

Long-term Outcomes of Modified Indiana Pouch Urinary Diversion in Pediatric-onset Lower Urinary Tract Disease

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What's known on the subject? and What does the study add?

Indiana pouch is effective in adults; pediatric long-term data are limited. Very long-term pediatric outcomes, highlighting high stone risk

Abstract

Objective: Lower urinary tract dysfunction in pediatric patients can lead to reduced bladder capacity, high intravesical pressures, hydronephrosis, renal deterioration, and incontinence. While bladder augmentation with clean intermittent catheterization is the standard approach, some patients are unsuitable candidates or experience failure. In these cases, continent urinary diversion using the modified Indiana pouch (IP) may provide protection of the upper urinary tract and social continence. Although the IP procedure has been extensively studied in adults, long-term data in pediatric populations remain limited. We present our 23-year single-center experience with pediatric-onset IP urinary diversion.

Materials and Methods: We retrospectively reviewed medical records of all patients undergoing IP diversion between January 1996 and January 2019. Twelve patients received the procedure; one adult with post-traumatic pelvic injury was excluded, leaving 11 with pediatric-onset pathology. Data collected included demographics, indication for surgery, perioperative complications, renal function, hydronephrosis, continence status, stone events, and long-term complications. Patients were followed with ultrasonography, biochemistry, and clinical evaluation every 3-6 months. Descriptive analysis was performed.

Results: Median age at surgery was 15 years (range 7-28), and median follow-up was 122 months (20-243). Indications included bladder exstrophy (n=9), neurogenic bladder secondary to radiotherapy for rhabdomyosarcoma (n=1), and complex urogenital anomaly with bilateral renal dysplasia (n=1). Early complications included five febrile urinary tract infections (45%), one late urinary tract infection, and three cases of hemorrhage requiring transfusion. Hydronephrosis improved in 66% of patients. Median preoperative glomerular filtration rate (excluding transplant cases) was 112 mL/min/1.73 m² (27.8-129), declining to 71.1 (20-123) at final follow-up; three patients developed chronic kidney disease, but none required dialysis. Nine patients (82%) achieved both daytime and nighttime continence, one achieved daytime continence only, and one remained incontinent. Stones developed in 7 patients (63%), requiring 16 surgical procedures: 5 open, 7 endoscopic, and 4 percutaneous. One open procedure resulted in pouch-skin fistula. Ostomy revision was required in three patients. No patient developed metabolic acidosis, B12 deficiency, or secondary malignancy.

Conclusion: The IP is a safe and effective long-term option for pediatric-onset lower urinary tract dysfunction, providing renal preservation and high continence rates. Stone formation is a frequent late complication, highlighting the importance of lifelong irrigation, surveillance, and multidisciplinary follow-up.

Keywords: Indiana pouch, pediatric, pediatric urology, reconstructive surgery

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Received: 21.06.2023 **Accepted:** 20.10.2025 **Epub:** 23.02.2026

Cite this article as: Aşçı A, Albayrak E, Altan M, Doğan HS, Tekgöl S. Long-term outcomes of modified indiana pouch urinary diversion in pediatric-onset lower urinary tract disease. J Urol Surg. [Epub Ahead of Print]

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Introduction

A healthy bladder has the capacity and compliance to store urine in a low-pressure environment while enabling voiding in a socially acceptable manner, thereby protecting the upper urinary tract and providing social continence. Patients with lower urinary tract pathologies may have low capacity, hypo-compliant bladders leading to higher filling pressures, upper urinary tract deterioration, and incontinence. This is a more prominent problem in the pediatric patient group. As children have a longer life expectancy, it is paramount to obtain a low-pressure urinary tract and protect renal function. Providing social continence is also crucial for the healthy psychosocial development of juveniles, as it has an immense impact on their quality of life (1).

Bladder augmentation techniques in conjunction with clean intermittent catheterization (CIC) may help these patients to protect the upper urinary tract and achieve social continence. Nevertheless, in some cases augmentation procedures may fail, or the present structure of the bladder may not be suitable for augmentation. A continent urinary diversion procedure may be performed in end-stage bladder dysfunction or lower urinary tract disease, where reconstruction is not feasible and preservation of continence is desired. The modified Indiana pouch (IP) method was described in 1991 and has gained popularity with long-term studies including many adult patients (2-4). As IP requires lifelong self-catheterization and follow-up, patient selection is crucial. Cognitive and manual ability to reliably perform CIC, or having a caregiver who can perform this task are essential. Adequate renal function is required as acid-base and fluid-electrolyte alterations can occur after continent urinary diversions. The patient must have a sufficient ileo-cecal segment without any contraindications such as short-bowel syndrome or inflammatory bowel disease. Normal liver function is also required, as patients with liver disease are more prone to metabolic derangements. The absence of severe comorbidities is likewise necessary. Finally, motivation for this procedure is important, especially in patients with pediatric-onset diseases as patients will commit to lifelong stoma care and CIC.

IP has been extensively performed in adult patients; however, literature on the utilization of the IP technique for pediatric patients is scarce. We aimed to share our 23 years of experience in continent urinary diversion using the IP procedure, performed for pediatric urogenital pathologies.

Materials and Methods

Hospital medical records were retrospectively examined to identify patients who underwent an IP, continent urinary diversion surgery between 01 January 1996 and 01 January

2019. A total of 12 patients received IP surgery in this period, and had long-term outcomes available, of which 11 had pediatric-onset urogenital pathologies. The other patient was an adult with a history of pelvic trauma and was excluded from the study. All surgeries were performed according to the original method proposed by the Indiana University group, which is also described in detail by Chowdhary et al. (5). All patients were hospitalized at least 2 days before surgery. Patients were started on a clear diet two days prior to surgery, and gut irrigation was performed on the day before surgery.

Nine patients had bladder exstrophy 1 had neurogenic bladder due to radiotherapy for paratesticular rhabdomyosarcoma, and 1 (patient A) had urogenital sinus anomaly, bilateral multicystic dysplastic kidney, and a hypoplastic bladder.

Hacettepe University Ethics Boards and Commissions, Non-Interventional Clinical Researches Ethics Board; acceptance number: GO15/621, date: 07.10.2015, date: 07.10.2015.

Statistical Analysis

All patients received concurrent cystectomy, while patient A, also received a bilateral nephroureterectomy. One patient with a history of bladder exstrophy had prior renal transplantation with a ureterocutaneostomy (patient B). In the postoperative period, patients were scheduled for follow-up examinations every 3 to 6 months; routine follow-up protocol included renal and urinary ultrasonography, serum biochemistry, along with evaluation of patients' symptoms. The patients' demographics, primary etiologies, preoperative and postoperative hydronephrosis status, postoperative early and late complications, stone complications, postoperative metabolic and continence status, were noted. Descriptive statistics were obtained using SPSS version 25.

Results

The median patient age at the time of surgery was 15 (7-28) years and the median follow-up period was 122 (20-243) months. Five patients developed febrile urinary tract infections (UTIs) within the first month, and one patient developed a late postoperative febrile UTI. There were no grade 3 or higher complications according to the modified Clavien-Dindo scoring system. Hemorrhage requiring transfusion occurred in three patients.

Nine patients (except Patients A and B) had preoperative hydronephrosis, which resolved completely in 2 patients (22%) and partially in 4 patients (44%) postoperatively. Preoperatively, 1 patient had grade 1, 2 patients had grade 2, and 6 patients had grade 3 hydronephrosis. At the last follow-up, two patients had no hydronephrosis, one patient had grade 1 hydronephrosis, five patients had grade 2 hydronephrosis, and one patient had

grade 3 hydronephrosis. All patients, except for four who had relatively shorter follow-up periods (22, 32, 85, and 96 months), developed stones in the pouch (63%). No association was found between hydronephrosis status and stone development. Only 3 patients experienced calculi within 5 years, whereas the other 4 presented 164 to 240 months after surgery. In these 7 patients, a total of 16 stone surgeries were performed, consisting of 5 open stone removals, 7 endoscopic stone removals (via the continent stoma), and 4 percutaneous stone surgeries. One open stone surgery led to the development of pouch-skin fistulae, and required a secondary surgery for repair.

One patient developed a stricture in the left ureterocolonic anastomosis and underwent a re-implantation at postoperative month 85. One patient in the early postoperative period required ostomy revision due to leakage. Two patients in the late postoperative period (postoperative 12 and 140 months, respectively) required ostomy revisions due to stenosis.

Only one patient had chronic kidney disease preoperatively, aside from two special cases: Patient A, who received a renal transplantation 6 months after IP surgery, and Patient B, who had already received a renal transplantation. The preoperative median glomerular filtration rate (GFR) was 112 mL/min/1.73 m² (range 27.8–129), excluding patients A and B. In the postoperative period, 3 patients eventually developed chronic kidney disease, but no patient required renal replacement therapy. At the last follow-up, the median GFR was 71.1 mL/min/1.73 m² (range 20–123), excluding data from patients A and B. All patients had normal liver function at the last visit. No patient reported chronic diarrhea, and no patient had significant electrolyte anomalies.

In the postoperative period, all patients performed CIC 4 to 6 times a day. Nine patients have achieved continence day and night via CIC. One patient achieved continence during the day but reported nighttime overflow incontinence requiring one to two urinary pads. Patient A remained incontinent day and night, and used a urostomy bag on the stoma tract.

Discussion

In adults, the IP procedure following cystectomy for malignant bladder conditions is widely performed (2–4). However, literature on the utilization of the IP for pediatric patients is scarce, with relatively few studies, short follow-up periods, and most studies published before the 2000s (6,7). In 2014, Chowdhary et al. (5) published the results of a 15-year prospective study on the IP procedure in children, shedding light on the subject by stating that the procedure is safe and reliable with favorable outcomes in the pediatric population. Nevertheless, their median follow-up period is only 24 months. Based on these findings, we aimed to share our 23 years of experience with IP in pediatric lower

urinary tract pathologies, with a median follow-up of 122 months.

Complications

The procedure proved to be safe, with no mortalities and no grade 3 or above complications according to the Clavien-Dindo classification. Five (45%) of the patients developed febrile UTI in the acute postoperative period, despite proper bowel preparation and perioperative antibiotic prophylaxis. They were successfully treated with wide-spectrum IV antibiotics and discharged accordingly. This rate appears high compared to adult IP (23%) and pediatric augmentation cystoplasty (29.4%) series (8,9). Chowdhary et al. (5) report 16.6% cases of febrile UTI during the early period in 12 pediatric IP patients. Our higher early infection rate may be caused by the local resistance profile of colonizing bacteria in the urinary tract. Türkiye has historically had high antibiotic use and higher rates of antimicrobial resistance compared to the rest of Europe (10,11). This may have rendered perioperative prophylaxis ineffective. It should also be kept in mind that the cohort consists of only 11 patients. Small sample size may have exaggerated the rate of infection. During follow-up, three patients required intervention due to ostomy complications (27%), similar to the literature's values (15–20%) (5).

Hydronephrosis improved in 66% of patients, supporting the goal of providing a low-pressure, high-capacity reservoir. Only three patients developed chronic kidney disease over a prolonged follow-up period, and none required dialysis. No patient developed vitamin B12 deficiency.

Continence

Continence outcomes were favorable: Nine patients (82%) achieved both daytime and nighttime continence, one achieved daytime continence with nighttime leakage, and one remained incontinent. This aligns with reported continence rates of 85–100% (5,6,12).

Stone Formation

Stone formation occurred in 63% of patients, which is higher than previous reports. Chowdhary et al. (5) reported no stones; Kaefer et al. (7): 14%; and Surer et al. (13) 26%. This discrepancy may be due to our longer follow-up period of 122 months, compared to 24, 62, and 72 months, respectively, in the previously mentioned papers. Another possible cause of our high calculi rates may be a lack of patient cooperation. Patients are advised to undergo routine saline irrigation of the pouch at least once a week. Over time, waning adherence may allow mucus aggregation and calculi formation. Symptoms such as new-onset incontinence should prompt evaluation for stones. In patients lost to follow-up, stones may reach excessive sizes, reducing pouch capacity.

Stone Surgery

Another important consideration in the management of pouch stones is the choice of surgical technique. Conventional open stone removal offers the advantage of complete clearance in a single session and is often used in patients with very large stone burdens (14). However, open surgery carries significant risks, particularly in patients with a history of multiple prior operations, including intestinal injury, wound complications, pouch-skin fistulae, and impaired healing, which can result in subsequent urine leakage.

Endoscopic management is increasingly favored as a less invasive alternative. Use of the stoma for stone fragmentation and extraction may be appropriate only when the calculi are small and the overall stone burden is limited, since excessive manipulation risks iatrogenic damage to the stoma and future catheterization difficulties. Our preferred approach is combined stomal entry with percutaneous access under direct endoscopic guidance after filling the pouch with saline. A similar technique has been reported by Öztürk et al. (15) with favorable outcomes and no major complications. Lam et al. (16) described a hybrid endoscopic-laparoscopic method with similarly safe results, and even transvaginal approaches have been explored in isolated cases (17).

Despite these advantages, endoscopic surgery has limitations. Longer operative times and the need for multiple sessions are not uncommon, particularly with large calculi. Because stones cannot be removed en bloc, residual fragments may persist and act as a nidus for recurrence. Szymanski et al. (18) found no significant difference in recurrence rates when comparing open, percutaneous, and endoscopic approaches. Nevertheless, our experience, consistent with the literature, suggests that minimally invasive techniques reduce morbidity and are effective even for large stone burdens, provided patients are monitored for recurrence.

Cancer Risk

Secondary tumor development is a recognized risk after urinary diversion procedures: ureterosigmoidostomy poses the highest risk (2.58% to 10%) (19,20). Nitrosamines, carcinogenic compounds that are produced when feces and urine are mixed, are blamed for this excessively high risk. Adenocarcinoma development in procedures where isolated gut segments are used is very scarce. Kälble et al. (19) found a risk of 0.14% in ileocecal pouch patients.

Some authors have speculated that secondary cancers developing in ileocecal pouches in patients who have been treated for bladder cancer, may be related to an underlying genetic predisposition, such as familial cancer syndromes (20,21). Whereas the underlying mechanism may still be a matter of debate, authors propose regular endoscopic evaluations of the pouch 10 years

after diversion or in the case of symptoms such as hematuria or difficulty with catheterization. In our clinical practice, routine yearly cystoscopy was advised for the patients after 10 years of follow-up. Fortunately, we did not have any secondary cancer development in our cohort. This may partly be related to the fact that none of the patients, excluding one patient with rhabdomyosarcoma, had primary malignant conditions, adhering to the hereditary cancer syndromes theory.

Study Limitations

We acknowledge several limitations to our study. Due to the nature and scarcity of the condition in the pediatric population, the number of patients we have is low. For that reason, only descriptive statistics were presented because it is not feasible to perform statistical analyses. Routine saline irrigation of the pouch was advised to all patients, but as we do not have the means to know how well they have complied with regular irrigations, we cannot make a conclusion as to the benefits of pouch irrigations for preventing stone formation. Saline irrigation of a pouch structure might not be as effective as it would be in a normal or augmented bladder. This study also, does not present any data on patients' subjective quality of life, but our clinical impression shows high patient acceptance of the procedure. Furthermore, we present objective data on the protection of the upper urinary tract and incontinence, which are the main aims of this surgery. Because of the chronic nature of the disease, patients' compliance with follow-up decreased over time, and patients became reluctant to continue follow-up and searched for medical advice as problems developed. All these limitations aside, we believe this study presents an acceptable number of patients who have received IP surgery for pediatric lower urinary tract disorders and includes a very long follow-up period.

Conclusion

In patients in need of reconstructive surgery due to pediatric-onset lower urinary tract pathologies, the IP procedure is safe and effective, providing renal protection and high continence rates. Most patients tolerate the procedure well, with acceptable complication rates. Stone formation is a significant long-term issue. Lifelong irrigation and vigilant management are essential. The surgery also seems effective in preserving renal functions in the long term.

Ethics

Ethics Committee Approval: Hacettepe University Ethics Boards and Commissions, Non-Interventional Clinical Researches Ethics Board; acceptance number: G015/621, date: 07.10.2015, date: 07.10.2015.

Informed Consent: Retrospective study.

Footnotes

Authorship Contributions

Surgical and Medical Practices: A.A., E.A., M.A., H.S.D., S.T., Concept: A.A., E.A., M.A., H.S.D., S.T., Design: A.A., E.A., M.A., H.S.D., S.T., Data Collection or Processing: A.A., E.A., M.A., H.S.D., S.T., Analysis or Interpretation: A.A., E.A., M.A., H.S.D., S.T., Literature Search: A.A., E.A., M.A., H.S.D., S.T., Writing: A.A., E.A., M.A., H.S.D., S.T.

Conflict of Interest: Serdar Tekgöl MD is section editor in Journal of Urological Surgery. He had no involvement in the peer-review of this article and had no access to information regarding its peer-review.

Financial Disclosure: The authors declared that this study received no financial support.

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