

Hourglass bladder after ileocystoplasty (case report)

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Narrowing of the anastomotic area between the bowel segment and urinary bladder is known as “hourglass bladder.” This report describes a rare case of hourglass bladder deformity occurring post-ileocystoplasty in a 14-year-old male. We describe the essential steps that should be taken during bladder augmentation to avoid such a complication, along with a discussion of the relevant literature. This case emphasizes the need to strictly adhere to the steps of augmentation cystoplasty to avoid this technical complication.

In 1959, Goodwin et al described various methods of anastomosing a patch of the ileum to the native bladder.¹ An important step involves opening the bladder wide enough to prevent the formation of a narrow anastomotic area between the bowel segment and urinary bladder (“hourglass bladder” deformity). Although such measures are usually taken, this deformity is still rarely reported in cases of bladder augmentation using techniques such as seromuscular enterocystoplasty or bladder autoaugmentation.^{2,3} However, we recently encountered an instance of this preventable deformity following ileocystoplasty.

CASE

A 14-year-old male child presented with vague abdominal pain and foul smelling urine. His medical history began with a posterior urethral valve, which was incised at 1 year of age. He subsequently developed valve bladder syndrome, and augmentation ileocystoplasty was performed at his local hospital at 4 years of age. Since then, he had maintained regular clean intermittent catheterization (CIC) per urethra nearly five times per day, with overnight drainage. The patient was continent between CICs, with no history of recurrent febrile urinary tract infection (UTI), and had stable renal function.

His physical examination was unremarkable. Renal function testing revealed normal serum urea, creatinine, and electrolyte levels, although urine culture was posi-

tive for *Escherichia coli*. Renal ultrasonography detected bilateral grade III hydronephrosis, based on the Society for Fetal Urology grading system,⁴ with both ureters dilated, along with a grossly dilated and trabeculated bladder (Figures 1A and 1B). A micturating cystourethrogram (MCUG) detected the classical hourglass appearance, with no vesicoureteric reflux (Figure 1C).

Cystoscopy revealed normal anterior and posterior urethra with a high bladder neck as well as a small native bladder with a narrow neck connected to a hugely dilated augmented intestinal pouch, which was filled with mucus and debris. A suprapubic catheter was inserted under cystoscopic monitoring as a temporary diversion, and a daily washout was performed.

After 8 weeks, exploratory surgery performed through the previous midline incision revealed a constriction ring between the ileal portion of the augmentation and the native bladder. The two segments were surgically disconnected completely from each other followed by trimming of the ileum edges with complete bivalving of the native bladder in the sagittal plane starting from the anterior bladder to 2 cm proximal to the trigone. An incision of approximately 10 cm was created at the midline of the anterior wall of the augmented bladder, and an anastomosis was performed between the revised pouch and the bivalve of the native bladder.

The patient had an uneventful postoperative course, and was discharged after 2 weeks with a strict CIC pro-

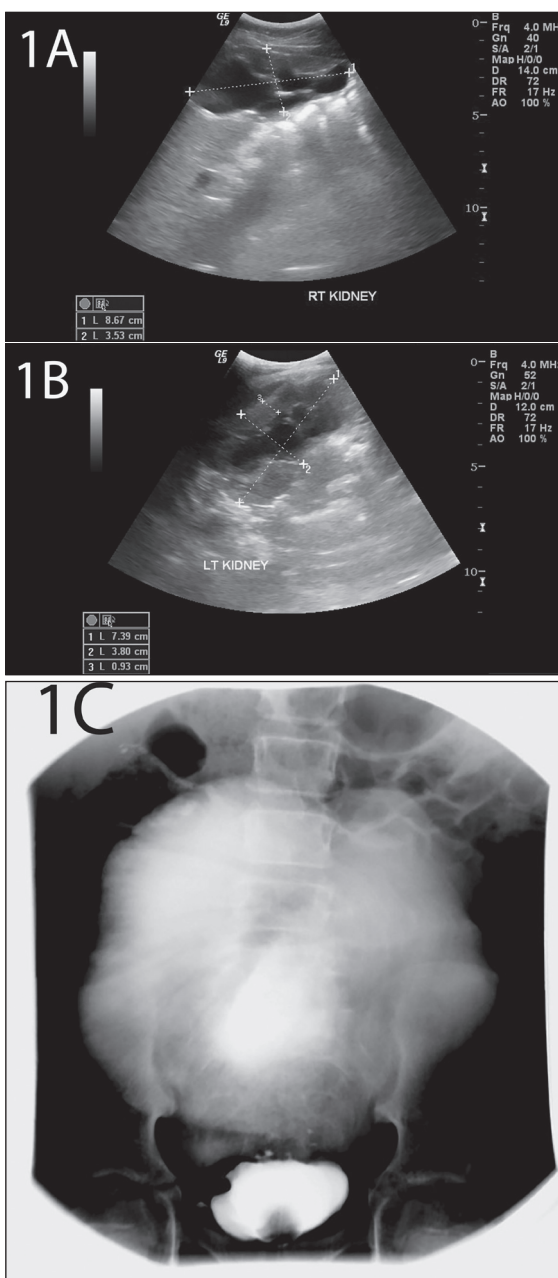


Figure 1. Images obtained at presentation. Renal ultrasonography showing bilateral grade III hydronephrosis (a,b). Micturating cystourethrogram showing a classical image of an hourglass bladder, with large augmented bladder capacity of 950 mL and no vesicoureteric reflux (c).

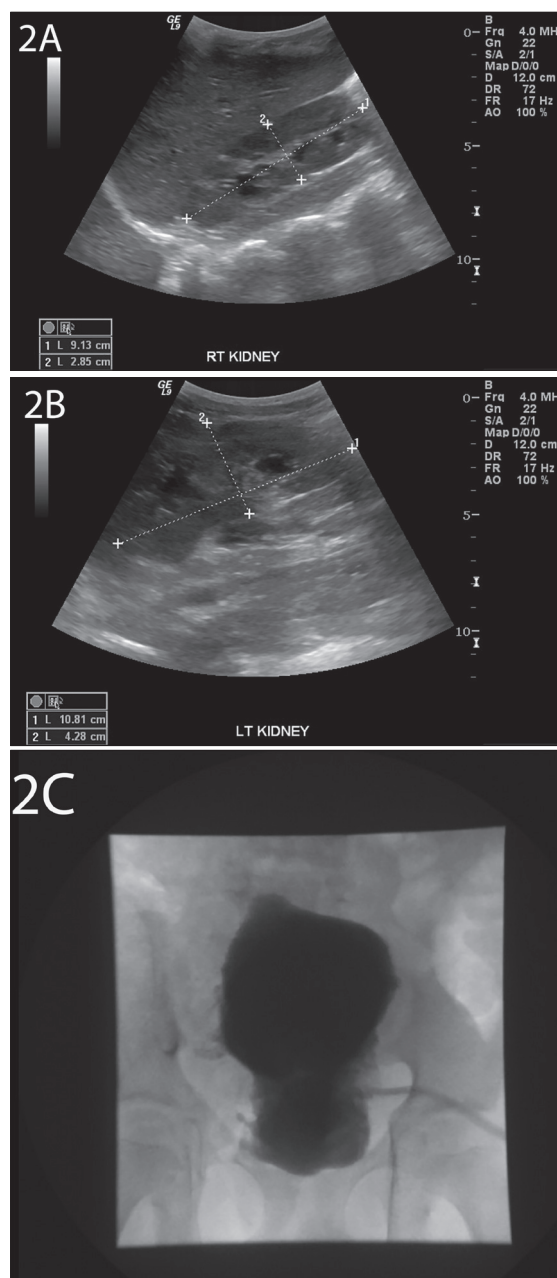


Figure 2. Images obtained 3 months after revision surgery. Renal ultrasonography showing bilateral hydronephrosis, grade 0 to I,(a,b). Micturating cystourethrogram showing the absence of the hourglass bladder and decreased bladder capacity (250 mL) with no vesicoureteric reflux (c).

to col of every 4 hours. At the 3-month follow-up visit, he was asymptomatic, had good compliance with CIC, and was completely continent. Ultrasonography revealed a reduction in the upper tract dilatation (Figures 1A and 1B), with MCUG revealing the absence of an hourglass bladder and decreased bladder capacity to

250 mL (Figures 1C). One year after surgery, the patient was still compliant with CIC, and his upper urinary tract was stable.

DISCUSSION

The bladder’s basic hydrodynamics and biomechanical

properties depend on the relationship between bladder shape, size, pressure, and tension, as expressed by the Laplace law, which explains the relatively constant intravesicular pressure exerted during bladder filling.⁵

Enterocystoplasty is a major reconstructive operation that seeks to protect the upper urinary tract and/or achieve urinary continence by reducing the bladder's storage pressure and increasing its volume, thereby altering the hostile native bladder environment. Hinman and Koff have documented the importance of detubularization and reconfiguration of the bowel segment during this procedure, as this ensures maximum gains in capacity and compliance.^{6,7} Unfortunately, not every augmentation is completely successful, and at least one-third of patients may require additional bladder surgeries. Thus, the success of enterocystoplasty is dependent on long-term follow-up, reliable family support, and life-long compliance with CIC per urethra/catheterizable channel.⁸

Aside from the complications associated with enterocystoplasty, the bladder has a great tendency to reform itself, occasionally converting the augmentation into a diverticulum with a relatively small opening. To prevent this, the bladder should be bivalved from the bladder neck ventrally to the trigone posteriorly. Alternatively, the "star" procedure may be used for thick-walled small bladders, which involves opening the bladder in both the sagittal and coronal planes. Occasionally, an upside-down U-shaped or a Boari flap modification for the initial bladder opening is used when a catheterizable stoma is positioned at the umbilicus. A supratrigonal bladder excision is the final option for thick bladders with severe tuberculosis and neuropathy.⁹

Although an inadequate native bladder opening is thought to be the major mechanism for such deformities, other theories include non-compliance with CIC, detrusor muscle inflammation or dysfunction, and uninhibited intestinal contractions.⁸ Regardless of the type of bladder opening that is used, the goal is the same: to provide a wide, defunctionalized bladder plate to which the bowel segment can be attached, thus preventing a narrow anastomosis or hourglass deformity that might allow the augmentation to become a diverticulum. Patients with this deformity have large volumes of post-void residual urine, due to improper bladder configuration and poor emptying capability.

An hourglass bladder deformity as a congenital anomaly is extremely rare, with only two previously reported cases.^{10,11} Hourglass bladder may occur in patients with mixed neurogenic urinary incontinence,¹² or more commonly in children following seromuscular colocolocystoplasty² and patients undergoing bladder autoaugmentation to treat neuropathic bladder, secondary to myelomeningocele.³ Treatment of this deformity can be safely performed using multiple Heinecke-Mikulicz incisions at the junction between the native and the augmented parts of the bladder.^{2,3} In this case, both parts were safely separated, and the enterocystoplasty was revised.

This case emphasizes the need to strictly adhere to the steps of augmentation cystoplasty to avoid this technical complication. Longer-term follow-up for such cases is necessary, and should include urine culture and abdominal ultrasonography. MCUG and urodynamic testing may be repeated if the ultrasonography reveals new hydronephrosis despite strict CIC compliance.

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