

## Case report

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# Emphysematous cystitis. A case report

*Cistitis enfisematosa. Reporte de un caso*

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### Abstract

The emphysematous cystitis refers to an uncommon entity generally secondary to low urinary tract infections producing gas around the bladder. It is associated with infections by *E. Coli*, *Enterobacter Aerogenes*, and *Klebsiella* as well as patient risk factors such as advanced age, diabetes and female gender. The diagnosis requires a timely management as well as directed antibiotic and associated comorbidities control. In the following case report a male patient is described in the hospital of San José de Bogotá in whom this pathology is diagnosed. In the following case report we are going to describe etiologies, diagnosis and therapy.

**Key words:** Cystitis, diabetes mellitus, type 2, urinary tract infections, anti-bacterial agents.

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### Resumen

La cistitis enfisematosa hace referencia a una entidad infrecuente, generalmente secundaria a infecciones de vías urinarias bajas, que por diferentes mecanismos producen gas alrededor de la vejiga. Esta patología se asocia a gérmenes (*E. Coli*, *Enterobacter Aerogenes*, y *Klebsiella*, entre otros), y factores de riesgo como género femenino, edad avanzada y diabetes. Este diagnóstico requiere un manejo oportuno, con antibioticoterapia dirigida y control de comorbilidades asociadas; a continuación, se reporta el caso de un paciente masculino de 81 años, en el hospital de San José de Bogotá, a quien se le diagnostica esta patología, con el respectivo aislamiento microbiológico, factores de riesgo, diagnóstico y manejo instaurado.

**Palabras clave:** cistitis, diabetes mellitus tipo 2, infecciones urinarias, antibacterianos.

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## Introduction

Emphysematous cystitis is characterized by the presence of gas between the wall and the lumen of the bladder, it is described as a complication of urinary tract infections.<sup>1</sup> It was initially described in animals by the year 1926<sup>1</sup>; later, by the year 1930, the presence of this pathology is evidenced in autopsy reports of female patients with associated diabetes.<sup>2</sup>

In emphysematous cystitis the most frequent etiologic agents are *E. Coli* and *Klebsiella Pneumoniae*; however, cases have been described with *Enterobacter aerogenes*, *Proteus mirabilis* and

*Streptococcus Spp*. The diagnosis is usually difficult since on many occasions its presentation is asymptomatic; however, clinically chills, flank pain, fever, pain on palpation and percussion at the costovertebral angle can occur; it should be highlighted the association of this pathology with glucosuria, complicated urinary tract infections and diabetes, the latter being the most frequent risk factor. In the results of the urinalysis, it is common to find the presence of hematuria, pyuria and bacteriuria, and in the urine culture the associated microorganism; given the importance of demonstrating the presence of gas at the level of the bladder, tomographic images play a vital role in the diagnosis of this entity.<sup>3</sup> For the treatment, the targeted antimicrobial approach and



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bladder catheterization are a fundamental part of the management. Due to the high risk of complications that can be generated by a late diagnosis, it is necessary to include this pathology within the complications of the urinary tract and take it into account as a differential diagnosis, although its incidence is low; hence the importance of the report of this case.

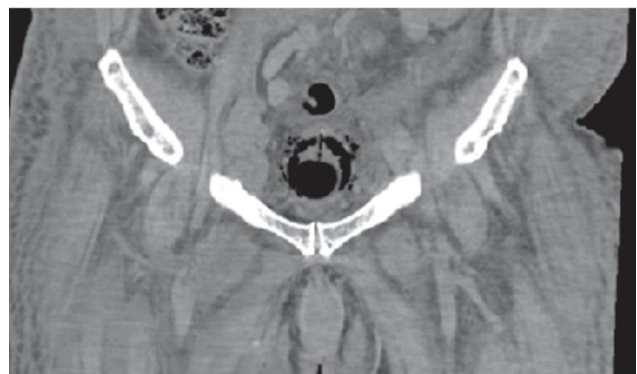
## Case presentation

An 81-year-old male patient, with a history of type II diabetes mellitus for 30 years, in outpatient management with glibenclamide, without allergies, or exposure to toxic substances or previous surgical history, who is admitted to the institution for resection of a mass in the right thigh and subsequent histopathological studies. The procedure was performed by plastic surgery, in which intraoperatively was evidenced involvement of deep planes (reaching the muscle) and lymphatic involvement until the base of the testicles, which is why they performed regional radical lymphadenectomy; given the large coverage defect, it was decided to perform a flap in the area. Postoperatively with no additional complications, except for an episode of urinary retention, and for this reason they considered leaving a urinary catheter indefinitely.

One week after the postoperative period, the patient presented clinical signs of a systemic inflammatory response, paraclinically with a leukocyte response and elevation of acute phase reactants, which is why in the search for an infectious focus they documented signs of local infection at the level of the flap, they started empirical antibiotic therapy with first generation cephalosporin (cefazolin), performed secretion cultures and awaited clinical evolution. After three days the clinical response was of torpid evolution; cultures of secretion of the flap and wound were received reporting polymicrobial flora with rescue of *Proteus mirabilis* of usual pattern, *E. coli* with a ESBL (extended-spectrum beta-lactamases) resistance pattern and *pseudomona aureginosa*. In context with the above, Plastic Surgery considered to carry out debridement and surgical lavage,

removing the flap. The patient was assessed by the Infectious Diseases service, where they considered that the isolated microorganisms are secondary to hospital stay, indicating antibiotic management with ertapenem and ciprofloxacin for 14 days.

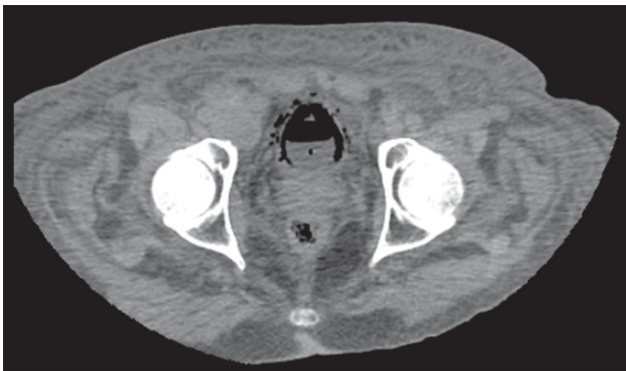
The pathology report was received, which evidenced a Merckel cell neuroendocrine tumor; then was assessed by Oncology, where they indicated to start outpatient chemotherapy, once the patient is discharged from the hospital. One week later the patient presented obvious hematuria and oliguria; the laboratory studies evidenced urinalysis with hematuria, proteinuria in the nephrotic range and active sediment, with renal function within normal parameters. Given the new clinical findings and the antecedent of diabetes, it was decided to request additional studies, and Ophthalmology ruled out diabetic retinopathy; in addition, the Nephrology team ruled out nephrotic syndrome, when evidencing urine of 0.3 g in 24 hours, hypoalbuminemia and normal lipid profile; likewise, globular morphology was found in 60% of eumorphic red blood cells, ruling out glomerulopathies. Investigating the origin of the hematuria, it was complemented with anatomical studies; indeed, starting with urinary tract ultrasound which reported good corticomedular differentiation with preserved kidney size; subsequently, an Uro-CT was requested to observe the lower urinary tract, showing thickening of the bladder wall, perivesical gas as seen in images 1, 2 and 3, and retroperitoneal lymph nodes conglomerates, findings compatible with emphysematous cystitis.



**Image 1.** Uro-CT in anterior sagittal section evidencing a vesicle reduced in size, with thickening of its walls and perivesical gas.



**Image 2.** Uro-CT in sagittal section evidencing particulate content within the bladder and perivesical gas.



**Image 3.** Uro-CT in axial section evidencing thickening of the bladder wall and perivesical gas.

Given the above finding, a urine culture and bacilloscopy were requested to determine the etiology, and additionally an evaluation by urology, ruling out an indication for surgical management. The serial urine bacilloscopies were negative and *E. coli* of ESBL pattern was isolated from urine culture; it was indicated to continue antibiotic management with ertapenem for 14 days and to continue with a urinary

catheter; later, macroscopic hematuria ceased and the urine output increased. In relation with the lymph node conglomerates described, it was considered a possible tumor compromise for subsequent follow-up by the service of oncology on outpatient basis. See the [paraclinical tests chart](#).

## Discussion

Grupper et al. (2007),<sup>4</sup> in a series of 53 cases between 1986 and 2006, describe regarding the emphysematous cystitis that the majority of patients were elderly women with diabetes *mellitus* (62.2%). The classic symptoms of urinary tract infection were present only in 53.3% of the cases. Abdominal tenderness and hematuria were observed in 65.6 and 82.3% of the cases, respectively. The plain abdominal radiography was very sensitive (97.4%), while the abdominal computed tomography was the most sensitive and specific diagnostic tool. A complicated course attributable to emphysematous cystitis was described in 18.8% of the cases. The exact mechanism that contributes to formation of gas in such cases is unknown. Several theories have been suggested, including the fermentation of glucose in urine, with an emphasis on the imbalance between gas formation and elimination.<sup>4</sup>

Even so, multiple risk factors are considered in the pathogenesis of emphysematous cystitis: 1) persistent hyperglycemia, which provides an excess of glucose to bacteria 2) fragility of the defense mechanism against infection due to the hyperglycemia 3) dysuria due to the diabetic nephropathy and 4) obstruction of the lower urinary tract, such as refractory and recurrent urinary tract infection, neurogenic bladder and benign prostatic hyperplasia.<sup>5</sup>

Emphysematous cystitis has a highly variable presentation and course, with considerable potential for complications. Wang (2010)<sup>6</sup> describes that emphysematous cystitis is an unusual infectious disease of the bladder and more than 50% of the cases of emphysematous cystitis have diabetes *mellitus*; Other risk factors include bladder outlet obstruction, neurogenic bladder, and patients receiving immunosuppressive therapy.

**Paraclinical tests chart**

Hemogram	Urinalysis	Metabolic	Globular morphology	Nitrogen compounds Electrolytes	Ultrasound	Uro-CT
Leukocytes 5,000	Density 1020	Albúmina 2,5 Albumin 2.5	Eumorphic red blood cells 60%	BUN 17	Both kidneys with good corticomedullar y differentiation	Kidneys of normal size, distended bladder with thickened walls, presence of gas inside, retroperitone al lymph nodes.
Neutrophils 2,900	pH 5	LDL Cholesterol 50	Dysmorphic red blood cells 40%	Creatinine 0.7	without focal lesions, right kidney 92*39*48 mm Parenchyma 11 mm and left kidney 90*44*42 mm.	
Hemoglobin 11.4	Leukocytes 500	HDL Cholesterol 43		Sodium 135	Parenchyma of 10 mm, no evidence of hydronephrosis or urolithiasis.	
Hematocrit 34.2	Nitrites positive	Triglycerides 115		Potassium 3.8		
Platelets 304,000	Sediment	Total Cholesterol 115		Phosphorus 2.2		
	Epithelial cells 0-2 x field			Calcium 6.9		
	Bacteria +++					
	Red blood cells 20-30 x field					
	24 HOUR PROTEINS 0.3 gr/24 hours					
	Volume 500 ml/24 hours					

The usual clinical symptoms are fever, chills, diarrhea, dysuria, and lower abdominal pain. In general, pyuria, hematuria, pneumaturia and leukocytosis are also observed. *E. coli* is the causative organism in the majority of cases, but *Klebsiella pneumoniae*, *Enterobacter aerogenes*, *Enterobacter cloacae*, *Citrobacter spp*, *Proteus mirabilis*, *Acinetobacter baumannii*, *Corynebacterium genitalium*, *Staphylococcus aureus*, *Aspergillus spp* and *Candida albicans* have also been reported.<sup>7</sup>

The prognosis for this condition is generally favorable; however, there have been reports of complications such as severe necrotizing cystitis requiring cystectomy and with a mortality rate of 20%.<sup>8</sup> Other complications due to late diagnosis can be bladder rupture, sepsis and acute abdomen, and

even ascent of the inflammatory process to the upper urinary tract causing emphysematous pyelonephritis, which increases morbidity and mortality.<sup>9</sup>

For the diagnosis of the EC and to rule out differential diagnoses such as vesicocolic fistula, intra-abdominal abscesses, adjacent neoplasms and emphysematous pyelonephritis, it becomes necessary to use imaging methods such as simple conventional abdominal radiography, abdominal ultrasound and computed tomography, where gas in the bladder, and thickening of the bladder walls can be evidenced.<sup>9</sup>

Given that in the majority of cases gram-negative microorganisms are the most commonly associated, the initial empirical antibiotic therapy should be

addressed to their coverage, using quinolones or cephalosporins, and once the result of the urine culture is obtained, the antibiotic therapy should be modified according to the antibiogram.<sup>10</sup>

In addition, the catheterization of the bladder as therapy is generally successful, with a complication rate lower than 20%<sup>8</sup>; as well as the management of comorbidities such as glycemic control,<sup>11</sup> this strategy reduces mortality without the need for surgical intervention and likewise preserves renal function. In case of complications, there are other rescue procedures such as percutaneous drainage, implantation of a ureteral stent, surgical debridement and cystectomy.<sup>12</sup>

The case reported here correlates with what is found in the literature from the point of view of microbiological isolation that corresponds to *Escherichia coli*, although ours is an *E. Coli* with an extended spectrum beta-lactamase (ESBL) resistance pattern, probably due to the complications noted, such as prior hospitalization for postoperative management of the skin flap and the use of antibiotics in this hospitalization. Also, it correlates with the literature in the presentation of the clinical picture, the history of diabetes as the most associated risk factor, and in the diagnostic approach, in which perivesical air could be documented by means of the Uro-CT, which is the test with higher sensitivity and specificity for the diagnosis of this pathology.

## Conflict of interest

The authors state they do not have any conflict of interest.

## Ethical responsibilities

### Protection of people and animals

The authors declare that no experiments were performed on human beings or animals for this research.

### Right to privacy and informed consent

The authors declare that patient data do not appear in this article.

## Author contributions

Rodolfo Torres Serrano: review, main researcher, bibliographic search.

Alejandro Dueñas: second author, structure of the article, writing.

Andrés Lamos, Cristian Rodríguez and Daniela Trujillo Hincapié: structure of the article, writing.

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