Case Report

Purple urine bag syndrome (PUBS)—an enigma!

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Abstract

Purple urine bag syndrome (PUBS) is an intriguing phenomenon featured by an alarming purple discoloration of the urine and the urine bag because of the mixture of indigo (blue) and indirubin (red). It is a secondary phenomenon to bacterial urinary tract infection (UTI) with sulfatase/phosphatase producing bacteria. This situation is thought to arise from a series of biochemical steps involving the metabolism of tryptophan in the gut to indigo and indirubin pigment in the urine. While there are multiple factors associated with PUBS, it occurs particularly in chronically debilitated, constipated, catheterized, elderly female subjects in the presence of alkaline urine. We report a case of 78-year-old man of multiple myeloma with acute kidney injury and other comorbid conditions, who was admitted for gastrointestinal bleed and developed *Escherichia coli*-related UTI, followed by the development of PUBS. He responded to the treatment with antibiotics and change of catheter and urine bag. PUBS is harmless without any serious consequences but an uncomfortable or troubling issue for patients and their families. Most patients who present with PUBS are largely asymptomatic. Treating the underlying UTI, reassurance, change of the urinary catheter, and urinary sanitation are fundamental to its treatment.

KEY WORDS: Urinary catheter, urinary tract infection, tryptophan metabolism, indigo, indirubin pigment

Introduction

Purple urine bag syndrome (PUBS) was first reported in literature by Barlow and Dickson in 1978.[1] This condition is characterized by purple discoloration of urine in the urine bag because of the presence of the pigments, indigo and indirubin. It is usually seen in chronically catheterized, constipated, bedridden elderly women. The risk factors associated with PUBS include female gender, increased urine alkalinity, high tryptophan content in the diet, chronic indwelling urinary catheter, urinary tract infection (UTI) and chronic renal failure.[2,3] This occurs by a series of biochemical reactions involving tryptophan when urine infected with certain bacterial strains interacts with catheter tubing or urine bag.[2] Although UTI is commonly encountered in everyday practice, the presence of a purple bag is an uncommon clinical entity. PUBS is considered to be benign and disappears after the treatment of UTI. However, it can be a cause of great anxiety to the patients and their relatives and an enigma to the nonskilled treating physician.

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Case Report

A 78-year-male patient with prior history of long-standing diabetes mellitus with diabetic nephropathy, chronic obstructive airway disease, and ischemic heart disease was admitted for passing black-colored stools, breathlessness, and chest pain of anginal nature of 15 days duration. He experienced seven to eight episodes of watery, black stools in a day. He was not on nonsteroidal anti-inflammatory drugs and antiplatelet agents in the recent past. He was also operated for plasmacytoma of abdominal wall 20 years back. On admission, he was very pale, lean, and dehydrated and showed pulse rate of 116/min, regular, and blood pressure of 110/60 mm Hg supine, right arm. There was no lymphadenopathy, clubbing, cyanosis, or icterus. Examination of all the systems was normal, except for mild splenomegaly in the abdomen. His hemoglobin was 4.3 g/dL, MCV 76 fL, and

RDW 21.6%. Peripheral smear showed hypochromic RBCs with polychromasia and poikilocytosis. Total leukocyte count was 3,500/mm^3 with 68% neutrophils and platelet count 90,000/mm^3. Biochemical parameters revealed normal serum bilirubin with slightly elevated ALT and AST (ALT, 53.7U/L; AST, 128.9 U/L; and ALP, 112 KA/L). Renal function tests showed blood urea 168.4 mg/dL and serum creatinine 4.2 mg/dL. He was catheterized on day 1 as he revealed difficulty in passing urine with urinary retention. His urine routine deposit and culture sent on day 1 was normal. USG abdomen showed grade III renal parenchymal disease with bilateral pleural effusion and mild ascites. His serum iron, folate, and vitamin B12 were within normal limits. Serum ferritin was 29 mg/dL. His serum protein electrophoresis showed increased gamma globulin fraction of monoclonal nature. He was treated with proton pump inhibitors, intravenous fluids, and ceftriaxone (1 g bd, intravenously). He was transfused with three units of compatible packed red cells. Ryle's tube aspiration showed coffee ground aspirate. He refused to undergo upper gastrointestinal endoscopy. On seventh day of admission, he developed high-grade fever (103°F) with rigors. His urine examination done on the eighth day revealed pH of 8.5 with protein 20 mg/dL, blood 1+, 12–15 WBCs/high power field (hpf), and 8–10 RBCs/hpf. Culture of the same sample was, however, sterile. He was started on intravenous piperacillin–tazobactum (2.25 g tid). On the same day, he noticed purplish discoloration of urine and urine bag, which later became dark purple colored although he had not taken any medication or substance known to cause purple discoloration of urine [Figure 1]. Spectroscopic examination of urine for methemoglobin was negative. His Foley's catheter and urine bag were changed on the tenth day, and the urine color was found to be normal [Figure 2]. His urinary infection responded to piperacillin–tazobactum, which was continued for 14 days. His melena subsided with proton pump inhibitors. He refused endoscopy and bone marrow examination and was discharged against medical advice on the 15th day. There was no recurrence till discharge. On the basis of the patient's history and the microbiology results, a diagnosis of PUBS was made.

Figure 1: Purple urine is noted in Foley's catheter and bag.

Figure 2: Normal urine color after the change of Foley's catheter and bag.

Discussion

Although first reported in literature in 1978, historically, PUBS dates back to 1812 when physicians treating King George III observed bluish discoloration in his urine[4] and urinary catheter and bag. It is a benign and rare clinical phenomenon, commonly observed in bedridden, constipated, elderly chronically catheterized female subjects. Here, the urine and the urine-collecting bag and the tubing turn purple in color within hours to days after catheterization.[5,6] Literature shows a prevalence rate of PUBS from 8% to 16% in various studies.[5] It is known to occur with alkaline urine.[6] Although referred to as syndrome, it is actually an uncommon phenomenon of urine discoloration mostly due to urinary tract infection (UTI).

Most authors believe that purple urine is a mixture of indigo and indirubin, which are derived from the metabolites of tryptophan, an essential amino acid in the body.[5–7] According to the most commonly accepted hypothesis, owing to altered gut motility or intestinal bacterial overgrowth associated with chronic constipation, tryptophan is exposed to intestinal flora before it can be absorbed. It undergoes deamination in the gastrointestinal tract to indole by intestinal bacteria, which is then absorbed into the portal circulation via the intestinal wall. The liver conjugates indole to indoxyl sulfate (or indican), which is excreted in the urine. Most of the excreted indoxyl sulfate is digested into indoxyl by indoxyl sulfatase produced by certain bacteria. Levels of the urinary excretion of indoxyl sulfate are generally increased above the normal range. In an alkaline environment, indoxyl turns into indigo (blue color) and indirubin (red color) pigments, which combine with catheter tubing to give the characteristic purple appearance. These series of steps are outlined in Figure 3. However, few case reports have recorded patients revealing purple urine bag but no indicanuria and the characteristic violet pigment can result owing to either a steroidal or bile acid conjugate.[6]

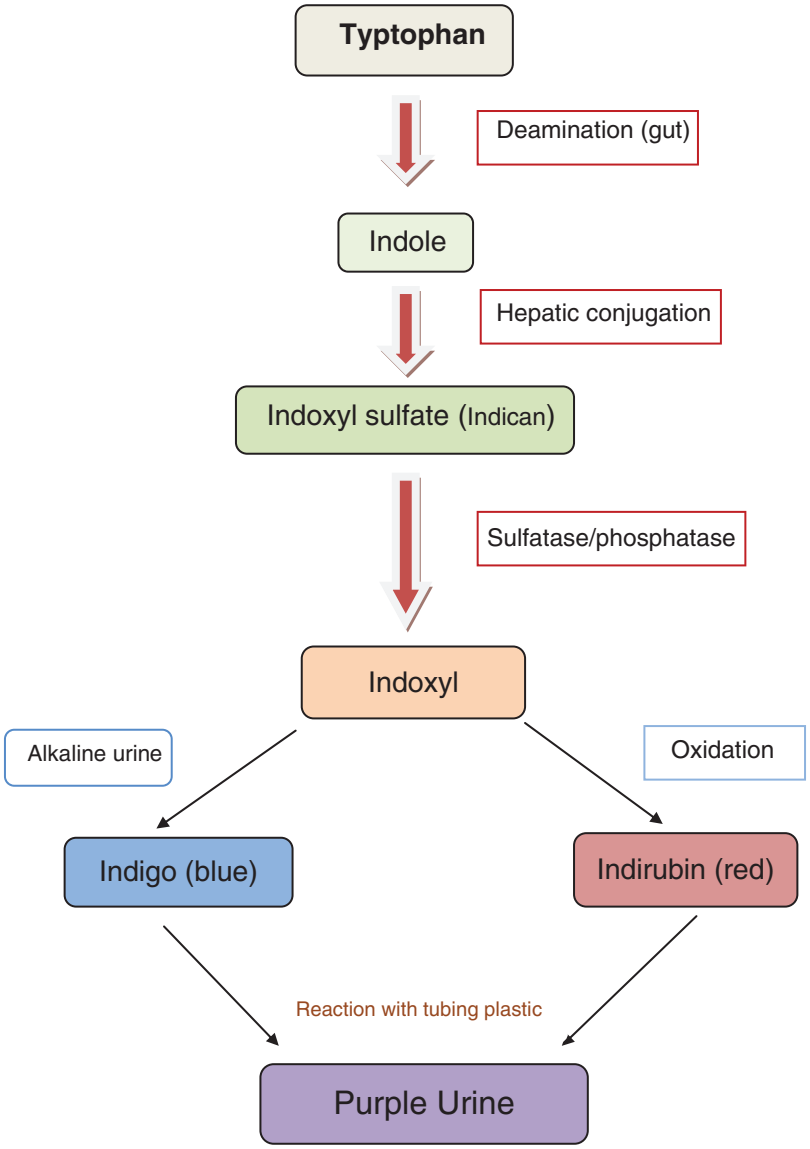

Figure 3: Proposed biochemical pathway of conversion of tryptophan to indigo and indirubin.

Catheter-associated UTIs are commonly caused by Gram-negative bacteria. Bacteria implicated in the causation of PUBS include *Providencia stuartii*, *Providencia rettgeri*, *Klebsiella pneumoniae*, *Proteus* species, *Escherichia coli*, *Enterococcus* species, *Morganella morganii*, and *Pseudomonas aeruginosa*.[7–9] These Gram-negative bacteria produce enzymes sulfatase and phosphatase, which increase the conversion of indoxyl sulfate into indoxyl and are, thus, important in the genesis of PUBS. Similarly, chronic constipation is generally related to bacterial overgrowth in the colon, which elevates the transformation of tryptophan in the gut into indole and is, then, absorbed into the portal circulation. Hence, the association of PUBS in chronically catheterized and constipated patients is common. In addition, presence of high tryptophan in the diet is required for the formation of the essential pigments.

Urinary discoloration is commonly seen in clinical practice and is owing to several reasons. There was no history of consumption of methylene blue in our patient, which is known to cause bluish discoloration to urine. Our patient was unique in the sense that he was a case of multiple myeloma and showed frequent, watery foul-smelling stools with melena rather than constipation. Presence of altered blood in the gut favors abnormal bacterial overgrowth. In addition, he developed acute kidney injury and dehydration on admission. His urine culture did not show growth of organisms probably because

he was on antibiotic before the culture was sent. Notably, when his urine was collected in the sterile container, it appeared normal and not purple. In the literature, this syndrome is typically described in female patients; however, our patient was a male.

According to Pillai et al.,[10] simultaneous presence of multiple risk factors increases the likelihood of development of PUBS. It is also reported in patients with chronic renal failure who are hemodialysis dependent,[11] those with nephrostomy tubes, in a patient with an ileal conduit and urinary diversion,[12] in patients with polyvinylchloride (PVC)-containing urine bags when compared with non-PVC urine bags,[3] and in cases of intussusception.[13] In 2008, Chung et al.[14] reported a case of PUBS with acidic urine.

Most patients who present with PUBS are asymptomatic. Although it has favorable prognosis, the phenomenon itself can be a cause of anxiety among patients and their families because of the sudden inexplicable discoloration of the urine and the urine bag. It generally disappears after treatment of underlying UTI by antibiotics and no special investigation is required. Thus, for asymptomatic patients, treatment should be aimed at the underlying medical problem rather than purple bag itself.[15] This syndrome signals underlying recurrent UTIs, caused by improper care of the urinary catheters and improper sanitation. Good urologic sanitation by proper care and recurrent alteration of urinary catheters may aid in avoiding this rare entity. Although the condition itself is benign, there have been case reports in the literature of PUBS being associated with higher mortality and morbidity because of the underlying UTI.[10]

This uncommon clinical condition can lead to misdiagnosis by untrained medical personnel and unnecessary investigations. It is, therefore, essential for a physician to be aware of this syndrome, recognize the risks of an underlying UTI, and initiate treatment for UTI as soon as possible, as the outcome may be fatal if it progresses to generalized septicemia. This is particularly so as patients reported to develop this condition are often elderly, unwell, and show significant comorbid conditions.

Conclusion

PUBS is an infrequent manifestation of UTI mostly seen in chronically catheterized, constipated elderly women. Its clinical course is usually benign and harmless without any serious consequences. Improvement in the care of urinary catheters prevents both PUBS and catheter-associated UTI.

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