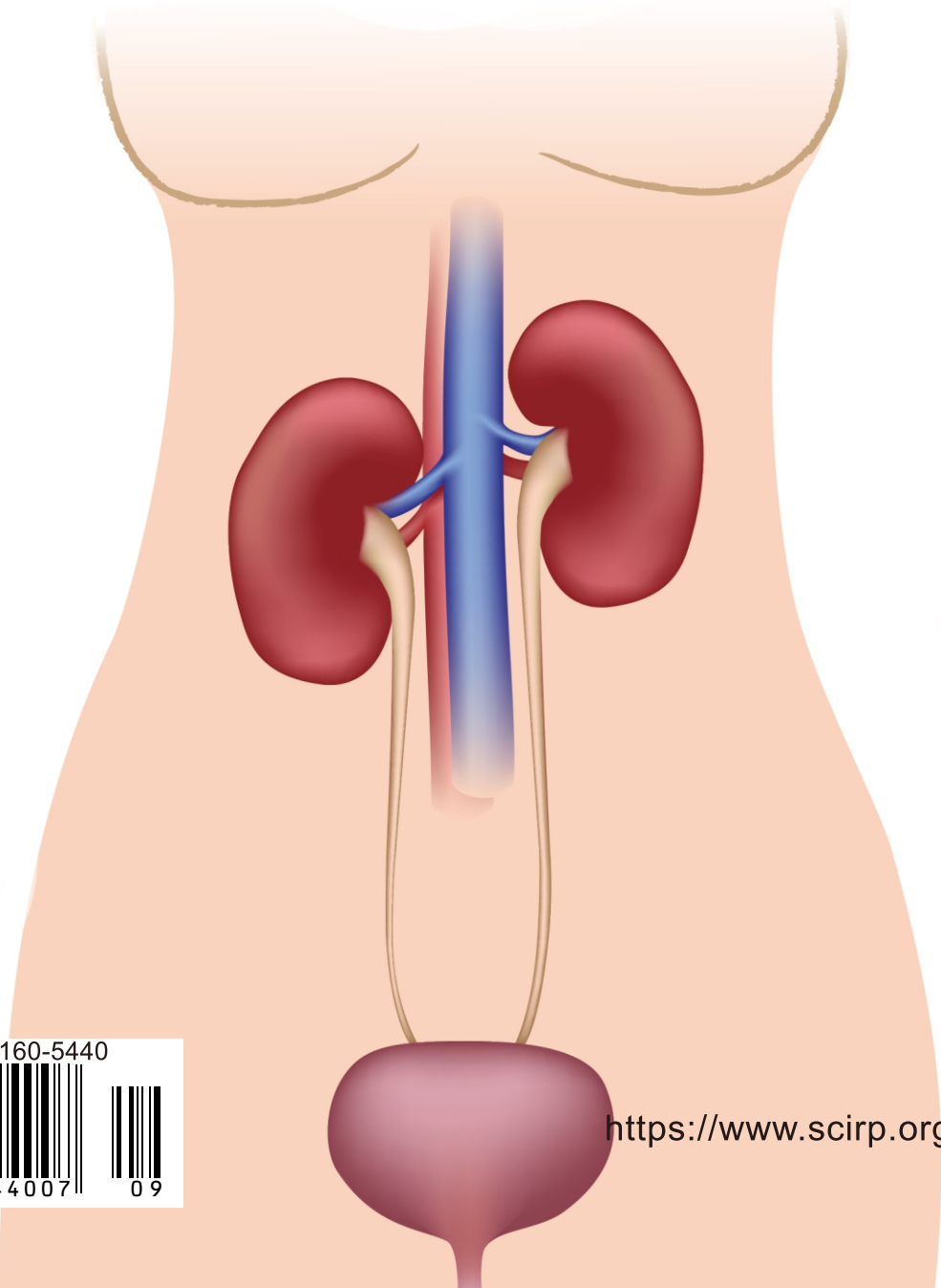


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# The Wooden Rod Foreign Body in the Urethra after Self-Dilatation of the Urethral Stricture: Case Report in the National Hospital of Ignace Deen Conakry (Guinea)

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## Abstract

A foreign body in the urethra is the presence of an object in the urethra. They are often secondary, either to an erotic curiosity, or to psychiatric disorders. The diagnosis remains clinically aided by imagery. Treatment is most often endoscopic, but in some cases the use of open surgery is necessary. We reported the case of an 80-year-old man with no history of behavioral disorders; a wooden rod was inserted into the urethra to empty his bladder. The care consisted of a meatoplasty under locoregional anesthesia. Upon discharge, the patient had a good urine flow.

## Keywords

Foreign Body, Urethra, Meatoplasty

## 1. Introduction

Self-insertion of a foreign body into the urethra is a rare condition, it is often reported in the literature as isolated cases [1]. This condition may arise as a result of satisfaction of erotic curiosity, mental disorders, or rarely in the context of an attempt to evacuate urine during acute urinary retention [2]. Several types of foreign bodies have been described in the literature. The diagnosis is most often clinical and management is generally endoscopic [3].

Although it is more common in patients with mental disorders, it has also been described in people in full possession of all their mental abilities. We report

through this clinical case a situation of self-insertion of a wooden rod in an 80-year-old patient, presenting with meatal stenosis without any associated mental deficit. Through this case, we underscore the lack of information on urology in the general population and the socio-cultural considerations making urological pathologies taboo subjects.

## 2. Observation

We had an 80-year-old patient, retired, residing in Gbessia port 1 located in the region of Conakry, who was admitted with complaint of acute urinary retention secondary to urethral outlet obstruction occurring 2 hours after self-insertion of a wooden rod into the urethra. History taking revealed a relevant past history of purulent urethritis, hypertension and self-dilatation of the urethra using rigid objects (wooden rods, etc.). There was no psychiatry past history.

Physical examination revealed a satisfactory general condition, normal-colored skin and conjunctivae, BP = 150/90 mmhg.

A palpable and mildly tender bladder globe, stenosis of the urethral meatus, induration along the anterior urethra from the navicular fossa to the root of the anterior third of the penis (**Figure 1**). The perineum was physically normal. On digital rectal examination, the prostate was slightly enlarged, smooth, elastic, painless and well limited.

An antero-posterior x-ray of the pelvis with **penile centering** did not allow for visualization of the foreign body.

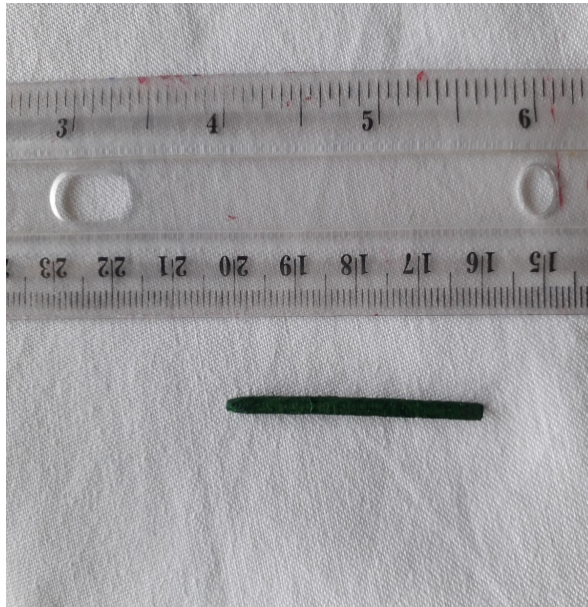
Urea and creatinin were normal.

The patient underwent urine drainage by transvesical catheter and meatoplasty associated with the removal of a wooden rod of about 4 cm (**Figure 2**). The urethral catheter was put to replace the cystostomy for one week, after the removal of the catheter the patient had a very good urine flow.

We reviewed the patient 6 months after meatoplasty with normal urine flow. Urine analysis and culture results were normal.



**Figure 1.** Stenosis of the meatus.



**Figure 2.** Foreign body.

### 3. Discussion

Most urinary tract foreign bodies are inserted during masturbation. This is however usually in the context of mental illness with self-aggressive impulsive acts [4].

Patients who are sometimes in full possession of their mental abilities use makeshift instruments for self-dilatation of the urethra. These instruments can remain or fragment in the urethra causing hemorrhagic, obstructive or infectious complications. Dysuria or urinary retention is the revealing symptoms of foreign bodies in the urethra [5]. The clinical and paraclinical diagnosis is based on history taking (notion of introduction of a foreign body into the urethra), physical examination (palpation of the foreign body along the urethra), radiography, computed tomography [3].

In our case, the diagnosis was purely clinical with palpation of the rod along the urethra. Antero-posterior pelvic X-ray didn't help for the diagnosis. Computed tomography was not done in this case.

Differential diagnosis with other urinary tract diseases can be difficult when patients do not admit that a foreign body has been self-inserted as could be the case in the event of mental illness or in the absence of a previous history of self-insertion foreign body [6]. Most foreign bodies from the urethra are removed by minimally invasive endoscopic techniques [1], and any manipulation or urethral catheterization should be prohibited until the exact type, shape, size, mobility and location of the object are determined. In some complex cases, such as that of our patient (anatomical abnormality of the associated urethra, calcified foreign body) [7] [8], surgery is certainly more invasive, but has the advantage of solving the problem without great morbidity. The foreign bodies are very varied, wooden rod as in the case of our patient, but also electric cables, tweezers, hair-

pins, pushpins, pebbles, paper clips [6].

#### 4. Conclusion

The self-insertion of foreign bodies into the urethra in patients in full possession of their mental faculties with urethral stenosis is carried out for the purpose of self-dilatation of the urethra. Treatment must systematically have two components, on one hand, the extraction of the foreign body and on the other hand the correction of the stenosis.

#### Conflicts of Interest

The authors declare no conflicts of interest regarding the publication of this paper.

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# Evaluation of Knowledge of Medical Students of University of Lomé (Togo) on the Contribution of Prostate Biopsy in the Detection of Prostate Cancer

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## Abstract

**Background:** Prostate cancer is the second leading cause of death from cancer in humans in most developed countries. In Togo, around 59.8% of prostate cancers are diagnosed at an advanced stage because of the insufficient screening in connection with the recent introduction of prostate biopsy and the late generalization of PSA (prostate specific antigen). **Objective:** This study aimed to assess the knowledge of students from the Faculty of Health Sciences of the University of Lomé on the contribution of prostate biopsy in the diagnosis of prostate cancer. **Results:** A total of 1017 students filled out the form, with an overall response rate of 95.3%. The range age of the subjects was 24.5 years, and a sex ratio (M/F) of 2.51. The majority of students were license (546, 53.69%). Five hundred and eighty-one (57.13%) of the students had not received any training in prostate cancer. Five hundred and eighty-two students (57.23%) had no knowledge of prostate biopsy. There is a statistically significant link between students' knowledge of the prostate biopsy and age (OR 1, 95% CI [0.49 - 2.03], p = 0.0001), sex (OR 1, 95% CI [0.33 - 1.08], p = 0.0003), the study cycle (OR 2.5, 95% CI [1.02 - 5.06], p = 0.0047) and the effectiveness of an internship in urology department (OR 1, 95% CI [0.61 - 1.31], p < 0.0001). **Conclusion:** Medical students have little knowledge of the place of biopsy in the detection of prostate cancer. Educational efforts

should focus on improving their knowledge so that they can make appropriate decisions in the future.

## Keywords

Prostate Cancer, Biopsy, Knowledge, Medical Students, Togo

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## 1. Introduction

Prostate cancer is the second leading cause of death from cancer in humans in most developed countries, and the global burden of this disease is increasing [1]. More than half of these prostate cancers are diagnosed before the age of 75 [2]. The constant decrease in specific mortality from prostate cancer observed in recent years in the USA and France through data from cancer registers, partly responds to the interest in individual screening for men aged 50 to 75 with a long life expectancy greater than 10 years [3] [4]. The diagnosis of prostate cancer is established by prostate biopsy and allows a real mapping of the prostate to be carried out [5] [6]. Many prostate cancers are detected based on high plasma levels of prostate specific antigen (PSA > 4 ng/mL), a glycoprotein normally expressed by prostate tissue [6]. However, because men without cancer have also been found with high PSA levels, a tissue biopsy is the standard of care to confirm the presence of cancer [7].

In Africa, prostate cancer is first cancer in both the occurrence and the number of deaths [8] [9]. It is also the leading human cancer in Togo, accounting for 74.63% of urological cancers [10]. In Togo, around 59.8% of prostate cancers are diagnosed at an advanced stage (Gleason score > 7) [11]. This is probably due to insufficient screening in connection with the recent introduction of prostate biopsy and the late generalization of PSA, but also to the lack of exhaustive registration of cases treated in a liberal environment. Medical students who are future general practitioners play an essential role in health prevention, because they are the first prescribers of PSA. The objective of this study was to assess the knowledge of medical students of the Faculty of Health Sciences at the University of Lomé on the contribution of prostate biopsy in the early detection of prostate cancer.

## 2. Methods

### 2.1. Study Plan and Sampling

A cross-sectional study was carried out among students regularly enrolled in the Faculty of Health Sciences (FSS) of the University of Lomé (UL) in the 2019-2020 school year account. Togo is a country of 56,600 Km<sup>2</sup>, with an estimated population of 7,200,000 located between Ghana in the west and Benin in the east. The total number of students regularly registered with the UL FSS was 1635. The minimum sample size (*n*) was calculated using the following formula:

$$n = t^2 \times p \times (1 - p) / m^2 .$$

$t$ : 1.96: value corresponding to the 95% confidence level.

$p$ : estimated prevalence rate = 0.4.

$m$ : desired degree of precision = 0.05.

$$n = (1.96)^2 \times 0.4(1 - 0.4) / (0.05)^2 = 368.79.$$

The minimum sample size  $n = 368.79$  or 369 students.

We enrolled 1017 of 1635 students, representing 62.2% of the total FSS student population. It took place from January 1 to March 31, 2020, duration of 3 months.

The selection criteria were the statute of regularly registered students and the agreement to participate in the study.

## 2.2. Data Collection

The data were collected using a self-administered anonymous structured questionnaire, divided into three (03) main parts. The survey questionnaire included an introductory first part detailing the objectives and methodology of the study. The second part sought information on the socio-demographic characteristics of the students, the notion of previous training or courses on prostate cancer and the experience of an internship in a urology department. The third part dealt with their level of knowledge on prostate biopsy. The knowledge was considered bad if the student hadn't any notion on prostate biopsy, intermediate if the conditions of realization were known and good if conditions of realization and signification of the result were known. The socio-demographic section focused on the personal characteristics of the students in the sample, such as age, gender, level of medical education. The section on prostate biopsy included 3 questions concerning the conditions for carrying out this examination. These were questions that subjects should answer with "Yes" or "No". The questionnaire was pre-tested on 50 students, without any particular problem.

## 2.3. Data Management and Analysis

Data was entered twice in Microsoft Excel to reduce data entry errors and then exported to Epi Info version 7 software. A descriptive analysis was carried out with a view to highlighting the characteristics of the different qualitative and quantitative variables. We used percentages for qualitative variables and means with their standard deviations for quantitative variables. The statistical tests used were the Paerson Chi-square test for the qualitative variables and the Student test for the quantitative variables. The significance threshold was set at 0.05.

## 3. Results

### 3.1. Epidemiological Data

**Table 1** shows the sociodemographic characteristics of the participants. The self-administered questionnaire was returned by a total of 1017 students, with an overall response rate of 95.3%. The average age of the subjects was 24.5 years



**Table 1.** Socio-demographic characteristics.

	<i>Value</i>
<b>Total</b>	<b>1017</b>
<b>Age (years)</b>	
1) Middle age	24.5
2) Range	17 - 32
3) ≤25	821/1017 (80.7%)
4) >25	196/1017 (19.3%)
<b>Sex</b>	
1) Male	727/1017 (71.5%)
2) Female	290/1017 (28.5%)
<b>Study cycle</b>	
1) Doctorate	357/1017 (35.1%)
2) Master	114/1017 (11.2%)
3) License	546/1017 (53.7%)
<b>Training/courses</b>	
1) Yes	581/1017 (57.1%)
2) No	436/1017 (42.9%)
a) ≤6 months	68/436 (15.6%)
b) Between 6 months and 1 year	33/436 (7.6%)
c) >1 year	335/436 (76.8%)
<b>Internship in a urology department</b>	
Yes	886/1017 (87.1%)
No	131/1017 (12.9%)

with extremes of 17 and 32 years. Eight hundred and twenty-one (821) or 80.73% of the students were 25 years of age or less. These were 727 (71.48%) male subjects and 290 (28.52%) female subjects, representing a sex ratio (M/F) of 2.51. According to the university course, 357 (35.10%) were in the Doctorate cycle, 114 (11.21%) in the Master cycle and 546 (53.69%) in the Bachelor cycle. Five hundred and eighty-one (581) or 57.13% of the students had received no training or course in prostate cancer. Sixty-eight (68) or 6.69%, 33 (3.24%) and 335 (32.94%) had received training or a course in prostate cancer, with a delay of less than 6 months respectively, between 6 months and 1 year, and more than a year. Eight hundred and eighty-six (886) or 87.12% of students had never completed an internship in a urology department.

### 3.2. Knowledge of the Prostate Biopsy

Five hundred and eighty-two (57.23%) students had no knowledge of the prostate biopsy. These were 510 students, or 93.40% of those in the Bachelor's cycle.



For students in the Master and Doctorate cycles, respectively 79 (69.29%) and 320 (89.63%) had knowledge of the prostate biopsy. Ninety-two (9.05%) students had previously requested a patient's prostate biopsy. Seventy-two (78.26%) had requested it in patients with abnormal prostate rectal examination associated with elevated PSA (prostate specific antigen), 15 (16.30%) with PSA elevation alone and 05 (5.44%) before only an abnormal digital rectal examination of the prostate. There is a statistically significant link between students' knowledge of the prostate biopsy and age (OR 1, 95% CI [0.49 - 2.03]),  $p = 0.0001$ ), gender (OR 1, 95% CI [0.33 - 1.08],  $p = 0.0003$ ), the study cycle (OR 2.5, 95% CI [1.02 - 5.06],  $p = 0.0047$ ) and the effectiveness of an internship in urology department (OR 1, 95% CI [0.61 - 1.31],  $p < 0.0001$ ). On the other hand, there is no statistically significant link between students' knowledge of the prostate biopsy and taking a course or training in prostate cancer (OR 1.72, 95% CI [0.12 - 2.95],  $p = 1.345$ ) (Table 2).

#### 4. Discussion

Our study has some limitations like any series based on opinion poll. The sincerity of the answers cannot be verified.

**Table 2.** Socio-demographic characteristics VS knowledge on prostate biopsy.

Socio-demographic characteristics	Univariate analysis (Knowledge of prostate biopsy)				
	n/N	%	OR	IC à 95%	P-value
<b>Sex</b>					<b>0.0003</b>
Male	330/727	45.4	1	-	
Female	105/290	36.2	0.5	[0.33 - 1.08]	
<b>Age</b>					
≤21 years	48/524	9.2	1	-	<b>0.0001</b>
>21 years	387/493	78.5	0.86	[0.49 - 2.03]	
<b>Study Cycle</b>					
License	36/546	6.6	1	-	<b>0.0047</b>
Master	79/114	69.3	3.4	[1.02 - 3.72]	
Doctorat	320/357	89.6	2.15	[1.83 - 5.06]	
<b>Follow up on your latest prostate cancer training/course</b>					
Less than 6 months	60/68	88.2	1	-	1.345
Between 6 months and 1 year	30/33	90.9	0.33	[0.12 - 1.95]	
More than 1 year	297/335	88.7	0.68	[0.45 - 1.22]	
None	48/581	8.3	1.72	[1.31 - 2.95]	
<b>Effectiveness of an internship in urology department</b>					
Yes	120/131	91.6	1	-	<b>&lt;0.0001</b>
No	315/886	35.6	0.72	[0.61 - 1.31]	

Prostate cancer is an adenocarcinoma that is most often sporadic (80% of cases), but can be hereditary (10% of cases) or familial (10% of cases) [12]. Prostate cancer in the initial stage shows intraglandular cancerous lesions or intraepithelial neoplasias that do not reach the prostate capsule [12]. Medical educators must teach students the nuances and uncertainties of detecting prostate cancer, and future physicians will ultimately need to integrate this knowledge into communication with their patients. Our study provides an overview of contemporary knowledge on the detection of prostate cancer in medical students. The PSA test has received negative press in recent years, which is why the controversy surrounding prostate cancer screening continues [1] [2] [3] [4]. Since a reduction in the incidence of the disease through effective primary prevention or the use of pharmacological treatments is not expected, at least in the short term, secondary prevention with PSA test seems to remain the most appropriate [13]. In addition, decisions regarding prostate cancer screening should be based on the preferences of an informed patient. The majority of students (80.73%) are 25 years of age or younger. None belonged to the group of subjects at risk of prostate cancer. Indeed prostate cancer is a cancer of the elderly, and screening is recommended in men from 40 years if there is a family history of prostate cancer or breast cancer in a first-time relative degree, and generally from the age of 50 in all other men [5] [6] [7]. We noted a clear male predominance with a sex ratio (M/F) of 2.51. Given that this is an exclusively male pathology, this explains a pronounced interest in the male gender.

Prostate biopsies allow the histological diagnosis of prostate cancer [14]. They are carried out by a urologist or a radiologist in a patient who presents a clinical suspicion with an abnormality in the rectal or biological touch (PSA > 4 ng/ml) [15] [16]. They are performed under ultrasound control with an 18 Gauge needle, on an outpatient basis or in a day hospital, under local, locoregional or general anesthesia [16]. An isolated elevation of the PSA level should not prompt immediately to perform a biopsy, this rate should be checked after a few weeks using the same dosage under standardized conditions including, no ejaculation, digital rectal examination, urethral manipulations or urinary tract infection [17]. A hemostasis check-up is required if a bleeding disorder is suspected. In the absence of clinical or imaging abnormalities (ultrasound or MRI), the 12-sample regimen is recommended [18]. In case of clinical or imaging abnormalities, additional directed biopsies are performed [19]. The indications for a new biopsy are linked to the increase or persistence of a high PSA level, suspect digital rectal examination, and the presence of atypical small acinar proliferation on the first series, the existence of 'a diagnostic doubt on a first series of biopsies [20] The second series must include 4 to 6 additional biopsies, in the anterior apex and the transition zone (anterior fibromuscular stroma) in search of anterior cancer [21]. Many authors have shown in clinical studies that the percentage of cancers detected exclusively by additional transitional zone biopsies is very low [22] [23]. For Bazinet, only 2.9% of cancers diagnosed by an extensive protocol (combining conventional peripheral biopsies and biopsies of the transition zone) are de-

tected exclusively by biopsies directed in the transition zone [23]. In practice, guidelines are not often followed; one of the main causes being the lack of knowledge of general practitioners about the recommendations [7]. This observation was made among medical students at the University of Lomé. In fact, the majority of participants (57.23%) had no knowledge of the prostate biopsy. There is a statistically significant link between the students' knowledge of the prostate biopsy and the effectiveness of an internship in a urology department (p-value < 0.0001; 95% CI [0.61 - 1.31]). It is desirable to continue this supervision in the field of these future physicians, in order to allow them to make informed decisions regarding the non-negligible risks of the prostate biopsy. On the other hand, there is no statistically significant link between students' knowledge of the prostate biopsy and taking a course or training in prostate cancer (p-value = 1.345; 95% CI [0.12 - 2.95]). This suggests a reinforcement of knowledge capacities on the prostatic biopsy of the latter during theoretical courses at the faculty [24] [25].

## 5. Conclusion

Prostate cancer is a common but heterogeneous condition. Confirmation of the diagnosis requires a prostate biopsy. This act involves risks including allergy, hemorrhage, or secondary infection. The patient's information must be prior to the biopsy procedure and delivered in consultation with the doctor indicating the biopsies. Doctors hardly follow these recommendations for lack of knowledge. The same observation was made among medical students at the University of Lomé. It is necessary to strengthen their knowledge of prostate biopsy in order to equip them for informed decision-making in the future.

## Conflicts of Interest

The authors declare no conflicts of interest regarding the publication of this paper.

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# Intra-Diverticular Bladder Tumor about a New Observation in the Urology Department of Chu Cocody—Abidjan, Côte d’Ivoire

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## Abstract

This case report is about a 61 years old woman who consulted for hematuria, the cystoscopy performed revealed an intra-diverticular bladder tumor. After tumor resection, an anatomopathological exam revealed the urothelial carcinoma pT1. A few months later she presented with a local recurrence of bladder tumor. We proposed her anterior pelvicectomy but she refused it and she was lost to follow-up. The aim of this study is to show the anatomopathological and evolutionary characteristics and the therapeutic difficulties of IDBT. These tumors are in the majority of cases infiltrating from the outset with a high potential for recurrence after transurethral resection of bladder (TURB). The standard treatments are TURB and total cystectomy.

## Keywords

Tumor, Bladder, Diverticulum, TURB

## 1. Introduction

Bladder diverticula are outpouchings of the bladder wall they are either congenital or acquired defects. They are usually thin-walled with a narrow neck and lack the muscular propria layer. A subset of these lesions, however, may be complicated with inflammation, calculus, infection, and malignancy [1]. Intra-diverticular bladder tumors (IDBT) are rare and pose diagnostic, therapeutic and anatomopathological difficulties. In 2% - 7% of patients with vesical diverticula, neoplasms develop within the diverticulum [2]. The absence of muscular in the diverticulum, and the direct contact between mucosa and the peri-bladder fat ex-

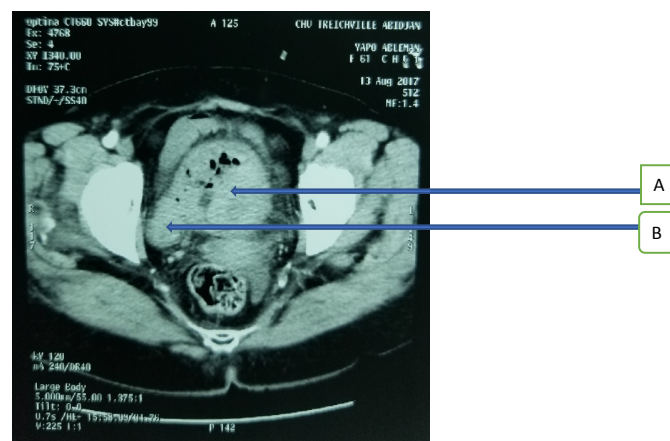
plains the different evolutionary profile of IDBT [3]. The aim of our study is to show the anatomopathological and evolutionary characteristics and the therapeutic difficulties of IDBT.

## 2. Observation

Mrs. Y.A.S., 61 years old, is a retired schoolteacher who was admitted to our department for total hematuria on 08/16/2017. She had presented for 3 months pelvic pain associated with intermittent terminal hematuria initially, which became total and permanent with numerous blood clots. This hematuria was associated with dysuria with an impact on the patient general condition such as asthenia, dizziness and palpitations. The physical examination found a conscious, cooperative patient with pale conjunctiva. Her blood pressure was 100/60 mmHg, a pulse of 100 bpm, her temperature was 37.4°C and a respiratory rate of 25 cycles/min. The examination of the lumbar fossa and hypogastrium was normal. The cervix was normal on vaginal examination and speculum.

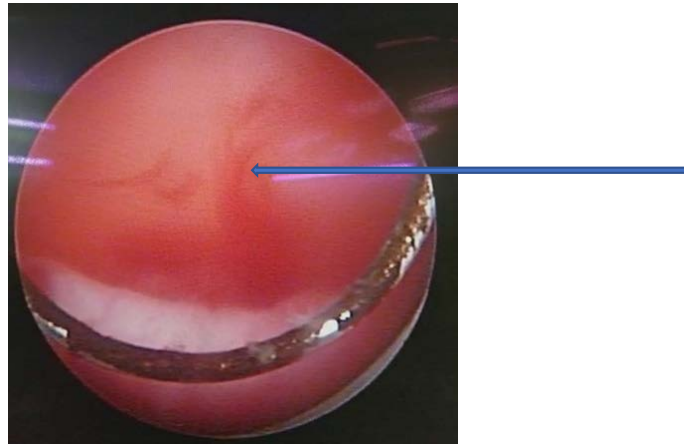
The tomography performed showed an isodense necrotic tissular process at the bladder measuring 88 × 63 mm. This image was similar to a usual bladder tumor. In fact, it was a large blood clot. There were no locoregional invasion, lymphadenopathy or bone lesions. There was the presence of a diverticulum at the right lateral wall of the bladder (**Figure 1**).

The renal biological assessment was normal with a blood urea level of 0.27 g/l and serum creatinine at 7 mg/l (62 mmol/l). The blood count showed severe anemia with a hemoglobin (hb) level of 3 g/dl. She was transfused with 3800 ml of concentrate erythrocyte and after transfusion the blood count level was 7.8 g/dl of hbb. An indication for transurethral resection of the bladder (TURB) has been made. It was for hemostatic and diagnostic purposes. During the operation, the cystoscopy found many clots which we aspirated with the Ellick pear. The exploration revealed healthy bladder lining as a whole and a diverticulum in the right lateral wall about 3 cm in diameter. Bleeding was observed within this diverticulum (**Figure 2**).



**Figure 1.** CT scan showing an isodense necrotic tissular process in the bladder (A) and a diverticulum in the right lateral wall (B).





**Figure 2.** Bleeding from the diverticulum on cystoscopy.

Exploration of the diverticulum revealed a sessile-based urothelial tumor about 1 cm diameter (**Figure 3**).

During the resection, there was an accidental perforation of the bladder inside the diverticulum which required the catheter to be maintained for 10 days. Pathological examination of the surgical specimen revealed papillary urothelial carcinoma G II pT1 (**Figure 4**).

During monitoring, a cystoscopy performed at 3 and 6 months found normal bladder mucosa, a diverticulum with healthy mucosa. The patient refused all adjuvant treatment and was lost to follow-up. Reappeared 15 months later with terminal hematuria, the cystoscopy performed found a recurrence tumor in the diverticulum and another urothelial tumor in the bladder dome. An indication for a previous pelvicotomy was made but the patient refused and was lost to follow-up.

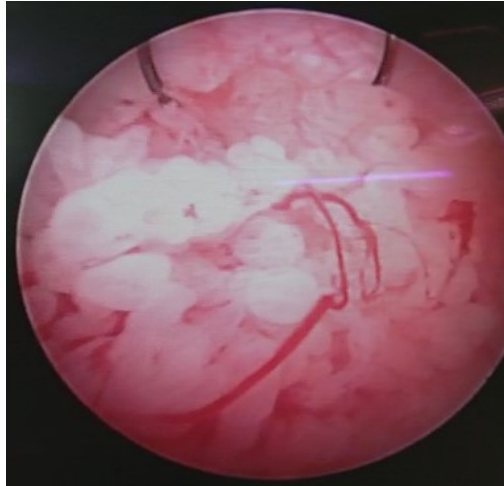
### 3. Discussion

The incidence of IDBT is between 0.8% and 13.5% with a male predominance and an average age between 60 and 70 years [4] [5]. IDBT represents 1.5% of bladder tumors [6]. This case is the first observed in the CHU Cocody urology department.

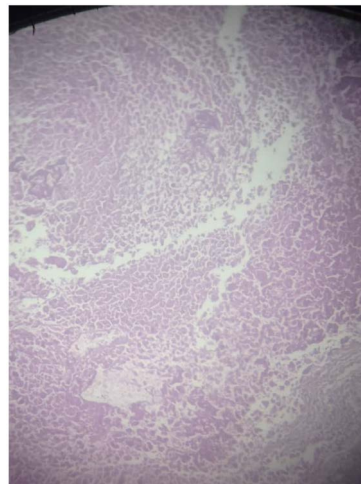
There is a well-documented relationship between bladder diverticula and bladder cancer. One study reported that when a diverticulum was present, the likelihood of neoplastic changes compared to a normal bladder. It was increased by 0.8% to 10% [7]. Urine stasis in the bladder diverticulum is thought to produce chronic mucosal irritation and prolonged exposure to urinary carcinogens, thus increasing the risk of malignant tumors of the epithelial lining of the diverticulum [6].

The revealing symptoms are either those of a bladder tumor such as hematuria (66% to 100% of cases), or those of urinary obstruction [3]. Our patient presents profuse hematuria, which led to severe anemia with respiratory distress.





**Figure 3.** Intra-diverticular bladder tumor with urothelial tumor appearance.



**Figure 4.** Image anatomopathological showing urothelial carcinoma pT1 GII.

Cystoscopy is the gold standard for diagnosis [3]. It is limited in the case of a diverticulum with a narrow neck where the cystoscope cannot penetrate [7]. The CT scan and MRI make it possible to diagnose and furthermore assess the tumor extension to the peri vesical fat [2] [8] [9]. In our case the CT scan did not allow us to make the diagnosis, it's rather the cystoscopy. This cystoscopy was made during the TURB indicated to stop the bleeding and to do a pathological diagnosis of a large bladder tumor seen on CT scan.

Among malignant tumors, urothelial carcinoma is the most common (78%), followed by squamous cell carcinoma (17%), a combination of transitional and squamous cell types (2%), and adenocarcinoma (2%) [10]. IDBT progresses from Ta-T1 stage to T stage  $\geq 3$ . T2 stage does not exist due to the absence of muscle in the diverticulum. Fifty-five (55) to 60% of tumors diagnose are made at stage T3 [6] [11]. The absence of muscle fibers exposes a high risk of bladder perforation during resection. This accident occurred during our operation, wich required the maintenance of the urinary catheter for 10 days. This is why some

authors advise doing only a simple cold forceps biopsy [5] [6] [12].

Therapeutically: Ta-T1 and low-grade IDBT can be treated by endoscopic resection alone or followed by BCG therapy in the event of associated in situ carcinoma. High-grade IDBT, unique and without associated in situ carcinoma, can be treated by diverticulectomy associated with pelvic lymph node dissection. Multiple high-grade IDBT, or those associated with in situ carcinoma, justify total cystectomy [13]. Baniel and al [4] in their series of 8 patients with a tumor considered to be superficial (5pTa and 3pT1) used the combination endoscopic resection—endovesical instillation. With an average follow-up of 42 months, they obtained the following results:

- pTa: No recurrence.
- pT1: 1 disease-related death, 1 local recurrence and 1 positive cytology without visible tumor were reported. Thus, although this therapeutic combination (TURB + endovesical instillation) seems a better treatment for pTa tumors of low grade tumors, it is not recommended for tumors infiltrating the chorion [3]. Diverticulectomy and partial cystectomy have not showed their effectiveness due to the tumor frequency  $\geq$  pT3 [11] [14]. Total cystectomy remains the standard treatment. Despite this procedure, the prognosis for these tumors is more unfavourable, linked to the high percentage of peri-bladder extension  $\geq$  pT3a [3]. The endovesical instillation wasn't performed in our patient due to the unavailability of products. She presented with recurrence tumor at the diverticulum and at the bladder dome.

The survival rates, in the literature, were 45% at 1 year, 20% at 3 years, and 15% at 4 years at the pT3a stage after total cystectomy [5] [9] [15].

#### 4. Conclusion

IDBT are rare, the clinical signs are those of a usual bladder tumor. Cystoscopy provides a definite diagnosis as other bladder tumors. Pathology is distinguished by the absence of stage T2 because there are no muscle fibers in the diverticulum. These tumors are in the majority of cases infiltrating from the outset with a high potential for recurrence after TURB. The standard treatment is TURB and total cystectomy.

#### Conflicts of Interest

The authors declare no conflicts of interest regarding the publication of this paper.

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# Nephrectomy in a Patient with Medullary Sponge Kidney Associated with Severe Nephrocalcinosis and Nephrolithiasis: A Rare Condition

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## Abstract

**Background:** Medullary sponge kidney (MSK) is a disturbance of renal development characterized by cystic dilation and diffuse precalyceal duct ectasia. The disease affects both genders in equal proportions and is generally diagnosed in adulthood, as a result of recurrent calcium nephrolithiasis and nephrocalcinosis. The most frequently encountered manifestations being renal colic, microscopic or macroscopic hematuria, and fever. The intravenous pyelogram is standard for diagnosis and metabolic workup is required to identify the underlying cause. The main goal of treatment is to prevent recurrence and disease progression. Though considered a benign condition, a nephrectomy may often be required in patients presenting late with irreversible complications and end-stage renal disease. **Aim:** To highlight and discuss the presentation and management of a rare case of nephrocalcinosis and nephrolithiasis secondary to the medullary sponge kidney. **Case presentation:** We report herein the case of a 56-year-old male with long-standing hematuria in whom a diagnosis of medullary sponge kidney disease was made and he underwent a left total nephrectomy. The postoperative course was uneventful. **Conclusion:** Nephrocalcinosis and nephrolithiasis are complications of MSK and can result in irreversible renal damage. A high index of suspicion is necessary for patients presenting with renal colic, recurrent urinary tract infections, or hematuria for prompt diagnosis and management.

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## Keywords

Medullary Sponge Kidney, Nephrolithiasis, Nephrocalcinosis, Nephrectomy

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### 1. Introduction

Medullary sponge kidney (MSK) or Cacchi-Ricci disease represents a disturbance in renal development characterized by cystic dilation and diffuse precalyceal duct ectasia. These dilations are between 1 and 7 mm in size, frequently containing small calculi, and the communication between the respective ducts and calyces may be stenotic [1].

The true prevalence of MSK in the general population is not known, but is estimated to be less than 0.5% - 1%. MSK is much more frequent (12% - 20%) in recurrent renal calcium stone formers [2].

It has been claimed that MSK results from the coexistence of multiple tubular function defects in different nephron segments which points to the abnormal development (polarization) of the tubular epithelium, probably as a consequence of an abnormal “ureteric bud–metanephric mesenchyme”, or receptor tyrosine kinase (RET)/glial cell line-derived neurotrophic factor (GDNF) interface [2].

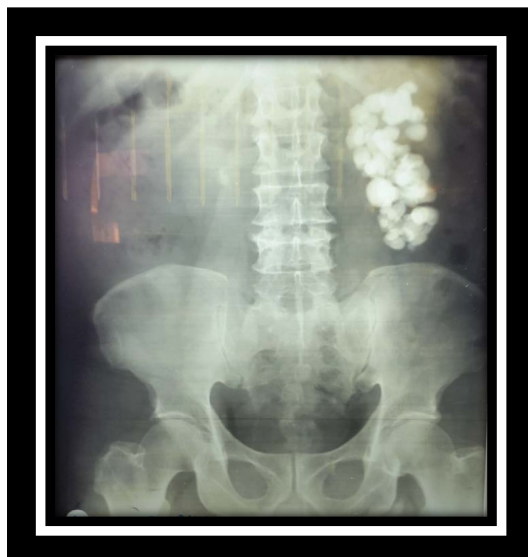
MSK affects both genders and is generally diagnosed in young adults. The disease is typically observed in recurrent renal stone formers [2]. MSK can be asymptomatic but can present as nephrocalcinosis, urolithiasis, and recurrent urinary tract infections (UTIs) [3]. Uncommonly, in a few patients, the clinical course of MSK may be dominated by pain, which may, or may not, be associated with the spontaneous passage of stones or pyelonephritis [2]. Indeed, microhematuria and macrohematuria are naturally very frequent, generally associated with stone passage, though rarely, patients may experience frank, persistent hematuria apparently unrelated to the passage of stones [2].

We present and share our experience in the management of a complicated case of MSK in a 56-year-old patient with long-standing hematuria.

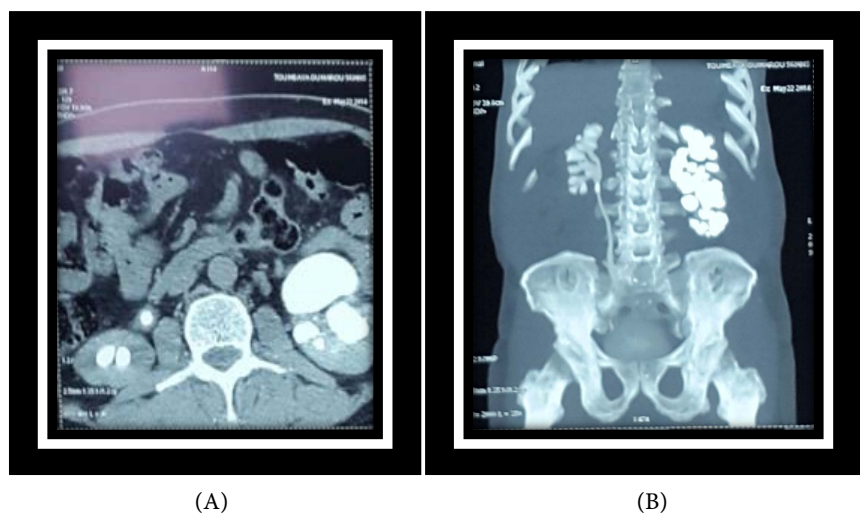
### 2. Case Report

A 56-year-old patient living in the northern region of Cameroon was referred for multiple lithiasis of the left kidney. He complained of multiple episodes of total hematuria associated with left renal colic. There were no urinary symptoms or fever. His past history revealed repeated hematuria for the last 31 years. He has type 2 diabetes and has been followed for 14 years on oral antidiabetic drugs. There was no notion of urinary schistosomiasis or smoking. Clinical examination showed stable vital parameters with a blood pressure of 130/85mmHg and heart rate of 78 beats/minutes. Physical examination was unremarkable. In the absence of scintigraphy, we were unable to assess differential renal function although serum creatinine (9.13 mg/l) and urea (0.33 g/l) were normal. A plain abdominal radiograph (Figure 1) showed multiple opacities on the left renal

area. CT Urogram (**Figure 2**) showed left nephrocalcinosis and right renal lithiasis. It also showed accumulation of contrast medium in dilated ducts within the left renal medulla with the characteristic “bouquet of flowers” appearance. The ureter was not visible on the left side, but there was no obstruction on the right. Calcemia (90.3 g/l) and natremia (148.1 mEq/l) were normal. In contrast, phosphoremia (51.87 IUI/l) and uricemia (72 mg/l) were slightly elevated. Urine culture was sterile and urine electrolytes were not checked. We carried out a left total nephrectomy (**Figure 3**) via a flank approach under general anesthesia. The operative findings included 61 stones of various sizes, arranged in layers in the medulla and cortex as well as diffuse ectasia of the precalyceal ducts. This was strongly suggestive of Cacchi-Ricci disease (**Figures 4-6**). The intervention was hitch-free and the postoperative course was uneventful. Neither stone analysis



**Figure 1.** Plain abdominal radiograph showing multiple calcifications in left renal area.

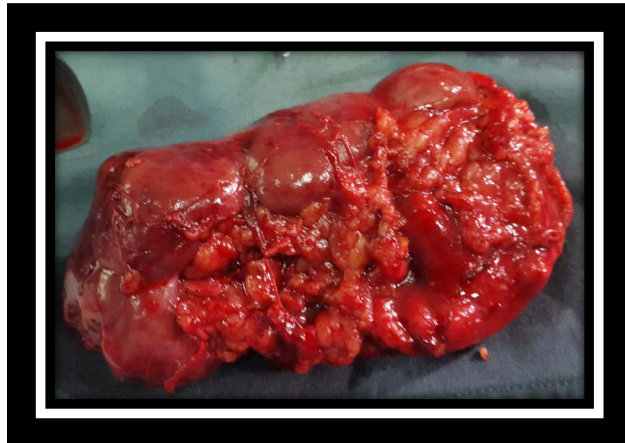


(A)

(B)

**Figure 2.** CT Urogram ((A) bilateral urolithiasis; (B) “Bouquet of flowers” appearance during the excretory phase).





**Figure 3.** Left kidney specimen after nephrectomy.



**Figure 4.** Nephrocalcinosis.



**Figure 5.** Precalyceal ectasia of left kidney.



**Figure 6.** 61 stones from left kidney.

nor histopathology of the nephrectomy specimen was performed. The patient was discharged on postoperative day five. The patient was jointly reviewed with the nephrologist. Follow-up visits were done at 6 and 9 months with assessment of renal function and ultrasound imaging of the urinary tract during each visit. The right renal calculi were managed conservatively with high fluid intake and urine alkalization using potassium citrate. Adverse prognosis was mitigated by strict follow up with regular control of renal function and screening for urinary tract infections as well as dietary modification.

### 3. Discussion

Theories behind the etiology of stone formation in MSK include urinary stasis in dilated tubules leading to increased crystal precipitation, renal leak, hypercalciuria resulting from impaired tubular function, and distal renal tubular acidosis and associated hypocitraturia resulting from impaired tubular function [4].

Any hypothesis on the pathogenesis of MSK should explain the concomitant occurrence of alterations in the ureteric bud derived pre-calyceal and collecting ducts (*i.e.* the “lower nephron”), such as “cysts”, nephrocalcinosis and acidification and concentration defects, as well as in the metanephric blastema-derived nephron (“upper nephron”), such as proximal tubular defects [3].

As a disease itself, MSK can coexist with other renal developmental anomalies since it is considered a renal developmental malformation itself. Abnormalities in developmental genes including GDNF and RET as well as the association with Multiple Endocrine Neoplasia (MEN 2A) can lead to several different renal pathologies, extending beyond MSK [5].

Most patients with MSK are asymptomatic. MSK may have a sporadic prevalence of 5/10,000 to 5/100,000 for some authors while other sources state that the epidemiology of MSK is difficult to predict. This may be the result of missed diagnosis as MSK is known to have an indolent course and often asymptomatic



presentation [5].

The diagnosis is highly suggested in light of investigations imposed by related complications such as renal stones, urinary tract infections, renal tubular acidosis or urine concentration defects. In some cases, MSK may be silent or take an indolent clinical course with an unremarkable history; very rarely though, the MSK patient may have a clinical course marked by colicky flank pain not necessarily associated with stone passage or macro haematuria [3].

The disease is clinically diagnosed in 0.2% to 20% of cases, the most frequently encountered manifestations being renal colic, micro- or macroscopic hematuria, and fever [1] [4]. Indeed, nephrolithiasis may also lead to frequent UTIs, pyelonephritis and hematuria and the differential diagnoses for MSK can be made more precisely when patients present with other renal manifestations, especially in groups at risk. These groups include patients between the age of 20 and 30, those with other renal malformations, high sodium diet, hyperparathyroidism, and family history of MSK [5]. It was recurrent hematuria and left renal colic that led our patient to consult. The diagnosis was however made late as the patient has had recurrent hematuria for the past 31 years.

Fabris *et al.* [3] reported that the disease affects both genders in equal proportions, is generally diagnosed in adulthood and results in recurrent calcium nephrolithiasis and nephrocalcinosis. Moreover, it was suspected that the diagnosis is made mainly in adulthood because of its progressive nature [3].

The vast majority of MSK patients develop medullary nephrocalcinosis which may be noticeable on ultrasound as renal pyramid hyperechogenicity of different intensity. Due to the small size of calcium salt deposits, they are usually not accompanied by acoustic shadows [6].

In patients with suspected MSK, further imaging techniques are required hence the ultrasound signs are not specific for this condition [7]. The gold standard for diagnosis of MSK remains IVU showing accumulation of contrast medium within papillary ducts.

However, other procedures such as CT, X-ray, and endoscopy can also be used [5]. Radiologic imaging makes it possible to confirm the diagnosis as well as identify complications as is the case when stones occupy the entire urinary tract. Indeed, it is a disease found in 12% to 20% of radiopaque renal stones composed predominantly of calcium oxalate. Nephrolithiasis can be observed in 70% of patients with MSK [5].

Typical radiographic images of MSK show collections of contrast media in dilated collecting tubules with the appearance of a papillary brush or bouquet of flowers [8] [9]. Contrast medium collection in papillary ducts leads to this classic image which when present displays either a pattern of bouquets, brush-like striation, or linear striations [8]. In our case, the first plain abdominal film done by the patient showed the characteristic “bouquet of flowers” appearance.

Our patient’s CT Urogram showed nephrocalcinosis and nephrolithiasis with complete obstruction on the left, but no obstruction on the right. However, some

authors stated that the replacement of IVU by CT imaging is a drawback in MSK management due to CT being unable to properly demonstrate the classic signs of MSK [8] [10] [11]. CT imaging has been proven to better establish the diagnosis of most urologic conditions, but this, unfortunately, does not apply to MSK. However, Multi-detector computed tomography can accurately diagnose MSK comparable to IVU [8].

As another tool for diagnosis, endoscopy has been shown to properly establish a diagnosis of MSK by observing renal papillae for diffuse damage and contour rounding causing a billowy appearance leading to distal papillae blunting [12].

Concerning biological analyses, the metabolic and phosphorus-calcium workups were done in our patient with no significant abnormalities. It has been demonstrated that MSK is usually associated with increased levels of urinary calcium along with normal urinary potassium and bicarbonate levels in most patients. Urine pH, ammonia and titratable acid are increased in MSK patients [12].

Mc Phail *et al.* [4] conducted a study that aimed to evaluate clinical and metabolic features in 56 patients with MSK. They found that stones were composed of calcium oxalate monohydrate, calcium oxalate dihydrate, calcium phosphate apatite, and uric acid. Metabolic profile was obtained for 26 of 39 (67%) stone-forming patients demonstrating abnormalities in 22/26 (84.6%). These included hypercalciuria, 58% (15/26); low urine volume, 35% (9/26); hyperuricosuria, 27% (7/26); hypocitraturia, 19% (5/26); elevated urine sodium, 15% (4/26); and hyperoxaluria, 12% (3/26).

Faced with calcification and total obstruction of the left kidney, nephrectomy was performed in our patient. It has been suggested that if MSK is not diagnosed early leading to long term irreversible complications, nephrectomy may be required [11]. Cheungpasitporn *et al.* [13] while studying the outcomes of living kidney donors with medullary sponge kidney found that donors with MSK had lower kidney stone-related events after nephrectomy compared with nondonors with MSK. Meanwhile, the right kidney of our patient was already a site for stone formation. This could be explained by the fact that MSK is a congenital disease that may affect both kidneys [14].

We found various types and sizes of stones in the removed kidney of our patient. However, these stones have not been analyzed due to an insufficient technical platform. Meanwhile, we identified staghorn calculi. This may suggest infectious episodes that may have gone unnoticed. Gambaro *et al.* argued that infectious episodes may complicate the renal stones, leading occasionally to secondary struvite stones and/or pyelonephritis, which are probably the most common causes of end stage renal disease in MSK patients [15].

The main goal of treatment of MSK is the prevention of recurrent urolithiasis and inhibition of progression of nephrocalcinosis. In all patients, oral potassium citrate supplementation and high fluid intake are indicated [16] [17].

According to Fabris *et al.* [3], after studying a cohort of 61 MSK patients, made the following recommendations: oral potassium citrate (2 - 4 g/day, *i.e.* 10 - 20 mmol/day divided into two or three doses) in patients who have MSK and at

least one urine abnormality (hypercalciuria, hypocitraturia, hyperuricosuria and/or hyperoxaluria).

Start with 2 g/day and gradually step up the dose in patients who fail to achieve the target citraturia level of 450 mg/24h, adding 1 g at a time until this citraturia level is reached, provided the urinary pH in a 24-H collection is <7.5 (to prevent the risk of exacerbating any calcium phosphate lithogenesis).

Patients are followed up once a month until the treatment's dosage has been finely adjusted, then 6-monthly [3].

Other prophylactic measures, such as increasing water intake, reducing dietary sodium and proteins, increasing vegetable and fruit intake, are also generally recommended [3].

Aside from recurrent episodes of nephrolithiasis, MSK can be considered a benign condition. Typical complications of nephrolithiasis are observed in patients including UTIs, pyelonephritis, and end stage renal disease, especially if urinary tract obstruction is not addressed timely [5] [13].

In case of urinary tract obstruction, the use of minimally-invasive procedures may be useful. As a last resort, surgical intervention consisting of open stone extraction or nephrectomy may be an option for patients who experience both recurrent stone formation and severe symptoms. Though stone removal is necessary to relieve symptoms, the actual prognosis of the disease does not change [3] [5].

Compliance with all these measures and collegial follow-up in a nephrology unit would delay or avoid progression to renal failure.

#### **4. Conclusion**

Nephrocalcinosis and nephrolithiasis are complications of MSK. Its asymptomatic character can often lead to diagnostic delay. Any telltale signs such as hematuria, urinary tract infections, or renal colic should lead to proper imaging to exclude Cacchi Ricci disease. Dietary and hygiene measures may be sufficient to avoid nephrectomy and delay chronic kidney disease. For this purpose, it is recommended that MSK patients with at least one stone risk factor (hypercalciuria, hypocitraturia, hyperuricosuria, hyperoxaluria) are considered to be at a higher risk for renal stone complications and should prophylactically be treated with potassium citrate [18].

#### **Informed Consent**

Thorough consent was formally obtained from the patient to report on this case.

#### **Authors' Contributions**

All authors contributed to the realization of this manuscript

#### **Conflicts of Interest**

The authors declare no competing financial or personal interests.

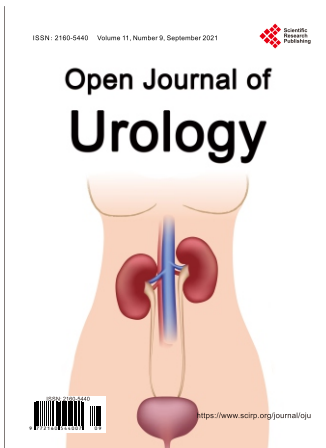
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