

A Rare Case of Purple Urine Bag Syndrome Presenting with Delirium and Associated with Acidic Urine and UTI due to *Serratia marcescens*: But do we Still know Enough?

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ABSTRACT

Purple Urine Bag Syndrome (PUBS) is a rare occurrence characterised by a striking purple to blue/violet discolouration of the urine bag and its tubing after long-term indwelling urinary catheterisation while the urine inside remain clear. This phenomenon is a warning sign of Urinary Tract Infection (UTI). However, unlike traditional UTIs, such patients can be non verbal and asymptomatic with purple/blue urine bag being the only signal for UTI. Tryptophan-Indigo hypothesis is advanced to explain the mystery behind this phenomenon. A rare case of PUBS in acidic urine in an octogenarian (80 years old) female patient, with right sided hemiparesis, hypertension, Post Stroke Aphasia (PSA) and delirium is presented here. Moreover, her urine culture yielded an opportunistic bacteria *Serratia marcescens* which is not a typical organism responsible for this condition. The bluish colour of urine bag disappeared after hydration, change of the catheter along with urine bag and antibiotic therapy. To the best of the author's knowledge, this is the first case of PUBS reported from the state of Assam. It is hoped that this case report will be valuable in raising awareness and educating healthcare providers about this rather unknown esoteric syndrome in this region.

Keywords: Chronic constipation, Indigo, Indirubin, Tryptophan, Urinary catheterisation, Urinary tract infection

CASE REPORT

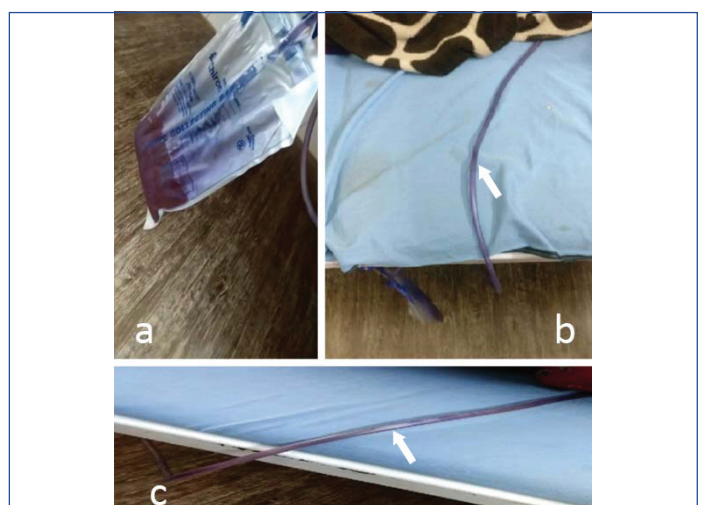
An 80-year-old euglycaemic hypertensive female patient was brought to Gynaecology Outpatient Department (OPD) in dehydrated and delirious state with right sided hemiparesis and constipation and had reddish to blue/violet discolouration of urine bag and tubing over past 2-3 days [Table/Fig-1]. Low grade fever existed during those few days together with decreased oral intake and this coincided with sudden progressive agitation with frequent moans and groans. Caregivers then also encountered a very stinking smell (similar to putrefied meat) while emptying urine bag. No food/drug history resulting into such discolouration or malodour was present. Patient was otherwise normal until recent ischaemic stroke involving left middle cerebral artery territory three weeks back and received treatment in another local hospital for two days. Taking discharge against medical advice, patient's caregivers continued amlodipine (5 mg), telmisartan (40 mg), atorvastatin (10 mg), clopidogrel (75 mg) at home with no change of her catheter/urobag prior to admission here.

Communication with patient was difficult due to delirious state and possible PSA. Examination revealed right sided hemiparesis, disorientation, hallucinatory behaviour with frequent inelible sounds. Patient was thin built, poorly nourished, afebrile, dehydrated, pale with blood pressure 138/80 mmHg. The presence of lower urinary tract symptoms could not be confirmed. While differently coloured urinary bag and tubing (reddish-to-blue/violet) was noted, the urine itself was not discoloured when collected for sampling [Table/Fig-1,2].

Urine analysis showed straw colour, turbid appearance, deposits (present), protein (2+), leucocyte esterase (2+), nitrite (positive) with specific gravity (1.020), pH (6.0), pus cells {20-25/High-Power Field (HPF)}, Red Blood Cells (RBC 2-4/HPF) and bacteria (plenty). Blood investigations revealed low haemoglobin (7.6 g/dL), White Blood Cell (WBC 9,600/mL) and elevated Erythrocyte Sedimentation Rate (ESR 65). Ultrasound abdomen revealed gall bladder microliths with gravels and cystitis.



[Table/Fig-1]: Urinary catheter drainage system with varying urine colour from dark brown to blue to violet at OPD (1a: 1 hour before admission) and shortly after admission (1b: 1 hour after admission).



[Table/Fig-2]: Dark brown discolouration of urine bag; a) To bluish discolouration of the urine bag tubing; b,c) (White arrows) Hours after admission.

Purple Urine Bag Syndrome (PUBS) were diagnosed and intravenous hydration was started along with empirical cefoperazone-sulbactam while awaiting urine culture report. Urinary catheter and drainage

bag were replaced, and bluish colour of urine bag and tubing got normalised by next day [Table/Fig-3]. The patient received risperidone 0.5/1 mg, lactulose, bisacodyl suppository (once), preprobiotics, multivitamins, blood transfusion along with physiotherapy. Whole blood was transfused because of her symptomatic anaemia limiting her effort in active physiotherapy due to tachypnoea and tachycardia and due to difficulty in availing Packet Red Blood Cell (PRBC). Delirium gradually resolved and the patient was later found to have PSA (mostly expressive). Informal assessment of PSA revealed that the patient understood short sentences and could respond appropriately in form of nodding, hand movements and facial expressions accompanied by some ineligious sounds. She had some difficulty in understanding long complex sentences. These deficits were expressed as "PSA mostly expressive".



[Table/Fig-3]: The colour of urine bag and tubing got normalised from second day of treatment following change of catheter and urine bag.

Urine culture grew significant amount of *Serratia marcescens* (>105 CFU/mL). Patient was discharged on request on day sixth with the addition of ciprofloxacin and gradual escalating dose of donepezil. There was no recurrence of PUBS in the last three months of the follow-up.

DISCUSSION

PUBS is a not well-known, visually stunning and baffling phenomenon characterised by intense purple-to-blue/violet discolouration of urine bag and its tubing while the urine inside remains clear. The process occurs over a period of hours/days usually with long-term indwelling urinary catheterisation and is seen majorly in elderly, institutionalised, poorly ambulant (bed or chair-bound) and constipated patients with other co-morbidities [1-3]. This condition also produces a strong peculiar-smelling malodour [1,2]. Thus, PUBS often creates panic situation [1,4] and it is essential to reassure and educate patients and caregivers [5]. Studies report a wide range of prevalence of PUBS between 8.3% to 42.1% in patients with long-term indwelling catheter [2,5,6]. Very few cases of PUBS have been reported in literature from the Indian subcontinent [7-9].

Several reports of this condition first surfaced in 1978 [3]. Dietary tryptophan is deaminated by intestinal microbiomal tryptophanase into indole [10]. Indole is conjugated in liver into indoxyl sulphate (indican) which is then excreted into urine (imparting a dark brown colour). Bacterial sulphatase and phosphatase enzymes convert indican to indoxyl which is oxidized in alkaline urine producing indigo (blue) and indirubin (red) pigments [11]. These two insoluble pigments may then react with plastic of catheter bag and produce a purple hue [4]. However, PUBS can develop without indicanuria [2,10] and the causative pigment can be a steroidal/bile acid conjugate [2,12].

Clubbed under "ABCDEFGH", major risk factors for PUBS comprise: A) alkaline urine pH; B) bedridden situation; C) constipation; D) dementia; E) end-stage renal disease; F) female gender; G) growth of bacteria; H) hygiene (long-term catheterisation) [11].

Other determinants include old age, immunocompromised status, dehydration, uraemia, high tryptophan diet and use of Polyvinyl Chloride (PVC) catheter and/or bags [2,4,11,13]. PUBS mostly occur in alkaline urine environment which favours catalysis of indoxyl [1,2,11]. But, alkaline urine is not a necessity as few reports of PUBS have surfaced with acidic urine [3,10,12,13] and same is also found in the present case. *Serratia marcescens* in a laboratory study model was found to raise the urine pH upto maximum of 6.4 [14]. Bacterial overgrowth, altered microbiota and increased time for bacterial deamination of tryptophan in chronic constipation/intestinal obstruction facilitate development of PUBS [2,4,11]. PUBS are rarely reported with diarrhoea [3].

Higher prevalence of PUBS in females is due to their predisposing anatomy for UTI [2-5,12]. Long-term catheterisation with poor hygiene and sanitation, predisposes to both UTI and PUBS [2,11]. However, there are reports of PUBS even without any urinary catheters [1,3]. High urinary bacterial load facilitates development of PUBS due to greater availability of sulphatases/phosphatases [12]. Yet, there can be lack of required enzymes even in the same bacterial species or insufficient quantity of pigments produced. Therefore, although UTIs are common in clinical practice, the occurrence of PUBS is rare as it depends on concurrent presence of multiple factors.

Certain bacteria (singly or mixed) namely *Proteus mirabilis*, *Klebsiella pneumoniae*, *Providencia stuartii* and *rettgeri*, *Escherichia coli*, *Enterococcus spp.*, *Pseudomonas aeruginosa*, *Morganella morganii* are commonly associated with PUBS [2,4,11,15]. The present case involved *Serratia marcescens* which lack sulphatase/phosphatase activity. It is mentioned in three articles of PUBS [5,11,16]. Amazingly, this gram-negative bacillus (often referred as "miracle" bacillus) synthesises bright-red pigment (Prodigiosin) which can cause red urine, red diaper syndrome and pseudohaemoptysis. Growing under extreme conditions (including antiseptics, disinfectants, soap/shampoo residue), this known opportunistic pathogen spread via contaminated medical instruments/devices, disinfectants, etc., and cause nosocomial infections of the respiratory and urinary tracts, bloodstream, etc., [17]. Thus, possible source of *Serratia* in the present case can be linked to previous catheterisation procedure at another hospital or unhygienic bathroom. Isolation of a peculiar combination of multidrug resistant bacteria *Corynebacterium urealyticum* and *Enterococcus faecium* in one recent case of PUBS calls for future research into understanding "Gut-UTIs axis" and role of gut and urinary dysbiosis in causing UTIs/PUBS [15].

Delirium is an acute illness associated with longer hospitalisation and increased mortality. Like PUBS, UTI is a common cause for delirium in elderly women and treatment of UTI also causes delirium to disappear as observed in the present case [18]. However, delirium is rarely reported with PUBS. Although PUBS is considered as benign [3,6,11], grave complications like Fournier's gangrene [6], severe sepsis [6], multidrug-resistant vulvar abscess, etc., have occurred.

Management of PUBS is chiefly targeted at treating the UTI and constipation, if present. Adequate sanitation and change of catheter help in resolving the PUBS and also prevents future occurrence. Antibiotic treatment for the underlying urinary infection is required to control and treat PUBS [19]. Physicians and healthcare staffs must be aware of this condition and should focus on the need for adequate urological sanitation and timely change of catheter. Even the patient caretakers should be advised for the same, as sudden change in colour of urine or urine bag may be alarming. PUBS along with UTI have greater mortality and morbidity than UTI alone [20]. Hence, early detection along with aggressive antibiotics regime can minimise the fatality.

The present case is a very rare case of PUBS associated with acidic urine and UTI due to *Serratia marcescens* while presenting with delirium. PUBS disappeared after hydration, change of urine catheter and bag and antibiotics in conformity with other studies

[3,12]. The author feels that management of any PUBS should be individualised on a case-by-case basis [6,11]. To the best of the author's knowledge, this is the first case of PUBS reported from the state of Assam.

CONCLUSION(S)

Proper urologic sanitation, reducing duration of catheterisation, timely catheter with urine bag replacement aseptically, treatment of UTI and constipation if present, is fundamental to manage and prevent PUBS. PUBS may prove to be fatal when left untreated in patients with significant co-morbidities. Extensive research is warranted to unravel mysteries and controversies underlying this enigmatic phenomenon.

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