Abstract. Emphysematous cystitis (EC) is a very rare urinary tract infectious disease that can be fatal if not treated. In general, it frequently occurs in diabetic women and is thought to be associated with gas-producing bacteria. Type 2 diabetes mellitus, immunosuppression, drugs (mostly steroids), neurogenic bladder and instrumentation are the major risk factors of this type of infection.

We present a case of emphysematous cystitis in a 53-year-old male, in which the patient does not have any classical risk factors associated with EC other than alcohol consumption. To the best of our knowledge, the only case in the literature where this type of infection develops without a classical risk factor and negative urine culture. It is also one of the rare EC cases that may be associated with alcohol use.

Keywords: emphysematous cystitis, alcohol, infection, infectious disease, urinary tract.

Conflict of interest statement: all the authors declared no competing interests.

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Introduction. Emphysematous cystitis (EC) is a rare urinary tract infection that typically presents with an air-fluid level in bladder resulting from air accumulation in the bladder wall and lumen [1]. This type of infection is most common among over 65 age in the geriatric population although it can affect people all of the ages. Type 2 diabetes mellitus, immunosuppression, drugs (mostly steroids), neurogenic bladder and instrumentation are the major risk factors of this type of infection [2]. The clinical presentation varies from being asymptomatic to having painful urination, dysuria, hematuria, fever, abdominal pain, urosepsis and, unfortunately, it can be fatal if not treated. The most common isolated pathogens are gas-forming bacteria such as Escherichia coli, Enterobacter aerogenes, Proteus mirabilis, Klebsiella pneumoniae, Streptococci, and Staphylococcus aureus [3, 4]. The first imaging modality for EC diagnosis is urinary system ultrasonography (USG). As a diagnostic tool, a computed tomography (CT) of the abdomen is superior to plain USG and urinary system graphy because it clarifies the extent and location of the gas collection and diagnoses other pathologies that may coexist [4, 5].

The purpose of the work was to report a rare medical case to increase clinical awareness.

Methods. This article is a case report and the data used are reviewed retrospectively. In our case, the patient was 53 years old male. He was a chronic alcohol user and his only comorbidity was hypertension. The patient was admitted to our polyclinic with painful urination with the sudden occurrence and no other symptoms. The patient had no pathological sign on physical examination. The laboratory tests: his serum biochemical parameters were normal, the Prostate-specific antigen level was 0,78 and urinalysis was positive for red blood cells and nitrite. Urine culture was negative for bacteria and fungi. Post voiding residual urine and peak flow were 58 and 24 respectively. Urinary system USG: there was a clear image of intramural and intraluminal gas (Fig.1).

The patient had no urological operation for any symptom and had no risk factors for this type of infection. His previous urine cultures were negative twice before being admitted to our hospital.

An abdominopelvic computed tomography was performed in order not to miss any coexisting pathology as the patient had no risk factors for this type of infection. Abdominopelvic computed tomography reported as the patient had only EC, and no other abdominal pathology such as bowel perforation, enterovesical fistulas, diverticulosis, Crohn disease or rectosigmoid colonic carcinoma (Fig. 2).
The patient was consulted on infectious diseases and immunology departments. The suggestion was not to use antibiotics as urine culture was negative. The patient was treated symptomatically, prescribed an antimuscarinic agent and 550 mg oral naproxen sodium once per day. A single dose of 40 mg intramuscular prednisolone injection was used to cease the progression of inflammation. We advised the patient not to take alcohol during the rest of the treatment.

Results. After 1 week of medical treatment, the patient came to our polyclinic for control. His lower urinary tract symptoms have resolved. He was appointed for another control for radiological evaluation.

After 2 months of medical treatment, the patient came to our polyclinic for his second control. We performed urinary system ultrasonography and found that the patient had a complete recovery of gas accumulation as an air-fluid level in the bladder lumen and wall were absent. The only remaining radiological pathology was minimal bladder wall thickening (Fig. 3).

Discussion. EC is an uncommon urinary tract infection that was first reported in a human body in 1961 [6] and characterized as the presence of gas in the bladder wall or bladder lumen. EC is common in especially >65 age population, other known risk factors are female gender and type 2 diabetes mellitus in patients [5]. Most of the time, gas-forming bacteria like E. coli or K. pneumonia are isolated in the urine cultures. According to Yoshimatsu et al; among their patients with EC, 52.2 % of the patients were male with the average age of 76.3 years [7]. Among them, there were 88.2 % of patients with positive urine cultures (in 47.8 % of samples E. coli was isolated) [7]. 87 % of these patients had type 2 diabetes mellitus and other urological disorders [7].

Rarely EC can occur after prostate biopsy, renal transplant, hysterectomy, colectomy, hemodialysis, rectum resection, chemotherapy [8]. EC symptoms and signs are similar to classical cystitis without emphysema which includes irritative voiding symptoms, abdominal pain, hematuria, fever, nausea, vomiting, pneumaturia, painful urination, urinary retention, and urosepsis [4, 8].

In our case, the patient was a non-diabetic middle-aged man with none of the risk factors mentioned above and had overall good health status. We could not clearly identify the etiology of the infection in this patient because he had no gas-producing bacteria in the urine culture and there were no known risk factors but alcohol consumption.

A single dose of intramuscular prednol, followed by a daily NSAID tablet and antimuscarinic has resolved his symptoms in one week, and all radiological evidence was absent in two months. Since the only etiological factor of this patient is alcohol consumption, this suggests that removal of this single risk factor might have improved the prognosis.

To the best of our knowledge, the only case in the literature where this type of infection develops without a classical risk factor and negative urine culture is this one [9]. It is also one of the rare EC cases that may be associated with alcohol use. Alcohol metabolism is known to modulate the cellular response to LPS in the liver and independently generate endogenous inflammatory inducers. Thus, both alcohol and LPS act simultaneously to influence the inflammatory response in the liver and other organs, including bladder [10, 11]. Aasems Jacob et al. and M. Al-Assiri et al. identified the alcohol-related EC in a patient with pancreatitis. In their study, the simultaneous effect of alcohol on multiple organs was observed [12, 13].

Conclusions. We suggest that EC can occur in the absence of gas-producing bacteria, without any classical risk factors, and may initially manifest itself as mild lower urinary tract symptoms such as painful urination in our case.
On clinical approach, even if there are no other known risk factors for EC in patients with chronic alcohol consumption and suddenly occurring irritative lower urinary tract symptoms, EC is one of the most important diseases that should be kept in mind as it can be fatal if not treated early and properly.

**Conflicts of interest.** The authors declare no conflict of interests.

**References:**


**Contribution:**

**A. Sancı:** concept, design, writing manuscript;

**E. Aydoğ:** data collection and analysis;

**M. C. Karaburun:** literature search;

**E. Süer:** supervising, analysis and interpretation.