Case Report

An 85-year-old man presented to his general physician with an acute painful scrotal swelling. He had an indwelling transurethral catheter because of urinary retention. The general physician diagnosed this swelling as an acute epididymitis and treated the patient with 4 weeks of ciprofloxacin. Treatment reduced but did not resolve the symptoms. Consequently, the general physician took a urine culture and restarted ciprofloxacin. In the absence of improvement, treatment was switched to amoxicillin clavulanate. After 4 days of treatment with amoxicillin clavulanate, urine culture showed >10^5 c.f.u. ml⁻¹ of Escherichia coli susceptible to trimethoprim, nitrofurantoin and fosfomycin. The isolate was resistant to amoxicillin, amoxicillin clavulanate and ciprofloxacin. Treatment was switched to trimethoprim and continued for 3 weeks without success. During these 3 weeks, the indwelling tranurethral catheter was exchanged. Finally, the patient was referred to our department. On physical examination, the right epididymis was still swollen, rigid and painful on palpation. Again, urine was cultured but did not yield any bacteria on the blood agar plate and the chromogenic medium (Oxoid) after 48 h of incubation in 5 % CO2. The Gram stain, however, showed 4+ leukocytes and 4+ small Gram-positive rods (on a scale from 0 to 4+). Incubation of the blood agar plates was prolonged and showed small colonies after 5 days. Routine identification methods [VITEK2 and API Coryne (bioMérieux)] did not return an acceptable result (probability scores were too low for a reliable identification). Finally, the strain was isolated and identified as Actinobaculum schaali by PCR techniques (sequence analysis of the 16s ribosomal DNA). Based on these results, treatment was started with amoxicillin 500 mg three times a day for 3 weeks. After this treatment, the patient was completely asymptomatic. Physical examination showed soft epididymis on both sides and no pain on palpation.

Inflammation of the epididymis causes pain and swelling that is almost always unilateral and relatively acute in onset. In some cases, the testes are also involved in the inflammatory process, called an epididymo-orchitis. In young men, epididymitis is mainly associated with sexually transmitted diseases such as chlamydia infection and gonorrhoea. However, the majority of cases occur in elderly men and are mainly caused by common urinary tract pathogens (members of the Enterobacteriaceae family). Bladder outlet obstruction and urogenital malformations are risk factors for this type of infection.

In this case report, A. schaali, present in high numbers in the urine culture, was considered to be the causative pathogen of the epididymitis. The genus Actinobaculum was first described by Lawson et al. (1997), and is closely related to the genera Actinomyces and Arcanobacterium. To date, four species of Actinobaculum have been described: A. suis, A. massiliense, A. urinale and A. schaali (Lawson et al., 1997; Reinhard et al., 2005; Bank et al., 2010). A. suis is known to cause urinary tract infections and abortions in sows (Wolde-meskel et al., 2002). A. massiliense and A. urinale have been identified as a cause of chronic cystitis in elderly women (Greub & Raoult, 2002; Hall et al., 2003). A. massiliense might also be involved in superficial skin infections (Waghran, 2004). It is thought that Actinobaculum species are part of the commensal flora of the human genital or urinary tract (Lawson et al., 1997).

A. schaali is a CO2-dependent, Gram-positive coccoid rod. Because of its similarity to normal bacterial flora on skin and mucosa in Gram stains, A. schaali is easily considered part of the normal flora. A. schaali grows very slowly and only forms visible colonies on agar plates after 3 to 5 days of incubation. Most laboratories, however, use urine culture procedures that incubate culture media only for 1 or 5 days, thus failing to detect the presence of A. schaali. Furthermore, urine cultures are usually not incubated under anaerobic conditions or with additional CO2 (5 %), which contributes to the failure to detect A. schaali since it is a CO2-dependent rod.

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Actinobaculum schaali is a Gram-positive coccoid rod that causes various infections in humans and is easily overlooked in cultures. A. schaali has long been thought to be of low prevalence and limited invasive potential, causing benign cystitis in elderly patients with underlying urological conditions. Here, we report the first case of epididymitis caused by this bacterium.
However, recent studies show otherwise. Bank et al. (2010) showed that 22% of 252 urine samples from patients over 60 years of age were positive for *A. schaalii* using real-time PCR. In addition, recent studies have described cases of skin abscesses, urosepsis, osteomyelitis, endocarditis and necrotizing fasciitis caused by *A. schaalii*, indicating that this bacterium, previously considered harmless, has invasive potential (Haller et al., 2007; Sturm et al., 2006; Vanden Bempt et al., 2011; Hoenigl et al., 2010; Beguelin et al., 2011). Our case report adds epididymitis to this list.

The European Association of Urology Guidelines (Grabe et al., 2012) prefers fluoroquinolones as a first-choice treatment for epididymitis. Only a few studies have measured the penetration of antimicrobial agents into the epididymis and testes in humans. Fluoroquinolones were chosen to be the drug of choice because these antibiotics penetrate well into the tissues of the urogenital tract and Enterobacteriaceae members are commonly susceptible (Grabe et al., 2012). However, *A. schaalii* has been claimed to be partially resistant to quinolones. Nielsen et al. (2010) isolated 76 strains of *A. schaalii*, all of which were resistant to trimethoprim and ciprofloxacin, but susceptible to ampicillin and cefuroxime. Cattoir et al. (2010) isolated 48 strains, all of which were resistant to ciprofloxacin, whereas 90% and 96% remained susceptible to levofloxacin and moxifloxacin, respectively. All isolates were resistant to metronidazole and only 15% were susceptible to co-trimoxazole. Reinhard et al. (2005) isolated nine strains susceptible to penicillin, cefuroxime, amdinocillin, nitrofurantoin, tetracycline and clindamycin. Reduced susceptibility was seen for ciprofloxacin and gentamicin. Regarding these results, it has to be kept in mind that susceptibility testing in *A. schaalii* has not been standardized and differences in testing procedures are likely to exist.

Overall, treatment with β-lactams has been used successfully in several cases. Unfortunately, the optimal duration of antimicrobial drug treatment with β-lactams is not clearly defined, although it has been suggested that a prolonged course of several weeks may be required (Reinhard et al., 2005; Bank et al., 2010; Haller et al., 2007; Hoenigl et al., 2010; Beguelin et al., 2011). Excluding other causative pathogens by appropriate techniques (Gram stain and culture) before and during prolonged treatment remains important.

Concluding, this case report shows that *A. schaalii* can cause epididymitis, which is therapy resistant to standard empirical treatment for this disease. The literature has already shown that infections with *A. schaalii* are more prevalent than previously thought. *A. schaalii* is easily overlooked due to its resemblance with normal urinary tract flora, its slow growth rate and its CO₂ dependency. Infection with *A. schaalii* should be suspected in patients with clinically documented urinary tract infections who do not respond to treatment with ciprofloxacin or co-trimoxazole and who show lack of growth after normal culture procedure. In addition, unexplained chronic pyuria, showing many Gram-positive rods in the Gram stain without any growth after 2 days on standard culture media, should trigger microbiologists to be aware of the presence of *A. schaalii*. In that case, urine samples should be incubated for 5 to 7 days on blood agar media under anaerobic conditions or in 5% CO₂.

References


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