Emphysematous cystitis: mortality, risk factors, and pathogens of a rare disease

Andreas Schicho, Christian Stroszczynski, Philipp Wiggermann

Department of Radiology, University Hospital Regensburg, Regensburg, Germany

Abstract

Although high mortality rates have been reported for emphysematous pyelonephritis (EP), information on emphysematous cystitis (EC), which is less common, is sparse.

Here, we report one new case of severe EC and 136 cases of EC that occurred between 2007 and 2016, and review information about the characteristics, diagnosis, treatment and mortality of these patients, and the pathogens found in these patients.

The mean age of the 136 patients was 67.9±14.2 years. Concurrent emphysematous infections of other organs were found in 21 patients (15.4%), with emphysematous pyelonephritis being the most common of these infections. The primary pathogen identified was *Escherichia coli* (54.4%). Patients were mainly treated by conservative management that included antibiotics (n=105; 77.2%). Ten of the 136 patients with EC died, yielding a mortality rate of 7.4%. Despite the relatively low mortality rate of EC compared with that of EP, a high degree of suspicion must be maintained to facilitate successful and conservative management.

Introduction

Emphysematous urinary tract infections (UTIs) are potentially life threatening. Although mortality rates of approximately 24% have been reported for emphysematous pyelonephritis (EP),[1] information on emphysematous cystitis (EC), which is less common, is sparse. The first case of pneumaturia, which was most likely a symptom of EC, was reported in *Curiosities of Nature* by Raciborsky in 1671.[2] Here, we report one new case of severe EC and 136 cases of EC that occurred between 2007 and 2016, and review information about the characteristics, diagnosis, treatment and mortality of these patients, and the pathogens found in these patients.

Case Report

A 45-year old patient was admitted to our hospital with new diagnosed hepatic lesions of unknown dignity. Contrast-enhanced computed tomography (CT) of the abdomen revealed an advanced emphysematous cystitis involving the perirectal space (Figure 1A), and accompanying non-emphysematous, absceding prostatitis (Figure 1B). He had no fever, tachycardia or tachypnea, white blood cell count (WBC) was 13.9/nL, C-reactive protein (CRP) was 30.0 mg/L. He had a history of traumatic spinal cord injury L3 with intermittent self-catheterization, and arterial hypertension. The identified pathogen was *Klebsiella pneumoniae*. Despite the urgent need for i.v. antibiotics, the patient dismissed himself from inpatient care on oral Co-Amoxiclav. He did not show up in our hospital for a short-term follow-up appointment.

Discussion and Conclusions

Searching Medline for *emphysematous cystitis* and *cystitis emphysematosa* we found 140 articles; among the search results, 106 articles reported a total of 136 cases of EC. We recorded the following information for these cases, using Thomas *et al.*[3] as a reference: patient age, sex, and diabetes status; concurrent emphysematous infections involving other organs; urinary pathogens; white blood cell count (WBC); C-reactive protein (CRP) levels; treatment; imaging modality; and outcome. Incomplete data were allowed.

The mean age of the 136 patients was 67.9±14.2 years (ranging from 11 to 96; the mean age for men was 65.7 years; the mean age for women was 69.3 years). A total of 83 patients were female (61.0%) and 46 were male (33.8%; the sex was not available for 7 patients). A total of 86 patients had diabetes (63.2%); there were 58 diabetic women (69.9%) and 26 diabetic men (56.5%). In 37 cases, plain radiography was used as the diagnostic tool (27.2%); CT (computed tomography) was used in 98 cases (72.0%), ultrasound was used in 9 cases (0.7%), and cystoscopy was used in 5 cases (0.4%). The mean WBC was 13.6±10.7 10^6/L (n=78) and the mean CRP level was 48.0±77.6 mg/dL (n=32) on admission. Concurrent emphysematous infections of other organs were found in 21 patients (15.4%), with emphysematous pyelonephritis being the most common of these infections. The primary pathogen identified from urinary cultures was *Escherichia coli* (54.4%; Table 1). Patients were mainly treated by conservative management that included antibiotics (n=105; 77.2%) and bladder drainage, with additional glycemic control in diabetic patients. Surgical treatment was reported to be necessary in 11 patients (8.1%), and included cystectomy and nephrectomy in concurrent EP. Ten of the 136 patients with EC died,

Table 1. Patients' characteristics and pathogens identified in these patients by urinary culture.

Patients	No.	%
Men	46	33.8
Women	83	61
Diabetic	86	63.2
Women with diabetes mellitus	58	33.8
Men with diabetes mellitus	26	14.7
Overall mortality rate	10	7.4
Pathogen		
Escherichia coli	74	54.4
Klebsiella pneumoniae	16	11.8
Enterobacter spp.	4	2.9
Group D Streptococcus	2	1.5
Enterococcus spp.	2	1.5
Citrobacter spp.	2	1.5
Klebsiella aerogenes	1	0.7
Proteus mirabilis	1	0.7
Acinetobacter baumannii	1	0.7
Corynebacterium genitalium	1	0.7
Candida spp.	1	0.7
Aspergillus spp.	1	0.7

Correspondence: Andreas Schicho, Department of Radiology, University Hospital Regensburg, Franz-Josef-Strauss-Allee 11, D-93053 Regensburg, Germany.
Tel.: +49.941.944.7413 - Fax: +49.941.944.7469.
E-mail: andreas.schicho@ukr.de

Key words: Pathogens; emphysematous cystitis; mortality; urinary tract infection.

Conflict of interest: the authors declare no potential conflict of interest.

Received for publication: 11 December 2016.
Accepted for publication: 28 February 2017.

Clinics and Practice 2017; 7:930
doi:10.4081/cp.2017.930

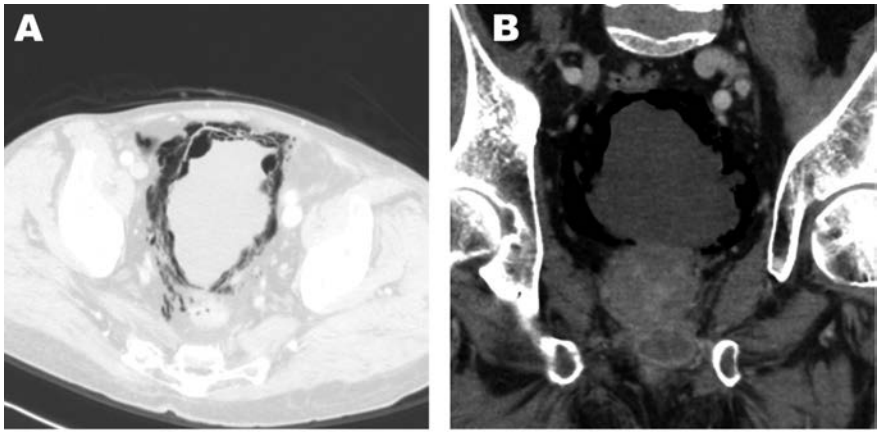

Figure 1. Contrast-enhanced computed tomography of the abdomen: A) emphyesematous cystitis involving the perirectal space; B) emphysematous cystitis with accompanying absceding prostatitis.

yielding a mortality rate of 7.4%.

EC is most often found in diabetic women who are in their 6th or 7th decade of life. Consistent with the female predominance of EC, we found a female-to-male ratio of 2.2:1 among the reported EC patients with diabetes; diabetic women showed an incidence rate of 91.5 per 1000 person-years for UTIs in general, whereas the rate was 28 in men.[4] Although most cases of EC are successfully treated by medical management that is built on an appropriate antibiotic regimen, more advanced cases of EC require surgical treatment. With a mortality rate of 7.4%, EC is markedly less life threatening than is EP. Although the pathogenesis of emphysematous UTIs, including EC, is not yet fully understood, gas-producing bacterial fermentation is required. Immunological host defense impairments, such as neurogenic bladder, immunosuppression, and diabetes, favor bacterial colonization of the urinary tract. Additionally, the increased availability of fermentation substrate in diabetic urine makes diabetic patients especially prone to emphysematous UTIs.

Despite the relatively low mortality rate of EC compared with that of EP, a high degree of suspicion must be maintained to facilitate successful and conservative management.

References

1. Falagas ME, Alexiou VG, Giannopoulou KP, Siempos II. Risk factors for mortality in patients with emphysematous pyelonephritis: a meta-analysis. J Urol 2007;178:880-5; quiz 1129.
2. Taussig AE. Pneumaturia, with report of a case. Boston Med Surg J 1907;156:769-74.
3. Thomas AA, Lane BR, Thomas AZ, et al. Emphysematous cystitis: a review of 135 cases. BJU Int 2007;100:17-20.
4. Hammar N, Farahmand B, Gran M, et al. Incidence of urinary tract infection in patients with type 2 diabetes. Experience from adverse event reporting in clinical trials. Pharmacoepidemiol Drug Saf 2010;19: 1287-92.