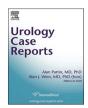
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## Inflammation and infection

# Emphysematous epididymitis following hydrocelectomy

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Acute epididymo-orchitis is an inflammatory process caused by bacterial infection. Emphysematous epididymitis is an extremely rare manifestation characterized by gas within the epididymal tissues. We report a case of emphysematous epididymitis following hydrocelectomy in a patient with a history of spinal cord injury and chronic bacteriuria. The diagnosis was made by clinical and laboratory data with imaging demonstrating foci of gas within the epididymal structures. We hypothesize that intermittent catheterization may have contributed to bacterial translocation into the adjacent cord structures and development of infection. High level of suspicion leading to early diagnosis, aggressive antibiotics and adequate debridement are required.

#### Introduction

As an immuno-privileged site that also has local innate immunity against invading pathogens, the seminal tract is highly resistant to infection. Emphysematous epididymitis is an extremely rare inflammatory process characterized by the presence of gas within the epididymal tissues. Few case reports of this entity exist, with no certain mechanism of pathogenesis. We report a case of emphysematous epididymitis following hydrocelectomy.

## Case presentation

A 39-year-old male patient underwent a right hydrocelectomy for worsening scrotal swelling that interfered with self-catheterization. The patient had a history of T12 complete spinal cord injury with no infraumbilical sensation, neurogenic bladder managed by self-intermittent catheterization, and chronic bacteriuria. He had recently been treated for a symptomatic *Klebsiella pneumoniae* and *Enterococcus faecalis* urinary tract infection prior to hydrocelectomy. The surgery was uncomplicated and the right testis was noted to be normal in appearance at time of procedure.

Four days after surgery the patient presented to our emergency department with recurrent scrotal swelling. He was insensate in the scrotum but complained of fever and bilateral groin and lower abdominal discomfort. Vital signs revealed temperature  $97.4^{\circ}F$ , heart rate 122bpm, blood pressure 126/64 mmHg, and respiratory rate 16. Exam revealed a significantly swollen and tense scrotum up to 15cm, and an intact surgical incision without evidence of superficial infection.

Laboratory studies indicated a hemoglobin of 13.9g/dL, elevated white blood cell count of 13,400/ $\mu$ L, absolute neutrophil count of 9700/ $\mu$ L, and a C-reactive protein level of 28.3mg/dL. A scrotal ultrasound noted large right hydrocele, a 1.2cm cystic structure in the right epididymal head and epididymal hyperemia concerning for possible developing abscess (Fig. 1). Doppler showed normal blood flow to the testis.

The patient was admitted and started on vancomycin and ertapenem based on previous culture sensitivities. Over the following 12 hours, his groin pain worsened and sinus tachycardia persisted. He remained afebrile. His scrotum had continued to enlarge to the point of near dehiscence of the surgical incision. A CT abdomen/pelvis revealed significant inflammatory changes involving the right epididymis and testicle extending through the inguinal canal, foci of gas within the epididymis, and a large surrounding hydrocele with mild enhancement of the wall (Fig. 2). Given this constellation of findings, the patient was taken urgently to the operating room for debridement.

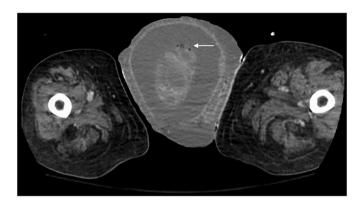
The surgical incision dehisced during sterile preparation, with drainage of serous fluid. The tunica vaginalis was opened along the previous incision and the right testis and epididymis were abnormal in appearance: firm and significantly enlarged with reactive swelling. The tissues were woody and poorly vascularized with areas of erosion. The individual structures were unable to be dissected due to edema and a necrotic rind that had formed. The spermatic cord was thickened and firm to the level of the external ring. Intraoperative ultrasound could not clearly distinguish the testis from the epididymis. Some pulsatile flow was seen in the proximal spermatic cord, but no blood flow was observed in the distal cord or testis/epididymis. A right orchiectomy was performed (gross specimen, Fig. 3).

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**Fig. 1.** Scrotal ultrasound epididymis (transverse image) showing finding of a large right hydrocele with a 1.2 cm cystic structure in the right epididymal head and epididymal hyperemia concerning for possible developing abscess.



**Fig. 2.** Computed tomography (CT), axial section of scrotum demonstrating foci of gas within the epididymis and a large surrounding hydrocele with mild enhancement of the wall.

Microscopic examination of the specimen showed marked edema, congestion, and granulation of the hilar, paratesticular, and spermatic cord soft tissue with focal acute abscess formation (Fig. 3 inset). There was marked testicular and epididymal atrophy with stromal fibrosis and limited spermatogenesis. Tissue cultures of the specimen revealed *Klebsiella pneumoniae* and *Enterococcus faecalis* with identical susceptibility patterns to pre-operative urine culture isolates. The patient improved clinically following orchiectomy with resolution of pain and tachycardia, and he was discharged on post-op day 4 and completed a 7-day course of vancomycin and ertapenem per Infectious Disease recommendations.

## Discussion

Epididymitis is the most common infectious process in the scrotum. Emphysematous epididymitis, characterized by the presence of gas within the tissue, is rare with few case reports in the literature.  $^{1-4}$ 

Studies of gas-forming bacteria suggest that impaired transport of gas and catabolic end products away from inflammatory sites triggers a pathway of mixed acid fermentation of glucose, usually by *E. coli* or *K. pneumoniae*, that manifests clinically as an emphysematous infection.

Our patient's intraoperative tissue cultures grew the same organisms (including Klebsiella) his urine had been colonized with prior to surgery, despite a course of culture-directed antibiotics.

We hypothesize that intermittent catheterization in the setting of chronic bacteriuria may have led to translocation of the bacteria into the



**Fig. 3.** Gross pathology with edema and necrosis of cord structures. Inset shows Gram stain of cord structures with presence of infiltrating gram-negative rods.

adjacent urinary structures including the prostate and spermatic cord. Subsequent manipulation of the cord structures at the time of hydrocelectomy may have set the stage for local inflammation and tissue ischemia that initiated the gas-producing infection by the colonizing *K. pneumoniae*.

Emphysematous epididymitis is distinct from necrotizing infections such as Fournier's gangrene. The latter are usually polymicrobial infections, with *Streptoccocus*, *Staphylococcus*, and Enterobacteriaceae being the most commonly isolated organisms. The rapidly progressive disease involves a series of host-bacterial and bacterial-bacterial synergistic interactions which initiate and propagate a cytokine cascade that results in endothelial breakdown, disseminated micro-thrombosis, and fascial necrosis. Importantly, the testes and cord structures are usually spared in Fournier's gangrene, which is theorized to be due to their separate blood supply from the surrounding soft tissue.

Imaging is critical to prompt diagnosis of emphysematous epididymitis, as this facilitates identification of gas within the affected tissues, which is pathognomonic. On scrotal ultrasound, gas is identified as hyperechoic foci but must be differentiated from other potentially hyperechoic lesions (e.g. suture, calcifications, shrapnel). Computed tomography(CT) imaging is the preferred modality, with high sensitivity and specificity for the identification of gas. Magnetic resonance imaging (MRI) is less-utilized due to its time-consuming nature, high cost, and equivalent performance to CT.

Early diagnosis, aggressive antibiotics and adequate debridement remain the mainstay of treatment. To our knowledge, this is the first case report of emphysematous epididymo-orchitis in a patient with a history of spinal cord injury and neurogenic bladder managed by intermittent catheterization.

### Conclusion

The treatment of emphysematous epididymitis is invariably surgical, as demonstrated in this case by the necessity of exploration and debridement after clinical failure of broad-spectrum intravenous antibiotics. The use of ultrasound and computed topography (CT) imaging as adjuncts are essential to early recognition and effective treatment.

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None.

## **Declaration of competing interest**

None.

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