no significant complications.

The surgical pathology report subsequently noted a 9.5 cm in length by 0.9 cm in diameter appendix with a moderately dilated, pink-tan lumen. No gross evidence of acute appendicitis was reported. Firm, fibrous serosal adhesions were noted on both the right kidney and the appendix, but the two organs were separated before reaching pathology in order to be placed into individual specimen containers so no comment

Abbreviations: ADPK, Dautosomal dominant polycystic kidney disease; UTI, urinary tract infection.

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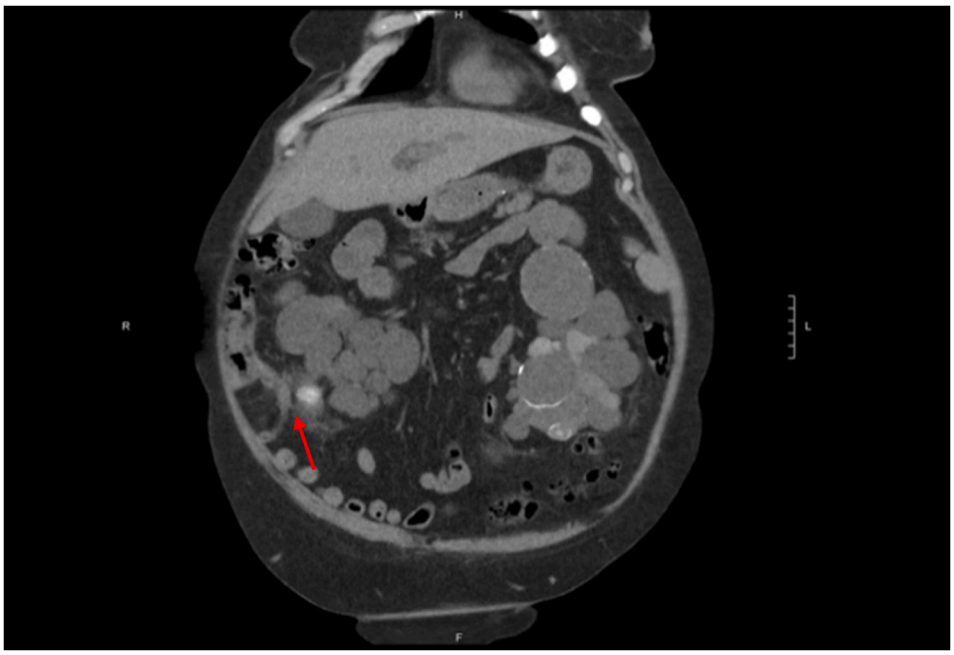

Fig. 1. Coronal CT showing appendix sweeping adjacent to a right sided renal cyst.

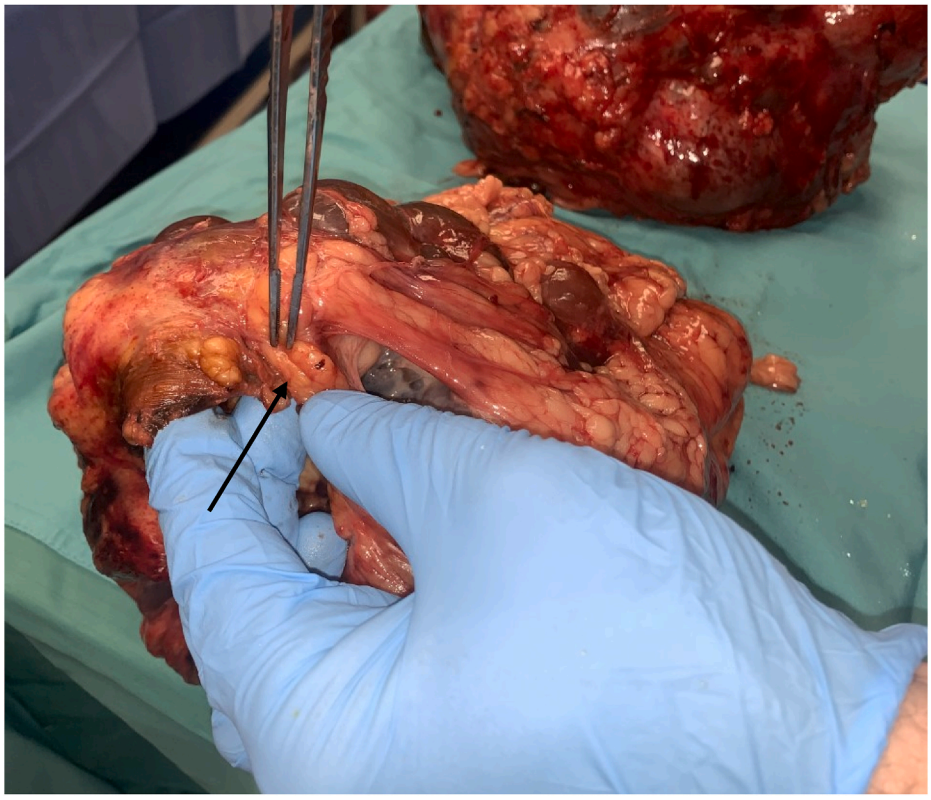

Fig. 2. Gross image of appendix adhered to right kidney.

on a fistulous tract could be made. A peri-appendiceal organized hematoma was noted on the gross specimen, that the pathologist stated was most likely due to shearing of the blood vessels in the fibrous adhesions between the right kidney and appendix, leading to local hemorrhage and a subsequent inflammatory reaction.

Discussion

We presented a rare Case of a reno-appendiceal fistula in a patient with ADPKD discovered during open bilateral nephrectomy. Reno-alimentary fistulas are usually a consequence of chronic, inflammatory disease of the urinary or gastrointestinal tract, but can also be due to trauma or iatrogenic. Traditionally, most cases of reno-alimentary

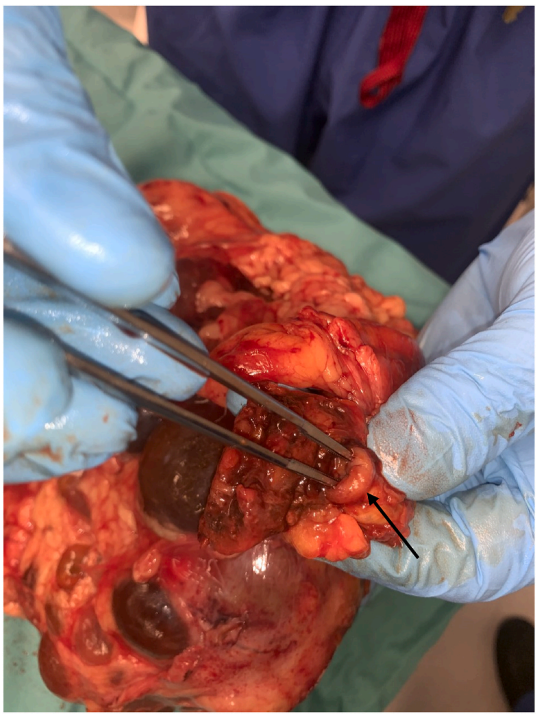

Fig. 3. Gross image of appendix adhered to right lower pole renal cyst.

fistulas originated from renal disease, particularly xanthogranulomatous pyelonephritis.[1] With the advent of minimally invasive surgery, there has been a rise in cases of reno-alimentary fistula development following percutaneous nephrolithotomy.[2] The most common locations of reno-alimentary fistulas are reno-colic, which make up about 60% of the reported cases, followed by reno-duodenal, with reno-jejunal and reno-ileal connections being exceedingly rare.[3,4] Medical management alone is rarely successful and operative intervention is almost always required.[3]

Most patients with a reno-alimentary fistula present with renal or gastrointestinal complaints such as flank or abdominal pain, fever, nausea and vomiting, and voiding symptoms like recurrent UTIs, hematuria, pneumaturia, and/or fecaluria.[4,5] While our patient did not present with any of these symptoms, surgical exploration showed a visible connection between the appendix and a right lower pole renal cyst. Lack of urinary symptoms can also be explained by the patients long-standing hemodialysis history, leading to minimal urine production. It is possible that the patient was asymptomatic because the fistulous tract between the two organs had yet to become fully patent, although, a lumen was observed intraoperatively. It is also possible that the renal cyst had successfully contained the contents of the fistulous tract and prevented seeding of gastrointestinal organisms into the kidney. Given the patients prior history of an infected right lower pole cyst, this leads the authors to believe that the fistula originated from renal inflammation surrounding that episode.

Moreover, the presence of appendiceal dilation noted on the surgical pathology report opens the possibility that the fistula could have been appendiceal in origin, potentially from a sub-clinical event of appendicitis, as no account of acute appendicitis was noted in the patient's record. It is also possible that the presence the fistula and local inflammatory response surrounding the right kidney subsequently led to the appendiceal dilation. While the exact origin of the fistula is unknown, it can be assumed that chronic inflammation of either or both organ systems contributed to the development of this fistulous tract.

Conclusion

This is a rare Case report of a reno-appendiceal fistula in a patient with ADPKD. The fistulous tract was incidentally discovered during open bilateral nephrectomy and necessitated an appendectomy for complete removal of the tract. The etiology of the fistulous tract was likely secondary to chronic inflammation in the kidney, appendix, or both. To the best of the authors' knowledge, this has never been described in the literature.

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Consent

Obtained.

Author statement

Madison Hill: Writing – original draft, Visualization, Project Administration, Formal Analysis. Natalia Arias Villela: Writing – Reviewing and Editing. John LaMattina: Investigation, Resources. Yvonne Rasko: Conceptualization, Investigation. Michael Phelan: Supervision, Project Administration, Conceptualization, Methodology.

Declaration of competing interest

None declared.

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