

Thigh Swelling: A Rare Presentation of Tuberculosis

Joana Cachão, Denise Banganho, Hugo Teles, Sara Carmo, Eduarda Vicente

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Abstract

Tuberculosis cold abscess, also known as tuberculous gumma is a rare manifestation of cutaneous tuberculosis. A 15-year-old female was admitted for a tumefaction in the left thigh noted three weeks before. Six months prior, latent tuberculosis was diagnosed, and isoniazid was prescribed. The left thigh was enlarged with a soft, rounded mass with 8 cm of greater diameter and well-defined edges, slightly painful to palpation without fluctuation. Routine investigation revealed a sedimentation velocity of 50 mm in the first hour and magnetic resonance revealed a liquid collection of heterogeneous content suggesting a cold abscess. Needle aspiration was performed, and polymerase chain reaction identified *Mycobacterium tuberculosis*. Antitubercular therapy was started with four drugs for two months, maintaining isoniazid and rifampicin for eight months. The authors pretend to highlight the rarity of cutaneous involvement in tuberculosis. The basic principles of the treatment are the same as in pulmonary tuberculosis, but overall duration can be prolonged.

Keywords: Adolescent; Antitubercular Agents/therapeutic use; Mycobacterium Infections; Mycobacterium tuberculosis; Thigh; Tuberculosis, Cutaneous/diagnosis; Tuberculosis, Cutaneous/therapy

Introduction

Tuberculosis is a worldwide public health problem, affecting all age groups. Pulmonary tuberculosis is the most frequent form of tuberculosis in pediatric age. Extrapulmonary tuberculosis accounts for nearly 10% of tuberculosis' cases.¹ Tuberculous lymphadenopathy and tuberculous meningitis are the most frequent extrapulmonary forms of tuberculosis in pediatric age.² Cutaneous tuberculosis is a rare cause of extrapulmonary tuberculosis, affecting about 2% of the infected patients.³ Tuberculous cold abscesses, also known as tuberculous gumma, are rare and atypical

forms of cutaneous tuberculosis manifestations (1.3% to 2%) that result from hematogenous dissemination of latent mycobacteria. Tuberculous cold abscesses are mostly reported in children with malnutrition or immunosuppression.^{3,4} The diagnosis of tuberculosis means that a recent transmission occurred inside the community. Therefore, any person who has contacted with a patient with active tuberculosis should be screened to identify either latent infection or active disease by *Mycobacterium tuberculosis*. The treatment should be performed in both situations. Contacts with higher risk of developing disease are family members and children under 5 years, with severe malnutrition and immunosuppression. The immunosuppression can be caused either by a congenital immunodeficiency or by an acquired immunodeficiency such as the infection by human immunodeficiency virus (HIV) or by therapy (immunosuppressants).^{3,5}

Case Report

A 15-year-old female, originally from Bulgaria and living in Portugal for three years was admitted with a tumefaction on the left thigh noted three weeks before, without previous history of local trauma or wound. The patient denied any concomitant symptomatology, as fever, anorexia, asthenia, weight loss, nocturnal hyper sweating or cough. There was no history of recent travel. The patient also denied smoking, alcohol consumption, drug use or risky sexual behavior. Her vaccination status was unknown. She was living in a rural area with her parents and her three siblings, and they were a low socioeconomic index family. Following the detection of maternal pulmonary tuberculosis six months before, the patient was diagnosed with latent tuberculosis through the positive interferon-gamma release assay (IGRA) test (she was asymptomatic at that time and the chest radiography was normal). She started treatment with isoniazid for six months and her parents were responsible for the daily medication administration and its observation. However, the treatment was irregularly

Serviço de Pediatria, Hospital de São Bernardo, Centro Hospitalar de Setúbal, Setúbal, Portugal

Corresponding Author

Joana Cachão

joanacbc@hotmail.com

Serviço de Pediatria, Hospital de São Bernardo, Centro Hospitalar de Setúbal, Rua Camilo Castelo Branco, 175, 2910-446 Setúbal, Portugal

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performed, despite the monthly follow-up consultation. On examination, the patient had a good general condition. She was hydrated, feverless, eupneic and the pulmonary auscultation was normal. The upper third of the external face of the left thigh was enlarged with a soft, rounded mass, measuring 8 cm of greater diameter, of well-defined edges, slightly painful to palpation without fluctuation, change of skin color above the mass or other inflammatory signs (Fig. 1). Satellite regional adenopathy was not palpated nearby the swelling described or in other locations. The remaining examination was normal, with a body mass index of 22.6 kg/m² (percentile 50-85) and a stage of Tanner with a breast development B5 and pubic hair development P5. Routine investigation revealed hemoglobin 13.2 g/dL, leukocytes 8.1 x 10⁹ cells/L (60% neutrophils, 29% lymphocytes), platelets 215 x 10⁹ cells/L, an increased erythrocyte sedimentation rate (50 mm/h) and a C-reactive protein of 0.2 mg/L.

Ultrasonography of the left thigh showed a collected, liquid and heterogeneous image, 11 x 23 mm, with no Doppler signal, in line with a hematoma with some degree of organization or an organized infection. The patient was admitted for the continuation of the etiological investigation. The magnetic resonance of the left thigh revealed a liquid collection with a reniform morphology, with a deep component to the tendon of *fascia lata*, insinuating postero-superiorly to the superficial plane of the gluteus muscles and antero-inferiorly to the superficial plane of the vastus lateral muscle, with heterogeneous content and regular walls, slightly thickened, captant, suggestive of a cold abscess (Fig. 2).

The patient was submitted to needle aspiration of the described tumefaction, resulting in drainage of a thick and hematic-purulent fluid. The cytochemical test revealed a glucose concentration of less than 5 mg/dL, proteins 6.0 g/dL and lactate dehydrogenase 4441 U/L; the cell count was not available due to the viscosity of the drained fluid, but there was an apparent predominance of neutrophils (> 90%). No resistant acid-fast bacilli were identified, but the nucleic acids amplification by

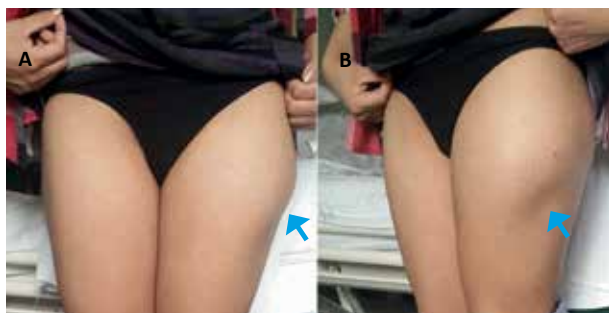


Figure 1. Tumefaction on the left thigh (arrows): front-view (A) and side-view (B).

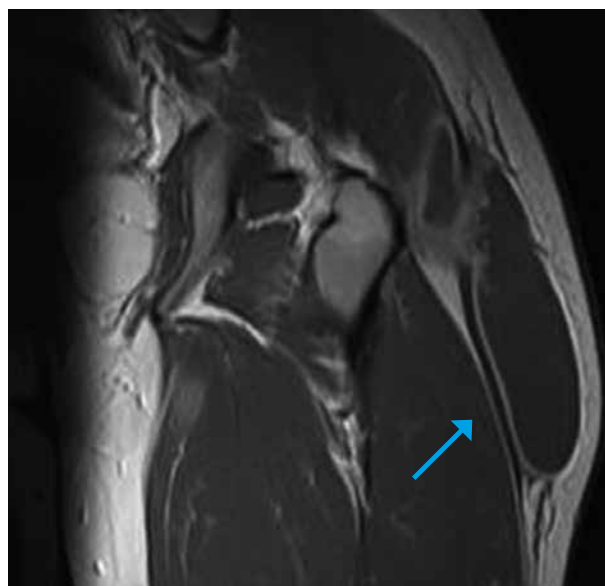


Figure 2. Magnetic resonance (coronal plane) of the left thigh, with an arrow pointing the liquid collection suggestive of a cold abscess.

polymerase chain reaction (PCR) of *Mycobacterium tuberculosis* was positive. A chest radiography was performed to investigate the presence of pulmonary tuberculosis, but no alterations were seen and the mycobacteriological study of the sputum (three samples obtained by nebulization with hypertonic serum) was negative for acid-fast bacilli and *Mycobacterium tuberculosis* PCR. A thoraco-abdomino-pelvic computed tomography was performed, which revealed a nodular formation in the posterior basal segment of the left lower lobe, along the azygos, of well-defined edges, solid and homogeneous appearance, with about 25 mm of larger longitudinal axis. No alterations were detected after ophthalmology examination. At the 11th day of hospitalization, after the diagnosis of tuberculous gumma, an empirical quadruple antitubercular therapy with isoniazid (300 mg/day), rifampicin (600 mg/day), pyrazinamide (1500 mg/day) and ethambutol (800 mg/day) was started, with the addition of pyridoxine to prevent the development of peripheral neuropathy. The patient accomplished the treatment described for two months in the hospital. A decrease of the dimensions of the tumefaction was noted immediately after the first needle aspiration. However, during the next weeks the size of the tumefaction increased, so a new needle aspiration was performed at the 40th day of therapy. The liquid drained had the same characteristics as before and the *Mycobacterium tuberculosis* PCR remained positive. The bacteriological study as well as the cultures in Lowenstein-Jensen and BACTEC™ medium of the liquid obtained from both needle aspirations and the sputum, did not reveal any growth of mycobacteria. While the patient stayed at the hospital, no signs or

symptoms of drug toxicity were observed. Laboratory evaluation (with renal, hepatic and thyroid functions) performed after two months of therapy was normal. Blood culture performed at admission, as well as serologies for HIV, syphilis, hepatitis B and C were negative. Serum results for immunoglobulins A, G and M were normal. No personal or family history of recurrent and/or severe previous infections, immunodeficiencies or autoimmune pathologies was detected.

The patient was discharged with isoniazid and rifampicin, in a directly observed therapy (DOT) regimen with supervision of the center for pneumological diagnosis of the residence area and follow-up in pediatric consultation. The treatment was maintained for eight months with a slow decrease of the dimensions of the described tumefaction. At the end of the treatment a residual and painless swelling, with no associated cutaneous alterations, was present in the upper third of the external face of the left thigh.

Discussion

Cutaneous tuberculosis in pediatric age is a diagnostic challenge, since it can present itself in a variable form, often with atypical symptoms and signals.³ Cutaneous tuberculosis can be classified into exogenous or endogenous sources.⁶ Exogenous spread, which is less common, occurs after direct inoculation of *Mycobacterium tuberculosis* through a cutaneous lesion or trauma in individuals naive to *Mycobacterium tuberculosis* infection. The etiology of such spread is usually secondary to needle stick injuries, injectable therapy and bacillus Calmette-Guérin (BCG) vaccine. These are unlikely in the patient of our case report, due to the anamnesis and the examination.

Endogenous spread is typically found in patients who have had prior infection with *Mycobacterium tuberculosis*,⁷ as the patient of our case report. This may be by direct extension of the disease from underlying infection or present after hematogenous spread,^{4,8} typically seen in tuberculous gumma (the likely cause in this case). Tuberculous gumma is usually described as a lesion with fluctuation, it can be single or multiple in the trunk, extremities or scalp and may be associated with cutaneous ulcer, which was not present in this case.^{3,7,8} It typically appears in malnourished children, in the presence of HIV infection, neoplastic disease or immunosuppressive therapy.⁷ However, tuberculous gumma can be diagnosed in immunocompetent individuals, as described in the literature,⁹ and may arise when the immune system is weakened.^{4,10}

The most widely accepted classification system for cutaneous tuberculosis is based on the mechanism of propagation, but the concept of bacterial load has also been added to the classification. In multibacillary forms mycobacteria are easily identified in cultures and in paucibacillary forms the cultures may not reveal any growth. In about 10% to 50% of extrapulmonary tuberculosis cases, there is concomitant pulmonary involvement, even if the chest radiography is normal, so pulmonary involvement should always be excluded.¹ During the investigation of our patient, there was a nodular formation in the posterior basal segment of the left lower lobe, which may probably correspond to the primary tuberculous complex. Mycobacterial culture is the gold standard test for diagnosing tuberculosis, but it is time-consuming (it can take up to eight weeks to get the definite result) and a negative result does not exclude the presence of tuberculosis since its sensitivity is low (0%-40%). In addition to this, the biological material obtained in extrapulmonary forms of tuberculosis is usually paucibacillary, particularly if it is a fluid,¹ a fact observed in our case report with negative culture results and acid-fast bacilli research. The PCR test allows a faster diagnosis than by culture and reveals a greater sensitivity in the detection of mycobacteria in extrapulmonary forms of tuberculosis, which are often paucibacillary; however, a negative PCR result does not exclude the diagnosis.¹ In children with latent tuberculosis, treatment with isoniazid monotherapy is recommended for six months. However, extending the treatment to nine months maximizes its efficacy and effectiveness, and the risk of toxicity is low.⁵ Several factors may compromise treatment compliance, such as socio-economic and cultural problems, poor accessibility to health care services, lack of information about the disease and the importance of accomplishing the treatment, non-supervision of the treatment and difficulties in communicating with health professionals.⁵ In our case report, the patient's family was immigrant, had difficulties in understanding the Portuguese language and was residing in a rural area, distant from primary health care; on the other hand, the treatment of latent tuberculosis was irregularly performed, under supervision of the patient's parents. In children with cutaneous tuberculosis, the therapeutic regimen recommended is the same as that for pulmonary tuberculosis, because the bacillary load is usually lower in cutaneous tuberculosis than in pulmonary tuberculosis.^{7,11,12} The therapeutic regimen of cutaneous tuberculosis consists in two months of quadruple therapy (isoniazid, rifampicin, pyrazinamide and ethambutol), followed by four months of double therapy (isoniazid and

rifampicin) and includes DOT.^{3,8,13,14} However, the second phase of the treatment may be prolonged according to clinical evolution.¹⁵ Usually after the beginning of the treatment, tuberculosis cutaneous lesions start to regress; however, its evolution may be slow and may justify repeated needle aspirations for the drainage of the content, as occurred in our case report, or even surgical excision.^{9,10,16} Although the clinical diagnosis of cutaneous tuberculosis is rare and not always evident due to its often nonspecific presentation, it should always be considered in the differential diagnosis of cutaneous lesions, particularly in children with known history of tuberculosis.^{7,13} The early diagnosis and the correct treatment of tuberculosis in pediatric age may contribute to the long-term change in the impact of this disease on public health.

WHAT THIS CASE REPORT ADDS

- Tuberculous gumma is a form of cutaneous tuberculosis caused by hematogenous spread of *Mycobacterium tuberculosis*.
- This case highlights the importance of a high suspicion rate for the diagnosis of cutaneous tuberculosis due to its rarity and atypical manifestation.
- A high suspicion rate combined with a needle aspiration allows the diagnosis of this pathology.

Conflicts of Interest

The authors declare that there were no conflicts of interest in conducting this work.

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Consent for publication

Consent for publication was obtained.

Confidentiality of data

The authors declare that they have followed the protocols of their work centre on the publication of patient data.

Awards and presentations

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Tumefação da coxa - uma apresentação rara de tuberculose

Resumo:

Os abscessos frios tuberculosos, também conhecidos por gomas tuberculosas, são formas raras de manifestação de tuberculose cutânea. Adolescente do género feminino, 15 anos, internada por tumefação na coxa esquerda com três semanas de evolução. Seis meses antes, tinha sido diagnosticada com tuberculose latente e medicada com isoniazida. Apresentava uma tumefação na coxa esquerda, arredondada, com limites bem definidos, 8 cm de maior diâmetro, mole, ligeiramente dolorosa à palpação, sem flutuação. Analiticamente destacava-se velocidade de sedimentação de 50 mm na primeira hora. A ressonância magnética revelou uma coleção líquida de conteúdo heterogéneo sugestiva

de abscesso frio e identificou-se *Mycobacterium tuberculosis* por reação em cadeia da polimerase após punção aspirativa. Cumpriu terapêutica antibacilar quádrupla durante dois meses, tendo mantido isoniazida e rifampicina durante oito meses. Pretende-se sublinhar a raridade do envolvimento cutâneo na tuberculose. Os princípios básicos do tratamento são comuns à tuberculose pulmonar, mas a sua duração total pode ser prolongada.

Palavras-Chave: Adolescente; Antituberculosos/uso terapêutico; Coxa; Infecções por *Mycobacterium*; *Mycobacterium tuberculosis*; Tuberculose Cutânea/diagnóstico; Tuberculose Cutânea/tratamento