A Case of Emphysematous Cystitis with Bone Metastasis Infiltrating from Hepatocellular Carcinoma: Case Report and Literature Review of the Disease

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Abstract

Emphysematous cystitis (EC) is a rare infection of the bladder caused by gas-forming bacteria. EC was first reported in 1671 and was described as ‘presenting with pneumaturia’ infectious emphysema of the bladder was first reported at autopsy in the late 1800s. Since then, many papers have reported on infectious emphysema—mainly in the urological field; but existence of infectious emphysema of the bladder has not been reported in the internal medicine and nephrology fields, and it seems that it has not been recognized. We witnessed a case of EC that was accidentally discovered, and we decided to report upon it, adding literature considerations. In addition, based on a literature search, many cases of EC develop with insufficient control of blood glucose as part of diabetes; furthermore, most cases of EC are lighter, but others may become severe. The progression of EC in this case is another example that illustrates the importance of managing diabetes to prevent the onset of infectious diseases.

Keywords: Emphysema cystitis; Pneumaturia; Case report; Literature review of disease

Introduction

Emphysematous cystitis (EC) is a very rare urinary tract infection (UTI) caused by gas-forming microbes. The presence of air within the urinary tract was first reported in 1671 in the case of a man who presented with pneumaturia. Infectious emphysema of the bladder was first reported from an autopsy performed by Eisenlohr in the late 1800s; it was later defined as “cystitis emphysematosa” by Bailey in 1961 [1-4].

Case reports of EC have been accumulating in recent years, but there are few reports describing onset and causes of onset. In each case, urological conditions such as urinary retention and neuropathic bladder are present, but glycemic control has not been reported; nonetheless, many consider it to be involved.

Here, we decided to conduct a review of case reports from the last 10 years (since 2008) to understand the epidemiology and pathology of EC.

Case report

A 67-year-old male, who was HCV (Hepatitis C virus infection) negative and HBV (Hepatitis B virus infection) negative presented with hepatocellular carcinoma infiltrating to the thoracic spinal cord (T11, T12) (no detailed description here). He also had type 2 diabetes mellitus and a neurogenic bladder, and had recently fallen down at home and broken a bone in his left ankle. He was admitted to the hospital to receive analgesic therapy for osteocopic pain caused by bone metastasis and for rehabilitation. One month after admission, he fell down in the hospital and suffered a fracture of the lumbar spine, and could no longer walk on his own.

He suddenly developed abdominal distension, nausea, and vomiting three months after admission. The results of urine testing showed a urinary protein concentration of 100 mg/dl and a urine occult blood reaction of 3+. Microscopic examination of urinary sediment revealed 20-29 red blood cells per high power field and >100 white blood cells per high power field, but the peripheral blood test showed normal renal function (serum creatinine concentration of 0.50 mg/dl, estimated Glomerular Filtration Rate (eGFR) of 124 ml/min, and serum urea nitrogen of 27.5 mg/dl). The patient had developed a urinary tract infection caused by Klebsiella pneumoniae.

An abdominal computed tomographic (CT) scan (Figures 1 and 2) revealed a characteristic accumulation of air in the wall and lumen of the urinary bladder. The patient was diagnosed with emphysematous cystitis, which was improved by urinary drainage, intravenous fluid transfusion (1 L/day), and intravenous antibiotic therapy (ceftriaxone, 1 g/day) for 6 days. CT scan was a sensitive method for detecting early signs and confirming the diagnosis.

We successfully treated this urinary infection, but the patient died from underlying disease six months after these events.
Figure 1: Computed tomography of a patient with emphysematous cystitis.

Figure 2: CT image of a patient with emphysema cystitis.

Review of EC literature

We searched PubMed for the term “emphysema cystitis”. The specific search criteria used on February 8, 2019 were “emphysema cystitis” [Text Word], “English” [Language], and “2008” [Data-Publication] (Figure 3). Of the retrieved papers, case reports were requested and then carefully screened based on the “title & abstract” and the “full text”, after which 39 papers remained. From these 39 papers, we created an evidence table and investigated the characteristics of emphysema cystitis. According to our survey, the percentage of countries with reported cases was as high as 46.2% (18 reports) in the West, 38% (15 reports) in Asia including Japan, and 7.7% (3 reports) in Africa and the Middle East (Table 1). The underlying disease was diabetes in 52.8% of cases (20 of 36 cases) and chronic kidney disease (CKD) including end-stage renal disease in 13.2% of cases (5 of 36 cases). Among the diabetic cases, 34% (13 of 36 cases) had an unknown glycemic control status, but 23% (9 of 36 cases) had insufficient glycemic control. Symptoms at onset (Table 2) included abdominal pain (44.7% or 17 cases), followed by fever (42.1% or 16 cases), and impaired consciousness (18.4% or 7 cases) (Table 2). As for a diagnostic method, 97% (37 cases) were easily diagnosed by CT. The most common causative bacteria were E. coli, which was found in 73% or 28 cases. 42% of cases (16 cases) did not cause complications and 78.9% (30 cases) improved, indicating a good prognosis, but 2.6% (1 case) developed sepsis, indicating that some cases were difficult to treat. In addition, death was the outcome for 13.2% (5 of 36 cases) (Table 1) [5-41].

Table 1: Clinical characteristics of emphysema cystitis cases.

<table>
<thead>
<tr>
<th>Age</th>
<th>72.0 ± 14.7</th>
<th>Age</th>
<th>15 cases</th>
</tr>
</thead>
<tbody>
<tr>
<td>Sex</td>
<td>Female</td>
<td>6 cases</td>
<td></td>
</tr>
<tr>
<td></td>
<td>Male</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Nations</td>
<td>Western country</td>
<td>18 cases</td>
<td></td>
</tr>
<tr>
<td></td>
<td>Asia</td>
<td>15 cases</td>
<td></td>
</tr>
<tr>
<td></td>
<td>Africa, Middle eastern countries</td>
<td>3 cases</td>
<td></td>
</tr>
<tr>
<td>Underlying diseases</td>
<td>DM</td>
<td>20 cases</td>
<td></td>
</tr>
<tr>
<td></td>
<td>Type2 DM</td>
<td>9 cases</td>
<td></td>
</tr>
<tr>
<td></td>
<td>Type1 DM</td>
<td>3 cases</td>
<td></td>
</tr>
<tr>
<td></td>
<td>Unknown DM</td>
<td>3 cases</td>
<td></td>
</tr>
<tr>
<td></td>
<td>CKD</td>
<td>5 cases</td>
<td></td>
</tr>
<tr>
<td>Blood glucose control status</td>
<td>Well controlled</td>
<td>1 cases</td>
<td></td>
</tr>
<tr>
<td></td>
<td>Poor controlled</td>
<td>9 cases</td>
<td></td>
</tr>
<tr>
<td></td>
<td>Unknown controlled</td>
<td>13 cases</td>
<td></td>
</tr>
<tr>
<td>Symtoms</td>
<td>Abdominal pain</td>
<td>17 cases</td>
<td></td>
</tr>
<tr>
<td></td>
<td>Fever</td>
<td>16 cases</td>
<td></td>
</tr>
<tr>
<td></td>
<td>Disturbance of conscious</td>
<td>7 cases</td>
<td></td>
</tr>
<tr>
<td></td>
<td>Dysuria</td>
<td>5 cases</td>
<td></td>
</tr>
</tbody>
</table>

Figure 3: Result of literature search for ‘Emphysema Cystitis’.
A previous literature review by Grupper and colleagues found that 62.2% of patients were elderly diabetic women and that classic symptoms of UTI were present in only 53.3% of cases. Abdominal tenderness and hematuria were noted in 65.6% and 82.3% of cases, respectively. Plain abdominal x-ray was highly sensitive (97.4%), while abdominal CT was the most sensitive and specific diagnostic tool [42].

The exact mechanism of gas formation in emphysematous infections is unclear. It is presumed to be due to the presence of aerobic gas-forming organisms such as enterobacteraeae that rapidly ferment glucose and produce carbon dioxide. In nondiabetic patients, it has been proposed that urinary lactose or tissue proteins may serve as substrates for the gas formation [43,44].

The prognosis for EC patients is favorable if the disease is promptly diagnosed and treated. However, the clinical course can be severe, progressing to rupture of the urinary bladder and emphysematous pyelonephritis that may require surgical procedures such as nephrostomy or nephrectomy.

We were able to find two review papers on EC. One reported on 23 cases of emphysema cystitis in a single institution. According to Yoshimatsu, of the 23 cases, 52.2% involved male patients, with an average age of 76.3 years. Co-existing diseases included: neuropsychiatric condition 69.6%, malignant tumor 65.2%, diabetes mellitus 43.5%, and urologic disorder 43.5%. The initial systematic symptoms were: hematuria 52.2%, urinary retention 47.3%, and fever 34.8%. In addition, 28.6% were blood culture positive cases and 88.2% were urine culture positive cases. Among these culture positive cases, 47.8% were positive for E. coli and 11.6% for Klebsiella pneumoniae. In treatment descriptions, antibiotics and urinary catheters were included in 60.9% of cases, and only antibiotics in 17.4%. In addition, death was observed in 17.4% of the cases. The second review paper was the only one to address emphysema cystitis in Japan, to the extent we have examined it here. According to Amano, PubMed and Ichuushi searches for articles submitted between 2006 and 2013 identified 72 cases of emphysematous cystitis published in the foreign literature and 30 cases published in the Japanese literature. The average age was similar to what we found: 68 years old abroad and 72 years old in Japan. The proportion of males was 30% in Japan and 31% overseas. The rate of coexistence with diabetes was almost the same 66% overseas and 70% in Japan. The percentage of cases resulting in death was slightly lower in Japan: 12% overseas and 3% in Japan. In addition, it was reported that the proportion of cases requiring surgical intervention was relatively high in Japan: 5% overseas versus 7% in Japan. As for the identification of pathogens, the percentages were E. coli 64% and K. pneumoniae 20% in Japan, compared to E. coli 56% and K. pneumoniae 10% overseas [45,46].

As described above, epidemiological considerations were very similar to our review. In addition, we could not identify the relationship between the malignant tumor and emphysema cystitis found in this case within the scope of the search.

### Table 2: On the causative bacteria of Emphysema cystitis.

<table>
<thead>
<tr>
<th>Causative bacteria</th>
<th>Cases</th>
</tr>
</thead>
<tbody>
<tr>
<td><em>E. coli</em></td>
<td>28 cases</td>
</tr>
<tr>
<td><em>Klebsiella pneumoniae</em></td>
<td>4 cases</td>
</tr>
<tr>
<td>Fungus</td>
<td>2 cases</td>
</tr>
<tr>
<td><em>Enterococcus cloacae</em></td>
<td>1 case</td>
</tr>
<tr>
<td><em>Enterococcus aerogenes</em></td>
<td>1 case</td>
</tr>
<tr>
<td>Group B streptococcus</td>
<td>1 case</td>
</tr>
<tr>
<td>unknown</td>
<td>1 case</td>
</tr>
</tbody>
</table>

*DM: diabetes mellitus, CKD: chronic kidney disease

### Methods of diagnosis ‘emphysema cystitis’

<table>
<thead>
<tr>
<th>Methods of diagnosis</th>
<th>Cases</th>
</tr>
</thead>
<tbody>
<tr>
<td>CT</td>
<td>37</td>
</tr>
<tr>
<td>XP</td>
<td>6</td>
</tr>
<tr>
<td>US</td>
<td>1</td>
</tr>
</tbody>
</table>

*CT: Computed Tropography, XP: X-ray Photograph, US: Ultrasonography

### Discussion

EC is a relatively rare condition, which is characterized by gas collection within the bladder wall. EC was first identified in postmortem examination in 1888. The collected gas is believed to consist of carbon dioxide produced by the fermentation of glucose or albumin by micro-organisms infecting the bladder. The common causative organisms are Escherichia coli, Enterobacteraeogenes, and *Klebsiella pneumonia*. The clinical presentation is nonspecific and ranges widely from asymptomatic urinary tract infection to urosepsis and septic shock at the onset of disease. The majority of patients had a history of diabetes or neurogenic bladder.

Discussion

EC is a relatively rare condition, which is characterized by gas collection within the bladder wall. EC was first identified in post-mortem examination in 1888. The collected gas is believed to consist of carbon dioxide produced by the fermentation of glucose or albumin by micro-organisms infecting the bladder. The common causative organisms are Escherichia coli, Enterobacteraeogenes, and *Klebsiella pneumonia*. The clinical presentation is nonspecific and ranges widely from asymptomatic urinary tract infection to urosepsis and septic shock at the onset of disease. The majority of patients had a history of diabetes or neurogenic bladder.
Conclusion

EC is a relatively rare form of complicated UTI characterized by the presence of gas within the bladder wall and lumen. The predisposing risk factors and conditions include older age, female gender, and severe DM.

Clinicians should be aware of the clinical presentations of EC. Based on our findings, suspicion of emphysematous cystitis should be aroused by abdominal pain in diabetic patients, with or without a clinical presentation suggestive of UTI, especially when hematuria is present. In such cases, we suggest conducting a plain abdominal x-ray for screening, followed by an abdominal CT when urinary bladder gas is present.

Acknowledgement

The authors state that they have no Conflict of Interest (COI).

References


