

A Case Report

Obstructive nephropathy secondary to meatal stenosis: a rare cause in adults; a case report and literature review.

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Abstract-

Obstructive uropathy is a well-documented reversible cause of acute kidney injury; however, it can lead to chronic kidney disease if not identified and treated early. The common causes of urinary tract obstruction in adults are stones and malignancy. Congenital causes and meatal stenosis are rarely seen in adults. We report a 34-year-old male who complained of abdominal swelling with evaluation revealing evidence of renal impairment and an abdominopelvic CT scan showing bilateral hydronephrosis. Clinical examination revealed an adult circumcised phallus with a pinhole meatus. Urodynamic studies confirmed obstruction to urine outflow. He subsequently had meatoplasty done but renal impairment persisted due to long-standing obstruction.

Keywords: Obstructive uropathy, obstructive nephropathy, meatal stenosis, chronic kidney disease

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Introduction

Obstructive uropathy refers to the structural or functional changes in the urinary tract that impede normal urine flow. Obstructive nephropathy on the other hand, is defined as renal disease caused by the impaired flow of urine or tubular fluid. Obstructive nephropathy accounts for 7 – 12% of cases of chronic kidney disease.^(1–3) Malignancies and calculi in the urinary tract are the common causes identified in adults whereas congenital malformations and meatal stenosis are reported more commonly in children. Meatal stenosis is as an abnormal narrowing of the urethral opening (meatus). It is notably more common in circumcised males, occurring in 5 – 20% of them, and is exceedingly rare in uncircumcised children.⁽⁴⁾ Prompt identification of meatal stenosis in childhood with relief of obstruction and definitive treatment with meatotomy or meatoplasty is required to prevent long standing renal damage. We report a young male with chronic kidney from obstructive nephropathy secondary to meatal stenosis which was discovered in adulthood.

Case Description

A 34-year-old male presented to our nephrology outpatient clinic on referral from a private facility due to laboratory findings of elevated urea and creatinine and abdominopelvic computed tomographic (CT) scan findings of bilateral hydronephrosis. He had presented to the hospital of referral with a one-month history of progressive abdominal swelling. He also endorsed occasional abdominal pain, early satiety and constipation. He had no leg or facial swelling, shortness of breath, urinary symptoms, vomiting or diarrhea. He had no significant past medical history and was not on any medication.

Clinical examination including vital signs was normal except for an obviously asymmetrically distended abdomen with right flank, suprapubic and left iliac fossa fullness with a bimanually palpable right kidney.

His laboratory results were unremarkable except for a urea of 95mg/dl and creatinine of 3.6mg/dl. Abdominopelvic CT scan showed bilaterally enlarged kidneys (right > left) with multiple cystic masses of varying sizes communicating with each other in keeping with bilateral hydronephrosis (Figure 1). It also demonstrated thinning of the renal cortex with a dilated fluid-filled ureter up to the vesico-ureteric junction with thickened bladder walls. The liver was displaced anteriorly and the inferior vena cava was displaced medially. No calculus was seen.

An immediate urology consult was requested and during evaluation by the urology team, examination of his external genitalia revealed an adult circumcised phallus with a pinhole meatus. It was impossible to pass the smallest adult urethral catheter available in the urology clinic (size 14-French foley's catheter) through the urethral meatus. A diagnosis of obstructive nephropathy secondary to meatal stenosis was made. He had a uroflowmetry done which revealed a maximum flow rate of 7.9ml/s and minimum flow rate of 4.9ml/s. Urine analysis revealed a cloudy yellow urine with 2+ protein, 2+ blood, 2+ Leukocyte esterase with 10-15WBCs/hpf, 6-10 RBCs/hpf and no growth on culture.



Figure 1: Abdominopelvic CT scan of the patient (sagittal view) showing A) enlarged right kidney with hydronephrosis and B) enlarged left hydronephrosis.

He subsequently had meatoplasty with slow decompression of the urinary bladder during which about 7.6L of urine was drained over an 18 hour period following surgery. The drained urine was noticed to be turbid. Abdominal swelling thereafter subsided and he was discharged home 36 hours post-surgery to be followed up on an outpatient basis.

Repeat maximum flow rate on uroflowmetry one-month post-surgery was 14.7ml/s however serial follow-up abdominopelvic ultrasound scans showed persistent grade 4 hydronephrosis with significant cortical thinning. He also developed stage 1 hypertension as well as persistent proteinuria for which he was initiated on an angiotensin receptor blocker which was well tolerated with good blood pressure control. Serial renal function panels showed creatinine persistently between 3.3-3.5mg/dl as at 5 months post-surgery.

Discussion

Meatal stenosis is a rare cause of obstructive nephropathy presenting in adulthood but is more frequently diagnosed in children. It is notably more common in circumcised males and circumcision of male infants is widely practiced in a variety of populations. The reasons for this practice tend to be based on beliefs about its hygienic, cultural or spiritual benefits, rather than any documented medical advantages of the procedure. Circumcision is very prevalent among the Jewish and Islamic cultures⁽⁴⁻⁷⁾ and a study in Nigeria reported a circumcision rate of 87% among neonates.⁽⁸⁾ Our patient was a circumcised male.

A 5 year prospective study of children with renal disorders reported 17 cases of obstructive uropathy of which 47% was due to meatal stenosis following circumcision.⁽⁹⁾ Morris et al, in a systematic review and meta-analysis that evaluated the risk of meatal stenosis following circumcision however reported that the risk of meatal stenosis after circumcision was low although, there was weak evidence suggesting a non-statistically significant higher prevalence in circumcised males.⁽¹⁰⁾ Other less common documented causes of meatal stenosis are lichen sclerosus (balanitis xerotica obliterans), prolonged catheterization, previous hypospadias repair and rarely, it can be congenital.^(4,5,11-15)

Studies also suggest that meatal stenosis more commonly complicates circumcision done via conventional dissection surgery with ligation of the frenular artery compared to those done via the use of plastibell devices.^(16,17) While meatal stenosis is majorly a disease of males, probably because its causes are most commonly found in males, there are however anecdotal reports of meatal stenosis in females. Bueschen et al, reported the case of a 2-month old female with severe bilateral hydronephrosis from meatal stenosis.⁽¹⁸⁾ Meatal ischemia following damage to the frenular artery at circumcision in addition to traumatic meatitis of the unprotected post-circumcision urethral meatus are the two most cited mechanisms for the development of meatal stenosis.^(6,19,20) Meatitis, an inflammation of the meatus secondary to ammoniacal diaper irritation, has also been suggested as the underlying cause of secondary meatal stenosis. The lack of protection by the foreskin in the circumcised male has been said to correlate with an increased incidence of meatitis. This inflammation leads to a loss of the delicate epithelial lining in the ventral meatus, leaving a narrow orifice at the tip of the glans with consequent reduction in urinary flow which if not detected and corrected early will lead to obstruction.

Obstruction results in biochemical, immunologic, hemodynamic, and functional changes within the kidney. A cascade of events occur which lead to release of angiotensin II, cytokines, and growth factors including transforming growth factor- β and tumor necrosis factor- α . Some mediators are produced directly from the renal tubular and interstitial cells while others are generated by fibroblasts and macrophages. Progressive and permanent changes to the kidney occur via tubulointerstitial fibrosis, tubular atrophy and apoptosis as well as interstitial inflammation. In patients with unilateral obstruction or renal agenesis, enlargement of the contralateral kidney occurs, however, a reduction in compensatory growth occurs with age and an increase in the number of nephrons or glomeruli does not occur, despite enlargement. Urinary stasis can also lead to infection, renal scarring, calculi formation and sepsis. The degree and duration of obstruction, presence of renal cortical thinning, age of patient, and baseline renal function affect the likelihood of renal recovery. Two phases of recovery may occur: tubular function recovery and glomerular filtration rate (GFR) recovery. Full recovery of GFR can occur with relief of acute complete obstruction whereas longer periods of complete obstruction are associated with a reduced likelihood of recovery of GFR.^(21,22) Our patient likely had obstruction for over 30 years, and had significant cortical thinning and is unlikely to have significant recovery of his GFR.

Obstructive nephropathy can be asymptomatic; however, patients frequently experience obstructive and irritative urinary symptoms as well as symptoms suggestive of renal impairment. Obstructive symptoms include weak stream, hesitancy, intermittency, terminal dribbling, straining to void and feeling of incomplete voiding while irritative symptoms include frequency, urgency, nocturia, dysuria and urge incontinence. Patients may also present with recurrent

urinary tract infections. Hematuria can also occur especially in patients in whom the etiology of obstruction is a malignancy or calculi. The index patient appeared to have been completely asymptomatic despite objective evidence of poor urine stream. It is likely that because the meatal stenosis occurred very early in life, he grew up assuming his urine flow was “normal” because that was what he was used to.

Physical examination may be completely normal. Patients with upper urinary tract obstruction may have flank tenderness. Long-standing obstruction may result in an enlarged palpable kidney in children. Lower urinary tract obstruction causes a distended, palpable bladder. Hypertension frequently ensues from sodium and water retention whereas hypotension can occur due to polyuria and volume depletion especially following relief of the obstruction. A rectal examination and, in women, a pelvic examination should be performed because they may reveal prostatic enlargement or a local malignancy.⁽²³⁾ Meatal stenosis may be asymptomatic, or can present with any of the aforementioned symptoms as well as deflection of the urinary stream upward, difficult-to-aim urine stream, narrow and high velocity urine stream, penile pain at the initiation of micturition and prolonged urination. Examination findings consistent with meatal stenosis are a pinhole meatus, usually smaller than a 5-French feeding tube in an infant and an 8-French feeding tube in children aged 1 – 6years.^(6,19,20) Diagnosis is usually on visualization of the meatal opening and is based on a change in the elliptical shape of the meatal opening to a circular shape because of fibrosis or scarring, with visually apparent narrowing of the urethral meatus.⁽²⁴⁾ Our patient, however had none of these symptoms, he had symptoms that were likely due to the pressure effects of his large kidneys on the adjacent organs. Urinary tract obstruction must therefore be excluded in all patients with otherwise unexplained renal insufficiency.

Urinalysis may reveal haematuria, proteinuria and pyuria, all of which were present in the index present. Full blood count may reveal leucocytosis in patients with urinary tract infection or anaemia depending on the extent of renal involvement. Renal ultrasonography is the imaging modality of choice to exclude UTO due to its non-invasiveness and availability. A computed tomography scan is however indicated when calculi are suspected, ultrasonography results are equivocal, or when the cause of the obstruction cannot be identified on ultrasound.

When UTO from meatal stenosis is detected early, renal imaging findings may be normal,⁽²⁵⁾ however in a review of 87 circumcised males between the ages of 4 and 8, with severe meatal stenosis, ultrasonography showed hydronephrosis, hydroureter, and bladder wall thickening with both a full and empty bladder, and this latter finding was seen in 82% of the patients.⁽⁶⁾ In another study of 132 neonatally circumcised boys aged 5 to 10 years who had no urinary symptoms, 20.4% of them had severe meatal stenosis and ultrasonography revealed thickening of the bladder. Bilateral hydronephrosis (pyelocaliceal) was found in three cases necessitating the performance of a voiding cystourethrogram which demonstrated vesicoureteral reflux (VUR) in these patients.⁽²⁰⁾ Our patient had no urinary symptoms but had abdominal imaging findings in keeping with obstruction of the urinary tract which most likely began in childhood. Uroflowmetry is a diagnostic non-invasive objective test used to assess bladder and sphincter function and to confirm obstruction. A maximum flow rate (Qmax) less than 11ml/s is indicative of bladder outlet obstruction.⁽²⁶⁾ The reported patient had a Qmax of 7.9ml/s which improved to 14.7ml/s one-month post-surgery.

The management of obstructive nephropathy is hinged on prompt relief of obstruction, definitive management of the underlying cause and treatment of complications. Obstructive uropathy and nephropathy frequently coexist, and their management requires close collaboration between nephrologists and urologists.⁽²³⁾ Acute upper UTO is usually relieved by the insertion of a nephrostomy tube. Chronic upper UTO is relieved by the insertion of a ureteric stent or a pyeloplasty. Lower UTO is usually treated by insertion of a urethral catheter or a suprapubic catheter.⁽²³⁾ Patients with complications are to be managed as appropriate, for example, the nephropathy in our patient was managed conservatively, if there had been indications for renal replacement therapy, it should be offered to the patient. Meatotomy or meatoplasty is the definitive treatment for meatal stenosis. During meatotomy, the ventrum of the meatus is crushed and then divided with fine-tipped scissors, whereas in meatoplasty, in addition to this, stitches are placed to help keep the meatus open.⁽²⁷⁾ The patient had a meatoplasty done. Meatoplasty has been found to be associated with a lower rate of repeat surgery.⁽²⁸⁾

Several interventions to prevent meatal stenosis following circumcision of male infants have been studied. There have been reports that the application of lubricant jelly to the glans penis after circumcision was effective in reducing

the risk of meatal stenosis.^(5,29) Some other studies also documented a reduction in the occurrence of meatal stenosis when topical hydrocortisone was applied post circumcision.^(7,12)

Conclusion

This case highlights an uncommon and preventable cause of obstructive nephropathy in adults: meatal stenosis, which has been reported to complicate male circumcision, a common procedure in our environment. It also brings to fore the possibility of late presentation of a childhood cause of obstructive nephropathy and the need to examine the external genitalia in all young patients with abdominal and urinary complaints especially when they are males.

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