IDCases 27 (2022) e01410

Contents lists available at ScienceDirect

IDCases

journal homepage: www.elsevier.com/locate/idcases

Case report

Emphysematous cystitis due to *Streptococcus salivarius* infection in a patient with a neurogenic bladder

Shuhei Okada, Yasushi Ichimura, Masahiro Iinuma*

Department of Urology, Mito Medical Center, 280 Sakuranosato, Ibaraki-machi, Higashi-Ibaraki-gun, Ibaraki-ken 311-3193, Japan

ARTICLE INFO

Article history:
Received 18 October 2021
Received in revised form 17 January 2022
Accepted 17 January 2022

Keywords:
Emphysematous cystitis
Streptococcus salivarius
Neurogenic bladder
Urinary tract infections
Putamen hemorrhage

ABSTRACT

Streptococcus salivarius (*S. salivarius*) is an oral commensal bacterium that rarely causes disease. Here, we report a case of emphysematous cystitis due to *S. salivarius* infection in a patient with a neurogenic bladder. A 56-year-old woman was hospitalized and managed for left putamen hemorrhage. Afterward, she developed poor oral intake. Although the patient was afebrile, laboratory test results suggested an inflammatory response. Urinalysis revealed pyuria and hematuria. Abdominal computed tomography revealed a thickened urinary bladder wall and intraluminal gas. Additionally, she was diagnosed with a neurogenic bladder as she had approximately 200 mL of residual urine. The patient was diagnosed with emphysematous cystitis, and *S. salivarius* was isolated from urine culture specimens. The patient's condition improved immediately after treatment, which included bladder drainage and administration of appropriate antibiotics. We could not find any report on *S. salivarius* causing urinary tract infections, such as emphysematous cystitis. Accordingly, we report this case along with a review of the literature.

Introduction

Streptococcus salivarius (*S. salivarius*) is a viridans streptococcus, that is relatively commonly isolated from the tongue, palate, and saliva. It has low pathogenicity and is occasionally isolated from the intestine or vagina. We could not find any report on *S. salivarius* causing urinary tract infections. Herein, we report a case of emphysematous cystitis due to *S. salivarius* infection, along with a review of the literature.

Case presentation

A 56-year-old female patient, who had been generally healthy, was admitted to the Department of Neurosurgery at our hospital and managed for a left putamen hemorrhage on February 5, 2020. There was no relevant family, medical, or medication history. She gradually recovered through conservative treatment and was scheduled to be transferred to another hospital for further rehabilitation. However, she developed poor oral intake around February 19, 2020, but was afebrile. The patient was referred to our department on February 22, 2020, following routine blood sampling and an abdominal computed tomography (CT) scan. The blood sample revealed an elevated inflammatory response, and the scan showed a thickened bladder wall and intraluminal gas.

Physical examination revealed a blood pressure of 135/82 mmHg, pulse rate of 106/min, SpO_2 of 96% (room air), and body temperature of 97.3 °F. Suprapubic tenderness was observed on abdominal palpation; however, bilateral costovertebral angle tenderness was not detected.

Laboratory findings

Elevated inflammatory response and renal dysfunction were observed. A complete blood count revealed a white blood cell count of 28,600/μL; the C-reactive protein (CRP) level was 47.2 mg/dL, while blood urea nitrogen and serum creatinine levels were 97.0 mg/dL and 2.49 mg/dL, respectively. Urinalysis revealed hematuria and pyuria. These findings are shown in Table 1.

Culture

Only *S. salivarius* was detected in the urine culture (10^6 CFU/mL). Four days were required for confirmation, and no drug resistance was observed. *Peptostreptococcus anaerobius* was detected in the blood cultures; however, it was thought to have resulted from contamination. This is because only one of the two sets of blood cultures was positive, and confirmation of findings took seven days. Notably, images of the culture media are unavailable.

* Corresponding author.
E-mail address: iinuma.masahiro.nv@mail.hosp.go.jp (M. Iinuma).

https://doi.org/10.1016/j.idcr.2022.e01410
2214-2509/

Table 1
Laboratory findings on February 22, 2021.

TP	7.3	g/dL	WBC	28,600	/μL
ALB	3.7	g/dL	RBC	4.88×10^6	/μL
AST	16	U/L	Hb	17.1	g/dL
ALT	37	U/L	Ht	50.6	%
ALP	485	U/L	MCV	103.7	fL
LDH	388	U/L	MCH	35.0	pg
T-BIL	1.3	mg/dL	MCHC	33.8	g/dL
BUN	97.0	mg/dL	PLT	306×10^3	/μL
CRE	2.49	mg/dL	PT	18.2	sec
CK	38	U/L	PT(INR)	1.43	
Na	143	mmol/L	APTT	49.8	sec
K	5.5	mmol/L	D-dimer	4.7	μ./mL
Cl	104	mmol/L			
CRP	47.2	mg/dL			

TP: total protein; ALB: albumin; AST: aspartate aminotransferase; ALT: alanine aminotransferase; ALP: alkaline phosphatase; LDH: lactate dehydrogenase; T-BIL: total bilirubin; BUN: blood urea nitrogen; CRE: creatinine; CK: creatine kinase; Na: sodium; K: potassium; Cl: chloride; CRP: C-reactive protein; WBC: white blood cell; RBC: red blood cell; Hb: hemoglobin; Ht: hematocrit; MCV: mean corpuscular volume; MCH: mean corpuscular hemoglobin; MCHC: mean corpuscular hemoglobin concentration; PLT: platelet; PT: prothrombin time; INR: international normalized ratio; APTT: activated partial thromboplastin time.

Imaging

A plain CT was performed after inserting an indwelling bladder catheter; therefore, the bladder was collapsed. The bladder wall was thickened compared to the previous CT scan performed on admission. In addition, perivesical fat stranding was observed (Fig. 1); however, perirenal fat stranding and hydronephrosis were not observed bilaterally. No other abnormal findings were observed. The bladder was considered the focus of inflammation, and air bubbles were observed around the bladder wall. Although iatrogenic gas contamination could not be ruled out, emphysematous cystitis was most suspected because of a severe inflammation localized to the bladder. Since other diseases could also cause gas contamination into the bladder, colovesical fistula and vesicovaginal fistula were ruled out based on CT findings. Since an abscess continuous from the umbilicus to the dome of the bladder was not observed, a urachal abscess was similarly ruled out.

Urination

Communication with the patient was difficult; however, according to her family, she experienced no disturbance on urination before admission. In addition, the CT scan on admission showed no thickening or deformation of the bladder wall. In our hospital, acute care physicians occasionally perform a whole-body CT scan regardless of chief complaints; however, it is not routine in many centers.

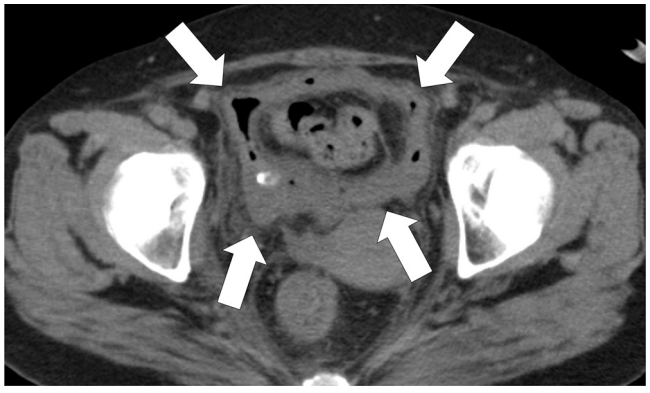

Fig. 1. Computed tomography after urethral catheterization. Arrows indicate the thickened bladder wall and air bubbles around the bladder wall (the axial slice).

An indwelling bladder catheter was inserted on February 5, 2020, and removed on February 10, 2020. On the day of catheter removal, the residual urine volume was 291 mL, which was measured only once. Afterward, her medical record did not report lower abdominal distension. However, nurses only checked whether or not the diaper was wet with urine, and the residual urine volume was not measured. The residual urine volume was approximately 250 mL when an indwelling bladder catheter was re-inserted on February 22, 2020. Therefore, we speculated that the residual urine volume was more than 200 mL after removing the indwelling bladder catheter. There was no history of diabetes mellitus or pelvic surgery. Therefore, increased residual urine volume was supposedly caused by a neurogenic bladder associated with the left putamen hemorrhage.

Diagnosis

Based on the above findings, we diagnosed the patient with emphysematous cystitis due to *S. salivarius* infection. Neurogenic bladder associated with left putamen hemorrhage is supposedly one of the causes of this disease.

Clinical course

Pyuria drainage was achieved by re-inserting an indwelling bladder catheter. Meropenem (2 g/day) and clindamycin (1200 mg/day) were administered intravenously from February 22, 2020. The dose of meropenem was reduced from 3 g/day to 2 g/day due to renal dysfunction. The inflammatory response and renal function gradually improved and were almost normalized on March 1, 2020 (white blood cell count: 6800/μL, C-reactive protein: 0.94 mg/dL, blood urea nitrogen: 9.1 mg/dL, and serum creatinine: 0.50 mg/dL). On March 2, 2020, we changed the antibiotic to levofloxacin (500 mg/day), based on antibiotic susceptibility testing. On March 15, 2020, levofloxacin was discontinued, and no relapse of the inflammatory reaction was observed (Fig. 2). The patient was transferred to another hospital for further rehabilitation on March 24, 2020. Since she could not perform intermittent self-catheterization, we inserted an indwelling bladder catheter. After hospital discharge, the patient was scheduled to be examined for neurogenic bladder at our department.

Discussion

Emphysematous cystitis is caused by gas-forming bacteria. The mean age of emphysematous cystitis occurrence is approximately 66 years, and it occurs twice as often in women than in men [1]. Diabetes mellitus and urinary stasis are well-recognized risk factors [2].

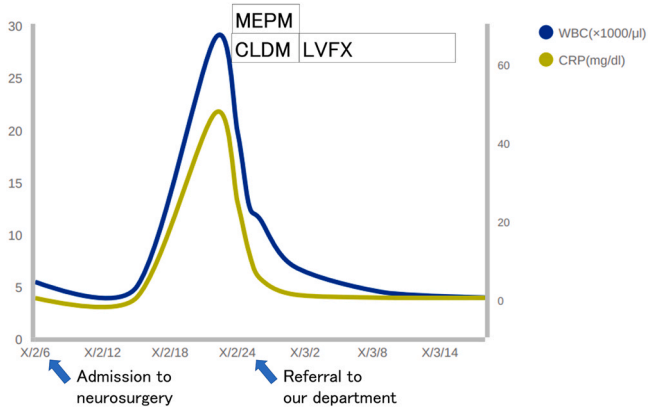

Fig. 2. Clinical course post-admission. MEPM, meropenem hydrate; CLDM, clindamycin phosphate; LVFX, levofloxacin hydrate.

In emphysematous cystitis, *Escherichia coli* and *Klebsiella pneumoniae* are often isolated from urine cultures [1]. As for the genus Streptococcus, group D streptococcus has been reported to cause emphysematous cystitis [1]. To the best of our knowledge, there are no reports of *S. salivarius* as the causative pathogen of urinary tract infections, such as emphysematous cystitis [3]. *S. salivarius* is a viridans streptococcus, which has low pathogenicity. It is relatively commonly isolated from the tongue, palate, and saliva, and is occasionally isolated from the intestine or vagina [4–6]. Its presence in the vagina is considered to inhibit the growth of Group B Streptococcus [4]. According to previous reports, it causes bacteremia, meningitis, endocarditis, sinusitis, and liver abscesses [5–10]. There are two possible mechanisms by which *S. salivarius* causes bacteremia: (1) iatrogenic contamination associated with an invasive procedure, such as spinal anesthesia, and (2) invasion associated with disruption of the gastrointestinal mucosa [5,11,12]. In this case, these mechanisms were unlikely because the patient did not have bacteremia. The route of infection was supposedly retrograde, with *S. salivarius* arising from feces or vaginal discharge. However, this was not verified since culture tests of feces and vaginal discharge were not performed. In addition, iatrogenic contamination of *S. salivarius* was possible through the saliva of medical staff during the insertion of an indwelling bladder catheter on admission. However, this is considered unlikely because all emergency room staff at our hospital wear personal protective equipment. As mentioned above, since plain CT was performed after inserting an indwelling bladder catheter, we could not deny the possibility of gas contamination. The collection of gas in the bladder wall, which is characteristic of emphysematous cystitis, was difficult to recognize because the bladder was collapsed. In addition, according to the medical record, the indwelling bladder catheter was inserted at 11:00 am, and the plain CT scan was performed at 2:00 pm. Thus, approximately 3 h had passed, during which the intramural and intraluminal gas may have decreased. However, emphysematous cystitis was suspected because of strong inflammation localized to the bladder. Although emphysematous cystitis is characterized by cobblestone and beaded-necklace appearance on abdominal X-ray [13], such findings may not be observed when an indwelling bladder catheter is inserted. Hence, CT is useful for establishing diagnoses, as in this case. Besides emphysematous cystitis, pyocystis (bladder abscess) also presents with strong inflammation localized to the bladder. However, pyocystis is unlikely in this case because it has been reported in oliguric patients, such as patients with supravesical urinary diversion or hemodialysis-dependent patients [14]. Although there is no report of emphysematous cystitis due to *S. salivarius* infection [1,2,15,16], *S. salivarius* can lead to gas formation. The urease system in *S. salivarius* breaks down urea to ammonia and CO_2 [17]. Since substances other than glucose are supposedly substrates for gas formation, it is feasible that urea was a substrate for gas formation in this case. The initial management of emphysematous cystitis consists of drainage of the bladder by catheter placement, appropriate antibiotics administration, and treatment of predisposing conditions, such as glycemic control [15]. If these treatments are ineffective, surgical treatment, including cystectomy, may be required [15]. The overall death rate from emphysematous cystitis has been reported to be approximately 7%; however, if emphysematous pyelonephritis occurs, the mortality rate increases to 14–20% [1,15]. Therefore, early detection of emphysematous cystitis is desirable. Additionally, we considered what could have been done to diagnose this case earlier. Emphysematous cystitis can vary from asymptomatic cases to sepsis. The frequent chief complaints of patients with emphysematous cystitis are suprapubic pain, gross hematuria, fever, and pneumaturia [15]. In this case, gross hematuria was not observed, and the patient had been afebrile. Due to the acute left putamen hemorrhage, it was difficult for the patient to complain of suprapubic pain. If pneumaturia existed, it was difficult to recognize because she was incontinent. In addition, even if she had residual urine retention, it was difficult to complain because of the acute left putamen hemorrhage. Therefore, in the acute phase of cerebrovascular disease, residual urine measurement and urinalysis after removing an indwelling bladder catheter should be performed periodically to prevent emphysematous cystitis.

Conclusion

We reported a case of emphysematous cystitis due to *S. salivarius* infection in a patient with a neurogenic bladder. We could not find any reports of *S. salivarius* causing urinary tract infections, including emphysematous cystitis. Infectious diseases can be detected by clinical signs and blood work in afebrile patients as well. In the acute stage of cerebrovascular disease, it may be difficult for a patient to complain of bladder irritability or suprapubic pain. Therefore, emphysematous cystitis should be understood not only by urologists but also by neurologists.

Ethical approval

This study has been approved by the institutional review board of Mito medical center.

Funding

This research did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors.

CRediT authorship contribution statement

Shuhei Okada: Conceptualization, Investigation, Writing – original draft. **Yasushi Ichimura:** Conceptualization, Investigation. **Masahiro Iinuma:** Writing – review & editing, Supervision, Project administration.

Declaration of interest

None.

Acknowledgments

None.

Approval of research protocol

Not applicable.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Registry and registration number

Not applicable.

References

[1] Schicho A, Stroszczynski C, Wiggermann P. Emphysematous cystitis: mortality, risk factors, and pathogens of a rare disease. Clin Pract 2017;7:54–5. https://doi.org/10.4081/cp.2017.930

[2] Adeyemi OA, Flaherty JP. Emphysematous cystitis. Cureus 2020;12:e11723https://doi.org/10.7759/cureus.11723

[3] Facklam R. What happened to the streptococci: overview of taxonomic and nomenclature changes. Clin Microbiol Rev 2002;15:613–30. https://doi.org/10.1128/CMR.15.4.613-630.2002
[4] Patras KA, Wescombe PA, Rösler B, Hale JD, Tagg JR, Doran KS. Streptococcus salivarius K12 limits Group B Streptococcus vaginal colonization. Infect Immun 2015;83:3438–44. https://doi.org/10.1128/IAI.00409-15
[5] Legier JF. Streptococcus salivarius meningitis and colonic carcinoma. South Med J 1991;84:1058–9. https://doi.org/10.1097/00007611-199108000-00031
[6] Elsawy AM, Faidah HS, Redwan EM. Streptococcus salivarius meningitis in immunocompetent: a case report. Int Arch Med Microbiol 2018;1. https://doi.org/10.23937/iamm-2017/1710004
[7] Nilson B, Olaison L, Rasmussen M. Clinical presentation of infective endocarditis caused by different groups of non-beta haemolytic streptococci. Eur J Clin Microbiol Infect Dis 2016;352:215–8. https://doi.org/10.1007/s10096-015-2532-5
[8] Radosz-Komoniewska H, Kapp-Burzyńska Z, Kłaptocz B, Wilk I, Ekiel A. Tlenowa i beztlenowa flora bakteryjna w przewlekłym zapaleniu zatok przynosowych u dorosłych [Aerobic and anaerobic bacterial flora in chronic sinusitis in adults]. Med Dosw Mikrobiol 1997;49:89–94.
[9] Ruiz-Tovar J, Gamallo C. Streptococcus salivarius causing multiple liver abscesses in a patient with situs inversus. Surg Infect 2012;132:130–1. https://doi.org/10.1089/sur.2011.082
[10] Kamachi S, Otsuka T, Tsuji C, Nakashita S, Ide Y, Mizuta T. A case of multiple liver abscesses associated with Streptococcus salivarius in a patient with chronic periodontitis. Nihon Shokakibyo Gakkai Zasshi Jpn J Gastro-Enterol 2014;111:1602–8.
[11] Veringa E, van Belkum A, Schellekens H. Iatrogenic meningitis by Streptococcus salivarius following lumbar puncture. J Hosp Infect 1995;29:316–8. https://doi.org/10.1016/0195-6701(95)90283-x
[12] Idigoras P, Valiente A, Iglesias L, Trieu-Cout P, Poyart C. Meningitis due to Streptococcus salivarius. J Clin Microbiol 2001;39:3017. https://doi.org/10.1128/JCM.39.8.3017.2001
[13] Middela S, Green E, Montague R. Emphysematous cystitis: radiological diagnosis of complicated urinary tract infection. BMJ Case Rep 2009;bcr05.2009:1832. https://doi.org/10.1136/bcr.05.2009.1832
[14] Elsayed M, Finn A, Dapaah-Afriyie K. Unusual organism causing pyocystis in an immunosuppressed haemodialysis patient. BMJ Case Rep 2016;bcr2015214264. https://doi.org/10.1136/bcr-2015-214264
[15] Amano M, Shimizu T. Emphysematous cystitis: a review of the literature. Intern Med 2014;53:79–82. https://doi.org/10.2169/internalmedicine.53.1121
[16] Thomas AA, Lane BR, Thomas AZ, Remer EM, Campbell SC, Shoskes DA. Emphysematous cystitis: a review of 135 cases. BJU Int 2007;100:17–20. https://doi.org/10.1111/j.1464-410X.2007.06930.x
[17] Zlatkovic N, Aleksić G, Gašić K. Biology of oral streptococci. Microbiol Spectr 2018;6. https://doi.org/10.1128/microbiolspec.GPP3-0042-2018