A surprise during hernia surgery: inguinoscrotal megaureter

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ABSTRACT

Background. Ureteroinguinal herniation is a rare occurrence that is typically diagnosed during the surgical repair of inguinal hernias.

Case Presentation. We present the case of a 4-year-old male who underwent inguinal hernia repair, during which a megaureter was discovered within the hernia sac. The surgical intervention included high ligation of the hernial sac and repositioning of the ureter back into the retroperitoneum. Postoperative investigations confirmed a diagnosis of primary non-refluxing and nonobstructive megaureter.

Conclusion. Although ureteral herniation is rare in infants, it is crucial to remain vigilant about the possibility of encountering the ureter during hernia repair to prevent potential ureteral injuries. Additionally, any associated urinary tract anomalies should be thoroughly investigated and ruled out.

Key words: hernia, ureteroinguinal hernia, herniation of the ureter, child.

Inguinal hernia repair is one of the most frequently performed surgical procedures worldwide.1 While inguinoscrotal hernias typically encompass various intraperitoneal organs such as the small intestine, colon, appendix, and ovaries, ureteric herniation into the inguinal canal is exceedingly rare due to the ureter's retroperitoneal location. To date, fewer than 150 cases have been reported in adults, and the condition is even more uncommon in the pediatric population, with only 12 documented cases in children.2 The literature reveals that most ureteral inguinal hernias are discovered during surgical interventions rather than in preoperative evaluations.3 In this article, we present the case of a 4-year-old male who underwent inguinal hernia repair, during which a megaureter was discovered within the hernia sac.

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Case Presentation

A 4-year-old male patient presented to the pediatric surgery clinic complaining of right-sided inguinal swelling. Physical examination identified a reducible mass in the right inguinal region, with both testes appropriately located in the scrotum, and no other remarkable findings. The patient's medical history was free of comorbidities, and pre-operative blood tests were within normal limits. Based on the typical presentation, he was scheduled for a right inguinal hernia repair under general anesthesia, without the need for preoperative imaging.

During the surgical procedure, an open exploration of the right inguinal canal was performed, which revealed a peritoneal sac with a herniated luminal structure behind the sac (Fig. 1). Initially, the nature of this structure was unclear, including whether it represented an anatomical variation or was part of a direct hernia. Aspiration was performed using an injector to elucidate the identity of the luminal structure, and the presence of a

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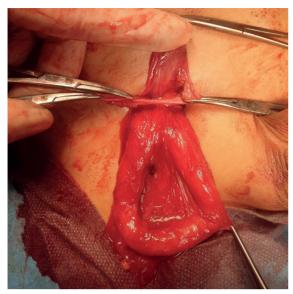


Fig. 1. Ureteral herniation showing the ureter behind the peritoneal sac

urine-like fluid indicated that the structure was the ureter. The ureter was found to originate from the retroperitoneum, with a portion looping within the inguinal canal. The surgical intervention included high ligation of the hernial sac and repositioning of the ureter back into the retroperitoneum. Furthermore, the internal inguinal ring was narrowed to prevent recurrence. The patient was successfully discharged on the same day, a few hours following surgery, without any postoperative complications.

Postoperative assessments were planned for further investigation of the patient's condition. system ultrasonography revealed a minor degree of hydronephrosis, along with bilateral dilatation of the ureters, measuring 15 mm in diameter on the left and 12 mm on the right with normal bladder. Voiding cystourethrography (VCUG) ruled out vesicoureteral reflux (VUR). During the micturition phase, the posterior urethra was observed to be normal, thereby eliminating the diagnosis of posterior urethral valves (PUV). Additionally, diuretic scintigraphy using Mercapto-acetyltriglycine (MAG-3) was performed to exclude obstructive causes of the megaureter. The MAG-3 renography revealed

normal split renal function (right: 48%, left: 52%), with evidence of bilateral megaureter and slow urinary drainage. Near complete drainage was observed after the diuretic injection, with no obstructive urinary pattern detected. The calculation of the half-time ($T_{1/2}$) was recorded as 13.5 minutes on the right and 15.2 minutes on the left. These findings led to a diagnosis of primary non-refluxing and nonobstructive megaureter. Given the absence of any history of urinary tract infections, the patient was recommended for intermittent follow-up in the outpatient clinic. A written informed consent was obtained from the parents of the patient for the publication of this case report.

Discussion

Inguinal hernia is a common diagnosis, often requiring surgical intervention.¹ However, inguinal herniation of the ureter is a rare occurrence, primarily seen in obese, middleaged men.⁴ In infants, this condition is extremely rare, with only 12 cases reported in the literature (as summarized in Table I).^{2,3,5-14} In adults, risk factors for ureteral inguinal hernias include male sex, age over 50, collagen synthesis disorders, a history of kidney transplant, and obesity.⁴ Although risk factors have not been identified in the pediatric population, it is noteworthy that all reported cases have involved male infants, with no instances documented in females to date.

Ureteroinguinal hernias are classified into two main types: paraperitoneal (80%) and extraperitoneal (20%). Paraperitoneal hernias involve a true hernia sac that pulls the ureter into the inguinal canal through traction. In contrast, extraperitoneal hernias lack an associated sac and are thought to arise from incomplete differentiation of the ureter from the Wolffian duct or adhesion between the primitive ureter and the genitofemoral ligaments.^{8,15} Notably, numerous cases of extraperitoneal hernias are associated with renal and urinary tract malformations, such as wandering kidney and transverse renal ectopia. Extraperitoneal hernias

Table I. Reports of cases of inguinal ureteral hernia in children

No	Author	Year	Age/Sex/Side	Type	Associated urinary anomaly	Management
1	Cianci et al. ⁵	2024	2 mo/M/R	Paraperitoneal	Hydroureteronephrosis, VUR	Reduction to the
						retroperitoneal space
2	Delgado-Miguel	2024	2 mo/M/L	Paraperitoneal	Hydroureteronephrosis, VUR	Reduction to the
	et al. ⁶					retroperitoneal space,
						ureteroneocystostomy
3	Hosoda et al. ²	2022	14 y/M/L	Paraperitoneal	A past medical history of left	Inguinal exploration
					inguinal hernia surgery	
4	Wishani et al. 7	2021	4 mo/M/L	Paraperitoneal	Primary obstructed megaureter	Reduction to the
						retroperitoneal space
5	Cao et al. 8	2018	12 y/M/L	Extraperitoneal	Cloacal exstrophy, cross-fused	Transureteroureterostomy
					pelvic kidney	
6	Bosschieter et	2018	3 mo/M/R	Paraperitoneal	Hydroureteronephrosis, VUR	Ureteroneocystostomy
	al. ³					
7	Handu et al. 9	2012	18 mo/M/R	Paraperitoneal	Solitary kidney	Ureteroneocystostomy with
						appendiceal interposition
8	Sripathi et al. 10	2011	10 mo/M/L	Paraperitoneal	VUR	End ureterostomy
9	Burgu et al. 11	2010	4 mo/M/L	Paraperitoneal	Posterior urethral valve	End ureterostomy
10	Powell et al. 12	1985	4 wk/M/L	Paraperitoneal	Megaureter	Reduction to the
						retroperitoneal space
11	Morris et al. 13	1977	6 wk/M/B	Paraperitoneal	Multicystic dysmorphic kidney	Not mentioned
12	Jewett et al. 14	1953	9 y/M/L	Extraperitoneal	Hydroureteronephrosis	Transureteroureterostomy

B: Bilateral, L: Left, M: Male, R: Right, VUR: Vesicoureteral reflux.

are more frequently associated with coexisting anomalies compared to paraperitoneal hernias, according to the literature. 9,11,12 Additionally, extraperitoneal herniation can be an acquired condition due to retroperitoneal fat prolapse and is reported to be relatively more common after renal transplantation. 4 Among the 12 reported pediatric cases in the literature, 2 were of the extraperitoneal type. 8,14 The patient presented here was diagnosed with the paraperitoneal type.

In both types of ureteroinguinal hernias, symptoms are often nonspecific, with the most common presentation being distension of the inguinal region due to ureteral slippage.^{2,9} The majority of patients are asymptomatic, and routine radiological studies are typically not conducted preoperatively.³ Consequently, the diagnosis of ureteroinguinal hernia is often missed until surgical exploration, as in the presented patient. If an ureteroinguinal hernia is encountered during routine herniotomy, the presence of associated urinary tract

anomalies should be investigated. In the case presented, no prior diagnosis related to the urinary system was known, as the patient had no previous symptoms. However, postoperative investigations revealed the primary nonobstructive, nonrefluxing megaureter. The literature reports extraperitoneal hernias are more likely to present with symptoms such as back pain or hernia incarceration due to ureteral obstruction or strangulation, compared to paraperitoneal hernias.8,14 Although preoperative diagnosis is challenging, the possibility of an inguinal ureteral hernia should be considered in cases of inguinal herniation, especially if accompanied by symptoms and signs of ureteral obstruction such as hydronephrosis or hydroureter.

Despite their rarity, surgeons should remain vigilant for the possibility of a ureteroinguinal hernia to avoid inadvertent ureteral injury during herniotomy. Cases of ureteral injury during hernia surgery have been documented in the literature.⁹ Once the diagnosis is confirmed

during surgical exploration, various treatment options exist based on the specific presentation of the herniated ureteral loop.^{3,6,9} Most experts advocate for repositioning the ureter back into the retroperitoneum. However, in cases of ureteral redundancy or injury, resection and anastomosis are recommended.8,14 Endureterostomy has been reported as a management option for extremely dilated ureters, with subsequent plans for reimplantation.^{10,11} The laparoscopic approach in these cases remains controversial. In cases involving adult patients, it has been reported that although laparoscopy may offer enhanced visualization of the ureter entering the inguinal canal, managing hernia repair and achieving vascular control of the ureter may pose challenges.4 However, there is currently no literature on laparoscopic repair of ureteroinguinal hernia in children for comparative analysis in this regard.

In the present case, the standard open approach was chosen. Given the lack of information regarding the patient's etiology, a further postoperative investigation was planned, and a reduction of the ureter into the retroperitoneum along with hernia repair was performed. However, intraoperative identification of the herniated structure proved challenging. It is possible that a laparoscopic approach would have facilitated a clearer understanding of the anatomy and aided in the diagnostic process during the procedure.

Conclusion

Even though ureteral herniation in infants is rare and sporadic, it is important to consider the possibility of encountering the ureter during hernia repair, and precautions should be taken to prevent ureteral injury. Additionally, potential associated urinary tract anomalies should be thoroughly investigated and ruled out.

Ethical approval

Informed consent was obtained from the parents of the child.

Author contribution

The author confirms contribution to the paper as follows: study conception and design: GG, data collection and literature review: GG, draft manuscript preparation; GG. The author reviewed the results and approved the final version of the manuscript.

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Conflict of interest

The author declares that there is no conflict of interest.

REFERENCES

- Bay-Nielsen M, Kehlet H, Strand L, et al. Quality assessment of 26,304 herniorrhaphies in Denmark: a prospective nationwide study. Lancet 2001; 358: 1124-1128. https://doi.org/10.1016/S0140-6736(01)06251-1
- Hosoda T, Hijikata K, Ishioka S. Rare case of inguinal ureteral hernia in a child diagnosed by drip infusion pyelography-computed tomography. Int J Surg Case Rep 2022; 94: 107007. https://doi.org/10.1016/j. ijscr.2022.107007
- 3. Bosschieter J, van Roij K, Ahmadi N, Bokenkamp A, van der Horst HJR. Kidney blow out due to inguinal herniation of the ureter in an infant. Urol Int 2018; 100: 488-490. https://doi.org/10.1159/000449015
- Malde S, Bilagi P, Marsh H. Obstructive uropathy due to uretero-inguinal hernia: an uncommon occurrence. Indian J Urol 2013; 29: 355-356. https:// doi.org/10.4103/0970-1591.120126

- Cianci MC, Tocchioni F, Mantovani A, Ghionzoli M, Morini F. Unexpected pediatric uretero-inguinal hernia: case-report and literature review. Urology 2023; 176: 178-182. https://doi.org/10.1016/j. urology.2023.02.015
- Delgado-Miguel C, Muñoz-Serrano AJ, Aguado P, Fuentes E, Díez R. Ureteroinguinal herniation with consecutive ureteral stricture in a 2-month-old infant: case report. European J Pediatr Surg Rep 2024; 12: e16-e19. https://doi.org/10.1055/s-0044-1779253
- Wishahi M. Congenital primary obstructed megaureter presenting as inguinal hernia in an infant. Clin Case Rep 2022; 10: e05677. https://doi. org/10.1002/ccr3.5677
- 8. Cao K, Turner M, Mishrah P, Cuckow P. The rare case of an extraperitoneal ureteral herniation into the inguinal canal of a 12-year-old boy. Ann R Coll Surg Engl 2019; 101: e32-e34. https://doi.org/10.1308/rcsann.2018.0176
- Handu AT, Garge S, Peters NJ, Kanojia RP, Rao KL. Undiagnosed ureteroinguinal hernia with solitary kidney in a child with ureteric injury during herniotomy. J Pediatr Surg 2012; 47: 799-802. https:// doi.org/10.1016/j.jpedsurg.2012.01.006

- Sripathi S, Rajagopal K, Kakkar C, Polnaya A. Case report-inguinoscrotal ureteral hernia diagnosed on micturating cystourethrography. Indian J Radiol Imaging 2011; 21: 199-201. https://doi. org/10.4103/0971-3026.85368
- 11. Burgu B, Reşorlu B, Süer E, Soygür T. Scrotal hernia of the ureter in association with posterior urethral valves. J Pediatr Urol 2010; 6: 87-88. https://doi.org/10.1016/j.jpurol.2009.05.008
- 12. Powell MC, Kapila L. Bilateral megaureters presenting as an inguinal hernia. J Pediatr Surg 1985; 20: 175-176. https://doi.org/10.1016/s0022-3468(85)80296-7
- Morris LL, Caldicott WJ, Savage JP. Inguinal herniation of the ureter. Pediatr Radiol 1977; 6: 107-108. https://doi.org/10.1007/BF00973532
- Jewett HJ, Harris AP. Scrotal ureter: report of a case.
 J Urol 1953; 69: 184-187. https://doi.org/10.1016/ S0022-5347(17)68048-0
- 15. Pollack HM, Popky GL, Blumberg ML. Hernias of the ureter. An anatomic-roentgenographic study. Radiology 1975; 117: 275-281. https://doi.org/10.1148/117.2.275