

Actinomyces Causing Recurrent Perianal Fistulae

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ABSTRACT

Actinomyces is a rare but easily curable infection that should be considered in the differential diagnosis of perianal fistulizing disease. We present the case of a 26-year-old woman with complex perianal fistulae, including trans-sphincteric and suprasphincteric fistulous tracts and a rectovaginal fistula, requiring multiple abscess drainages, seton placement, and antibiotic courses, with little improvement. After extensive investigation, *Actinomyces* spp. was identified in anal cytology. The patient underwent a 6-week course of intravenous penicillin followed by oral amoxicillin, with remarkable improvement. This case illustrates the importance of pursuing less common diagnoses in refractory complex perianal disease, such as actinomyces.

INTRODUCTION

Actinomyces is a rare disease caused by *Actinomyces* spp., a gram-positive filamentous bacillus that often colonizes the human urogenital and digestive tracts. The mechanisms through which *Actinomyces* spp. becomes pathogenic are not completely understood, but it has been proposed that tissue injury may be the initiating event leading to its deep penetration and proliferation.¹ The most common infection sites are cervicofacial, osteoarticular, respiratory, urogenital, and gastrointestinal. Diagnosis is challenging and often delayed, which may in part be explained by the specificities of its isolation because it requires prolonged culture in an appropriate medium. Gram staining and histopathologic examination are usually more sensitive in the diagnosis, the latter possibly revealing the presence of yellowish granules containing bacilli and inflammatory cells, the so-called sulfur granules.¹ Primary anorectal and perianal actinomyces are particularly rare forms of the disease.²⁻⁵

CASE REPORT

A 26-year-old black woman, born in Cape Verde and living in Portugal since the age of 3 years, presented with anal and vaginal suppuration. She had a known history of two deliveries, including a cesarean section in 2005 and a vaginal delivery in 2011 without episiotomy. During the second trimester of her second pregnancy, she had a Bartholin's cyst requiring surgical drainage. She was using oral contraceptives and denied previous use of intrauterine devices. She was a smoker (6–7 cigarettes/day) and did not drink alcohol. She denied recent travels to Africa.

The patient was first admitted in 2013 for complaints of anal and vaginal suppuration lasting 2 weeks, without fever or other accompanying symptoms. Perianal inspection revealed the presence of an external fistulous orifice in the posterior perineum, close to the left labium majus. On rectal examination, there was a palpable induration of the left anterior rectal wall. Her gynecological examination was unremarkable.

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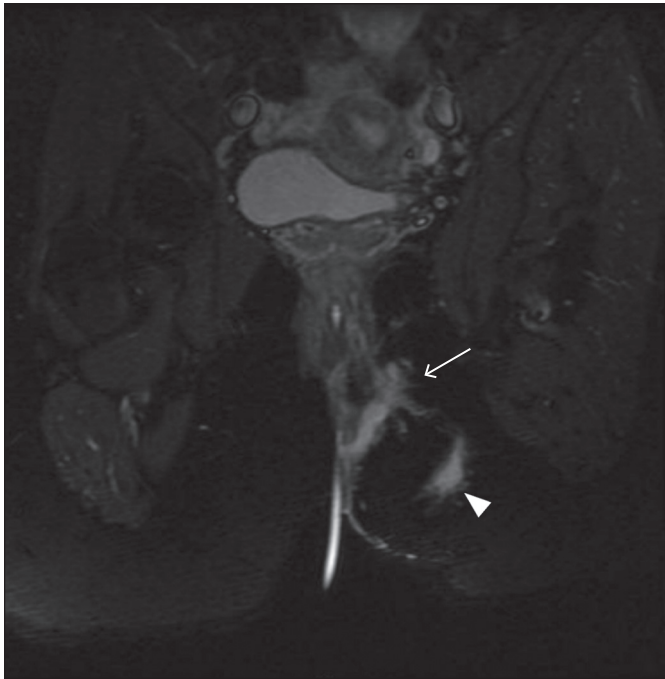


Figure 1. Coronal pelvic magnetic resonance image before antibiotic therapy, depicting a short, trans-sphincteric component of the complex perianal fistula dividing in the ischioanal space (arrow), with significant densification of surrounding fat (arrowhead).

A pelvic computed tomography excluded the presence of abscesses or other pelvic masses. The patient was put on antibiotics and underwent rectal examination under anesthesia, where no abscess was found. Two fistulous tracts were identified and treated with seton placement, including one tract involving the vagina. Biopsies of the indurated rectal wall were taken, and pus was collected for analysis. Pelvic magnetic resonance imaging 1 month after hospital discharge revealed multiple fistulous tracts, including trans-sphincteric and suprasphincteric tracts and a rectovaginal fistula (Figure 1).

An extensive work-up to exclude inflammatory bowel disease (IBD) was done, including an upper endoscopy, colonoscopy with ileoscopy, small bowel capsule endoscopy and computed tomography enterography, which were unremarkable. Endoscopic biopsies of the colon, terminal ileum, and stomach did not show any significant findings. Anti-*Saccharomyces cerevisiae* antibodies titer was also negative. Cytology and cultural exams of both rectal and vaginal exudates and biopsy of the rectum were negative for neoplastic cells and microorganisms, including *Mycobacterium tuberculosis*. Serologies of *Chlamydia trachomatis* and human immunodeficiency virus were also negative, as was a VDRL test for syphilis.

In the following 3 years, the patient underwent multiple abscess drainages, fistulae treatment by seton placement, and empiric antibiotic courses, with no overall improvement (Figure 2). This situation had a high impact on the patient's



Figure 2. Photograph showing several setons in situ, the most anterior corresponding to the rectovaginal tract (arrowhead). The most posterior external orifice shows significant granulation tissue (white arrow). A scar from abscess drainage is visible close to the left labium majus (black arrow).

quality of life, eventually leading to clinical depression. Despite high suspicion of inflammatory bowel disease, a complete endoscopic investigation again was negative. In 2016, repeated anal cytology revealed the presence of bacterial

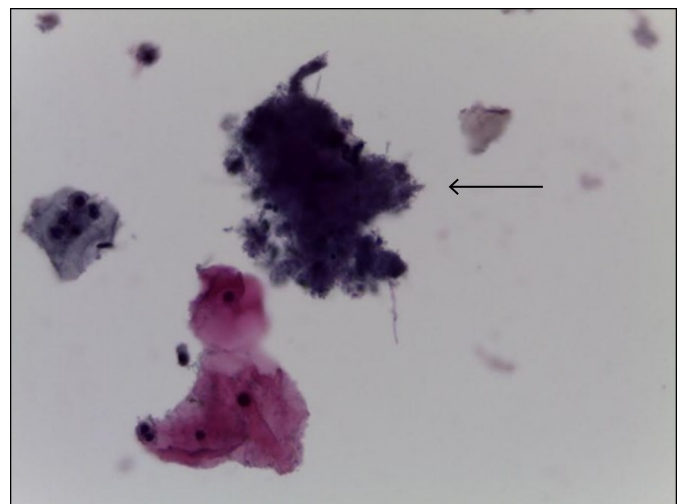


Figure 3. Anal cytology (papanicolau stain, 40x) identified filamentous bacteria compatible with *Actinomyces* species (arrow).



Figure 4. Perianal examination showing significant improvement of the perianal fistulae after removal of setons, with most orifices being closed, no signs of new fistulous tracts, and no exudation. The fistulous tract involving the vagina is still visible, but with no exudation (arrowhead). The anal sphincter complex has preserved morphology and tonus (arrow).

colonies compatible with *Actinomyces* spp. (Figure 3). The patient was then hospitalized and completed a 6-week course of intravenous (IV) penicillin G, after which she was started on oral amoxicillin, which is to be maintained for 12 months. This resulted in a remarkable clinical improvement, allowing for the removal of setons (Figure 4). The reevaluation magnetic resonance imaging, performed after only 5 weeks of antibiotic therapy, showed almost complete resolution of the fistulous tracts (Figure 5). Currently, after 6 months of oral amoxicillin, the patient is clinically well, with no perianal suppuration, and reports a significant improvement in her quality of life. Although notably reduced, the vaginal fistula is still patent and will possibly require surgical correction.

DISCUSSION

Perianal actinomycosis is a very rare disease that has only been described in isolated case reports and small series.²⁻¹⁵

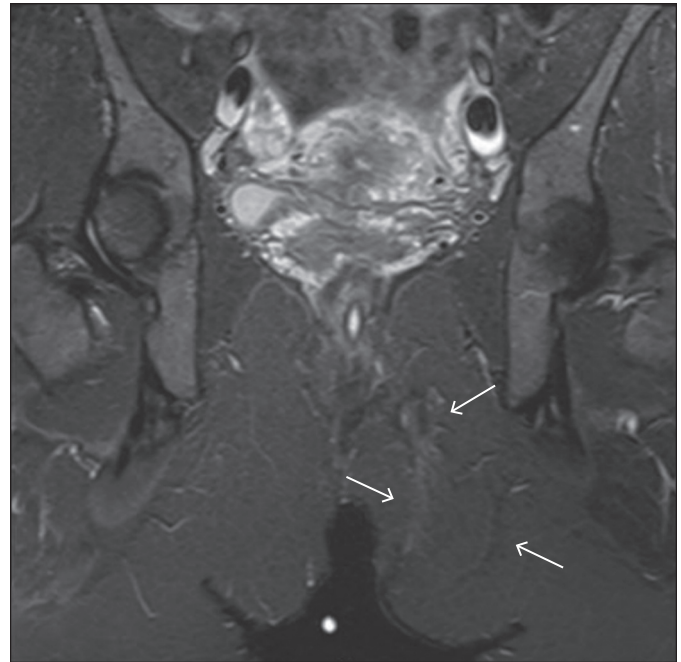


Figure 5. Coronal pelvic magnetic resonance image after 5 weeks of penicillin, showing almost complete resolution of the fistulous tracts (arrows).

Although several species of *Actinomyces* have been implicated in this condition, the most frequently isolated is *Actinomyces israelii*.^{1,4,12} It usually presents as recurrent perianal fistulizing disease, but it may also manifest as a painful mass.^{4,7,8,14,16} Perianal actinomycosis has a clear male predominance (male to female ratio is 9.5:1) and is more frequent in the setting of predisposing conditions, namely human immunodeficiency virus infection, diabetes mellitus, or alcoholism.^{2,4,9,10} The use of intrauterine devices was been implicated in pelvic actinomycosis in women.¹ Differential diagnoses include inflammatory bowel disease, tuberculosis, malignancy, hydradenitis suppurativa, and sexually transmitted diseases such as lymphogranuloma venereum and syphilis.^{4,17} Although several antibiotic regimens may be used, the gold standard remains intravenous penicillin, specifically 10–20 million units per day of penicillin G over 2–6 weeks, divided 4 times a day, followed by therapy with penicillin V or amoxicillin for 6–12 months.^{5,16}

Our case of complex perianal fistulizing disease with a severe and protracted course is particularly rare because the patient was female with no identifiable predisposing condition for perianal actinomycosis. To our knowledge, she is the youngest patient reported with this diagnosis. Although Crohn's disease was suspected, the absence of gastrointestinal involvement after extensive investigation did not allow for a definite diagnosis and deferred the beginning of biologic therapy, as no clear evidence of isolated perianal Crohn's disease exists to date.¹⁸ This led to the continuous search for

other etiologies and ultimately to the identification of a rare but readily treatable disease. The delayed diagnosis was probably attributable to the difficulties inherent to the isolation of *Actinomyces*, requiring an anaerobic atmosphere and incubation period of up to 15–20 days, which is not a standard procedure. Indeed, only repeated anal cytology was able to identify the microorganism.¹

DISCLOSURES

Author contributions: M. Ferreira Cardoso, C. Carneiro, L. Carvalho Lourenço, and A. Costa wrote the manuscript. C. Graça Rodrigues, S. Alberto, A. Alagoa João, R. Rocha, and V. Geraldes revised the manuscript. All authors approved the final version of the manuscript. C. Carneiro is the article guarantor.

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