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Case Report

A hyperechoic bladder-ring appearance as pathognomonic finding for emphysematous cystitis – A case report and literature review ☆,☆☆

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ABSTRACT

An emphysematous cystitis s a rare urinary tract infectious disease and fatal unless any treatments. The case was a 97-year-old female presented with knee pain after falling with co-existence of macroscopic hematuria and pyuria. The ultrasonography examined on the same day showed a hyperechoic bladder-ring appearance bordering bladder wall which was re-was re-confirmed by computed tomography by air bubble collection on the circumferential bladder wall. We conclude and emphasize as the leaning point that emphysematous cystitis is raised as a differential diagnosis when ultrasonography showed the pathognomonic as hyperechoic bladder-ring appearance bordering bladder wall.

Introduction

Emphysematous cystitis (EC) is a relatively rare and potentially life-life-threatening entity characterized by the collection of gas in the bladder. Its mortality rate is reported 10.4% [1]. It is commonly seen in older female with Diabetes Mellites (DM) seen in 50%-70% patients with EC [2]. This is an infectious disease caused by gas-producing microorganisms such as Escherichia coli, Klebsiella pneumoniae, Enterobacter aerogenes, Clostridium perfringens under predisposing factors of immunosuppression [3]. In this case report, we presented non-diabetic older female with a radiological finding in ultrasonography to be consider as a pathognomonic sign of EC.

Case report

A 97-year-old female presented with knee pain after falling. She admitted with a diagnosis of osteoarthritis of the knee. At presentation, her serum laboratory data of creatinine level was as high as 1.7 mg/dL and treated with infusion with rhabdomyolysis. On fourth day, her temperature was 37.1 °C. and

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Fig. 1 – The ultrasonography of the presented case showed the pathognomonic hyperechoic bladder-ring appearance around the bladder wall.

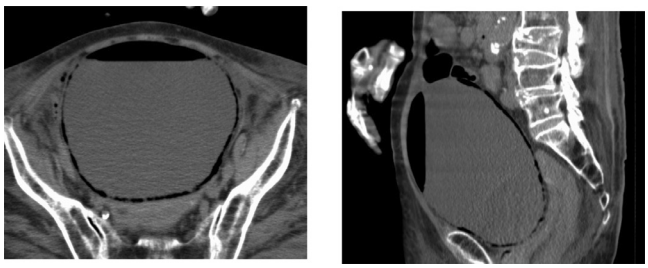

Fig. 2 – The CT scan of the presented case: the coronal section(left) and sagittal section (right) showed air bubble collection in the bladder wall.

serum creatinine level increased to 3.5 mg/dL 2 days later. The results of laboratory test at admission and on the 2nd day (shown in parentheses) were; white blood cell (WBC) count was 8,550 (43,790) / μL, hemoglobin was 11.9 (9.5) gram / dL, platelet count was 16.8 $\times$ 104 / μL, C-reactive protein on the 2nd day was 22.8 mg / dL, Hb A1c was 5.5 %, Urinalysis: WBC $>$ 100 / high power field (HPF), and red blood cell was 50-99 / HPF. Her urine analysis showed co-existence of macroscopic hematuria and pyuria that were consistent with findings of white blood cell and red blood cell counted $>$ 100 in high power field. Escherichia coli (*E. coli*) was grown in the urinary bacterial culture. Her urinary culture in aerobic and anaerobic conditions, *E. coli* was grown (10^7 / mL). The urethral balloon catheter was indwelled and the ultrasonography (US) was examined. The US finding showed a specific finding that "hyperechoic bladder-ring appearance" bordering along bladder wall on the sagittal plane (Fig. 1). The computed tomography (CT) taken on the same day showed an air bubble collection in the bladder wall on the sagittal and transverse planes (Fig. 2). From these radiological findings and laboratory data, she was diagnosed the emphysematous cystitis. Since this day, she showed hypotensive with septic shock and received anti-septic treatments with broad-spectrum antibiotics (tazobactam/ piperacillin, Zosyn; TAIHO Pharmaceutical Co., Ltd, Tokyo/Japan) and catecholamine for seven days until vital signs were settled down to within normal range. The specific finding of US disappeared on 18th day. She discharged and return to nursing home.

Table 1

Table 1 – The clinical profiles of three case with EC experienced in our hospital.

	1	2	3
Age	97	89	82
Sex	female	Female	Male
DM	-	-	-
Urine culture	*E. Coli* 3+	*E. Coli* 3+	K. pneumoniae
Blood culture	*E. Coli* 1+	-	-
Treatment	TAZ	CMZ	MEPM
Outcome	Survived	Survived	Survived
Radiological modalities	US, CT	CT	CT

Abbreviations, CMZ: cefmetazole, MEPM: meropenem, CT: computed tomography, DM: diabetes mellitus, *E. coli*: Escherichia coli, TAZ: Tazobactam, US: ultrasonography, -: absent

Discussion

EC is the rare urinary tract infectious disease and life-threatening [1]. The most common symptom is reported macroscopic hematuria followed by fever, abdominal pain [4]. The causative microorganisms are bacteria or fungus. Escherichia coli is the most common causative microorganisms followed by Klebsiella pneumoniae [4]. The gas collection in the bladder is a result of activity of gas-producing microorganisms under multifactorial etiology of impaired host responses [5]. The air in the bladder is located within or in the bladder wall. This air is identified as carbon dioxide (CO_2) produced through natural fermentation of sugar or protein by aerobic glycolysis process of bacteria or fungus. EC is commonly associated with DM, urinary outlet obstruction, neurogenic bladder, indwelling urethral catheters, or impaired immune function [6]. Another study reported hyper–echogenicity observed among young adults (8 female, 150 male, mean age 17 years) in the Schistosomiasis Control Project of Yemen with Schistosoma haematobium urinary tract infection [7]. Differences between this and our case might exist in causative organisms and existence of air collection in the bladder. Whereas 50%-70 % patients with EC had DM and high blood sugar concentration must be supportive to the bacterial growth, our case did not have glucose metabolic disorders. Her oldest age of 97 years old might be related with causative factor of opportunistic infection. We list all possible complications in the clinical course of patients with EC such as hydronephrosis, bladder necrosis and rupture [8], and sepsis and septic shock [9]. We have experienced another two similar EC cases in a single hospital. Among them, the diagnostic finding for EC was observed in US in the presented case and another two were diagnosed by not US, but CT with similar finding. Instead of plain abdominal X-ray, CT scout film did show in the retrospective read-

ing the similar air collection at the same site as US did show. From these considerations, plain abdominal X-ray also might be of importance for EC diagnosis. From these considerations, a plain abdominal X-ray also might be informative for EC diagnosis. To our best knowledge, the hyperechoic bladder-ring appearance as the pathognomonic finding for EC is the first nomenclature in the English literature. From this case report, we would draw the clinical warning that US might be the first radiological modality for patients who are suspected of the diagnosis of EC with hyperechoic bladder-ring appearance.

Conclusion

In this case report, we highlight the importance and utility of ultrasonography to characterize emphysematous cystitis with hyperechoic bladder-ring appearance as the first choice of diagnostic modality in radiology.

Declaration of Competing Interest

All authors have no conflicts of interest to disclose.

Author contributions

AT: manuscript writing, YF: manuscript editing and images contribution, TA: manuscript editing

Dear editor of radiology case reports journal

We present an interesting case of emphysematous cystitis (EC) with ultra-sonographic images (USs). In this article, especially USs showing pathognomonic “hyperechoic bladder-ring appearance bordering long bladder wall”. This finding must be identical to EC and it seems equivalent to diagnostic value. We acknowledge that radiology case reports is the most suitable journal for submitting of this interesting and educational case.

Patients consent

Informed consents are obtained from the patients.

Supplementary materials

Supplementary material associated with this article can be found, in the online version, at doi:10.1016/j.radcr.2021.05.051.

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