



## Primary bladder neck obstruction (PBNO) in children in a resource-limited setting

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### ABSTRACT

PBNO although rare when missed may have the debilitating effect of obstructive uropathy in children. The gold standard for its diagnosis requires videourodynamics and electromyography (EMG) studies which are not available in nearly all hospitals in developing countries. We present a case demonstrating the usefulness of readily available imaging in diagnosis of PBNO as well as demonstrate effectiveness of alpha blockade in children. MCUG and cystoscopy should suffice in cases of suspected PBNO in neurologically normal children to inform trial of therapy in resource limited settings.

### 1. Introduction

Primary bladder neck obstruction (PBNO) is a functional obstruction caused by delayed or incomplete opening of the bladder neck during the voiding phase of micturition impeding urine flow. This occurs in the absence of other causes of anatomic obstruction, neurological disease or increased striated sphincter activity. It is not uncommon but is rarely suspected in children as the cause of lower urinary tract symptoms (LUTs) unresponsive to empiric medical and behavioural treatments.<sup>1</sup> We present a case of a 1 and half year-old child who presented with persistent lower urinary tract symptoms whose investigations revealed features of PBNO and was successfully treated with alpha-1-blockers.

### 2. Case

A 1<sup>1</sup>/<sub>2</sub>-year-old male child, born at term out of an uneventful pregnancy presented to our unit with a 4-month history of difficulty initiating urination which was associated with swelling of the lower abdomen, crying and irritability. This was followed by dribbling of urine after which child would get short-lived relief. This would happen several times during the day and night. The child had presented at peripheral facilities where he had been treated with several courses antibiotics without improvement. Several days leading to the time of presentation

at our facility, the child required intermittent catheterization each time to pass urine.

Clinical evaluation in our unit was unremarkable including a normal neurological examination. Ultrasound of KUB showed a normal bladder outline without upper tract dilation. Urinalysis, urea, creatinine and electrolytes were normal. During an attempt at Micturating cystourethrogram the child was completely unable to pass urine despite a full bladder (Fig. 1). The child underwent urethra-cystoscopy which revealed a tenting and a narrowing of the bladder neck (Fig. 2) and no other anatomical defect in the urethra and bladder.

The child responded well to prazosin with immediate resolution of LUTs and bladder distention but developed intense painful prolonged erections. Owing to lack of availability of terazosin, the child was moved to 2.5 mg of alfuzosin daily whereupon resolution of LUTs was sustained without occurrence of erections.

In subsequent follow-ups the child did well clinically, had normal renal functions and a repeat KUB-Ultrasound was normal without post void residual (PVR).

### 3. Diagnosis

PBNO may present with a variety of symptoms including voiding symptoms, storage symptoms, and/or pelvic pain and discomfort. Some

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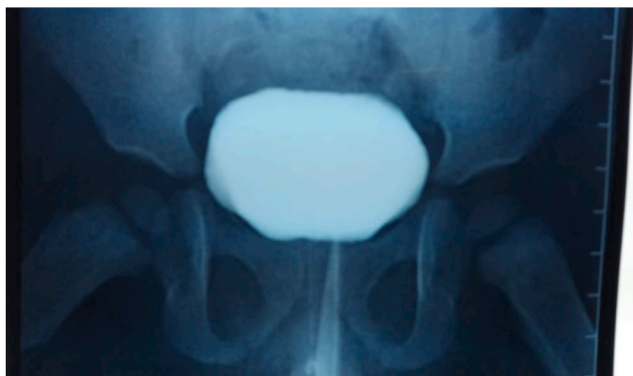
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**Fig. 1.** Attempt at Micturating cystourethrogram (MCUG) during which the child was unable to pass urine (We note that our facility has no capacity for fluoroscopy/VUDS).



**Fig. 2.** Urethroscopy showing tenting and narrowing of bladder neck.

patients present with acute retention following prior symptoms of lower urinary tract dysfunction as in the case of our patient.

PBNO is a videourodynamics (VUD) diagnosis. Pressure flow studies are utilized to diagnose obstruction and the addition of fluoroscopy allows for identification of obstruction specifically at the bladder neck, and, very often, incomplete bladder emptying is seen. Owing to a lack of fluoroscopy, the patient underwent MCUG during which the patient was completely unable to void with the MCUG showing failure of the bladder neck to open. At cystoscopy there were no posterior urethral valves but the bladder neck was noted to be narrowed with tenting rather than funneling and opening. We believe that a combination of these two should suffice in resource-strained settings.

#### 4. Treatment

The aim of treatment is to relieve bladder neck obstruction. Options for treatment include medical therapy with alpha-blockers, botulinum toxin injections and bladder neck surgery. Conservative management may be tried in those with mild symptoms and includes; watchful waiting with regular clinical evaluation and urodynamic studies so long as renal function is preserved, patient has a normal PVR and mild non-

bothersome symptoms.

Although an off-label use, medical treatment is hinged on  $\alpha$ -1-adrenoceptor blockade which primarily relax the smooth muscle of the bladder neck.<sup>1-3</sup> Although there are no randomized controlled trials on the use of alpha-adrenergic blockers, observational studies have shown significant improvement in IPSS scores, PVRs and maximal flow rates.<sup>2,4</sup> Owing to the rarity of PBNO diagnosis, there are few studies that have been conducted on the use of  $\alpha$ -blockers in children. In a study of 51 children treated with alpha-blockers following diagnosis of PBNO, 85% of the children reported significant symptom relief, a significant improvement measured on EMG lag time, and maximum and average flow rates; the medication was well tolerated.<sup>2</sup> The authors note that although improvement in uroflow parameters was seen after 4–6 weeks (when they took the first assessment after initiating therapy), a significant number of patients did not experience symptomatic improvement until 6 months.

Botulinum toxin injection has been shown to improve LUTs in PBNO but require multiple, probably annual injections for sustained effects and was reported to be acceptable to patients. Botulinum injection at the bladder neck in children with secondary bladder neck obstruction following PUV ablation did not show benefit.

Children may also benefit from bladder neck incision, although concerns remain due to the consequence of retrograde ejaculation for younger patients. In response, superficial bladder neck incision (SBNI) limited to a unilateral 2–3 mm has been utilized. Hennus et al. demonstrated preservation of antegrade ejaculation and concluded that SBNI in boys with severe infravesical obstruction can be done safely with preservation of antegrade ejaculation.<sup>5</sup> In their study, Hennus et al. followed up 40 males who had SBNI at a median age of 4.7 years and 19.6 years at follow-up. All had antegrade ejaculation, two (5.4%) had moderate incontinence and 8 (22%) had moderate lower urinary tract symptoms.

#### 5. Conclusion

PBNO is rarely diagnosed in children with voiding dysfunction but a high index of suspicion should prompt videourodynamics or fluoroscopic cystometry. In low resource settings, an MCUG may demonstrate failure of opening and funneling of the bladder neck and will rule out other causes. Cystoscopy where available is a good adjunct for evaluating other causes of bladder outlet obstruction. Alpha-1-adrenergic blockade maybe a good initial option for PBNO in children under two years.

#### Human and animal rights and informed consent

This article does not contain any studies with human or animal subjects performed by any of the authors. Informed consent was obtained from patient's parent.

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