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## CASE REPORTS

# Extrapulmonary Tuberculosis – a Rare Cause of Difficult Evolution in a Case Of Extensive Anoperianal Suppuration

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## Abstract

Extrapulmonary tuberculosis is very rare, representing around 15% of all tuberculosis (TB) cases. Intestinal localization of extrapulmonary TB occurs in 1% of the cases and is usually associated with HIV infection. The last portion of the ileum and the ileocecal region are most frequently involved, while perianal localization is extremely rare and has clinical and paraclinical aspects similar to Crohn's disease, thus leading to difficulties in establishing the diagnosis. We present the case of a 41-year-old patient who came to the proctology department with perianal pain and purulent discharge. He was diagnosed by clinical examination and colonoscopy with multiple simple and complex perianal fistulas complicated with a supralelevator abscess. Multiple biopsies were taken and the result was necrotizing granulomatous inflammation. The Ziehl-Neelsen test confirmed the suspicion of perianal TB.

**Keywords:** tuberculosis, perianal, fistula, extrapulmonary

## Rezumat

Tuberculoza extrapulmonară este foarte rară, reprezentând în jur de 15% din toate cazurile de tuberculoză. Localizarea intestinală reprezintă 1% din cazurile de TB extrapulmonară și apare de obicei la pacienții infectați HIV. Ileonul terminal și regiunea ileocecală sunt cel mai frecvent implicate în cazul tuberculozei intestinale; localizarea perianală este extrem de rară și prezintă aspecte clinice și paraclinice similare cu Boala Crohn, ducând astfel la dificultăți în stabilirea diagnosticului de certitudine. Prezentăm cazul unui pacient în vârstă de 41 de ani ce se prezintă la medic pentru durere perianală și scurgeri purulente. Acesta este diagnosticat clinic și colonoscopic cu multiple fistule perianale simple și complexe complicate cu abces supralevatorian. S-au prelevat multiple biopsii cu rezultat de inflamație granulomatoasă necrotizantă. Testul Ziehl -Neelsen a confirmat suspiciunea de TBC.

**Cuvinte cheie:** tuberculoză, perianal, fistula, extrapulmonară

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## INTRODUCTION

TB is an infectious disease with mainly respiratory transmission that is still quite widespread throughout the world (especially in low-income countries) despite today's wide availability of antibiotics.<sup>1</sup> The causative agent is an acid-alcohol-resistant bacillus called *Mycobacterium Tuberculosis*. Intestinal involvement represents 1% of TB cases, with perianal localization accounting for less than 0,7% of the intestinal TB cases (mostly in HIV-infected patients) often being misdiagnosed as Crohn's disease or other granulomatous disease.<sup>2,3,4</sup>

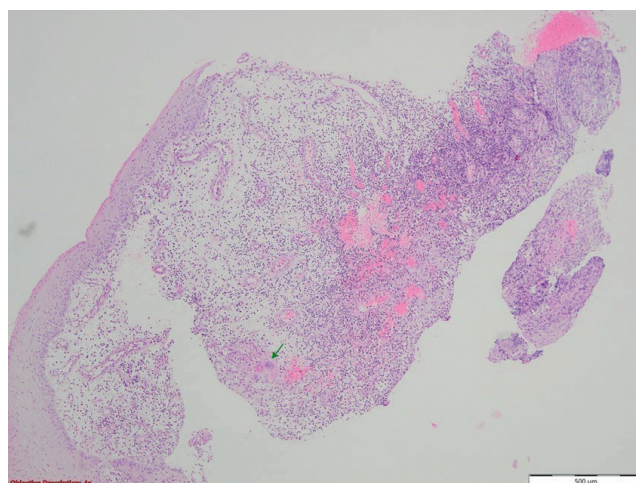
## CASE REPORT

We present the case of a 41-year-old patient with purulent perianal discharge and continuous ano-perianal pain. From the patient's history we note multiple operations in the anoperianal area for abscesses and fistulas during the last 3 years. Following the clinical examination, the anoscopy and the colonoscopy, the patient is diagnosed with a chronic anal fissure at 12 o'clock in left lateral decubitus complicated with a complex posterior perianal horseshoe fistula complicated with a supralelevator abscess, and two other fistulas located at 12-1 o'clock and 12-3 o'clock in left lateral decubitus.

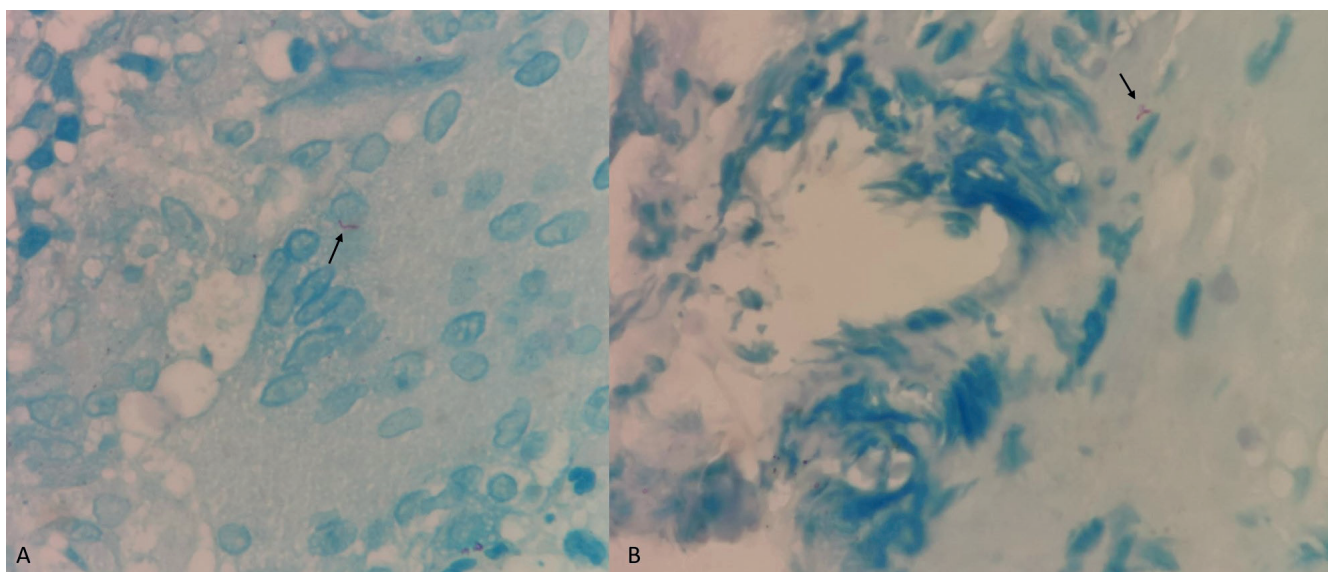
Surgical intervention is decided and fistulotomies for simple fistulas (12-1, 12-3) and partial fistulotomy with drainage seton placement (for the supraleva-

tor abscess) are performed. Multiple biopsies are taken from the fistulous tracts.

The histopathological examination describes fragments of fibro-conjunctival-adipose tissue showing several granulomas consisting of epithelioid histocytes, multinucleated Langhans giant cells, some of the granulomas showing eosinophilic, granular amorphous necrosis. The histopathological aspect concludes the presence of a necrotizing granulomatous inflammation in the biopsies from the fistulous tract, therefore the diagnosis of perianal TB is suspected and the Ziehl-Neelsen staining is performed, with a positive result thus confirming the diagnosis.



**Figure 2.** Fistulous tract lined by squamous epithelium, with underlying granulation tissue that includes a focal aggregate of multinucleated histiocytes (→); HE, 40x



**Figure 1 (A, B).** The presence of acid-alcohol-resistant bacilli is evident – the microorganisms are staining in red (→) on a blue background; Ziehl-Neelsen stain, 1000x

## DISCUSSION

Tuberculosis affects the ano-perianal region very rarely (less than 1% of intestinal TB) and occurs mainly in men, usually after the age of 40, aspects also observed in our patient.<sup>5</sup>

In most cases, perianal tuberculosis is associated with pulmonary TB and is much more common in immunocompromised patients (HIV-infected patients) and can occur as a secondary outbreak of pulmonary TB or by direct implantation on the pre-existing suppuration, enhanced by the low immunity of the host.<sup>6</sup> In this case the patient had no lung lesions at the time of diagnosis and the ELISA test for HIV was negative.

This pathology manifests with perianal pain, purulent discharge and determines multiple complex perianal fistulas that can persist and recur despite adequate surgical management, thus generating a complex and non-specific clinical picture that delays the diagnosis of TB.<sup>7</sup> In this case the diagnosis was made promptly after patient's presentation to the clinic and specific treatment was initiated quickly; the patient has not been detected with disease relapse during follow-up after this surgical intervention. In this case the imagistic findings regarding the anoperianal lesions are missing since clinical examination and anoscopy made the diagnosis, making additional investigations become unnecessary.

Differential diagnosis of perianal TB includes cutaneous amoebiasis, ulcerative colitis, herpes simplex syphilis, deep mycosis and venereal lymphogranuloma, but the most difficult differential diagnosis is with Crohn's disease considering the clinical, the colonoscopic and the histological characteristics superimposed with perianal TB; it was proposed that ASCA, antibodies often present in the serum of patients with Crohn's disease, can help in differentiating these two conditions, but there are studies that invalidated its use.<sup>8,9,10</sup>

Multiple diagnostic methods have been implemented in the case of TB. The most recent and, according to some studies, the most sensitive, is through the PCR technique that manages to diagnose tuberculosis in approximately 48 hours; however, the simplest and fastest diagnostic method remains biopsy of the lesions and histopathological analysis of the slides, which has an accuracy of over 70%, method used in our case as well.<sup>5</sup>

Gross examination of the resection specimens reveals different aspects, depending on the evolution

stage of the infection, and it is correlated with the histopathological aspect.

Macroscopically, tuberculosis appears as unique or multiple confluent, firm, grey-colored nodules, but once the infection evolves, white-yellow cheesy areas can be observed, which represent the gross expression of the caseous necrosis<sup>10</sup>.

Tuberculosis represents a specific type of inflammation. Microscopic examination reveals necrotizing granulomas (Koster follicle), which include four pathognomonic elements: uninucleated (epithelioid) and multinucleated histiocytes (Langhans cells), lymphocytes (at the periphery) and eosinophilic, acellular, granular areas of caseous necrosis (central). The granulomas tend to confluence and can have a variable morphology since the only mandatory elements are the epithelioid cells and the peripheral rhyme of lymphocytes<sup>11,12,13</sup>.

Special stains should be performed, in order to highlight the Koch bacillus.

Examination of Acid Fast-Bacilli under a fluorescent microscope (auramine-rhodamine stain) determines a gold-yellow stain of the microorganisms, on a dark background. A positive result at this staining must be mandatory confirmed by Ziehl-Neelsen stain. At this staining, Mycobacterium tuberculosis appears as red, granular bacilli on a blue background.

Some of the lesions can be paucibacillary, and that is why a negative Ziehl-Neelsen staining (after performing multiple slides on the same paraffin block or on different blocks) doesn't exclude the infection<sup>10</sup>.

## CONCLUSION

Perianal TB is a pathology that must be taken into consideration in patients with complex perianal fistulas with long term evolution despite adequate local treatment, including the immunocompetent individuals without a known history of pulmonary infection. Raising the suspicion of TB and subsequently biopsying these perianal lesions is very important in the early diagnosis of this disease, which can have an unfavorable evolution in the absence of specific treatment.

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