Contents lists available at ScienceDirect

# **Urology Case Reports**

journal homepage: www.elsevier.com/locate/eucr



Joshua Makary<sup>a,b,\*</sup>, Peter Galloway<sup>c</sup>, Pascal Mancuso<sup>b,d</sup>

<sup>a</sup> The University of Sydney, Sydney, NSW, Australia

<sup>b</sup> Campbelltown Hospital, Sydney, NSW, Australia

<sup>c</sup> John Hunter Hospital, Newcastle, NSW, Australia

<sup>d</sup> Liverpool Hospital, Sydney, NSW, Australia

ARTICLE INFO

Keywords: Fistula Renal calculus Sepsis Nephrectomy

### ABSTRACT

In this case report we explore the clinical journey of a 66-year-old female patient that developed a ureteroduodenal fistula. This urological complication is extremely rare and to our knowledge, this is only the third female described in the literature with this finding. We discuss the risk factors associated with uretero-duodenal fistula and its management in the acute setting.

# 1. Introduction

Uretero-duodenal fistula (UDF) is an exceedingly rare complication in the field of urological surgery. In this report we discuss the rare finding of UDF in a female patient that presented acutely with severe urosepsis.

### 2. Case presentation

### 2.1. Background

A previously well 66-year-old female patient was initially referred to our urology department in March 2019, after presenting to her local emergency department with lower abdominal pain. The patient's vital signs were within the normal range and urine/blood investigations unremarkable. A CT abdomen/pelvis (Fig. 1 and Fig. 2) was also requested and incidentally revealed a large 35mm right sided pelvi-ureteric junction (PUJ) calculus and features of a xanthogranulomatous pyelonephritic (XGP) kidney. Given the mild nature of the abdominal pain, lack of associated flank pain or urinary symptoms and unremarkable blood investigations, the patient was deemed safe for outpatient management with a plan to return for elective surgery. Management options were discussed with the patient and she was consented for an endoscopic procedure.

### 2.2. First procedure

Subsequently, the patient underwent a ureteropyeloscopy and lasertripsy surgery in June 2019. Urine culture prior to the procedure revealed mixed growth. During the case, polyps were observed along the length of the right ureter in addition to pus in the renal pelvis. Due to the poor visibility and increased risk of sepsis, a decision was made to discontinue and plan for a repeat procedure. Prior to completion of the case an intraoperative retrograde pyelogram (RPG) revealed no evidence of fistula and a ureteric stent was inserted. The patient recovered well postoperatively and was discharged with a course of oral antibiotics in a stable condition. Ureteric biopsies of the polyps taken at the time of surgery, later revealed only inflammatory changes.

### 2.3. Second procedure

The repeat pyeloscopy procedure conducted in August revealed only a small volume of renal calculi that were subsequently treated with lasertripsy, there were no reportable complications. A ureteric stent was also inserted and intraoperative RPG was unremarkable. The ureteric stent was removed a week later in the outpatient clinic.

### 2.4. Acute presentation

The patient later presented a month later in September with severe sepsis, characterised by hypothermia, hypotension, tachycardia and tachypnoea. Urgent investigations revealed elevated inflammatory

https://doi.org/10.1016/j.eucr.2021.101969

Received 13 November 2021; Received in revised form 2 December 2021; Accepted 5 December 2021 Available online 6 December 2021 2214-4420/© 2021 Published by Elsevier Inc. This is an open access article under the CC BY-NC-ND license (http://creativecommons.org/licenses/by-nc-nd/4.0/).







<sup>\*</sup> Corresponding author. Campbelltown Hospital, Therry rd, Campbelltown, NSW, 2560, Australia. *E-mail address:* Joshua.Makary@health.nsw.gov.au (J. Makary).



Fig. 1. CT abdomen/pelvis (initial CT scan, March 2019). Large right PUJ calculus seen. Loss of tissue plane between duodenum and right proximal ureter can be appreciated.



Fig. 2. CT abdomen/pelvis (initial CT scan, March 2019). Classic "Bear paw" sign associated with right XGP kidney.

markers, moderate renal impairment and a positive urinalysis (leukocytes, nitrites and blood all positive, urine culture revealed mixed growth). Blood cultures taken revealed *Proteus mirabilis*. A CT KUB also done at the time revealed significant right sided peri-nephric stranding, in addition to the previous findings.

After initial resuscitation and treatment for sepsis, an emergency cystoscopy with RPG and stent insertion were conducted. RPG revealed a uretero-duodenal fistula (Fig. 3), with contrast seen tracking from the proximal ureter to the D2 portion of the duodenum. Given the critical condition of the patient and the operative findings, an urgent transfer to a tertiary hospital intensive care unit was organised.

# 2.5. Third procedure

To achieve source control of the patient's sepsis, a right sided nephrectomy was conducted with the assistance of the general surgery team. Initially a laparoscopic approach was employed to achieve bowel



Fig. 3. RPG reveals contrast draining in to duodenum. Proximal curl of the ureteric stent can also be seen in the right renal pelvis.

mobilisation and identification of the ureter. However, conversion to open surgery was later required to achieve adhesiolysis and haemostasis, no resection of duodenum was required. Post-operative management included input from both general surgery and urology services. Day 3 post operatively the patient was tolerating fluids and progressed to a full diet by Day 5. After an uneventful post-operative recovery, the patient was discharged home.

### 2.6. Follow-up

6 months post-nephrectomy a follow-up appointment confirmed that the patient continued to remain well, with nil urinary symptoms or pain issues. A repeat CT abdomen/pelvis revealed no abnormal collections in the nephrectomy bed.

#### 3. Discussion

Since the first reported case in 1918, there have been only 16 cases in the English language that have reported on this topic.<sup>1</sup> A review of prior UDF cases reveals relevant risk factors that are in keeping with the known and well-established causes of fistula development in general. In relation to UDF, risk factors that were identified included chronic infection, duodenal pathology, trauma and iatrogenic causes.<sup>2,3</sup>

Although the patient in our case is only the third known female patient to have UDF, there are similarities to the previously reported cases in the literature.<sup>4,5</sup> The first female patient described with UDF was also treated with nephroureterectomy and fistulectomy.<sup>4</sup> Evidence of chronic infection in the form of a right XGP kidney, and also a concurrent large PUJ calculus, likely contributed to UDF development in our patient. The subsequent instrumentation of this chronically infected urinary system may also have increased the risk of fistula formation.

### 4. Conclusion

In this case an XGP kidney and presence of a large PUJ calculus contributed to the increased risk of fistula development post ureteric instrumentation. The acute management was guided by sepsis control principles, which ultimately necessitated a nephrectomy.

### Funding

Nil source of funding to declare.

# Urology Case Reports 41 (2022) 101969

# Declaration of competing interest

The authors declare that they have no conflict of interest.

# References

- Davis EG. Duodeno-ureteral fistula of spontaneous origin: report of case. J Am Med Assoc. 1918;70(6):376–378.
- Patel B, Yande S, Kumar S, Talaulikar A. Ureteroduodenal fistula. J Surg Case Rep. 2015;2015(7):rjv065.
- Choorit T, Attawettayanon W, Chalieopanyarwong V. Uretero-duodenal fistula as a result from ureteropelvic junction obstruction correction: case report. J Health Sci Med Res. 2020.
- Shiff BM, Welling RE, Rege P. Rare case of left-sided ureteroduodenal fistula. *Dig Surg.* 2000;17(3):283–284.
- Srivastava DN, Singh DK, Wishard NJ. Spontaneous duodeno-ureteric fistula. *Australas Radiol.* 2002;46(4):416–417.