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URINARY TRACT INFECTION WITH ATYPICAL MYCOBACTERIA

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ABSTRACT

We present a case of disseminated atypical mycobacterial (*Mycobacterium intracellulare*) infection of the urinary tract. The patient had anhydrous ectodermal dysplasia and an unrelated defect in cell-mediated immunity. The infection resulted in a lengthy ureteral stricture with resultant hydronephrosis and diminished kidney function. A review of the literature revealed only 13 previous cases of atypical mycobacterial infection of the urinary tract. It is important to distinguish between simple colonization by these organisms and actual infection. Differentiation between atypical mycobacterial infection and urinary tuberculosis is important in determining the therapeutic regimen.

Although first described in 1918 it was not until Runyon's classification of atypical mycobacteria in 1959 that these organisms came under closer scrutiny. Runyon categorized the many varieties of atypical mycobacteria into 4 groups, based upon color and growth characteristics.¹ Of these groups I and III (the photochromogens and the non-photochromogens, respectively) are those most pathogenic in humans.² The vast majority of cases represent pulmonary involvement but systemic dissemination also is known to occur.^{3,4} Nonetheless, genitourinary tract infection has been reported only rarely, with only 13 cases reported previously in the literature, some of which were described incompletely.

Herein we describe a child with anhydrous ectodermal dysplasia in whom a ureteral stricture developed secondary to infection by *Mycobacterium intracellulare* (Battey's bacillus). We relate the features of our case to those previously reported.

CASE REPORT

A 14-year-old white boy was hospitalized with increasing pedal edema, ascites and diarrhea 5 days in duration. Bloody stools developed 3 days before presentation. There were no complaints referable to the urinary tract.

When the patient was 33 months old anhydrous ectodermal dysplasia was diagnosed. He had had numerous hospital admissions for soft tissue infections, including meningitis, pneumonia, otitis media and cellulitis. Immunologic testing revealed recently a defect in cell-mediated immunity, unrelated to the ectodermal dysplasia.⁵

Three years ago disseminated *M. intracellulare* developed, as evidenced by positive cultures of the urine, bone marrow, liver, lymph nodes, cerebrospinal fluid, stool and blood. Hepatosplenomegaly had been present continuously since then. Numerous antimicrobials had been tried and found ineffective, including isoniazid, para-aminosalicylate, ethambutol, streptomycin, kanamycin, capriomycin, erythromycin, ethionamide, rifampin, levamisole, trimethoprim-sulfamethoxazole and pyrimidine. Parenteral transfer factor was begun 4 months before hospitalization with some subjectively assessed reduction in the hepatosplenomegaly. However, cultures remained positive. At hospitalization the patient was receiving oral transfer factor, clofazimine and streptomycin.

The family history is significant, since his brother died when he was 2 years old of diarrhea and dehydration. At autopsy acid fast organisms were present in the gastrointestinal tract. These organisms were not characterized further.

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The patient was an afebrile, cachectic-appearing young boy, small for his age. The teeth were pointed and ridden with cavities, the hair was sparse and white, and the skin was dry and atrophic, with numerous scars and hypopigmented areas secondary to healed subcutaneous abscesses. The abdomen was more protuberant than usual and a fluid wave was believed to be present. Hepatosplenomegaly was palpably evident. There was a moderate amount of pitting pedal edema.

Laboratory values showed only a mild hepatic dysfunction unchanged from previous values. Electrolytes were normal except for slight hyponatremia. White cell count was 5,000 with 18 per cent bands. Creatinine was 0.4 mg./dl. and blood urea nitrogen was 11 mg./dl. Urinalysis was normal.

The diarrhea abated soon after hospitalization, although testing for occult blood remained intermittently positive. There was no gross melena. Coronavirus and *M. intracellulare* were cultured from the stool. An upper gastrointestinal barium examination revealed possible nodularity and thickening of the small bowel wall, as well as slow passage of barium. A gallium scan demonstrated abnormal uptake in the left upper quadrant and mid epigastrium, probably within the small bowel. These findings were interpreted as compatible with intestinal mycobacterial involvement.

During the third month of hospital stay hematuria, pyuria and an increase in serum creatinine to 0.8 to 0.9 mg./dl. occurred. Urine output remained normal. An abdominal ultrasound and a computerized tomographic body scan confirmed the presence of massive hepatosplenomegaly. The computerized tomographic scan also showed an unexpectedly large left kidney with hydronephrosis. A subsequent excretory urogram verified the left hydronephrosis and a normal right kidney, ureter and bladder. Retrograde pyelography revealed the source of the left pyelocaliectasis to be a 2.5 cm. long stricture of the left mid ureter (see figure). Cultures of urine taken from the left renal pelvis yielded a *Pseudomonas* species. Unfortunately, cultures for mycobacteria were not performed at this time.

The patient was treated for the *Pseudomonas*. Since then urine cultures for bacterial and mycobacterial growth had been negative. Serum creatinine remained at 0.8 mg./dl. and mild hematuria, pyuria and proteinuria persisted. Subsequently, the patient died. Postmortem pathologic examination revealed diffuse foci of atypical mycobacteria with poorly organized cellular reaction. Organisms were present in both kidneys and in the strictured region of the left ureter.

DISCUSSION

The table summarizes the findings in 14 cases, including our own, of atypical mycobacterial genitourinary involvement. As indicated in the table information concerning some of these

Summary of reported cases of genitourinary atypical mycobacterial infection

References	Age—Sex	Clinical Findings	Radiologic Findings	Organism/Source
Wood and associates ³	54—M	Orchitis, prostatitis, epididymitis	Bilat. hydronephrosis and nephroureterolithiasis	Photochromogen/sputum
Lattimer and Boyes ⁷	2 children	—	—	Photochromogen/sputum, urine
Faber and associates ⁹	27—F	Hematuria	Irregular, dilated calix	M. intracellulæris/urine, sputum
Lester ⁶	2 pts.	—	—	M. kansasii/urine
Kilbridge and associates ⁴	62—M	Fever, pancytopenia	—	Photochromogen/renal granuloma
Merckx, J. J., Karlson, A. G. and Carr, D. T.: Mayo Clin. Proc., 38: 271, 1963	—	Testicular swelling, epididymitis	—	Photochromogen/—
Newman, H.: J. Urol., 103: 403, 1970	52—F	Pyuria, dysuria	Ureteral obstruction	M. intracellulæris/urine
Hepper, N. G., Karlson, A. G., Leary, F. J. and Carr, D. T.: Mayo Clin. Proc., 46: 387, 1971	64—M	Epididymitis	—	M. kansasii/epididymitis
	57—M	Hematuria, pyuria	Prostatic calculi	M. kansasii/prostate
Pergament, M., Gonzalez, R. and Fraley, E. E.: J.A.M.A., 229: 816, 1974	62—F	Hematuria, dysuria	Calcified, non-functioning lt. kidney; rt. ureteral stricture	M. intracellulæris/urine
Listwan and associates ¹¹	40—M	Fever, decreased renal function	—	M. kansasii/kidney
Present case	14—M	Hematuria, pyuria, decreased renal function	Ureteral stricture	M. intracellulæris/urine, stool, cerebrospinal fluid, lymph nodes, liver, bone marrow, blood

Left retrograde pyelogram demonstrates 2.5 cm. long ureteral stricture adjacent to 4th lumbar vertebra. Contrast material flows proximal to stricture revealing moderate pyelocaliectasis.

cases is scant. In others, such as those reported by Lester,⁶ and Lattimer and Boyes,⁷ culture of *M. kansasii* in the urine was not associated with evidence of urinary tract disease. This is significant in that mycobacterial colonization and infection appear not to be synonymous. In 1970 Klotz noted 306 cases of urinary colonization with atypical mycobacteria, none of which was reportedly associated with observable urinary tract disease.⁸ Thus, we believe that adequate documentation of atypical mycobacterial infection must include either positive culture of genitourinary tissue specimens and/or positive urine cultures in conjunction with some combination of radiologic, pathologic or laboratory evidence compatible with granulomatous genitourinary disease. In view of Klotz's study the occurrence of these

combinations of findings would seem exceedingly rare.

Several facets of our case are uncommon with respect to those previously reported. The patient was only 14 years old. Lattimer and Boyes referred to 2 children in whom urine culture yielded group I atypical mycobacteria but no supporting evidence of genitourinary disease was found.⁷ In no other reported case was a patient <27 years old.⁹ In discussing the reasons for the rarity of urinary tuberculosis in children, Ehrlich and Lattimer have cited the usual lag between initial cortical dissemination and clinical or radiologic manifestations.¹⁰ Our patient possessed a defect in cellular immunity, which may have hastened the progression of the disease. While decreased immunocompetence appears to have a critical role in our case this is not generally true of others in the literature. Of the 13 cases reported only 2 had a recognized immune deficiency, which was an unexplained pancytopenia.^{4,11} Pancytopenia has been described in other instances of disseminated mycobacterial infections as well but it is not ascertained whether this represents cause or effect of this disease.¹²

Finally, the radiographic manifestations of our case are typical of those usually associated with genitourinary tuberculosis. Although *Pseudomonas* superinfection was present it is not reasonable to expect that this was responsible for renal non-function and the exceedingly long left ureteral stricture. Rather, the length and irregularity of the stricture are characteristic of granulomatous infection and healing, while the renal non-function may be attributed to obstruction and/or the effects of diffuse renal parenchymal infection.¹¹ The postmortem examination, revealing the presence of atypical mycobacteria in the kidneys and region of the ureteral stricture, supports this contention.

Although unusual, genitourinary infection by atypical mycobacteria is significant and potentially life-threatening. It is important to differentiate correctly this entity not only from pyogenic infections but from tuberculosis as well, because relative resistance of these organisms to conventional antituberculous therapy may necessitate the institution of more heroic measures.

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