

CASE REPORT

Purple urine bag syndrome in urinary tract infection. Case report from Bolivia

Síndrome de la bolsa de orina púrpura en la infección del tracto urinario. Reporte de caso en Bolivia

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ABSTRACT

Purple bag syndrome is a rare manifestation of urine discoloration associated primarily with chemical reactions caused by the indoxyl sulfatase/phosphatase activity of some bacteria or as secondary metabolites of food or supplement intake, whether oral or parenteral. Its presence causes confusion, leading to unnecessary laboratory tests and treatments. Is reported the clinical case of a 49-year-old oncology patient admitted to the trauma service for a pathological fracture and urinary tract infection. She was fitted with a urinary catheter, which contained purple urine. Laboratory tests were requested to identify the causative bacteria. The catheter was replaced, and antibiotic treatment was started, leading to resolution of the clinical picture.

Keywords: Urinary Infections; Amino Acids; Sulfur; Hydrogen-Ion Concentration.

RESUMEN

El síndrome de la bolsa de orina púrpura, es una manifestación rara en la coloración de la orina asociada principalmente a reacciones químicas por la actividad indoxil sulfatasa/fosfatasa de algunas bacterias o como metabolitos secundarios a la ingesta de alimentos o suplementos por vía oral o parenteral. Su presencia causa desconcierto llevando a realizar pruebas laboratoriales y tratamientos innecesarios. Se reporta el caso clínico de paciente femenina oncológica de 49 años de edad internada en el servicio de traumatología por fractura patológica e infección urinaria, portadora de sonda vesical, la misma que contiene orina de coloración púrpura, se solicitan laboratorios para identificación de la bacteria causal, se realiza recambio de sonda e inicia con tratamiento antibiótico remitiendo el cuadro clínico.

Palabras clave: Infecciones urinarias; Aminoácidos Sulfúricos; Concentración de Iones de Hidrógeno.

INTRODUCTION

Purple Urine Bag Syndrome (PUBS) is a rare but visually striking medical condition characterized by the violet or purple coloration of urine contained in the bag and tubes of the urinary catheter.^(1,2) Although formally described by Barlow and Dickson in 1978, a historical observation has been reported in the case of King George III in 1812, suggesting the presence of the phenomenon before its identification as a syndrome.^(1,3) Despite its alarming appearance, PUBS is considered a benign condition with a good prognosis when properly managed.^(4,5) However, its striking presentation can cause alarm and anxiety in healthcare professionals unfamiliar with the condition, as well as in patients and their families.⁽⁶⁾ A prevalence of 8,3 % to 16,7 % has been reported in patients with long-term urinary catheterization. The overall incidence in patients with long-term indwelling catheters can vary widely, but is in the range of 8 % to 9 %.⁽⁷⁾ The pathogenesis of PUBS is based on complex biochemical reactions that convert ingested tryptophan into indigo (blue) and indirubin (red) pigments in the urinary tract through bacterial activity.^(8,9) Several risk factors are associated with the development of PUBS, including advanced age (particularly common in women),^(4,10) long-term indwelling urinary catheterization,⁽¹¹⁾ urinary tract infections (UTIs),⁽¹²⁾ prolonged immobility or bed rest,^(10,13) alkaline urine, asymptomatic bacteriuria, diets rich in tryptophan, cognitive impairment, constipation, which promotes bacterial proliferation and tryptophan degradation; and chronic or acute kidney disease that compromises the elimination of indoxyl sulfate.⁽¹⁴⁾ Although PUBS is primarily a visual manifestation, it is an important clinical indicator of an underlying urinary tract infection. If not diagnosed and treated promptly, this condition can progress to serious complications, such as urosepsis, especially in vulnerable populations with multiple comorbidities; early diagnosis and prompt treatment are crucial to prevent adverse outcomes.⁽¹⁵⁾

Due to its multifactorial nature and rare manifestation, this report aims to describe the conditions in which this syndrome manifested itself, thereby contributing to the knowledge of its occurrence in clinical management.

CLINICAL CASE

A 49-year-old female cancer patient diagnosed with a pathological fracture of the L4-L5 lumbar vertebrae and a subtrochanteric fracture of the left hip due to a fall during hospitalization referred to the traumatology service, leading to partial immobility in the supine position with continuous traction. Osteoporosis was found during hospitalization, and cancer was under investigation based on tumor markers: Carcinoembryonic antigen (CEA) 195,83 ng/ml; acute renal failure with glomerular filtration rate (GFR) 24 ml/min/1,73 m² and urinary tract infection, treated with pregabalin, tramadol, tapentadol, nalbuphine, duloxetine, erythropoietin, olive oil, N-acetylcysteine, sodium heparin and low molecular weight heparin, ondansetron, lactulose, iron saccharate, slow K (potassium chloride), and Foley catheter with spontaneous diuresis.

Ten days after insertion of the Foley catheter, purple-colored urine was observed in the bladder bag (figure 1a).

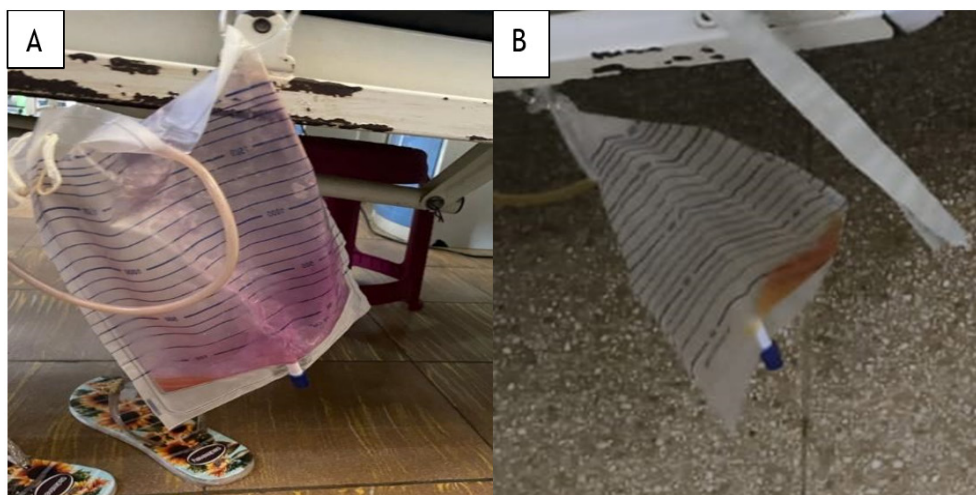


Figure 1. A: Foley catheter bag, 10 days after placement, with purple urine. **B:** Foley catheter bag, 48 hours after starting antibiotic therapy

A leukogram was taken, showing leukocytosis of 12 500/mm³ with a predominance of neutrophils of 8700/mm³. Urinalysis (purple-colored urine, alkaline reaction pH = 8,0; positive proteinuria, negative glucosuria, negative ketones, positive nitrites, microscopic examination with 2 to 4 red blood cells per field, 4 to 6 leukocytes per field, regular bacteria, abundant mucous filaments, with abundant amorphous and triple

phosphate crystals), microbiology and antibiogram requested (Table 1). Laboratory tests were performed, including hemoglobin (Hb), erythrocyte sedimentation rate, cyturia, vaginal exudate, and blood chemistry, which yielded normal results.

Table 1. Laboratory Tests			
Diagnostic Test	Result	Reference Value	Interpretation
HEMATOLOGY			
Leukocytes	12 500/mm ³	5,000-10 000/mm ³	Leukocytosis
Neutrophils	8 700/mm ³	1 800-7 200/mm ³	Neutrophilia
Hemoglobin	Normal	-	No abnormalities
Erythrocyte sedimentation rate	Normal	-	No abnormalities
URINALYSIS			
Appearance	Purple	Light yellow	Altered
pH	8,0	4,6-8,0	Alkaline
Proteins	Positive	Negative	Proteinuria
Glucose	Negative	Negative	Normal
Ketones	Negative	Negative	Normal
Nitrites	Positive	Negative	Bacteriuria
Red blood cells	2-4 per field	0-2 per field	Slightly elevated
White blood cells	4-6 per field	0-5 per field	Within normal range
Bacteria	Moderate amount	Scarce	Increased
Mucous filaments	Abundant	Scarce	Increased
Amorphous phosphate crystals	Abundant	Scarce	Increased
Triple crystals	Abundant	Scarce	Increased
TUMOR MARKERS			
CEA (Carcinoembryonic antigen)	195,83 ng/mL	<3,0 ng/ml	Very high
KIDNEY FUNCTION			
GFR (Glomerular Filtration Rate)	24 ml/min/1,73m ²	>90 ml/min/1,73m ²	Severe renal failure
OTHER TESTS			
Pyuria	Normal	-	No abnormalities
Vaginal discharge	Normal	-	No abnormalities
Blood chemistry	Normal	-	No abnormalities

Chest X-ray without alterations. Abdominal ultrasound showing: liver, gallbladder, pancreas, aorta, and spleen without alterations. Kidneys of normal size, echogenicity, and position, appearing to have an uncomplicated double excretory system (no pyelocaliceal dilatation), as well as bilateral double ureter (both upper thirds visible). Bladder with good capacity, thin walls, no focal lesions, intravesical dilation of both ureters confirmed, more pronounced on the left, which increases with Valsalva maneuver due to vesicoureteral reflux extending to the distal segments, and is also accompanied by slight dilation of the excretory system. Uterus of normal size, homogeneous texture, with IUD in endometrial cavity. Adnexa without alterations. No ascites.

Given the results obtained and the patient's risk factors, it was decided to start antibiotic therapy with Imipenem 1 gram every 12 hours for 5 days. Based on the results of the antibiogram and urine culture, abundant Gram-negative bacilli with extended-spectrum beta-lactamases (ESBL) positive were found, identified as "*Klebsiella pneumoniae*" sensitive to Imipenem, with high resistance to semisynthetic penicillin's, sulfa drugs, cephalosporins, and quinolones.

Two days after starting antibiotic therapy, there was spontaneous diuresis of normal appearance, with the discoloration of the urine subsiding (figure 1b). As the patient is an oncology patient, she is receiving palliative care and treatment for her various pathological conditions. After reviewing the probable causes of the PUBS manifestation, it was found that the drugs administered to the patient do not correspond to the evidence of causes of purple urine coloration reported in other PUBS reports.

DISCUSSION

Purple Urine Bladder Syndrome (PUBS) is a rare and visually striking condition that can cause alarm in both patients and healthcare personnel if not properly recognized. Despite its appearance, it is generally a benign condition. The characteristic purple coloration of PUBS is the result of a specific biochemical cascade involving tryptophan metabolism.⁽¹⁷⁾ Among the coincidences with reported cases of this manifestation, urinary tract infection and bladder catheterization stand out. It is likely that intestinal bacteria metabolized the tryptophan consumed in the diet to produce indole.⁽¹⁸⁾ Indole, when absorbed and transported to the liver through the

portal circulation, could be conjugated into indoxyl sulfate, which is a colorless substance excreted in the urine.⁽¹⁹⁾ However, the presence of bacteria that produce enzymes such as sulfatases and phosphatases in the urinary tract converts indoxyl sulfate to indoxyl. Indoxyl is then oxidized in an alkaline urinary environment, transforming into two pigments: indigo (blue) and indirubin (red).^(4,12,16) The mixture of these two pigments produces the purple coloration that adheres to and stains the plastic material (usually polyvinyl chloride or PVC) of the collection bag and urinary catheter present in the clinical case.^(20,21)

The urinary pH of 8,0 reported in the present case is consistent with the alkaline environment necessary for pigment oxidation.⁽¹³⁾ Urine culture isolated *Klebsiella pneumoniae*, a microorganism known to produce sulfatase and phosphatase enzymes, which are related to PUBS.^(2,16) Other pathogens associated with PUBS include *Escherichia coli*, *Proteus mirabilis*, *Providencia* spp., *Morganella morganii*, *Enterococcus* spp., *Citrobacter* spp., and *Pseudomonas aeruginosa*.⁽²²⁾

The risk factors most frequently associated with PUBS include advanced age (more common in the geriatric population, although our 49-year-old patient shows that it can occur in younger adults with multiple risk factors), female sex (higher incidence in women, possibly due to a shorter urethra that facilitates the ascent of germs from the intestinal flora, a factor present in our patient),⁽³⁾ chronic or prolonged urinary catheterization (a major factor, especially in long-term catheters; our patient had a Foley catheter for 10 days, which is sufficient time to develop bacteriuria, as PUBS can develop within hours or a few days after catheter placement), immobility or bed rest (bedridden patients are more susceptible; our patient was partially immobile due to fractures and continuous traction), chronic constipation (promotes bacterial overgrowth and increases the degradation of dietary tryptophan in the intestine; the patient reported constipation and was receiving treatment with lactulose), urinary tract infections or bacteriuria (PUBS is a manifestation of UTI or bacterial colonization of the urinary tract; our patient was diagnosed with UTI evidenced by positive nitrites, leukocytes, and bacteria in the urinalysis), chronic kidney disease or renal failure (potential risk factor; our patient had acute renal failure), diets rich in tryptophan (may increase the substrate for biochemical conversion; although the specific diet is not detailed, the patient was receiving nutrition), and the use of immunosuppressants (associated with an increased risk of UTI and PUBS; our patient was receiving cancer treatment, which often involves immunosuppressive agents).⁽¹⁷⁾

The diagnosis of BSUP is essentially visual. The appearance of purple urine in the catheter collection bag is the main clinical sign. In many cases, such as that of our patient, the urine was purple in color, but the patient may be asymptomatic or have subtle symptoms of a UTI. The patient in this case report, despite findings of leukocytosis and positive nitrites in the urinalysis, did not report symptoms characteristic of a UTI, highlighting the importance of recognizing discoloration as an indicator of an underlying infection. Diagnostic laboratory tests include a complete blood count, urinalysis, urine culture, and renal function assessment to confirm UTI and identify the responsible microorganism. All of these studies were performed in the case presented, which allowed *Klebsiella pneumoniae* to be identified as the causative agent and its resistance profile for treatment to be determined. The management of PUBS focuses on identifying and treating the underlying urinary tract infection, along with supportive measures. This case reports the initiation of antibiotic therapy with Imipenem, replacement of the urinary catheter, and resolution of the discoloration two days after starting antibiotic treatment, demonstrating a favorable response to management.

Regarding catheter type, a systematic review and meta-analysis by Gauhar et al. examined the incidence of catheter-associated urinary tract infections (CAUTIs) between coated and uncoated catheters. Although no significant overall difference in the rate of CAUTIs was found, a subgroup analysis revealed that the risk of CAUTIs was significantly lower with coated catheters in patients who required long-term catheterization (>14 days).⁽²³⁾ Although the patient reported in this case only had the catheter for 10 days, which is not considered long-term, the duration may have contributed to the proliferation of bacteria. In these patients, coated catheters should be considered for future management, along with proper maintenance, to reduce the risk of CAUTIs.

CONCLUSIONS

The patient's case is an example of PUBS, where risk factors (female sex, immobility, constipation, UTI, renal failure, and catheterization) align with the underlying pathophysiology. Rapid resolution with appropriate treatment, which included targeted antibiotic therapy and catheter replacement, underscores the importance of early recognition and precise management to avoid unnecessary complications.

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CONSENT

The patient's consent was obtained for the performance of this study.

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CONFLICT OF INTEREST

The authors declare that there is no conflict of interest.

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