

RADIOLOŠKI NALAZ PNEUMOMEDIJASTINUMA KAO RETKE KOMPLIKACIJE EMFIZEMATOZNOG PIJELONEFRITISA

PRIKAZ SLUČAJA

CASE REPORT

RADIOLOGICAL FINDING OF PNEUMOMEDIASTINUM AS A RARE COMPLICATION OF EMPHYSEMATOUS PYELONEPHRITIS

Ksenija Mijović¹, Nevena Stanišić¹, Dragan Mašulović^{1,2}, Dušan Šaponjski¹, Nikola Bogosavljević³, Dragan Vasin¹, Milica Mitrović Jovanović¹

¹ Univerzitetski klinički centar Srbije, Centar za radiologiju i magnetnu rezonancu, Beograd, Srbija

² Univerzitet u Beogradu, Medicinski fakultet, Srbija

³ Institut za ortopediju „Banjica“, Beograd, Srbija

¹ University Clinical Center of Serbia, Center for Radiology and Magnetic Resonance Imaging, Belgrade, Serbia

² University of Belgrade, Faculty of Medicine, Serbia

³ Institute for Orthopedic Surgery “Banjica”, Belgrade, Serbia

SAŽETAK

Uvod: Emfizematozni pijelonefritis je retka, akutna i životno ugrožavajuća nekrotizirajuća infekcija bubrega, uzrokovana fakultativno anaerobnim bakterijama, koje proizvode gas, kao što su *Escherichia coli*, *Klebsiella* i *Proteus*, a javlja se najčešće kod pacijenata koji boluju od diabetes mellitus-a. Pneumomediastinum je redak nalaz u ovom oboljenju, ali uz odgovarajuće kliničke i laboratorijske nalaze, kod pacijenata sa radiološkim nalazom pneumomediastinuma, potrebno je posumnjati i na eventualno postojanje emfizematoznog pijelonefritisa.

Prikaz bolesnika: Prikazujemo slučaj pacijentkinje srednjeg životnog doba, sa simptomima i laboratorijskim nalazima koji ukazuju na infekciju, bubrežnu insuficijenciju i hiperglikemiju, a koja se lečila od diabetes mellitus-a. Radiološkim pregledima je dijagnostikovana izražena i uznapredovala nekrotizirajuća infekcija bubrega, sa destrukcijom parenhima i intraparenhimskim inkluzijama gasa, koje su propagirale u perirenalni i pararenalni prostor. Postavljena je dijagnoza emfizematoznog pijelonefritisa sa pratećim perirenalnim apscesom i pneumoretroperitoneumom, uz kolekcije gasa, koje su dostizale medijastinum. Zbog ovako izražene infekcije, pacijentkinja je podvrgnuta hitnom operativnom lečenju, pri čemu je učinjena levostrana nefrektomija. Uprkos brzom i iscrpnom tretmanu, došlo je do smrtnog ishoda u postoperativnom periodu.

Zaključak: Pneumomediastinum je retka komplikacija retroperitonealnih procesa, i kada je prisutan, ukazuje na ekstenzivnost samog oboljenja, najčešće kao izrazito loš prognostički znak. Ovim prikazom slučaja želimo da ukažemo na ozbiljnost ove bubrežne infekcije, sa nedvosmislenom potrebom za hitnim reagovanjem, i da skrenemo pažnju na radiološke znake koji su neuobičajeni, ali bi morali da pobude sumnju na podmluku i ozbiljnu retroperitonealnu infekciju.

Ključne reči: infekcije bubrega, pneumoretroperitoneum, pneumomediastinum.

ABSTRACT

Introduction: Emphysematous pyelonephritis is a rare, acute, and life-threatening necrotizing renal infection caused by gas-producing facultative anaerobes, such as *Escherichia coli*, *Klebsiella*, and *Proteus*, most commonly in the setting of underlying diabetes mellitus. Pneumomediastinum is a rare imaging finding in this disease, and a high index of suspicion is required for diagnosing emphysematous pyelonephritis in patients presenting with pneumomediastinum, in the right clinical setting.

Case presentation: We present the case of a middle-aged female patient with symptoms and laboratory findings indicating infection, renal failure, and hyperglycemia, and a personal history of diabetes mellitus. The imaging procedures revealed findings of a severe and advanced necrotizing renal infection with parenchymal destruction and intraparenchymal gas collections extending into the perirenal and pararenal spaces. We established the diagnosis of emphysematous pyelonephritis accompanied by perirenal abscess formation and pneumoretroperitoneum, with gas collections propagating into the mediastinum. In the setting of such a severe form of infection, the patient underwent immediate surgery with left nephrectomy. Despite prompt and intensive treatment, the patient unfortunately succumbed to the disease during the postoperative period.

Conclusion: Pneumomediastinum is a rare complication of retroperitoneal processes, and, when present, indicates their extensiveness, often being an ominous prognostic sign. By presenting this case, we aim to highlight the severity of this form of renal infection and the unequivocal need for immediate response, as well as to emphasize the significance of imaging findings, which are somewhat unusual, but should raise suspicion of an insidious and serious retroperitoneal infection.

Key words: renal infection, pneumoretroperitoneum, pneumomediastinum

Autor za korespondenciju:

Ksenija Mijović

Centar za radiologiju i magnetnu rezonancu, Univerzitetski klinički centar Srbije

Pasterova 2, 11000 Beograd, Srbija

Elektronska adresa: mijovicksenija@gmail.com

Corresponding author:

Ksenija Mijović

Center for Radiology and Magnetic Resonance Imaging, University Clinical Center of Serbia

2 Pasterova Street, 11000 Belgrade, Serbia

E-mail: mijovicksenija@gmail.com

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UVOD

Emfizematozni pijelonefritis (EPN) je retko, akutno i životno ugrožavajuće oboljenje bubrega koje se definiše kao nekrotizirajuća infekcija sa partikulama gasa unutar parenhima bubrega, sabirnog sistema ili u perirenalnom prostoru [1]. Uglavnom se javlja kod pacijenata sa nekontrolisanim diabetes mellitus-om i češći je kod osoba ženskog pola. Najčešći uzročnici su gram-negativni fakultativni anaerobi, kao što su *E. Coli*, *Klebsiella* i *Proteus* [2].

Oboljenje može biti asimptomatsko ili se prezentuje nespecifičnim simptomima, kao što su generalizovana slabost, groznica, lumbalni bol, ili može imitirati intestinalnu opstrukciju i perforaciju digestivnog trakta i, zajedno sa nespecifičnim laboratorijskim nalazima, dijagnoza se često kasno postavlja [3,4,5]. Kompjuterizovana tomografija (CT) je najpouzdanija metoda za pravovremenu dijagnozu, evaluaciju rasprostranjenosti slobodnih gasnih inkluzija i tečnosti, kao i za praćenje toka bolesti [6].

PRIKAZ SLUČAJA

Pacijentkinja, starosti 56 godina, javila se u Urgentni centar Univerzitetskog kliničkog centra Srbije sa simptomima izražene malaksalosti, koja je bila praćena groznicom, dispnejom i opstipacijom. Ovi simptomi su trajali prethodnih 7 dana. Laboratorijski nalazi su pokazali leukocitozu ($26 \times 10^9/l$) sa neutrofilijom (88,6%), visoke vrednosti C-reaktivnog proteina (280,6 mg/l), izrazito visoku vrednost procalcitonina (49,2 ng/ml), povišene vrednosti azotnih materija: vrednosti uree – 29,7 mmol/l i kreatinina – 286 $\mu\text{mol/l}$, kao i nemerljivu hiperglikemiju i nakon primene insulina. Od hroničnih bolesti, u anamnezi je navela dijabetes.

Nativna radiografija abdomena pokazala je znake ekstraluminalnog gasa, u smislu pneumoperitoneuma, dok je ultrasonografijom abdomena vizuelizovana velika količina gasa u levom retroperitonealnom prostoru, koja je ograničavala vizualizaciju levog bubrega. Nalaz kompjuterizovane tomografije (CT) pokazao je uvećan levi bubrež, koji je bio dezintegriran parenhima, unutar kojeg su se videle male kružne i linearne inkluzije gasa sa propagacijom u perirenalni prostor, u čijem se posteromedijalnom aspektu prikazala i apscesna kolekcija sa hidroaeričnim nivoom (Slika 1). Celokupan nalaz je ukazao na postojanje EPN-a. Inkluzije gasa su se vizuelizovale i u levom retroperitonealnom prostoru, kao i u psoasnom mišiću, sa propagacijom u muskulaturu levog koksofemoralnog zgloba. Inkluzije slobodnog gasa su se vizuelizovale i kontralateralno u prednjem pararenalnom prostoru prekalvalno, sa retrokruralnom propagacijom u medijastinum (Slika 2). Dalja kranijalna ekstenzija inkluzija gasa dosegla je

INTRODUCTION

Emphysematous pyelonephritis (EPN) is a rare, acute, and life-threatening necrotizing renal disease defined as necrotizing infection with gas particles within the renal parenchyma, the kidney collecting system, or in the perirenal space [1]. It mainly occurs in patients with uncontrolled diabetes mellitus and is more frequent in women. It is most commonly caused by gram-negative facultative anaerobes, such as *E. coli*, *Klebsiella*, and *Proteus* [2].

The disease may be asymptomatic, or it may present with nonspecific symptoms, such as generalized fatigue, fever, lower back pain, or it may imitate intestinal obstruction and GI tract perforation and, combined with nonspecific laboratory test results, the result is that diagnosis is often established late [3,4,5]. Computerized tomography (CT) is the most reliable method for establishing a timely diagnosis, evaluating the distribution of gas inclusions and fluid, as well as for monitoring the course of the disease [6].

CASE PRESENTATION

A 56-year-old female patient presented at the Emergency Clinic of the University Clinical Center of Serbia with symptoms of extreme fatigue, accompanied by fever, dyspnea, and obstipation. These symptoms had been present the previous 7 days. Laboratory findings showed leukocytosis ($26 \times 10^9/l$) with neutrophilia (88.6%), a high level of C-reactive protein (280.6 mg/l), a very high level of procalcitonin (49.2 ng/ml), elevated levels of nitrogenous substances: urea – 29.7 mmol/l and creatinine – 286 $\mu\text{mol/l}$, as



Slika 1. Aksijalni presek CT pregleda u nivou gornjeg pola levog bubrega prikazuje apscesnu kolekciju u posteromedijalnom aspektu, sa hidroaeričnim nivoom

Figure 1. Axial CT section at the left kidney upper pole level demonstrating an abscess formation in the posteromedial aspect, with an air-fluid level.



Slika 2. Presek u aksijalnoj ravni prikazuje dezintegriran parenhim levog bubrega sa intraparenhimskim gasom i inkluzijama gasa koje propagiraju u perirenalni i pararenalni prostor. Gas se uočava i u kontralateralnom zadnjem pararenalnom prostoru, kao i u prednjem trbušnom zidu.

Figure 2. Axial plane showing destroyed left renal parenchyma with intraparenchymal gas and gas inclusions extending to the perirenal and pararenal spaces. Gas is also visible in the contralateral posterior pararenal space, as well as in the anterior abdominal wall.

nivo gornje torakalne aperture, dajući radiološku sliku pneumomediastinuma (**Slike 3 i 4**).

Intraoperativni nalaz kod naše pacijentkinje je ukazao na kompletnu destrukciju parenhima levog

well as extreme hyperglycemia, even after insulin administration. In her anamnesis, the patient stated diabetes as a chronic disease.

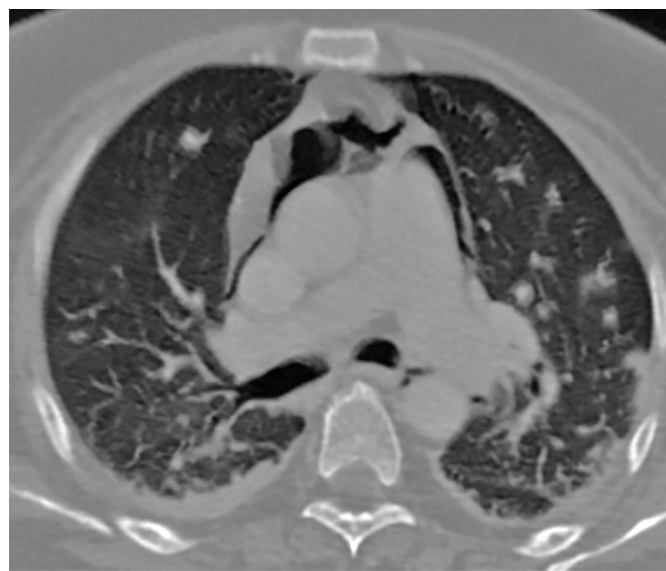
Native radiography of the abdomen showed signs of extraluminal gas, in the form of pneumoperitoneum, while abdominal ultrasonography showed a large amount of gas in the left retroperitoneal space, which limited the visibility of the left kidney. The computerized tomography (CT) finding showed an enlarged left kidney, with a disintegrated parenchyma, within which small circular and linear gas inclusions were visible that extended into the perirenal space, in whose postero-medial aspect an abscess formation with an air-fluid level was also visible (**Figure 1**). The complete finding indicated the existence of EPN. Gas inclusions were also visible in the left retroperitoneal space, as well as in the psoas muscle, with extensions into the musculature of the left coxofemoral joint. Inclusions of free gas were also visible contralaterally in the anterior pararenal space, precavally, with retrocrural extension into the mediastinum (**Figure 2**). Further superior extension of gas inclusions reached the level of the superior thoracic aperture presenting the radiological finding of pneumomediastinum (**Figures 3 and 4**).

The intraoperative finding in our patient revealed complete parenchymal destruction in the left kidney, which is why total left nephrectomy was performed. The pathohistological finding showed chronic



Slika 3. Reformacija u koronalnoj ravni prikazuje gas u perikavalnom prostoru, koji kranijalno propagira u bazalne delove medijastinuma. Inkluzije gasa se uočavaju i u mekim tkivima oko aortnog luka.

Figure 3. Coronal reformation showing gas in the pericaval space extending superiorly into the basal portions of the mediastinum. The gas inclusions are seen in the tissues outlining the aortic arch.



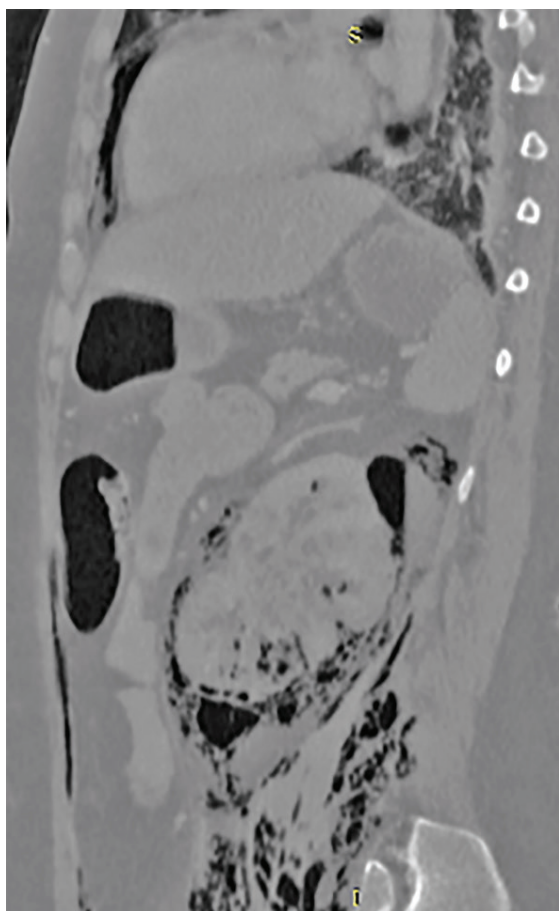
Slika 4. Aksijalni presek u nivou medijastinuma, subkarinealno, prikazuje gas koji ovičava velike medijastinalne vaskularne strukture kao i inkluzije gasa u prednjem medijastinumu.

Figure 4. Axial plane through the mediastinum at the subcarinal level shows gas outlining the great mediastinal vessels, as well as gas inclusions in the anterior mediastinum.

bubrega, usled čega je urađena totalna levostrana nefrektomija. Patohistološki nalaz je pokazao hronični pijelonefritis i perinefritis sa akutnom purulentnom egzacerbacijom i formiranjem apscesa, uz hemoragične infarkte parenhima. Pacijentkinja je, nažalost, podlegla bolesti u postoperativnom toku.

DISKUSIJA

Predstavljamo slučaj EPN-a sa pneumoretroperitoneumom i pneumomediastinomom, što predstavlja retku komplikaciju. Retroperitoneum, medijastinum i subkutana tkiva su u kontinuitetu i međusobno komuniciraju, povezana fascijalnim prostorima duž kojih se prostiru veliki krvni sudovi i vlakna dijafragme, što omogućava slobodnom gasu nastalom u bilo kojoj od ovih regija da nesmetano propagira u drugu regiju. Tako, retroperitonealni gas, nastao usled infekcije bakterijama koje proizvode gas, može dosegnuti medijastinum i subkutano tkivo sa nastankom pneumomediastinuma i subkutanog emfizema [7,8], što je i bio slučaj sa našom pacijentkinjom (Slika 5).



Slika 5. Sagitalna reformacija ukazuje na postojanje gasnih inkluzija, kako u levom retroperitoneumu, tako i u prednjem medijastinumu.

Figure 5. Sagittal reformation image depicting gas inclusions, both in the left retroperitoneum and the anterior mediastinum.

pyelonephritis and perinephritis with acute purulent exacerbation and abscess formation, with hemorrhagic parenchymal infarctions. Unfortunately, the patient postoperatively succumbed to the illness.

DISCUSSION

We present a case of EPN with pneumoretroperitoneum and pneumomediastinum, which is a rare complication. The retroperitoneum, mediastinum, and subcutaneous tissue are connected and in communication with one another, linked together by fascial tissue spaces along which large blood vessels and diaphragm fibers extend, which enables free gas developing in any of these areas to easily extend into any of the other regions. Thus, retroperitoneal gas, developing as the result of an infection caused by gas-producing bacteria, may reach the mediastinum and the subcutaneous tissue and result in the development of pneumomediastinum and subcutaneous emphysema [7,8], which was, indeed, the case with our patient (Figure 5).

Based on gas inclusion distribution and the degree of involvement of the kidney, EPN has been classified into three classes:

- Class 1: gas in the kidney collecting system
- Class 2: gas in the kidney parenchyma
- Class 3a: gas propagation into the perirenal space
- Class 3b: gas propagation into the pararenal space
- Class 4: bilateral EPN or the only kidney with EPN [9].

Based on this classification, our patient was categorized as Class 3b.

Mortality is high and may be as high as 80% in the absence of a surgical procedure [2]. When the parenchyma of the kidney is preserved, initial treatment is conservative, with possible percutaneous drainage or ureteral stenting. In case of diffuse and advanced infection with extensive parenchyma destruction, the indication for urgent surgery becomes quite clear [8].

EPN is a life-threatening infection caused by gas-producing microorganisms, which most commonly requires nephrectomy, and which may be complicated by propagation of gas inclusions along extraperitoneal tissues, rarely reaching the mediastinum. Therefore, the finding of pneumomediastinum of unknown cause with a certain clinical presentation warrants the inclusion of EPN into the differential diagnosis. As far as we could find, by researching available literature, pneumomediastinum is a rare complication of EPN and it correlates with an advanced stage of disease.

Conflict of interest: None declared.

Na osnovu distribucije gasnih inkluzija i zahvaćenosti bubrega, EPN je klasifikovan u četiri klase:

Klasa 1: gas u sabirnom sistemu bubrega

Klasa 2: gas u parenhimu bubrega

Klasa 3a: propagacija gasa u perirenalni prostor

Klasa 3b: propagacija gasa u pararenalni prostor

Klasa 4: bilateralni EPN ili jedini bubreg sa EPN-om [9].

Na osnovu ove klasifikacije, naša pacijentkinja je svrstana u Klasu 3b.

Mortalitet je visok, i može iznositi i do 80% bez hirurške intervencije [2]. Kada postoji očuvan parenhim bubrega, inicijalna terapija je konzervativna, eventualno sa perkutanom drenažom ili plasiranjem ureteralnih stentova. U slučaju difuzne i uznapredovale infekcije sa ekstenzivnom destrukcijom parenhima, indikacija za urgentnu hirurgiju postaje jasna [8].

EPN je životno ugrožavajuća infekcija uzrokovana mikroorganizmima koji proizvode gas, koja najčešće zahteva nefrektomiju, a koja se može komplikovati propagacijom gasnih inkluzija duž ekstraperitonealnih tkiva, retko dosežući medijastinum. Zbog toga, nalaz pneumomedijastinuma nepoznatog uzroka sa odgovarajućom kliničkom prezentacijom zahteva uključivanje EPN-a u diferencijalnu dijagnozu. Koliko smo mi saznali pretraživanjem dostupne literature, pneumomedijastinum je retka komplikacija EPN-a i korelira sa uznapredovalim stadijumom bolesti.

Sukob interesa: Nije prijavljen.

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