

Leprosy in 3-year-old twin children in Mato Grosso, Brazil: the importance of bacilloscopy in the diagnosis

Hanseníase em gêmeos de 3 anos em Mato Grosso, Brasil: a importância da baciloscopia no diagnóstico

Lepra en gemelos de 3 años en Mato Grosso, Brasil: la importancia de la baciloscopia en el diagnóstico

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ABSTRACT

Introduction: the clinical diagnosis of leprosy in children is particularly difficult.

Case Report: fraternal twins, three years old, were in contact with a father with lepromatous leprosy. Both patients have well-defined and irregular skin lesions previously treated as mycoses and a BCG scar. They were confirmed positive for *Mycobacterium* by histopathological analysis of the skin.

Discussion: especially, with less than five years, leprosy diagnoses are rare and difficult because they simulate other diseases. These diagnoses are epidemiological alarms for

endemic areas and show the importance of symptoms in children and tracking of patients' contacts.

Keywords: *Leprosy. Preschoolers. Bacilloscopy. Early Diagnosis.*

RESUMO

Introdução: o diagnóstico clínico da hanseníase em crianças é particularmente difícil. **Relato de Caso:** crianças gêmeas bivitelinas, com três anos de idade, eram contactantes de pai com hanseníase virchowiana. Os dois pacientes têm lesões cutâneas bem definidas e irregulares, anteriormente tratadas como micoses e uma cicatriz de BCG. Foram confirmados positivos para *Mycobacterium* por análise histopatológica da pele. **Discussão:** especialmente, com menos de cinco anos, os diagnósticos de hanseníase são raros e difíceis porque simulam outras doenças. Esses diagnósticos são alarmes epidemiológicos para áreas endêmicas e mostram a importância dos sintomas em crianças e o rastreamento nos contactantes dos pacientes.

Palavras-chave: *Hanseníase. Pré-escolares. Baciloscopia. Diagnóstico Precoce.*

RESUMEN

Introducción: el diagnóstico clínico de la lepra en niños es particularmente difícil. **Caso Clínico:** gemelos bivitelinos, de tres años de edad, estuvieron en contacto con el padre con la lepra virchowiana. Ambos pacientes presentaron lesiones cutáneas irregulares y bien definidas tratadas previamente como micosis y una cicatriz de BCG. Se confirmó su positividad para *Mycobacterium* mediante análisis histopatológico de la piel. **Discusión:** los diagnósticos de lepra especialmente con niños menores de cinco años, son raros y difíciles porque simulan otras enfermedades. Estos diagnósticos son alarmas epidemiológicas para zonas endémicas y muestran la importancia de los síntomas en niños y del seguimiento de los contactos de los pacientes.

Palabras clave: *Lepra. Preescolares. Baciloscopia. Diagnóstico precoz.*

INTRODUCTION

Leprosy is a chronic infectious disease caused by *Mycobacterium leprae*, and, less often, by *Mycobacterium lepromatosis*, which affects mainly the skin and peripheral nerves. This disease has a long period of evolution, which can be the main explanation for the low prevalence in children under

15 years old¹. Between the years of 2016 and 2020, 155,359 leprosy cases were registered in Brazil. In 2020, 3,382 new cases were registered in Mato Grosso (MT), with a prevalence rate of 9.59/100,000 inhabitants. One hundred of these patients were children under 15 years old, with a prevalence rate of 12.20. Considering the operational classification, 959 new cases were paucibacillary, and 2,423 were multibacillary². Leprosy cases in children are more frequent in regions of high endemicity when contacts aren't diagnosed and treated³.

The diagnosis is clinical^{1,4} and based on laboratory tests, but the response to clinical skin sensitivity tests is difficult in children. The histopathological test of skin lesions allows additional analyses, such as immunological, histological, and microbiological⁵. The presence of granuloma characterizes the histological aspects of skin lesions, dermal edema, lymphoplasmacytic infiltrate, and multinucleated giant cells. Fragments of skin patches are used to determine the presence, amount, and morphology of mycobacteria in patients. The staining Fite-Faraco is one of the most sensitive for skin biopsies, and the Ziehl-Neelsen method is the most used for elbow and earlobe lymph smears^{1,4}.

The present study aims to report cases of multibacillary leprosy in 2 fraternal twins patients. It also aims to discuss the genetic, immunological, and epidemiological peculiarities of the situation.

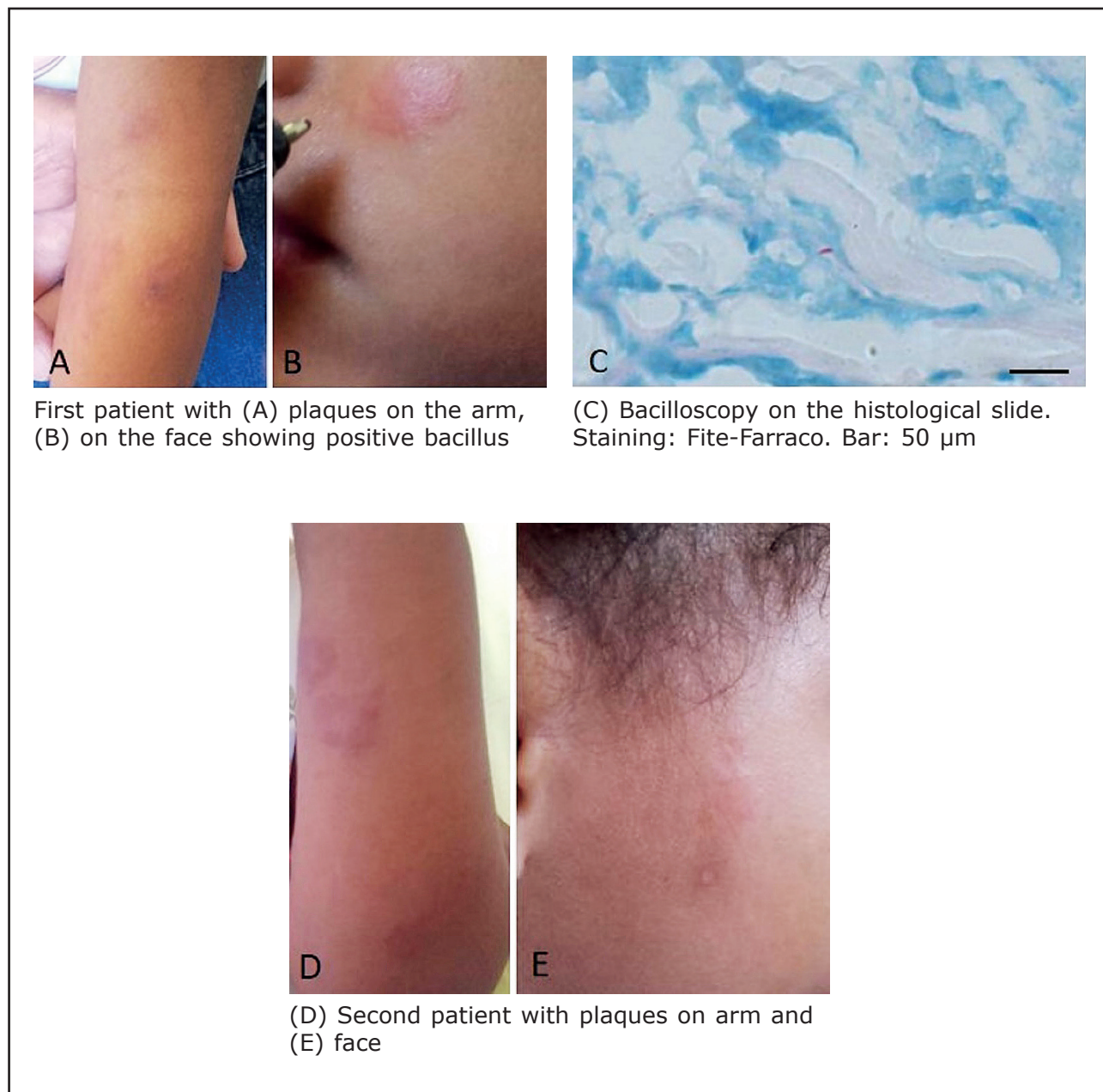
Case report

This case report describes two fraternal twins patients, 3 years old, under reaction conditions. The father was treated for lepromatous leprosy two years ago.

The first patient is female, premature, 3 years old, born and raised in Cuiabá, Mato Grosso, Brazil, presenting patches on the face and upper limb for three months. She was sent to University Hospital Julio Müller (HUJM) in November of 2019, Cuiabá. The child was eutrophic but presented skin lesions that were already evaluated. She was under treatment by medical indication, with ketoconazole ointment, without clinical improvement. On physical examination, she had several erythematous, hyperchromic, well-defined, and irregular patches in the aforementioned regions (Fig 1A and B). She had a scar from Calmette & Guérin bacillus (BCG) vaccine. The hypothesis of leprosy was considered, and the investigation continued with the histopathological analysis of the skin biopsy. The result, in Fite-Faraco staining, indicated a 4+ lymphoplasmacytic infiltrate, several granulomas, 2+ bacilloscopic index, and 50% of the bacilli with fragmented morphology (Fig 1C), suggestive of borderline leprosy in type 1 reaction. She began treatment with multidrug therapy for multibacillary leprosy, with a rapid reduction of lesions.

The second patient is male, premature, three years old, low weight, between p3-p15 on the growth curve, with extensive, hypochromic, well-defined, and irregular spots on the face and body (Fig 1D and E). He underwent a previous medical evaluation, and he was using ketoconazole ointment, also without improvement. He also had a scar from the BCG vaccine. There were no changes in nerves. Due to the family history, the same complementary investigation was chosen. The result, in Fite-Faraco staining, was the presence of diffuse lymphoplasmacytic infiltrate, bacilloscopic index 2+, 100% of fragmented bacilli, suggestive of borderline leprosy in type 1 reaction. He also started treatment with multidrug therapy.

Figure 1 – 3-year-old twin patients with leprosy



Source: Elaborated by the authors

DISCUSSION

This study is an alert about the high endemicity of the region of Cuiabá, MT, Brazil. The involvement, mainly in multibacillary cases, in pediatric patients is an alert to the high circulation of undiagnosed patients, as well as the active transmission of the disease. Younger children, due to the natural limitation of age, have shorter contact time with the bacillus than adults in the same situation. The situation also demonstrates a failure in combat actions, such as the contact screening test, which should have been carried out, according to the Brazilian Ministry of Health and the World Health Organization, at the time of diagnosis of the index case; in this case, the father, 2 years ago.

Clinical examination in young children, especially under 5 years old, is difficult. The importance of knowing the clinical manifestations of leprosy in pediatrics is fundamental for the correct diagnosis. Young children usually have nonspecific signs. They start with paresthesia or acroparesthesia, mainly in extremities, in addition to patches or macules, usually hypochromic or erythematous, this is associated or not with superficial alteration of sensitivity (thermal, tactile, and painful), they can alter sweating or hair growth and hair. In adolescence, they present more polar characteristics with nervous thickening and dermal infiltrations^{6,7,8}.

The immunological part is also very important. Despite the shorter contact time with the bacillus, children still have an immunological immaturity that can generate susceptibility. As they are siblings, the hypothesis of a component of genetic susceptibility is reinforced, with a diagnosis in three out of four people in the reported family. Currently, several genetic studies show that there is a genetic correlation with the expression of genes such as PARK2/PACRG, located on chromosome 6q25-q27, and NRAMP1, on chromosome 2q35, which confers susceptibility to develop the disease¹. Despite this, they are fraternal twins, which reinforces the importance of high contact with a multibacillary patient during the 3 years of life, and in 2 of these years, the father had already received treatment.

The clinical features of the children were subtle, but family history, the biopsy, and bacilloscopy allowed assertive diagnosis, characterizing multibacillary leprosy. Santino et al⁹ also identified a case in a 3-year-old child who had erythematous plaques, pain on palpation of ulnar nerves, and histopathological examination with bacilloscopy 3+ with fragmented bacilli.

CONCLUSION

Children are probably more susceptible to *Mycobacterium leprae* infection because they do not have a fully matured and developed immune system.



Diagnosis of leprosy in children is difficult due to varied clinical manifestations, raising the importance of complementary parasitological and molecular techniques. Immediate examination of all household contacts is mandatory to prevent household transmission and interrupt the transmission chain.

ETHICAL APPROVAL AND INFORMED CONSENT: *the present study was developed by the terms of Resolution 196/96 of the National Health Council and authorized by the Ethics Committee (CEP) of Júlio Muller University Hospital (CAAE Protocol No. 09292319.0.0000.5541, approved on 04/25/2019).*

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