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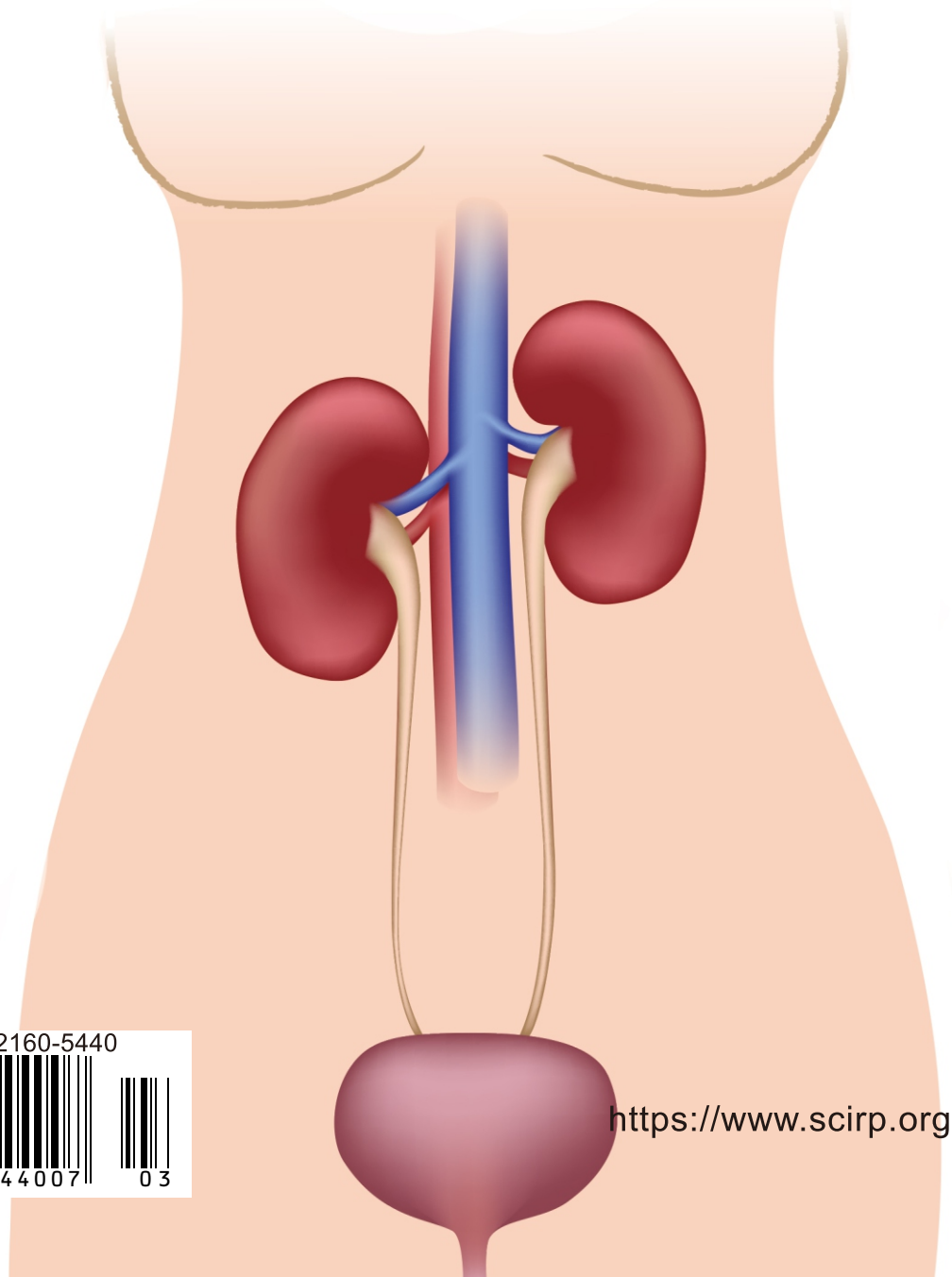

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Persistent Congenital Paraurethral Cyst, a Rare Cause of Dysuria in a Girl

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Abstract

Congenital paraurethral cyst rarely occurs. Its natural history is spontaneous regression, leading to conservative management adoption. We report an exceptional case of a 13-year-old female with a persistent congenital paraurethral cyst, leading to dysuria. We surgically excised the cyst, and the outcomes were unremarkable 20 months postoperatively.

Keywords

Congenital Paraurethral Cyst, Persistent, Dysuria, Girl, Surgical Excision

1. Introduction

Congenital paraurethral cyst (CPUC) is a rare condition, and its incidence is reported to range from one in 1038 to 7246 live female births [1]. It is due to ductal obstruction of paraurethral glands, Skene's glands [2]. The diagnosis is usually made in neonate, during the physical examination, with investigations needed only for atypical presentation to exclude differentials [3]. The natural history of CPUC is regression, as influence of maternal hormones decreases. Its persistence is exceptional [4] [5]. Management of CPUC during the neonatal period is conservative, when surgery (excision or drainage) is indicated for persistent or lately diagnosed CPUC [3]. We report a girl thirteen years old who presented dysuria due to a persistent CPUC.

2. Case Presentation

A thirteen-year-old girl was received in our department for dysuria and interlabial mass. The mother reported that soon after birth, a vulvar mass was identified in the patient. At the maternity, she was reassured that the mass would soon

regress, and no investigations were requested. However, the mass persisted, and since infancy, it progressively increased its size, but no medical nor traditional management was sook. During the past months, the patient experienced progressive dysuria, which worsened two weeks before the consultation in our department. Then parents consulted at a district medical center, where she was referred. The patient had no medical history; she had her menarche a year ago.

On physical examination, vitals were within normal ranges. An interlabial globular pink mass was identified. It was soft, not painful, and was located on the right side of the urethral meatus, distorted, and deviated on the left side. Its largest diameter was 20 mm (**Figure 1**). Assumed diagnoses were CPUC or prolapsed megaureter. An abdominopelvic ultrasound (US) was ordered and did not identify any urinary tract anomaly. A bacteriological examination of urines did not show any bacteria.

The patient was hospitalized at our department. Initial management included parenteral paracetamol (15 mg/kg/6hours) as analgesic, and placement of a Foley catheter to allow better emptying of the bladder. The same day, she underwent surgical excision of the cyst under general anesthesia, using cautery. The implantation of the cyst was sutured with 4/0 Vicryl using interrupted sutures (**Figure 2**). Postoperatively, intravenous paracetamol was continued, along with ibuprofen (10 mg/kg/8hours). The immediate postoperative course was uneventful, and she was discharged 24 hours later, with per oral paracetamol and ibuprofen for 7 days. Histology of the excised tissue revealed presence of a urothelial tissue with Malpighian metaplasia and unspecific sclero-inflammatory reaction. After 20 months of follow-up, no recurrence nor complication was noted, the physical examination was normal.

3. Discussion

Congenital paraurethral cyst is a rare congenital malformation of the urinary

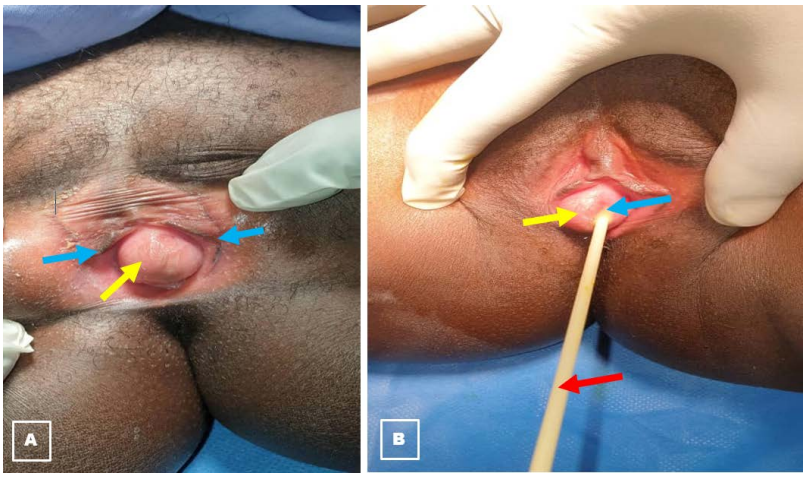

Figure 1. Physical examination. Before insertion of a Foley catheter (A), note the interlabial (yellow arrow) with retracted labia minora (blue arrows). After insertion of the catheter (B), the mass (yellow arrow) originates right to the urethral meatus (blue arrow).

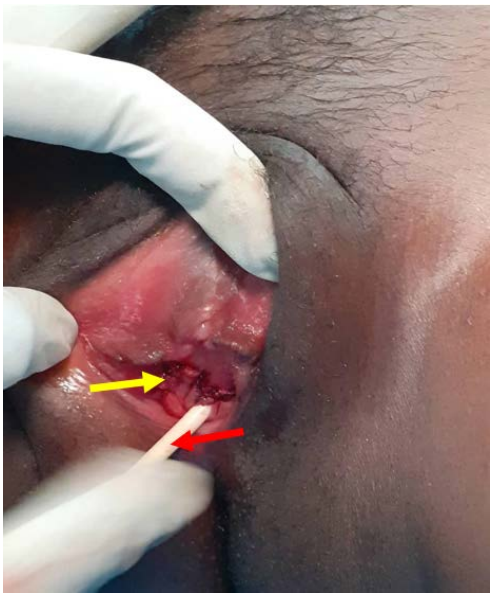

Figure 2. Intraoperative view. After surgical excision of the mass, no residual mass was identified (yellow arrow), as the Foley catheter (red arrow) remained inserted.

tract and an exceptional etiology of dysuria in girls [4]. It is secondary to obstruction of paraurethral glands, homologous to the male prostate, which drains into the female urethra through their ducts. Skene's glands are the largest two glands of 6 to 30 that secrete a mucoid material on sexual stimulation into the distal two-thirds of the female urethra [6]. Etiology of the ductal obstruction is still controversial. Advocated hypothesis include: influence of maternal estrogen, stenotic or inflammatory obstruction, and urothelial dislocation [5].

During the newborn examination, genitalia examination is mandatory. When properly performed, it allows the diagnosis of this congenital lesion which presents as an interlabial golden mass (due to its milky content), lateral to the urethral meatus, and non-painful [1]. The natural history of CPUC is regression as the child grows due to the reduction of maternal hormones impregnation, as suggested by authors supporting the hormonal hypothesis [7]. No additional investigations are necessary unless the patient presents atypically, such as in our case, where persistence up to 13 years old is exceptional. In such cases, differentials should formally be excluded before attempting any surgical option [1]. One of the possible differentials is prolapsed ectopic uretereterocele, which must be ruled out by urinary tract (UT) US [8]. In our case, no anomaly of the UT was detected by the US. Another possible differential is urethral prolapse, frequently encountered in African and African descent girls, as well as in our context [9]. In our patient, it was ruled out after clinical examination, as the mas was lateral to the urethral meatus. Other causes of interlabial mass must be ruled out: botryoidal rhabdomyosarcoma, imperforated hymen with subsequent hydrocolpos, hymenal cyst, Gartner duct cyst, and urethral polyp or diverticulum [1] [10].

Management of CPUC is controversial [7]. Considering its natural history and absence of symptoms, many authors favor conservative management with close

monitoring, especially in newborns. In a series of five patients, Japanese authors reported spontaneous regression from 76 to 304 days [1]. However, other authors praise operative management, arguing that interlabial mass in the neonate can be a stressful situation for parents, and its surgical removal guarantees the absence of recurrence [7]. Several operative options include needle aspiration, incisional drainage, marsupialization or surgical excision of the lesion [3] [11]. Optimal management of CPUP should consider both natural history and the possibility of persistence or complications, such as meatal obstruction, which occurred in our patient. Therefore, neonatal and asymptomatic CPUC should be managed conservatively, and persistent or symptomatic lesions should be operatively managed [1]. In our case, persistence until 13 years old and dysuria due to mass effect on the distal urethra and urethral meatus, we chose surgical excision. Outcome of CPUC is excellent, so that no recurrence was reported in the literature, neither after conservative nor surgical management [12].

4. Conclusion

Congenital paraurethral cyst is a rare condition. Usually asymptomatic and spontaneously resolving, it can be persistent and lead to dysuria, as shown in our patient. In such cases, surgical excision is the cornerstone of management, with excellent outcomes.

Consent for Publication

A written consent for publication was obtained from the patient's parents.

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Conflicts of Interest

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

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Distal Ureteric Dilatations Functioning as Urinary Reservoir in a Case of Ectopic Vesicae; A Case Report

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Abstract

Background: Bladder exstrophy is a rare congenital malformation of the genitourinary system, with an estimated incidence of approximately 1 per 50,000 live births. Clinically, patients do not have capacity to accumulate urine and urine continously leak. We present patient with partial storing capacity from the dialated distal ureters. A case of dialated distal ureters from an 8-year-old female patient with ectopic vesicae is described. The dialated ureters act as reservoir of urine where the patient is partially continent in the night time. These dialated ureters are the compensation for the literally absent bladder. During reconstruction, we observed that they can be used as an additional bladder volume reducing risk of reconstruction failure from tension

Keywords

Bladder Exstrophy, Ureteric Dilatations

1. Introduction

Ectopic Vesicae is also known as bladder exstrophy. Bladder exstrophy is a rare congenital malformation of the genitourinary system, with an estimated incidence of approximately 1 per 50,000 live births. The exstrophy-epispadias complex represents a severe midline abdominal birth defect that causes wide separation of the pubic symphysis, an abdominal wall defect and an anteriorly positioned open bladder and urethra [1] [2]. Many of the cases do not have any urine reservoir and continuously leak until some form of reconstructive surgery is done to make them store urine and have continence. Our case, however, had partial continence due to dilatation of the distal urethra functioning like urinary bladder. This situation has not been reported so fat to our knowledge and we be-

lieve that this report creates awareness of surgeons dealing with ectopic vesicae.

2. Case Presentation

History

An 8 year old girl presented with bladder extrophy otherwise called classic ectopic vesicae to our clinic in Ibex hospital located in Gondar town, Amhara region, Ethiopia.

She has this condition since birth and has never been surgically repaired. Parents report they have seen doctors several times earlier for possible reconstruction but she was turned down.

As in the case of all bladder extrophy patients, she had been wetting continuously and had never controlled her bladder function. Parents also reported that bladder was opened to the external environment over the abdomen since birth. From age 6 onwards parents were able to notice that she has some reduction in wetting in the night time but not in the day time. This was witnessed by the amount of diaper used; she uses more diapers in the night compared to that of the day. She quitted school due to the discomfort from the smell of urine and complaint of other children.

Physical Examination revealed that malnourished girl with mild physical growth retardation but no features of uremia. Vital signs were all with in normal range. The relevant finding is in the lower abdomen where we noticed complete bladder extrophy, separation of the clitoris and the labia majora. Both ureteric orifices were visible. No feature of bladder mucosal ulceration and changes in the mucosa was observed.

Complete blood count, BUN and creatinine were all with in normal limit. Pelvic radiography showed moderate separation of the symphysis pubis.

After optimization, surgery was performed to reconstruct the urinary bladder. The ureters are catheterized with nasogastric tube of 10 FR size. Upon ureteric catheterization, we found big reservoir of urine in both distal ureters (**Figure 1**). The ureteric catheters however could pass to the kidneys without difficulty. The distal dilatation of the ureters were about 25 - 30 ml. in volume each. It was also evident on Intravenous urography done before reconstruction (**Figure 2**). Patient was optimized to undergo major surgery including bladder closure and genital reconstruction. The procedure went smooth and there was no complication observed during postoperative period, urethral catheter was removed 7 days after the day of surgery

3. Discussion

This situation, dilatation of distal ureters, has been acting as urinary reservoir replacing some of the functions of the bladder to certain extent. The fact that she is less wetter in the night compared to the day can be explained by the fact that the ureteric reservoirs work better in the supine position due to gravity. Bladder closure was done successfully followed by trigone and genital reconstruction

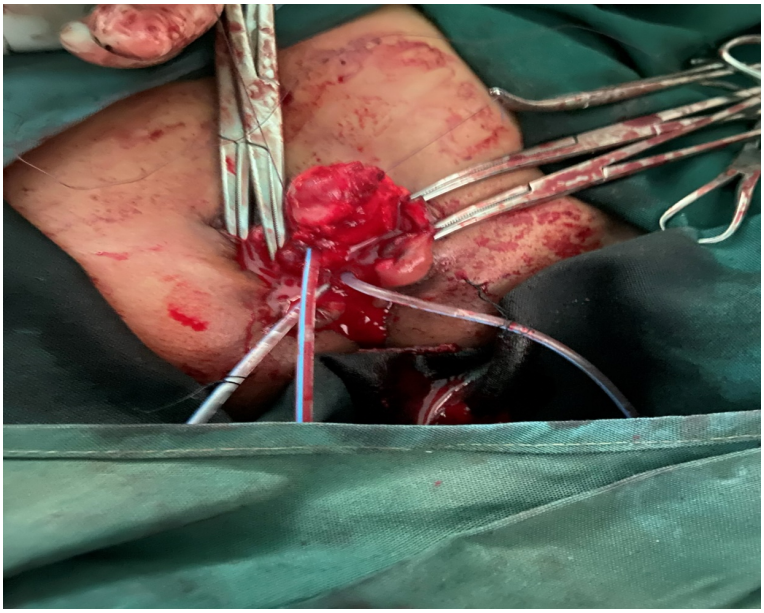

Figure 1. This figure shows introperative finding of the dilated distal ureters as indicated by insertion of 8 Fr infant feeding catheter.

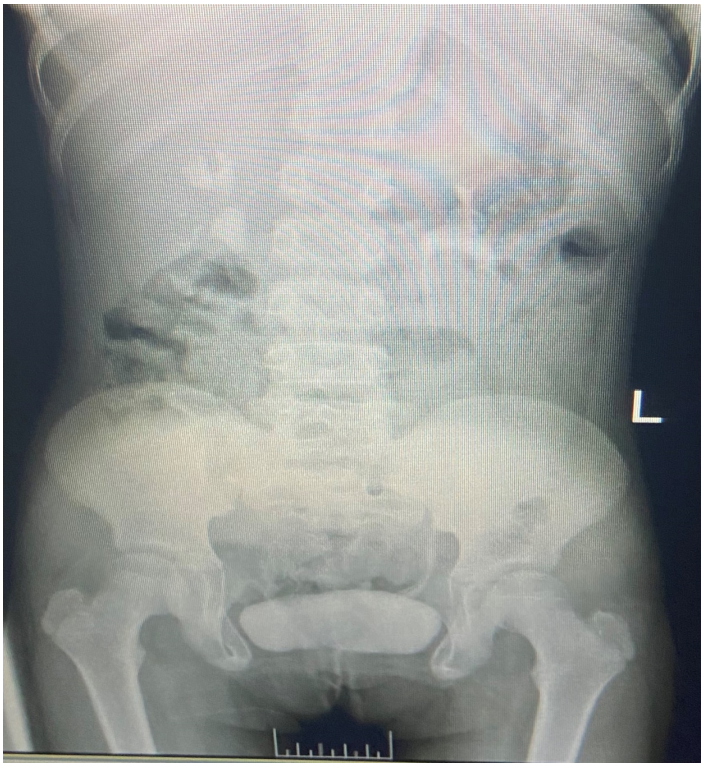

Figure 2. Intravenous Urography: showing the dialated distal ureters containing the Intra venous contrast. Picture was taken weeks before surgical closure of the exstrophied bladder.

within 3 months [3]. Reconstruction did not need a pelvic osteotomy or any complication seen on extra vesical flap development [4]. Follow up reveals she is better off in degree of dryness during day and night compared to those children with no dilatation of distal ureters.

4. Conclusion

This condition has never been reported so far to our knowledge. The dilated distal ureters compensate the reservoir function of the reconstructed bladder. They also prevent tension developing over the suture lines and avoid failure of bladder closure. Though they are now supporting the bladder function as an extra reservoir. In planning the reconstructive surgery, it may be useful to consider these volumes as an important augmentation and avoid extensive dissection to find increased bladder size. The complications that may arise in the future are not yet known and this patient needs close follow up and intervention in case this ureteric abnormality impedes urinary flow and causes upper tract deteriorations [5] [6].

Conflicts of Interest

The author declares no conflicts of interest regarding the publication of this paper.

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Urinary Sphincter Disorders of Neurological Origin: Prognostic Aspects in the Neurology Department of the Ignace Deen National Hospital

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Abstract

Introduction: Complications of Urinary sphincter disorders of neurological origin can be life threatening. The objective of this study was to describe the prognosis of urinary sphincter disorders during neurological conditions. **Methods:** This was a six-month analytical study conducted at the neurological unit of the Hôpital national Ignace Deen. Including patients with urinary sphincter disorders following a neurological condition; Chi-square, Fischer, and Student's t-tests were used for variables with a p value less than 0.10 and then included in a logistic model with a significance level set at 0.05 and a 95% confidence interval. **Results:** We collected 1081 patients among whom, 324 presented, that is to say a frequency of 30%, which concerned subjects aged 57.3 ± 16.4 years with a slight female predominance 50.3%. Urinary incontinence (80.6%) was associated with complications such as urinary tract infection with a high proportion of cerebral damage (92.3%). HIV infection (P = 0.015), bedsores (P = 0.049), and inhalation pneumonia (P = 0.001) were the main poor prognostic factors. **Conclusion:** Urinary sphincter disorders are elements of poor prognosis, both vital and functional, concerning elderly subjects with a predominance of urinary incontinence. HIV infection, bedsores, pneumopathy are poor prognostic factors.

Keywords

Urinary Sphincter Disorders, Incontinence, Retention, Neurology

1. Introduction

Neurological urinary sphincter disorders are defined as deficiencies in the vo-

luntary and/or involuntary motor control systems of the vesico-sphincter apparatus, either by a lack of activation and/or inhibition of the medullary, pontine and cortical centers [1].

The severity of Neurological urinary sphincter disorders is related to the risk of renal and bladder complications. These complications can be life threatening [2]. They are one of the deficiencies usually observed in spinal cord injury. Their complications have been for a long time a cause engaging the vital prognosis of the spinal man in the short term [3].

They represent elements of poor prognosis vital than functional, This is because these disorders are markers of severity of stroke [4].

The death rate in the initial phase of SUD is significantly higher in the incontinent patient, 52% [5].

Approximately 55.5% of cerebral palsy patients present at least one symptom of the lower urinary tract, of which urinary incontinence is the most frequent [6].

Neurogenic bladders are secondary to neurological diseases (spinal cord injury, myelitis etc...) [2].

It can be manifested by dysuria, detrusor hypocontractility and/or vesico-sphincter dyssynergy, accompanied by incomplete micturition with post-void residue, leading to irritative and infectious symptoms [7].

A urodynamic examination usually allows neuro-urology to specify the risk of deterioration of the upper urinary tract by detecting certain abnormalities at risk (bladder compliance disorder, bladder-sphincter dyssynergy), thus the management of these disorders requires an adapted framework, a multidisciplinary evaluation (urologist, neurologist, physical medicine and rehabilitation physician...) and specialized tools [2].

To date, despite the effectiveness of the treatment of these disorders, their evolution remains poorly known over time, so a longitudinal follow-up is essential for the implementation of therapeutic strategies [8].

The objective of this study was to evaluate the prognosis of bladder-sphincter disorders during neurological conditions.

2. Methods

We conducted an analytical, prospective study for a period of six (6) months from January 6 to June 6, 2020 in the neurology department of the Ignace Deen National Hospital in Conakry. In which we included, patients with urinary sphincter disorders following a neurological condition.

2.1. Clinical Data

The interrogation and clinical examination allowed us, to divide the patients into (02) two groups according to the type of Neurological urinary sphincter disorders presented.

Incontinence: it is an involuntary leakage of urine. This incontinence could

be:

- Stress urinary incontinence: involuntary leakage of urine during physical exertion, coughing and sneezing.
- Urgenturia: involuntary leakage of urine preceded by a sudden, compelling and irrepressible desire to urinate.

Retention: this is the inability to empty the bladder completely or partially.

2.2. Paraclinical Data

Biology workup: retroviral serology (VRS).

All our patients underwent at least one imaging procedure, either computed tomography (TDM) or magnetic resonance imaging (IRM), depending on the suspected etiology of Neurological urinary sphincter disorders, and radiography to look for signs of inhalation lung disease.

2.3. Etiological Diagnosis

Our etiologies were divided into three (3) groups according to the topography of the involvement, this allowed us to determine the predominance according to whether they are:

1) Cerebral;
2) Medullary;
3) Peripheral.

2.4. Therapeutic Data

Perineal rehabilitation:

Perineal rehabilitation or sphincter tone strengthening sessions were performed in all our patients.

- Patients are asked to close the sphincter muscles on two fingers of the examiner introduced into the perineum for one to two minutes.
- Is done by introducing a ball into the perineum that the patient must hold for one to two minutes.

2.5. Evolutionary Data

- Favourable: These are patients who have presented SUDs and who have improved without urinary catheter.
- Unfavorable: These are patients with persistent clinical signs of Neurological urinary sphincter disorders after rehabilitation with the occurrence of complications.

2.6. Duration of Hospitalization

This is the period between the patient's hospitalization and discharge from the hospital. We expressed it in days and calculated the average length of hospitalization and expressed the extremes.

Our collected data were entered using EPI data 3.1, Excel 2013 and analyzed

using SPSS software. We performed a descriptive statistics of the data set, qualitative variables were broken down into proportions expressed as a percentage and quantitative variables were expressed as mean plus or minus standard deviation and then recoded into two slices.

Chi-square, Fischer, and Student's t tests were used for variables with a p value less than 0.10 and then included in a multivariate logistic model with significance at 0.05. In this study, the following variables (pressure sores, HIV infection, and lung disease) were significant with $p < 0.05$ and a 95% confidence interval, allowing us to identify these predictors of poor prognosis.

3. Results

We collected 1081 patients, 324 of whom had Neurological urinary sphincter disorders, a frequency of 30% (Table 1).

Table 1. Distribution of patients by sociodemographic characteristics.

Socio-demographic characteristics	Workforce (N = 324)	Proportions (%)
Age (year)		
<15	6	1.8
>74	41	12.7
15-29	17	5.2
30-44	45	13.9
45 - 59	85	26.2
60 - 74	130	40.1
Medium	57.3 ± 16.4 ans	
Sex		
Male	161	49.7
Female	163	50.3
Sex-ratio		0.98
Provenance		
Urban	249	76.9
Semi-urban	61	18.8
Rural	14	4.3
Marital status		
Married	296	91.4
Single	11	3.4
Widowed	16	4.9
Divorced	1	0.3
Total	324	100.0

Urinary incontinence was predominant at 80.6% versus 19.4% for urinary retention (Figure 1).

Cerebral damage was the most common cause of death, accounting for 92.3% of all cases, with strokes being the most common (Figure 2).

HIV infection (P = 0.015), pressure ulcer (P = 0.049), and inhalation lung disease (P = 0.001) were the main poor prognostic factors (Table 2).

The average length of hospitalization was 11 days with extremes of 5 days and 17 (Table 3).

4. Discussion

Three hundred and twenty-four (324) patients had Neurological urinary sphincter disorders or (30%) thirty percent of cases.

This high frequency in our series could be explained by a high rate of patients

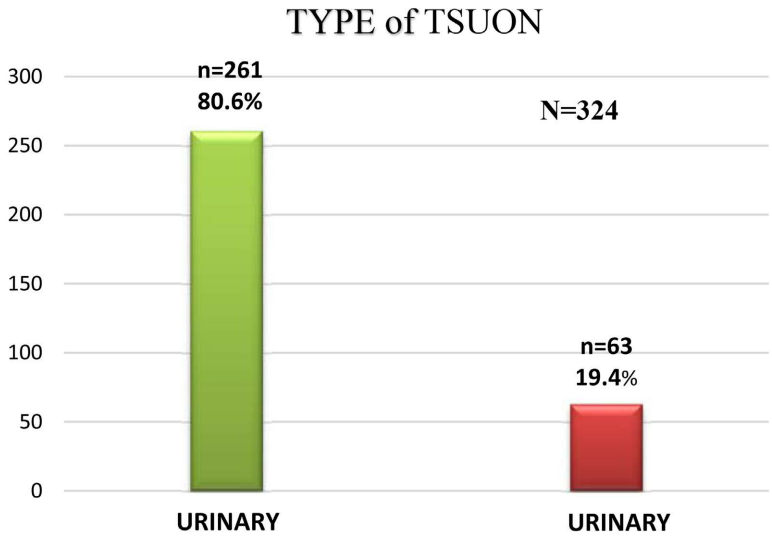

Figure 1. Distribution of patients according to clinical signs.

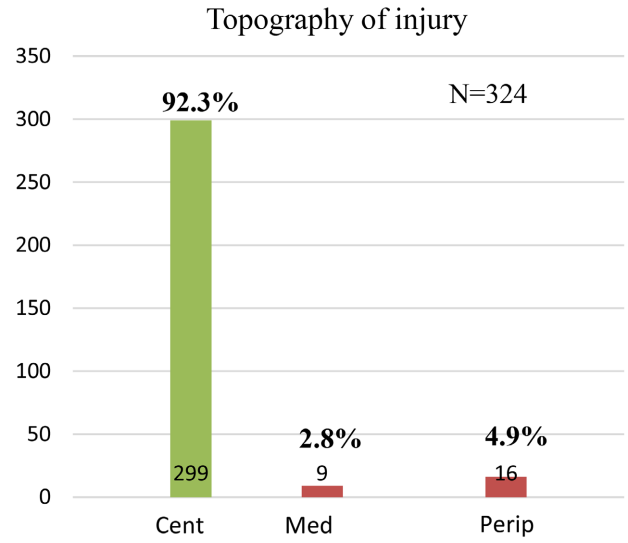

Figure 2. Distribution of patients according to the topography of the attack.

Table 2. Summary Chi-square test and the multivariate logistic regression model.

Unfavourable evolution	B	P-value	Odd-ratio	IC 95%
Recidivism				
No	−1.023	0.148	0.359	0.090 - 1.440
Yes	-	-	-	-
HIV infection				
No	−2.099	0.015	0.123	0.022 - 0.669
Yes	-	-	-	-
Respiratory disease				
No	−1.966	<0.001	0.140	0.050 - 0.394
Yes	-	-	-	-
Pressure sores				
No	−1.398	0.049	0.247	0.062 - 0.991
Yes	-	-	-	-

Table 3. Distribution of patients by length of hospitalization.

Length of hospitalization	Workforce (n = 324)	Proportions (%)
<10	139	42.9
10 - 20	154	47.5
>20	31	9.6
Medium	11.48 ± 6.76 days	

received for stroke. It confirms that found by Diagne N *et al.* [9] in Senegal in 2013, however contrasts with that of Phe V *et al.* [10] in France in 2013. These disorders affect elderly female subjects without significant predominance over the male sex. This result is lower than that found in France by Daviet J *et al.* [8] in 2004, who reported a higher age.

This disproportion could be explained by the coexistence of risk factors such as trauma in young subjects. However, it appears from our study that urinary incontinence was the most frequently encountered reason for consultation.

This is consistent with the literature in which urinary incontinence is predominant, especially during cerebral damage, although the mechanism of occurrence is poorly understood [11] [12]. This is in contrast to the study by Sakakibara R *et al.* [13], who reported a predominance of bladder emptying disorders. However, this urinary incontinence may frequently be present in the initial phase, but it is a medical priority only in rare cases, such as urinary incontinence by overflow during acute retention [11].

This result can be explained by an insufficiency of health structures in countries like ours and by the frequency of cerebral damage during Neurological urinary sphincter disorders.

Perineal rehabilitation and etiological treatment were performed in all our patients. For female urinary incontinence, perineal rehabilitation is the first-line treatment regardless of the type of incontinence [4].

The average length of hospitalization was 11 days with extremes of 5 days and 17 days; during this time, 82% of patients had their bladder catheter removed compared to 28% who had it removed.

This lack of bladder recovery could be explained by the occurrence of complications such as urinary tract infection, which were favored by the stasis favoring a predominant leukocyturia compared to post-traumatic hematuria, but also by the presence of urinary incontinence in the majority of cases [5].

However, the association of the variables (pressure ulcer, HIV infection, pneumonia) with Neurological urinary sphincter disorders was significantly unfavorable with p values < 0.05 (0.049; 0.015; 0.001) and a confidence interval of 95%, which allowed us to identify these predictive factors of poor prognosis.

La pose systématique d'une sonde urinaire sans l'évaluation initiale des fonctions vésico-sphinctériennes et la non-disponibilité de certains examens urodynamiques: flowmeter (allows to study the lower urinary tract in order to evaluate the quantity of urine excreted from the bladder and the time required for the emptying phase), cystometry (allows to record the bladder pressure at rest during the filling and then during the miction). These were our main limitations and difficulties.

5. Conclusion

The complications of Neurological urinary sphincter disorders can engage the vital prognosis. They concern elderly subjects with a slight female predominance, associated with urinary incontinence whose observed etiology was cerebral damage. The poor prognostic factors were HIV infection (P = 0.015), pressure ulcer (P = 0.049), and inhalation lung disease (P = 0.001).

Conflicts of Interest

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

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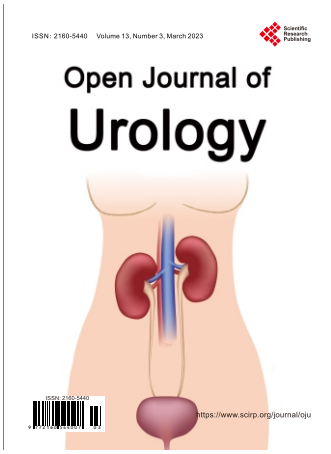

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