International Surgery Journal
Amry MAM et al. Int Surg J. 2026 Jul;13(7):1239-1243
https://www.ijsurgery.com
pISSN 2349-3305 | eISSN 2349-2902
DOI: https://dx.doi.org/10.18203/2349-2902.isj20262004

Case Report

Continent appendicovesicostomy in a patient with congenital obstruction of the renal pelvis and multiple kidney transplantations: a detailed case report

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Received: 10 May 2026
Accepted: 16 June 2026

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ABSTRACT

Congenital obstruction of renal pelvis, also known as ureteropelvic junction (UPJ) obstruction presents as a functional obstruction at the ureteropelvic junction primarily resulting from smooth muscle abnormalities of ureter and pelvis. Appendicovesicostomy commonly known as the Mitrofanoff procedure, is a reconstructive urological surgical technique that utilizes the appendix to create a continent catheterizable channel among the skin and bladder. A 30-year old patient visited the surgical department of Grodno regional clinical hospital with complaints of periodic urine leakage from a fistula in the umbilical region. The condition had been present for a prolonged period with a past medical history of congenital obstruction of renal pelvis and multiple kidney transplantations. Preoperative laboratory investigations revealed mild anemia, with mild decrease in hemoglobin levels, with normal leucocyte and platelet count. A diagnosis of a persistent fistula associated with appendicovesicostomy was established based on these findings from fistulography, CT and cystography. This case report presents a rare persistent umbilical fistula following a continent appendicovesicostomy in a patient with a history of congenital renal pelvis obstruction and multiple kidney transplants.

Keywords: Continental appendicovesicostomy, Kidney transplantation, Case report, Congenital obstruction of renal pelvis, Fistula

INTRODUCTION

Congenital obstruction of renal pelvis, also known as ureteropelvic junction (UPJ) obstruction is presented as a functional obstruction at the ureteropelvic junction primarily resulting from smooth muscle abnormalities of ureter and pelvis. This obstruction is generally results from functional narrowing of aperistaltic segment of ureter or structural integrity disruption during development involving the smooth muscle of ureter.[1] This is a significant urological condition responsible for impaired renal drainage, hydronephrosis and if left untreated, leads to development of end-stage kidney from progressive renal damage.[2] The prevalence of this anomaly varies; however, the overall congenital anomalies of the kidney and urethral tract is 3 to 6 births per 10,000 live births mostly caused by posterior urethral valves.[3] This condition remains to be a significant cause of paediatric and adult renal morbidity. The incidence of congenital obstruction of renal pelvis is around 1 in 1000 to 1500 newborns.

Thus, the requirement of the surgical intervention is necessary to preserve renal function and reduce the frequency of UPJ-related complications.[4] In addition, the current advances of renal transplantation have significantly elevated the prognosis of patients with end-

stage kidney disease secondary to congenital obstruction of renal pelvis, but complex challenges such as anatomical and functional variations present in patients remains to persist, thereby hindering the efficacy of the surgical intervention carried out. Appendicovesicostomy commonly known as the Mitrofanoff procedure, is a reconstructive urological surgical technique that utilizes the appendix to create a continent catheterizable channel among the skin and bladder.

This procedure promotes emptying of bladder in patients with neurogenic bladder dysfunction, complex urinary tract reconstruction, congenital anomalies of kidney and urethral tract and other conditions resulting in impairment of normal voiding of bladder. The primary relevance of this technique is the resultant abdominal stoma, which allows patients with severe bladder dysfunction to empty the bladder; thus, providing an alternative urethral catheterization, improve the quality of life, the benefit as a salvage procedure after multiple failed surgeries and when conventional urinary diversion methods are less efficient or carry higher morbidity.

However, the fistula formation remains to be an important complication following an appendicovesictomy, producing a crucial challenge in the postoperative management and prognosis of affected patients. This fistula can lead to the following complications such as urinary leakage, infections and impaired continence; thus, requiring further surgical management to improve the prognosis of the patient. The timely diagnosis of fistula formation and subsequent surgical management can improve the quality of life and surgical outcomes of the affected patient.

This case report highlights a unique illustration of a persevering umbilical fistula in a patient with a complicated urological and transplant history. The main aim for presenting this case is to elaborate the challenges and management plan related with fistula formation following continent appendicovesicostomy in the background of the multiple prior renal surgeries and the transplantations. The patient's medical history is a noteworthy for congenital obstruction of the renal pelvis and ureteral anomalies resulting in bilateral ureterohydronephrosis and chronic tubulointerstitial nephritis, which lead to bilateral nephrectomy and subsequent kidney transplantations complicated by graft failure and the requirement for dialysis.

Thus, these factors provided the seldom nature and complexity of this clinical case. The main goals of the surgical intervention and management in this case were to tackle the persistent fistula leading to urine leakage, restoring the urinary continence, and prevent complications such as infection or graft dysfunction. Therefore, this involved an elaborated diagnostic evaluation to verify the fistula's communication with the urinary bladder and careful planning of corrective surgery individualized to the patient's complex urological anatomy and transplant status.

This case highlights the significance of individualized management in patients with complicated urological histories and surgical procedures. This case report details the occurrence of fistula formation following continental appendicovesicostomy in a patient with congenital obstruction of the renal pelvis and multiple kidney transplantations, emphasizing the surgical strategies employed for its resolution.

CASE REPORT

The patient visited the surgical department of Grodno University Clinic with complaints of periodic urine leakage from a fistula in the umbilical region. The condition had been present for a prolonged period and it significantly affected his daily activities. From the patient's medical history, it was found that previously he had been diagnosed with congenital obstruction of the renal pelvis and congenital anomalies of the ureter which led to bilateral ureterohydronephrosis and chronic tubulointerstitial nephritis.

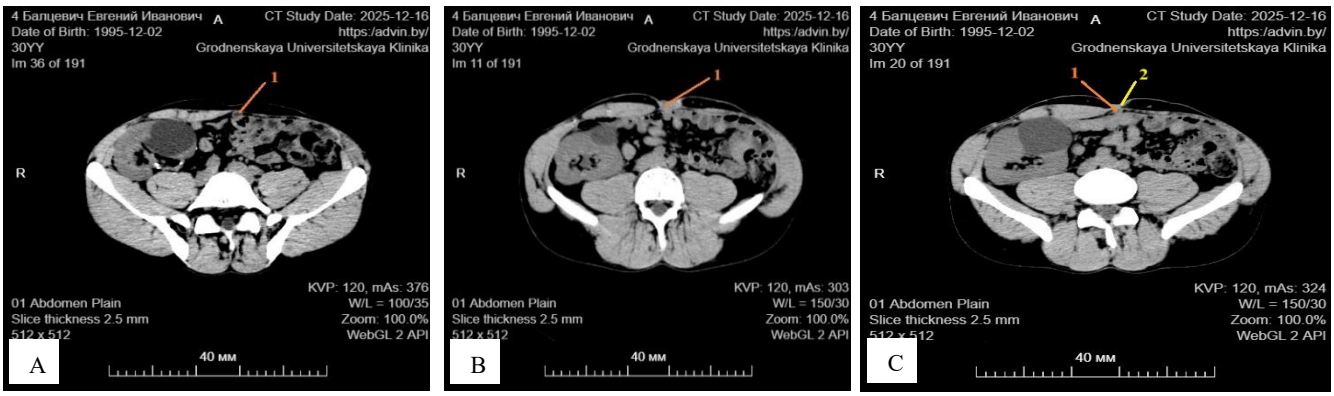

Figure 1: (A) abdominal CT showing the appendix (shown in arrow 1), (B) abdominal CT showing the appendix (shown in arrow 1) and skin (shown in arrow 2) and (C) abdominal CT showing the appendiculocutaneus fistula (shown in arrow 1).

Due to progressive renal failure, he had undergone bilateral nephrectomy in years 2010 and 2011 with deceased donor kidney transplantation in 2010. Later, in 2012 graft failure necessitated transplant nephrectomy and initiation of peritoneal dialysis, which was continued until 2014. A repeat kidney transplantation was successfully performed in 2014 and a continent appendicovesicostomy was created to facilitate urinary output. Additional findings included a bladder diverticulum and chronic kidney disease stage C2, with an estimated glomerular filtration rate of 65 ml/min/1.73 m^2.

Diagnostic workup

On admission, the patient's general condition was satisfactory, with stable vital signs. There were no signs of any local or systemic infection. Physical examination revealed a soft, non-tender abdomen without peritoneal irritation. A fistulous opening in the umbilical region with clear fluid discharge was found during local inspection. The surrounding skin appeared intact, without any erythema or maceration.

Lab findings

Preoperative laboratory investigations revealed mild anaemia, with mild decrease in haemoglobin levels, while leukocyte and platelet counts were both within normal limits. Biochemical analysis showed moderately elevated serum creatinine and urea levels, which was consistent with the patient's renal transplant status. Liver enzyme levels showed transient elevation and inflammatory markers including C-reactive protein had a mild increase shown in Table 1. Coagulation profile was within normal limits.

Table 1: CBC and biochemical parameters.

CBC and biochemical parameters	Values
White blood cells/109/l	6.23
Red blood cells/1012/l	4.07
Hemoglobin/g/l	124
Platelets/109/l	242
Hematocrit (%)	37.9
ESR/mm/h	15
C-reactive protein/mg/dl	7.4
Creatinine/mg/dl	148
Urea/mmol/l	9.6
Aspartate aminotransferase/u/l	42
Alanine aminotransferase/u/l	89

Imaging and instrumental findings

Instrumental diagnostic studies which included fistulography, CT and cystography was used to confirm the presence of communication between the fistulous tract and the urinary bladder. A diagnosis of a persistent fistula associated with appendicovesicostomy was established based on these findings shown in Figure A, B and C.

Surgical management

The preoperative planning was done based on the patient's complex surgical and urological history. Preserving the function of the transplanted kidney and minimizing the risk of infection, given the patient's ongoing immunosuppressive status was prioritized. A multidisciplinary team which consisted of urologists, transplant surgeons, anaesthesiologists and nephrologists was involved in the decision-making process to minimize any perioperative risks. Dense intra-abdominal adhesions due to multiple previous surgeries, potential difficulty in identifying and isolating the appendiceal part and the risk of bladder injury or postoperative urinary leakage was identified as challenges during the surgery.

The surgery was performed under general endotracheal anaesthesia. The patient was placed in a supine position, and the operative field was prepared and draped. An incision was made in the skin and subcutaneous tissue around the fistulous opening at the umbilicus. The appendiceal segment forming the appendicovesicostomy was identified and carefully mobilized by dissecting along the entire length of the appendix up to its insertion into the urinary bladder. After complete exposure of the tract, the appendicovesicostomy was fully excised. The bladder anastomotic site was then closed and sealed using watertight sutures. Additional seromuscular sutures were placed to reinforce the closure and ensure complete sealing to prevent leakage. The operative field was inspected to achieve haemostasis and confirm the integrity of the repair. The abdominal wall was then closed in layers, and an iodine-based antiseptic dressing was applied to the wound. A urinary catheter was inserted into the bladder to ensure adequate drainage during the early postoperative period. The procedure was completed without any intraoperative complications.

Postoperative course

Hemodynamic stability, urine output and wound condition was monitored closely. Analgesia, thromboprophylaxis, antibiotics and immunosuppressive therapy was continued. The catheter was removed once the bladder function returned to normal. On follow up the patient remained stable with complete resolution of symptoms.

DISCUSSION

This case highlights the complicated nature of managing a persistent umbilical fistula after a continent appendicovesicostomy in a patient with congenital obstruction of the renal pelvis followed by multiple kidney transplantations The rare nature of such cases in the literature highlights the clinical importance of this case report. On the contrary in the context typical fistula

cases, this patient's condition was augmented by previous bilateral nephrectomies, graft failures, and immunosuppressive therapy, all of which elevated the risk of postoperative complications and resulted in complicated surgical planning.

Compared to standard cases, this patient's history of bilateral nephrectomies and graft failures elevates the risk profile, resonating the findings in studies highlighting the increased perioperative risks in transplant recipients receiving urological reconstructions.[6] The surgical success achieved here, through complete excision of the appendicovesicostomy tract and hermetically sealed bladder by closure, duplicates the approaches depicted in recent literature supporting the careful surgical technique and improved closure minimizes the recurrence and leakage.[7] Contrastingly, some studies propose conservative management or partial tract revision for fistulas, particularly in less complicated patients, whereas this case emphasizes the requirement of definitive excision in a high-risk category patient.[8] Additionally, the interdisciplinary management, involves transplant surgeons and nephrologists alongside urologists, reflecting the current best practices for such complex cases but is rarely documented in simpler fistula cases.[9,10]

The novelty in this report is the detailed preoperative imaging approach merging both fistulography, CT and cystography to exactly map the fistula, which is highly recommended however not applied universally in all studies. This diagnostic precise surgical planning, is a point strengthen by recent studies recommending personalized interventions based on detailed anatomical assessment of the patient. Clinically, this case elaborates several significant lessons: initially, the requirement of individual patient-based treatment approach in cases with complex urological and transplant histories; secondly, the importance of advanced imaging techniques for precise anatomical localization before a surgical procedure; and thirdly, the essential role of multidisciplinary teamwork in managing high-risk surgical patients. These factors improve early recognition and proper surgical intervention utilized in similar cases, potentially declining morbidity caused by fistula formation.

For future cases, this report highlights the implementation of standard protocols for early detection of fistula complications following an appendicovesicostomy, particularly in transplant recipients. Furthermore, it is recommended to consider the complete tract excision with strengthened bladder closure as a definitive surgical solution for these kinds of surgical patients. To improve the long-term outcomes, timely postoperative monitoring is necessary to identify and neutralize complications.

CONCLUSION

This case report presents a rare persistent umbilical fistula following a continent appendicovesicostomy in a patient with a history of congenital renal pelvis obstruction and multiple kidney transplants. Thus, precise imaging with fistulography and cystography guided a successful surgical excision of the appendicovesicostomy tract and hermetically sealed the bladder by closure. The multidisciplinary management preserved graft function and restored urinary continence.

Recommendations

Future studies should focus on standard early detection protocols for high-risk surgical patients, non-invasive techniques, improve the imaging technique used, and long-term outcome studies to refine the surgical success achieved and patients' prognosis in similar complicated clinical scenarios.

Funding: No funding sources
Conflict of interest: None declared
Ethical approval: Not required

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Cite this article as: Amry MAM, Romualdovna OA, Direcksze NDKN, Dineshkumar PA, Hasnaa MAF. Continent appendicovesicostomy in a patient with congenital obstruction of the renal pelvis and multiple kidney transplantations: a detailed case report. Int Surg J 2026;13:1239-43.