

Case Report

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An Unusual Presentation of Purple Urinary Bag Syndrome: A Case Report

Juliano Alhaddad¹, Fares Kosseifi^{2,3}, Nehme Raad^{2,3}, Antoine Noujeim^{2,3}

¹Faculty of Medical Sciences, Lebanese University, Beirut, Lebanon

²Department of Urology, Faculty of Medical Sciences, Lebanese University, Beirut, Lebanon

³Department of Urology, Lebanese Hospital University Medical Center - Geitaoui, Beirut, Lebanon

Corresponding Author: Juliano Alhaddad, Beirut, Lebanon, julianohadd@gmail.com
Department of Urology, Lebanese Hospital University Medical Center - Geitaoui, Beirut, Lebanon
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Abstract

Background: The Purple Urinary Bag Syndrome (PUBS) is an uncommon and under-reported syndrome. It results from the purple discoloration of urine by tryptophan-oxidizing bacteria in a favorable alkaline milieu and usually affects patients with indwelling catheters. Other risk factors include female gender, chronic constipation, tryptophan-rich diet, and cognitive impairment. Although asymptomatic in the majority of cases, PUBS may be the sole and earliest sign of an aggressive urinary tract infection, especially in elderly patients where the presentation may be atypical and associated with resistant organisms, such as *Pseudomonas aeruginosa* and Vancomycin-resistant Enterococci (VRE). Proper treatment consists of catheter replacement and antibiotic therapy, which is indicated only in the setting of symptomatic infections.

Case Presentation: We report an unusual presentation of PUBS in a 79-year-old male patient with no classical risk factors. The patient had a supra-pubic catheter for urinary retention secondary to urethral strictures. Every month, by the time of catheter exchange, the patient developed an asymptomatic purple discoloration of the urinary bag, which was not investigated until he presented at our facility. Urine analysis revealed acidic urine. Appropriate antibiotic therapy was initiated after the development of urinary symptoms, and the catheter was replaced. No recurrent urine discoloration occurred.

Conclusion: This case represents the rare occurrence of PUBS in the setting of a suprapubic catheter in a male patient. It also highlights that even in atypical presentations, antibiotic therapy should be tailored to the clinical status of the patient and not the mere presence of bacteriuria. Raising awareness about this “esoteric” syndrome is essential for early diagnosis and proper management.

Keywords: purple urinary bag syndrome, PUBS, treatment, male, acidic urine, case report

Background

The Purple Urinary Bag Syndrome (PUBS) is a rare syndrome characterized by the purple discoloration of an indwelling urinary catheter and its collecting urinary bag. It was first reported by Dickson and Barlow in 1978, although the first description goes back to 1812, as King Georges III had purplish-bluish urine [1]. The major discrepancy in the reported incidence, which ranges between 9.8% and 42.1%, may be related to the absence of large studies and the limited number of patients enrolled in the few available ones [2,3].

PUBS is thought to be caused by an infection of the urinary tract with bacteria that can oxidize tryptophan metabolites [1]. It has been traditionally attributed to benign infections, but recent studies revealed an association of PUBS with resistant microorganisms such as Vancomycin-resistant Enterococci (VRE) and *Pseudomonas aeruginosa* [4,5]. Major risk factors for PUBS are represented by the ABCDEFGH rule: Alkaline urine, Bedridden-situation, Constipation, Dementia, End-stage renal disease, Female gender, Growth of bacteria (multiple urinary tract infections) and Hygiene [6,7].

The purple discoloration usually resolves by treating the infection along with catheter replacement (8). Male gender, acidic urine, and supra-pubic catheters are rarely associated with PUBS, and their constellation in the same patient with PUBS is even rarer (6,9).

We report the case of a 79-year-old man who lacked most of the predisposing factors but developed PUBS in acidic urine for 9 months, despite multiple catheter exchanges, which did not resolve until appropriate antibiotherapy was initiated.

Case Report

This is the case of a 79-year-old middle-eastern male patient presenting with refractory purple urine in his indwelling supra-pubic catheter. His past medical history is significant for coronary artery disease and idiopathic urethral strictures, diagnosed in 1998. The urethral strictures involved the anterior and posterior urethra, and the patient underwent an internal urethrotomy that was complicated by multiple recurrences requiring conventional cold knife and Holmium laser direct visual internal urethrotomies.

In December 2017, he underwent a coronary artery bypass graft that was complicated with

acute urinary retention in the post-operative course. Due to the inability to pass a Foley catheter, a suprapubic catheter relieved the retention, draining yellow colored urine with normal urinalysis. Since the patient was inoperable, the suprapubic catheter was left indwelling and changed monthly. Two weeks following the first suprapubic catheterization, the patient started noticing a purplish discoloration of his catheter and urinary bag. Seven to eight days following each catheter replacement, the purplish hue reappeared. The catheter was replaced eight times in various hospitals without the color change being addressed.

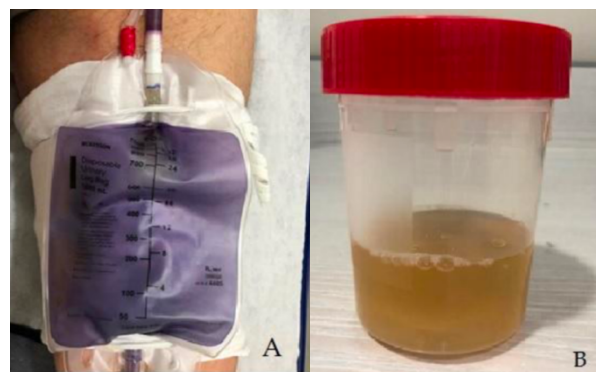


Figure 1: A) Figure showing the purple urinary bag and catheter, B) Figure showing the yellow and turbid urine that was in the bag emptied in a sampling cup

In August 2018, he presented to our emergency department to change the catheter, and the purple urinary bag was noticed (Fig. 1A). The patient denied any previous history of urinary tract infections (UTI), and previous urinary cultures were unavailable. Despite the purple discoloration of the surface of the urinary bag and catheter, once emptied in a sampling cup, the urine itself was of yellow turbid color and not purple (Fig. 1B). The patient denied any fever, chills, pain, or irritation from the suprapubic catheter. Furthermore, he did not have any constipation or high tryptophan diet. Urinalysis obtained from the bag revealed acidic urine (pH of 5.0) with numerous leucocytes and bacteria (Table 1). The urine culture from the suprapubic catheter grew a multi-sensitive *Escherichia coli* (100,000 Colony-forming unit/milliliter). The catheter was changed but no antibiotics were initiated because the patient was asymptomatic.

In September 2018, the patient started complaining from foul-smelling urine and persistent purple discoloration of the urinary bag, with ensuing social anxiety and public avoidance. Another set of urine analysis and culture revealed

Color	Purple	
Physical appearance	Turbid	
pH	5.0	4.8-7.4
Nitrites	Negative	
Proteins (mg/dL)	Negative	<10
Glucose (mg/dL)	Negative	<15
Ketones (mg/dL)	Negative	<5
Red Blood Cells (/HPF)	0-1	0-1
Leucocytes (/HPF)	Numerous	1-4
Casts	Negative	0
Bacteria	Numerous	0

Table 1: Urine analysis upon first presentation to our Emergency Department in August 2018 (dL: deciliter; HPF: High-Power Field; mg: milligram)

multi-sensitive *Pseudomonas aeruginosa*. Laboratory studies (including white blood count, C-Reactive protein, and creatinine) were unremarkable. Given the high rate of contaminated culture with *Pseudomonas*, and the absence of systemic signs of infection, we opted for outpatient treatment of an uncomplicated UTI with oral cefixime 400 milligrams daily for a total of 24 days.

The suprapubic catheter was replaced 5 days after the initiation of antibiotics. On his next monthly visit for catheter exchange, the urine bag was clear with no discoloration, and the patient reported that his urinary symptoms subsided a few days after his last catheter replacement (Fig. 2). The repeated urine culture was negative.



Figure 2: Color of the urinary bag after appropriate antibiotic therapy. After completion of the antibiotherapy, the urine retrieved its normal yellow color.

Discussion

Normal urine has a clear yellow color attributed to the presence of urochrome, a pigment derived from the degradation of hemoglobin [10]. Urine discoloration can be due to a benign process but may also indicate the presence of underlying pathology. The purple color in PUBS is thought to be due to the presence of dissolved indirubin (a red pigment) in plastic and indigo (a blue pigment) on its surface. Indirubin and indigo are generated via enzymatic degradation of indoxyl sulfate by bacteria colonizing the urinary tract. Indoxyl sulfate itself is derived from the hepatic metabolism of indol, which is produced by the intestinal flora using tryptophan as a substrate [11]. Algeria et al. report a case of PUBS following the initiation of tryptophan-containing nutritional supplements [12]. The mixture of indigo and indirubin in the plastic of the bag could explain the interesting finding in our patient, where the bag was colored in purple but the urine inside it was turbid yellow.

Besides a tryptophan-rich diet, risk factors include altered mental status, decreased motility, female gender, alkaline urine, and diabetes mellitus [1]. The main bacteria involved in PUBS are classical Gram-negative bacteria. Recently, cases due to Enterococci, Group B streptococci, and even VRE have been reported. VRE-induced PUBS can lead to a fatal urosepsis, as described by Bhattarai et al. [5]. This spectrum of severity can exist in the same patient; Lin et al. described a young paralyzed female patient who had PUBS on multiple occasions, ranging from asymptomatic infection to severe urosepsis requiring intensive care unit admission [4].

Most classical risk factors of PUBS are missing in our patient, a man with no cognitive impairment (mini-mental status examination score of 28), a regular diet, and no chronic constipation. Interestingly, his first urine analysis revealed acidic urine with a pH of 5.0, showing that PUBS is not limited to alkaline urine [9]. Whether the numerous urological interventions were a risk factor remains debatable. In one case series, the neurogenic bladder was the most common indication for catheterization associated with PUB, followed by posterior and anterior urethral stenosis. Urological intervention for the strictures was not associated with the recurrence of PUBS following treatment, but one-third of neurogenic bladder patients had a recurrence of the discoloration after antibiotic therapy [13]. Furthermore, the discoloration persisted before

the initiation of antibiotic therapy despite multiple catheter replacements. Therefore, PUBS eradication in this patient required both catheter replacement and antibiotherapy rather than solely exchanging the catheter.

However, the mere presence of PUBS without additional symptoms is not by itself an indication for urine culture or antibiotics treatment, as watchful waiting can lead to the resolution of the discoloration [11]. Antibiotics should be initiated in case of a symptomatic infection or ensuing social anxiety [14]. Even though our patients had PUBS on multiple occasions and a positive urine culture, an appropriate antibiotherapy wasn't initiated until the patient complained of bothersome foul-smelling urine that was a source of social anxiety to him. Moreover, although PUBS has been associated with urosepsis, in the absence of clinical and laboratory signs of a systemic infection, treatment as an uncomplicated UTI may be reasonable according to the microbiology prevalence, as was done in the case of our patient who received an oral cephalosporin.

Conclusion

PUBS is an underreported condition. Many young peers are unaware of this unique phenomenon which can be lifesaving if regarded as an early sign of a UTI, especially in atypical patients. Although most cases affect patients with an established risk factor, PUBS can occur in patients with no underlying risk factor and maybe the only alarm bell for the presence of an underlying infection. Nonetheless, antibiotics should only be initiated in the presence of symptoms and tailored to the patient clinical status accounting for the high rate of clinically insignificant catheter contamination.

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