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Uretero-appendicular fistula: A rare case mimicking ureterolithiasis in a child

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ABSTRACT

Uretero-appendicular fistulas represents a rare entity, closely related to appendicitis and are usually encountered intraoperatively. A literature review showed six published cases –only one of them involving a pediatric patient. We describe the case of a ten year-old girl with history of recurrent urinary tract infections (UTIs) since the age of three. Following a six-year asymptomatic period, she was referred for evaluation after a new diagnosis of UTI complicated by a right distal ureteral stone. The patient underwent ureteroscopy and laser stone fragmentation. During the endoscopic procedure a mature, well-epithelialized ureteral tract to the peritoneal cavity was encountered. The procedure was converted to open exploration through a right Gibson incision, where an appendicular phlegmon associated with an uretero-appendicular fistula with passing of faecoliths to the ureteral lumen was found. An appendectomy was performed along with primary fistula closure. Uretero-appendicular fistulas represent a rare entity. Our case highlights the relation between this type of fistula with appendicitis, its unexpected intraoperative diagnosis and the unusual presentation, as the patient did not present clinical findings compatible with acute abdomen.

Key Words: Uretero-appendicular fistula; appendicitis; ureteral stone; child.

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Introduction

Uretero-appendicular fistulas is a rare entity closely related to appendicitis. The reported etiologies include appendicitis, complicated radical hysterectomy, lymphoblastic leukemia, calculi and foreign objects [1-6]. The diagnosis is seldom considered pre-operatively, thus it is usually detected intraoperatively in an unexpected fashion. Here we present an unusual case in a child with recurrent urinary tract infections (UTIs) associated with obstructive hydroureteronephrosis without clinical findings consistent with appendicitis (or an acute abdomen). The patient underwent endoscopic exploration to address a right distal obstructive ureteral stone. During ureteroscopy and LASER stone fragmentation, an uretero-appendicular fistula associated with an obstructive intra-ureteral faecolith was encountered. An appendectomy was then performed, along with primary closure of the fistula.

Case report

A ten years-old female patient presented with recurrent febrile UTIs since the age of three. She had been previously admitted twice with a diagnosis of febrile UTI, yet no imaging studies were performed or antibiotic prophylaxis prescribed. After these two episodes, she remained asymptomatic for a six-year period without further UTIs or unexplained abdominal pain. At the age of nine, the patient presented with an episode of right flank pain associated with irritative urinary symptoms and a new episode of UTI was diagnosed. In the course of that year the girl had a total of three episodes of febrile UTIs, therefore additional studies were performed. Renal ultrasound showed SFU (Society of Fetal Ultrasound) grade II right hydroureteronephrosis. A ^{99m}Tc-Dimercaptosuccinic acid (^{99m}Tc-DMSA) scan reported bilateral multiple photopenic areas with normal differential function. Voiding cystourethrogram (VCUG) was negative for urinary reflux. Intravenous pyelogram showed a distal 9 mm ureteral calculus with associated with moderate to severe hydroureteronephrosis. Abdominopelvic computed tomography (CT) scan reported a right distal obstructive stone with ipsilateral hydroureteronephrosis. She was referred and treated as an out-patient in the general urology and pediatric nephrology clinics, undergoing metabolic screening and symptomatic medical treatment for her flank pain. Due to persistence of her right flank pain and associated with irritative urinary symptoms, she was referred to a quaternary care hospital for urolithiasis treatment.

The patient was admitted to the emergency room (ER) department in good general condition, abdominal pain was well controlled and had no signs of systemic inflammatory response. Physical examination revealed tenderness in right flank. No signs of peritoneal irritation were found. The rest of the clinical examination was normal. Laboratory findings revealed an elevated white blood cell count of 17.670/mm3 (Neutrophils: 90.7%). The hematocrit of 39.5% and platelets of 298.000/mm3 were noted. The blood urea nitrogen (18 mg / dL) and creatinine (0.48 mg / dL) levels were within normal limits. Urinalysis showed more than 20 CPF (Cells per field) leukocytes. Because of her clinical history of recurrent UTIs previously treated on another institution, broad-spectrum antibiotics were started based on the institutional infectious diseases policies.

The abdominopelvic CT scan was re-assessed and the aforementioned findings were confirmed [Figures 1-4]. Considering the dose

of radiation that a tomography would imply, an ultrasound of the urinary tract was done. The report showed worsening (grade IV) right hydroureteronephrosis with global increase in echogenicity. The left kidney was normal with no hydronephrosis or nephrolithiasis. The bladder could not be examined because of the presence of an indwelling foley catheter.

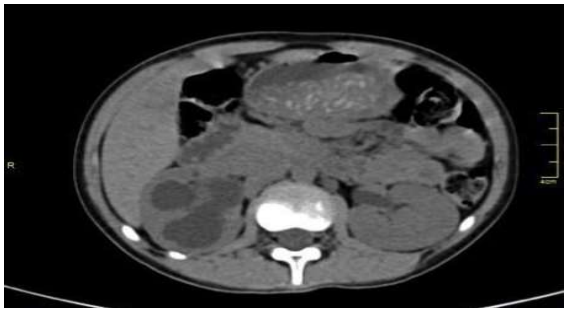

Fig. 1. Right pyelocalyceal dilatation is seen along with a decrease in the cortico-medullary relation and loss of renal parenchyma.

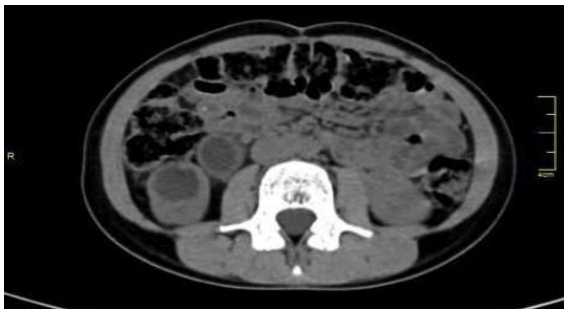

Fig. 2. Important right pelvic and proximal ureter dilatation is observed. Thickening of the right ureter's wall is also seen.

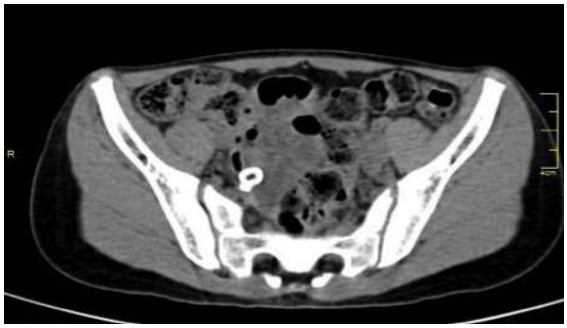

Fig. 3. A high-density image with central hypodensity corresponding to a faecolith is observed completely occluding the distal right ureter.

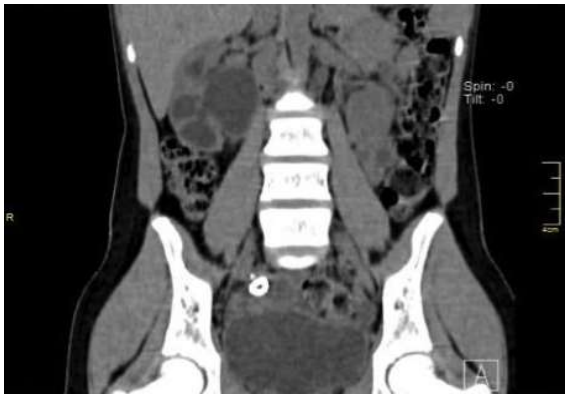

Fig. 4. An important right pelvic dilatation compromising the middle and inferior calyx is appreciated. Dilatation of the right ureter is seen until appearance of a high density image with central hypodensity underneath the iliac vessels. No bladder alterations are observed.

Given her worsening of hydronephrosis and recurrent UTIs, a new DMSA scan was sought in order to evaluate the function of the right renal unit, which showed a decrease on the uptake of the right kidney with a differential function of 21.6%. The left kidney was reported as normal without photopenic areas.

Once the infection was treated, as documented with a negative urine culture, the patient underwent ureteroscopy and laser stone fragmentation. The stone was found to be adherent to the ureteral wall. After partial blasting and removal of the fragments, an epithelialized ureteral communication to the peritoneal cavity was encountered. The lithotripsy was aborted and access to the proximal ureter secured with a double J stent [Fig. 5]. The child was then re-positioned supine, and an open procedure through a right Gibson incision was performed. The peritoneal cavity was explored and an appendicular phlegmon associated with an uretero-appendicular fistula (with passage of faecoliths into the ureteral lumen) was seen. An intra-operative consult to pediatric surgery was made and an appendectomy was performed.

Once the appendix was removed, a severe inflammatory and fibrotic process was seen at the right distal ureter. This phlegmon impeded its mobilization and dissection needed for an end-to-end uretero-ureterostomy. Therefore, we elected to perform a primary closure of the fistula. The edges were debrided and approximated with PDS 6-0 interrupted stitches. We considered that the patient had a high risk of prolonged urinary leak and/or urinary fistula, thus a peri-ureteral drain and a Foley catheter were left to straight drainage.

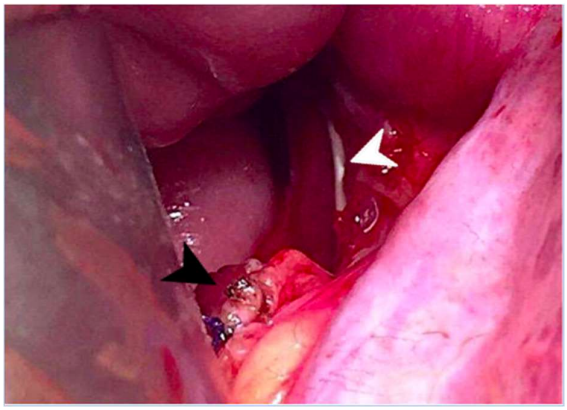

Fig. 5. Ureteral fistula after removal of the appendicular phlegmon. Note the ureteral stent in situ (White arrowhead) and the appendicular stump adhered to the ureter (Black arrowhead).

Her recovery was unremarkable. Pain was well-controlled and she experienced minimum output through Penrose drain. The Foley catheter was removed on postoperative day four. After catheter removal an increase in fluid drainage, which was consistent with urine leak due to elevated creatinine level. A new bladder catheter was placed and maintained for an additional three weeks, with expected subsequent decrease in drainage through the Penrose. Since the double J stent facilitates retrograde urinary flow, a VCUG was obtained as a minimally invasive way to document a retrograde pyelogram. The VCUG showed reflux without evidence of urinary leak at the closure site. The Foley catheter was removed followed by the Penrose drain after 48-hour observation. The patient was then discharged and awaits cystoscopy and stent removal as an outpatient.

Discussion

Uretero-intestinal fistulas are uncommon, [1,2] and can involve most of the gastrointestinal tract, including duodenum, jejunum, ileum, and colon [2]. Of those, uretero-appendicular fistulas are extremely rare [1,2]. A review of literature showed six published cases [1-6], and only one of them involved a pediatric patient [3]. The reported causes of uretero-appendicular fistula include appendicitis, [1,2] complicated radical hysterectomy, [5] lymphoblastic leukemia, [3] periureteritis associated with ureteric calculi [4] and a retained foreign object [6].

The most common symptoms associated with this entity involve right flank pain, right lower quadrant tenderness, fecaluria, recurrent UTIs, diarrhea and leakage of urine from the anus [1,2]. In our case, the patient's symptoms included right flank pain and recurrent UTIs. Notably, right lower quadrant tenderness or abdominal pain suggestive of an episode of appendicitis were never documented.

The most frequently reported mechanism for developing this type of fistula is appendicitis [1,2]. In our case the uretero-appendicular fistula most likely developed concurrently with an episode of appendicitis. The inflammatory process led to adhesion to the retroperitoneum located next to the distal third of the right ureter. This explains the findings during surgery, consistent with an inflamed retro-cecal appendix that drained into the ureter with a secondary peri-ureteral phlegmon. In addition, the patient received multiple antibiotics due to UTIs, helping to control the

local infection, and perhaps avoiding peritonitis and symptoms of acute abdomen. The formation of the fistula allowed the migration of a fecalith into the ureter, facilitating stone formation around the foreign body, and causing a right intraluminal obstructive ureteral process.

The diagnosis of urinary fistula depends on the location of the suspected injury [2]. Radiologic studies have been described as useful [7]. IV pyelogram, retrograde pyelogram and retrograde urethrogram are mainstays in the diagnosis of fistulas involving the urinary tract [7]. However, in most cases the uretero-appendicular fistula is often found intraoperatively after an initial diagnosis of suspected appendicitis [1,5]. In our case, we initially focused to her medical care, a complicated UTI secondary. The surgical approach was determined by the CT scan and clinical history of recurrent UTIs. The uretero-appendicular fistula was an intraoperative finding after seeing a ureteral lesion communicating to the peritoneal cavity while performing an endoscopic procedure.

Conclusions

The uretero-appendicular fistula is a rare entity. We underline the relation of this type of fistula with appendicitis, it's intraoperatively diagnosis and the unusual presentation of this case, since the patient did not present clinical findings compatible with acute abdomen. We consider important to emphasize the usefulness of the VCUG with a double J stent in place allowing direct visualization of the ureter in order to evaluate the integrity of the ureter, ruling out any potential urinary fistula.

Conflict of interest

None of the authors have any competing interests in the manuscript.

Consent

Verbal consent was obtained from the patient and her mother for the report of this case. Signed consent was not retrieved because the patient was sufficiently anonymized according to the ICMJE guidelines.

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