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Indonesian Journal of
Tropical and Infectious Disease

Vol. 12 No. 1 January – April 2024

Case Report

Purple Urine Bag Syndrome: a Rare Manifestation of Urinary Tract Infection

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Received: 3rd January 2023; Revised: 24th January 2023; Accepted: 6th September 2023

ABSTRACT

Purple urine bag syndrome (PUBS) is rare manifestation of urinary tract infection (UTI). Epidemiological study showed the prevalence of purple urine bag syndrome about 8.3%-16.7% worldwide. There are some factors which lead to the disease including female, long-term urinary catheter, bedridden or immobile for long time, constipation, and urinary tract infection. The mechanism of this condition involves the tryptophan in intestine that is degraded into indole. In the liver, indole is conjugated into indoxyl sulphate. This conjugate product then is excreted into urine by the kidney. In the infected urinary tract, some gram-negative bacteria produce enzymes called sulphatase and phosphatase. It converts the conjugated product, indoxyl sulphate into pigments, red indirubin and blue indigo. The two pigments-combination produces purple pigment which appears in urine. We present a-61-year-old female who has history of cerebrovascular accident who came to our emergency room with purple urine over the previous seven days.

Keywords: : Purple Urine Bag Syndrome, Urinary Tract Infection, Chronic Urinary Catheterization, Beta Lactamase, and Escherichia coli.

Highlights: an adequate antibiotic treatment of UTI bring the clinical improvement of purple urine bag syndrome. Urine culture may give the benefit in choosing appropriate antibiotic.

How to Cite: Toyyibah, I. D., Fajariya, R., Keswardiono, C. B., Queiroz, L. T. C., Wibowo, T. D P. Purple Urine Bag Syndrome: a Rare Manifestation of Urinary Tract Infection. Indonesian Journal of Tropical and Infectious Disease. 12(1). 67 - 72. April. 2024.

DOI: 10.20473/ijtid.v12i1.42151

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INTRODUCTION

Purple Urine Bag Syndrome (PUBS) is a rare manifestation Urinary Tract Infection (UTI) and first reported in 1978[1]. The unusual urine color can be distressing for the patient and families. Actually, the first case of purple urine bag syndrome was reported in 1812; this phenomenon happened to King George III.[2] Epidemiological study reported prevalence of PUBS 8.3%-16.7%.[3,4] Literature review by Yang et al.[5] collecting data from October 1980 to August 2016 showed 116 case reports with PUBS.[5,6] Female, elderly patient, constipation and chronically debilitated are considered to be the risk factor of PUBS.[3,7] This condition has been considered as benign condition and appropriate antibiotic remains to be suggested for its treatment.[5] We report a-61-year-old female with CVA pneumonia and PUBS as complication of UTI et causa prolong catheterization with *Escherichia coli* ESBL +.

CASE REPORT

A-61-year-old female with a background of cerebrovascular accident and chronic hypertension presented to emergency room with purple urine over the previous seven days (Figure 1). Due to neurogenic bladder, she had foley catheterization for one year which was changed every two weeks. For a few days, she lost appetite and did not drink enough water. There was no history of fever but she complained of pain on the tip of urethra. She was also suffering from melena and pneumonia. Her recent medication: adalat oros, candesartan, clopidogrel and citicoline.

The vital signs were stable while physical examination revealed pale conjunctiva, bilateral rhonchi, ascites and pitting edema in upper and lower extremities. Thorax photo showed pneumonia with pleural effusion. Laboratory test showed anemia (Hb 6.3, normal range 11.7-15.5), hypoalbuminemia (1.2, normal range 3.4-4.8), slight increase of creatinine serum (1.66, normal range 0.45-0.75) and BUN (40, normal range 4.6-23). Her urinalysis at the emergency room showed pH 8, Protein +3, leucocyte esterase 500, Leucocyte 25-30/HPF, erythrocyte 2-4/HPF and bacteriuria +. While urine sample was sent for culture and antibiotic sensitivity test, patient was treated by ceftriaxone 2x1 gram. The foley catheter was also changed. She received transfusion of PRC and albumin to treat anemia hypoalbuminemia.

After three days of admission, urine returned to yellow, but the symptom of UTI was not completely improved. Second urinalysis was conducted and it showed pH 7.5, albumin +2, leucocyte esterase 500, blood +1, leucocyte 10-15/HPF, erythrocyte 15-20/HPF, uric amorphous +, cast + and bacteriuria ++. Urine culture revealed significant growth of *Escherichia coli* ESBL+ (>10^5 cfu/ml) which was sensitive to amikacin, doripenem, ertapenem, meropenem, nitrofurantoin, minocycline and tetracycline. The patient was then given intravenous meropenem 3x1 gram for five days. After antibiotic was changed, the symptoms of UTI improved. The patient was discharged after 14 days of treatment.

Figure 1. Purple Urine in Urine Bag and Tubbing.

DISCUSSION

Purple Urine Bag Syndrome (PUBS) is an uncommon manifestation of UTI with prevalence 9.8% in patients with long term urinary catheter use.[8] The mechanism of PUBS involves a sequence reaction of dietary digestion and absorption of tryptophan in the intestine.[6,9] The tryptophan in the intestine is degraded by the bacteria and produces indole.[5,10] Indole transported to liver by hepatic circulation and hepatic enzyme converts indole into conjugate indoxyl sulphate. [1,5,9] The indoxyl sulphate is secreted into the urine by kidney.[5] The sulphatase and phosphatase produced by certain gram-negative bacteria in the urinary tract converts the indoxyl sulphate into blue indigo pigment and red indirubin pigment through the oxidation process.[1,5] The pigments combine causing purple staining of the urine. Not all the same species of bacteria produce sulphatase and phosphatase, but the bacteriuria is always present in all patients with PUBS even those without clinical symptoms of UTI[5]. Hepatic enzymes, bacterial urine oxidation, and the combination of indigo, a blue pigment, and indirubin, a red pigment, are the causes of the purplish discolorations of PUBS. The mechanism of purple urine bag syndrome us shown in Figure 2 below.

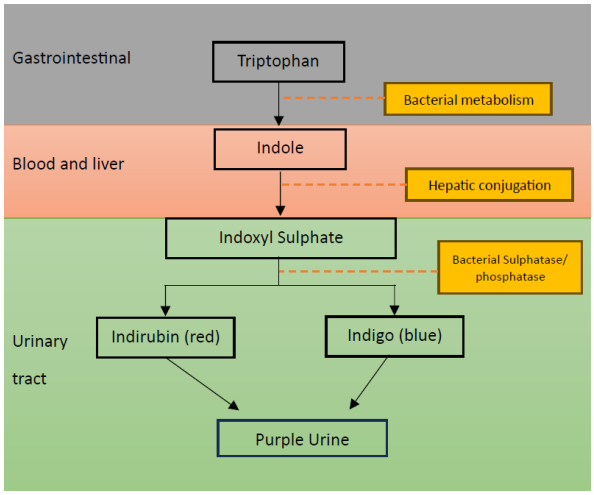

Figure 2. Mechanism of PUBS.

Based on the pathogenesis of PUBS, UTI is the factor developing the disease.[10] It is because the bacteria contribute to produce enzyme degrading indoxyl sulphate into the pigment so that the urine became purple.[5] All of the factors for UTI are also indirectly become risk factors for PUBS. Female has shorter urethra than male so that they are more likely to develop UTI.[4] Some of studies showed female obviously associated with PUBS.[5,10,11] The picture of PUBS in an old female patient are shown in Figure 3. In this presented case, the patient was an old debilitated female who complained pain on the tip of urethra which was a common symptom of UTI. She was on urine catheterization for a long time which is also the risk factor of PUBS. The condition of bedridden in this patient also has role in developing the disease. The bedridden condition is prone to reduce gut motility which is found relating to PUBS.[4] In this condition of patient, urine catheter cannot be removed and it is important to reduce the risk of infection by improve the hygiene.[12]

Figure 3. The PUBS in an Old Female Patient with Long-Term Urinary Catheter.[10]

The bacteria that are most commonly associated with PUBS are

Providencia stuartti and *rettgeri*, *Proteus mirabilis*, *Pseudomonas auruginosa*, *Klebsiella pneumoniae*, *Escherichia coli*, *Morganella*, and *citrobacter* species, Enterococci, and Group B Streptococci [2,12] The resistance organisms are infrequently reported.[11] The bacterium found in this patient was *E. coli.* The species of bacteria are the same with previous study, but in this case the bacteria produce extended-spectrum-beta-lactamase enzyme. The prevalence of extended spectrum beta-lactamase is high in Southeast Asia, Africa and Central America.[13] *E. coli* and *Klebsiella pneumoniae* are frequently organisms that produce its enzyme to became resistant to beta-lactam antibiotics including cephalosporin-third generation and penicillin, which are most commonly used due to broad spectrum activity and less toxicity.[14,15]

Urine culture and antibiotic sensitivity test have a significant role for the treatment in this presented case. Urine culture shows type of pathogen and sensitivity test helps the physician to consider antibiotic treatment especially when there is no improvement after empirical antibiotic. In this patient, the symptom of UTI did not improve with ceftriaxone as an empirical antibiotic. After she got cultured guide antibiotic, the symptom of UTI was diminished.

Alkaline urine is also considered as an associated factor because indoxyl turns into indigo and indirubin in alkaline condition.[4] Some cases, reported the patients with PUBS with alkaline urine.[1,6,8] The urinalysis of this patient revealed pH > 7. However, it had ever been reported PUBS in acidic urine and it has shown us that the pH is not a causative factor, but an associated factor.[4]

PVC plastic catheter is also reported to contribute in developing the disease[1,8]. PUBS is more frequent in patients with PVC urine bag than non-PVC urine bag. The interactions between the plastic urine catheter bag, the pigment produced by the bacteria and high bacterial load have a significant role in the disease[1].

There are many factors which contribute to develop the disease. Despite this, it is a benign ; purple urine bag syndrome is an indicate recurrent UTI due to improper hygiene[8]. Since there is no specific guideline for the disease, the management of UTI including antibiotic therapy is considered to be the important thing in PUBS treatment. Besides, good hygiene, changing the catheter regularly, considering non plastic catheter bag, treating underlying medical condition, and control of modified risk factors also bring about its resolution.

STRENGTH AND LIMITATION

The strength of this study is this was a rare case so it can give insight to manage a similar condition. The limitation of this study is the case presented is only a single case, so it is limited.

CONCLUSIONS

PUBS is rare manifestation of UTI and considered as a benign condition. There are many factors which contribute in developing the disease. Appropriate antibiotic treatment, good hygiene, good catheter care, treating underlying medical condition which precipitate to the disease and control of modified risk factors are the key in PUBS treatment.

ACKNOWLEDGMENT

We would like to thanks RSUD Syarifah Ambami Rato Ebu which supported this study.

ETHICAL CLEARANCE

This research was approved by the Health Reseach Ethics Committee of

UOBK RSUD Syarifah Ambami Rato Ebu Bangkalan with number: 0047/KEPK/XII/2023.

FUNDING

This research did not receive sponsors or specific funding.

CONFLICT OF INTEREST

All authors declare that there is no conflict of interest.

AUTHOR CONTRIBUTION

IDT involved in conceptualization, collecting data, writing manuscript. RF involved in conceptualization, data review, and supervision. CBK involved in conceptualization, data review, and supervision.

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