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Case Report

Giant Gluteal Mass : An Unusual Presentation of Anal Tuberculosis

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Abstract

Introduction: Anal tuberculosis is an extremely rare extrapulmonary form of tuberculosis even in highly endemic countries. Its non-specific symptoms often lead to a delayed diagnosis. Tubercular anal fistulae must be adequately recognized because they require a specific course of treatment.

Presentation of case: We present an unusual case of a large painless ball-shaped lump arising due to tuberculous infection of the anal and perianal region. The treatment course lasted 6 months. The patient showed signs of recovery after 2 weeks of treatment, and complete remission was achieved.

Conclusion: The rarity of anal and perianal tuberculosis and its nonspecific symptoms cause it to go unrecognized, delaying the specific treatment. It is recommended to evaluate for tuberculosis in case of atypical anal and perianal lesions, especially in highly endemic regions. Complex anal fistulae can heal after antitubercular therapy alone, especially when their surgical drainage can cause an extensive sphincter damage.

Keywords: Giant Gluteal; Mass; Anal; Tuberculosis

Introduction

Up to this day, tuberculosis is a continuing issue both in developing and developed countries as a result of large immigrant populations. Anal and perianal locations are extremely rare, forming less than 1% of all gastrointestinal tuberculosis cases. Clinical and radiological presentations can be misleading and cause the disease to go unrecognized. The present case has interesting clinical features, since the very peripheric and very large gluteal swelling was not associated with pain nor with inflammatory perianal symptoms.

Case Presentation

A 61-year-old man presented with a persistent massive painless swelling in inferior and external part of the right gluteal region of 7 months duration, which was rapidly progressing, associated with

increasing discomfort. He had no significant personal nor family medical history, especially no history of contact with Koch's infection. At presentation, he was of average build and afebrile. Physical examination revealed a visible heterogeneous non-tender soft but firm mass measuring 5 -6 cm in diameter, in the right gluteal region. There were no signs of inflammation in the overlying skin and no adenopathy was clinically identified (Figure 3).

Pre-operatory pelvic magnetic resonance imaging (MRI) revealed a transphincteric anal fistula complicated by a large peripheric heterogenous hyperintense collection of the right gluteal soft-tissue, measuring approximately 100x39x44 mm in size and containing air-fluid levels. There was no rectal wall thickening, and no pelvic lymphadenopathies were identified (Figure 1-3).

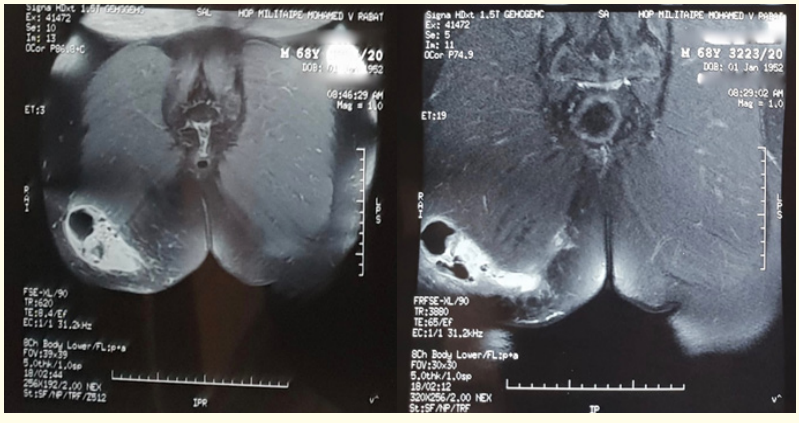

Figure 1: Axial T2-FS MRI cut on the level of proximal femur just distal to the lesser trochanter showing pattern of abscess extension distally.

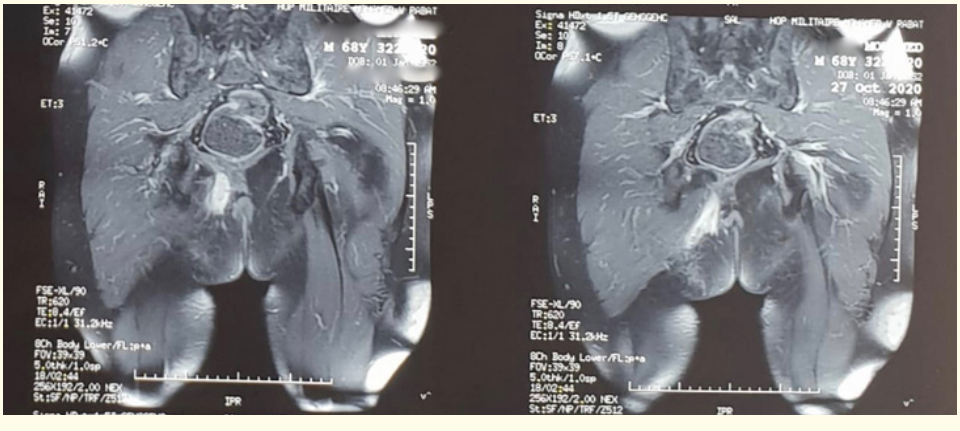

Figure 2: Coronal T2-FS MRI images of pelvic and bilateral femur showing extension of the abscess distally around the greater trochanter beneath the gluteus maximus muscle and tensor fascia lata.

Figure 3

The patient was admitted for surgical exploration. On the operating table, an exploratory puncture aspiration syringe did not bring anything and was therefore inconclusive (Figure 4,5).

The mass and surrounding tissue were excised. They contained dense purulent necrotic material and air (Figure 5). The clinical impression was that of a soft-tissue tumor involving the gluteal

Figure 4

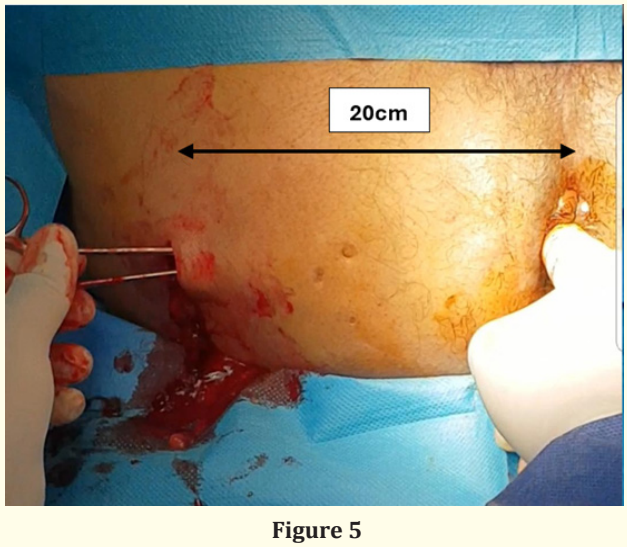

Figure 5

region. Along with this mass, there was a large fistulous track measuring approximately 20cm, that opened up in the anal lumen (Figure 6). The atypical collection was drained and left open and we collected a pus sample and a biopsy from the wall of the residual cavity. Since the internal orifice of the fistula was too far from the very peripheric collection, we were concerned about sphincter preservation and we decided not to proceed to the drainage of the fistula using a seton, since it was going to be too damaging. The patient was discharged after 2 days, with instructions on proper wound care and antibiotics. Post-operative period was uneventful.

Figure 6

Histopathological examination disclosed chronic inflammation with giant cell granuloma and did not identify any caseous necrosis (Figure 7).

Colonoscopy showed normal colorectal mucosa and biopsies were unremarkable. Fecal calprotectin was within the normal

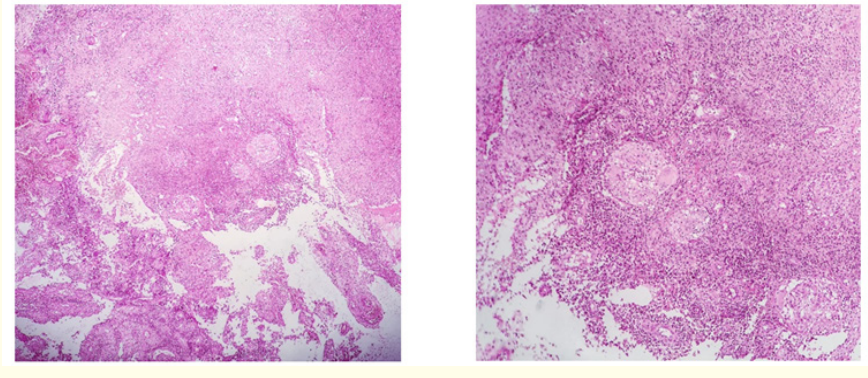

Figure 7: Haematoxylin-eosin stained slide (original magnification x40) showing granulomatous inflammation with Langhans giant cells.

range. The etiological diagnosis was suspected by histological examination of the surgically excised tissue and confirmed by a positive IGRA (Quantiferon) test result. There was no associated pulmonary tuberculosis.

Patient was started on antitubercular therapy, made of Isoniazid, Ethambutol, Rifampicin and Pyrazinamid daily. The plan was to continue this four drug regimen for a total of 4 months followed by a combination of Isoniazid and Rifampicin for a total of 2 months, while being followed as an outpatient. Overall, evolution was favorable, the gluteal wound and the fistula healed well.

Discussion

To this day, tuberculosis is still considered a major public health problem in Morocco [1].

In 2017, 30 897 new cases occurred (incidence rate: 88/100 000) and extrapulmonary tuberculosis, which can attack any organ with or without the lungs being affected [2,3], constituted 48 % of total tuberculosis cases[1]. Gastrointestinal tuberculosis forms 1% of extrapulmonary tuberculosis. The most common location is the ileocecal area, followed by the jejunum and the colon. This pattern can be explained by the abundance of lymphoid tissue in these regions and the affinity of BK where stasis is important [4,5]. However, tuberculous involvement of the anal and perianal region remains a very rare clinical entity even in countries where tuberculosis has a high prevalence (less than 1% of all gastrointestinal tuberculosis cases) [4-7]. Mycobacterium tuberculosis can reach the anal region mainly via ingestion of infected sputum or ingestion of bacilli from infected food products. The reactivation of latent loci formed during lymphatic or hematogenous dissemination of primary TB is another cause for perianal tuberculosis [8-11].

Anoperineal tuberculosis is a great mimicker. the disease often gets detected late because of its nonspecific symptoms and its slow insidious course. In fact, tubercular involvement of the anal and perianal region can occur in various clinical forms, which can be classified grossly as suppurative or ulcerative [12,13]. Anal suppurative pathologies are the most common clinical presentation, consisting mainly of anal fistulae, but also of anal abscess [14,15]. On the other hand, ulcerative skin manifestations such as painful erythematous and indurated perianal ulcers with a necrotic granular base have also been reported [16-20]. Constitutional symptoms related to tubercular infection can be present, in particular fever, chills, weight loss, anorexia and night sweats. In our patient, these symptoms were absent. he had a painless non-inflammatory swelling to begin with, which became uncomfortable in the last month due to its rapid progression in size.

A pelvic magnetic resonance imaging (MRI) is the modality of choice in order to further characterize and define the location and the extent of soft tissue growths [21]. Surprisingly, in our patient, the MRI scan showed an extensive inflammatory change in the deep subcutaneous tissues superficial to the right gluteus maximus muscle as well as a communication with the anal canal.

Nonspecific symptoms of perianal tuberculosis and similarities in clinical and radiological presentations with Crohn's disease, anal carcinoma or cryptoglandular fistula-in-ano make differential diagnosis challenging [22-25].

Since intestinal tuberculosis is a paucibacillary disease, demonstrating Mycobacterium bacilli using direct examination is difficult. Furthermore, the sensitivity of acid-fast bacilli culture ranges from 19% to 70% [26-28] with a turnaround time of 2 to 8 weeks [29].

A needle aspiration or excisional biopsy is mandatory for the differential diagnosis of anal tuberculosis. Histopathological diagnosis is usually suspected on the basis of the presence of epithelioid granulomas and can even be histologically confirmed in case of associated caseous necrosis, which is pathognomonic for tuberculosis. However, the presence of caseation is not constant and epithelioid granulomas can also be associated with Crohn's disease [30,31].

The newer tests like polymerase chain reaction (PCR) amplification, GeneXpert nucleic acid amplification test (NAAT) and interferon gamma release assays (IGRA) - Quantiferon - can also be used to confirm the diagnosis. However, a negative polymerase chain reaction (PCR) assays for the detection of Mycobacterium tuber-

culosis deoxyribonucleic acid (DNA) is not enough in order to exclude tuberculosis and a positive result is not always confirmatory. [32] Interferon gamma release assays (IGRA) – Quantiferon - is the most commonly used diagnostic technique in our hospital and is interesting in any BCG-vaccinated population because there are no false positives induced by the Bacillus Calmette-Guérin (BCG) vaccine. [33,34] Moreover, it has a specificity of 97.7% and a sensitivity of 85% in case of latent tuberculosis, especially in distinguishing it from Crohn's disease. Therefore, it appears to be a valuable tool for anoperineal tuberculosis diagnosis [35,36]. GeneXpert on tissue samples also has a high specificity for the detection of extrapulmonary tuberculosis and is therefore a valuable diagnostic tool [37,38].

Standard treatment of anal sepsis (consisting of a surgical drainage) in addition to antitubercular therapy remains the main recommended management strategy. Conventional anti-Mycobacterium regimens are combinations of Isoniazid, Ethambutol, Rifampicin and Pyrazinamid for two-months duration followed by Rifampicin and Isoniazid for four-months duration. An extension of this treatment course to 9-18 months is mandatory in case of complex fistulae or concurrent active tuberculosis [39-41]. However, anal fistula may sometimes heal after anti-tubercular treatment alone, as for our patient. Therefore, in case of complex tubercular fistulae, especially when surgery might involve an important anal sphincter damage, it could be more interesting to initially limit the treatment to antitubercular therapy and reassess the surgical indication according to the reponse to medical treatment.

Conclusion

Anal and perianal tuberculosis should be considered in case of persistent non-healing or recurrent anal abscess of fistula, especially in endemic TB areas. and. When microbiological identification is inconclusive or when histopathology doesn't detect caseation, the availability of new molecular techniques such as IGRA (Quantiféron) and GeneXpert are high-performing diagnostic tests. Accurate diagnosis is important in order to offer an appropriate course of treatment and reduce treatment delay. The availability of antitubercular therapy is associated with decreased morbidity and mortality. However, with the increasing incidence of multidrug-resistant bacilli, tuberculous complex anal fistulae could become a true therapeutic challenge in the near future.

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