



Case Report

Lepromatous leprosy with a suspected 30-year incubation period: A case report of a practically eradicated area

Niporn Jariyakulwong, MD^a, Narachai Julanon, MD^{b,c} and Charupong Saengboonmee, MD^{b,d,e,*}

^a Lamplaimat Hospital, Lamplaimat, Buriram, Thailand

^b Srinagarind Hospital, Faculty of Medicine, Khon Kaen University, Khon Kaen, Thailand

^c Division of Dermatology, Department of Medicine, Faculty of Medicine, Khon Kaen University, Khon Kaen, Thailand

^d Department of Biochemistry, Faculty of Medicine, Khon Kaen University, Khon Kaen, Thailand

^e Center for Translational Medicine, Faculty of Medicine, Khon Kaen University, Khon Kaen, Thailand

Received 2 November 2021; accepted 21 December 2021; Available online 1 February 2022



المخلص

أهداف البحث: يهدف هذا التقرير إلى توضيح وتذكير مقدمي الرعاية الصحية بأنه على الرغم من اعتباره قد تم استئصال الجذام في منطقة معينة، إلا أنه يمكن العثور على الإصابة به نتيجة فترة حضانة طويلة بشكل غير عادي.

طرق البحث: أبلغنا عن حالة من الجذام لأنثى تايلندية تبلغ من العمر 48 عاماً ظهرت عليها أعراض جلدية وعصبية كلاسيكية. تم إجراء الفحوصات الجسدية وإعداد اللطاخة الفلعية مع التلوين الصامد للحمض للوفاء بالمعايير التشخيصية لمنظمة الصحة العالمية. تم وصف الأنظمة العلاجية بناءً على توصية منظمة الصحة العالمية.

النتائج: تم تشخيص إصابة المريضة بالجذام بالنظر إلى استيفاء جميع العناصر الأساسية لمعايير التشخيص لمنظمة الصحة العالمية؛ الفحوصات الجسدية والميكروسكوبية. تقابل الأفات الجلدية عن طريق الفحص البدني نوع جذامي. أظهر تحضير اللطاخة الفلعية من الأفات وجود عصيات وفيرة صامدة للأحماض. كانت المريضة تعيش فقط في منطقة تم فيها استئصال الجذام عملياً ولم يتم الإبلاغ فيها عن حالات جديدة. وبالتالي، فإن أكثر عوامل الخطر المرئية لإصابتها هو الاتصال الوثيق قبل 30 عاماً مع أحد أفراد عائلتها المصاب بالجذام في نفس المنزل لمدة أسبوع.

الاستنتاجات: يوضح هذا التقرير حالة نادرة من الجذام مع فترة حضانة طويلة في منطقة غير موبوءة. تم الإبلاغ عن مثل هذه الحضانة الطويلة بشكل غير عادي فقط للقرود غير البشرية، ولكنها نادرة للغاية بالنسبة للبشر. تذكر هذه

المعلومات مقدمي الرعاية الصحية بأن الجذام ليس مرضاً في الماضي، ولا يزال برنامج المراقبة الدقيقة للجذام ضرورياً حتى في المنطقة التي تم استئصالها منها.

الكلمات المفتاحية: مرض هانسن؛ فترة الكمون؛ الجذام؛ العسوية الجذامية.

Abstract

Objectives: This report aims to demonstrate and remind healthcare providers that, despite being considered eradicated in a specific area, leprosy can still be found due to its unusually long incubation period.

Methods: A case of leprosy has been reported in a 48-year-old Thai woman who presented classic dermatological and neurological symptoms. A physical examination and slit smear preparation with acid-fast staining was performed to fulfil the diagnostic criteria of the World Health Organization (WHO). Therapeutic regimens were prescribed according to WHO recommendations.

Results: A patient was diagnosed with leprosy given that all cardinal signs of WHO diagnostic criteria were met after both physical and microscopic examinations. Physical examination of the skin lesions revealed the lepromatous type. Slit smear preparation of the lesions revealed abundant acid-fast bacilli. The patient has been living solely in an area where leprosy has been practically eradicated, with no new cases reported. The most likely cause of her infection is her close contact with a leprosy-infected family member 30 years ago, with whom she shared a house for a week.

* Corresponding address: Department of Biochemistry, Faculty of Medicine, Khon Kaen University, Khon Kaen, 40002, Thailand.

E-mail: charusa@kku.ac.th (C. Saengboonmee)

Peer review under responsibility of Taibah University.



Production and hosting by Elsevier

Conclusions: This report describes a rare case of leprosy with a long incubation period in a non-endemic area. Only non-human primates have had such an unusually long incubation period, which is extremely rare in humans. This information reminds healthcare providers that leprosy is not a disease of the past, and a careful surveillance program for leprosy remains necessary, even in eradicated areas.

Keywords: Hansen's disease; Latency period; Leprosy; *Mycobacterium leprae*

© 2022 The Authors. Published by Elsevier B.V. This is an open access article under the CC BY-NC-ND license (<http://creativecommons.org/licenses/by-nc-nd/4.0/>).

Introduction

Leprosy is a neglected tropical disease that is considered rare in most regions despite having relatively high incidence in some countries.¹ It is caused by infections with *Mycobacterium leprae* and *Mycobacterium lepromatosis*. Transmission occurs via droplets during close and frequent contact with untreated cases.¹ Both *M. leprae* and *M. lepromatosis* are slow-growing bacteria with incubation periods varying from 5 to 10 years, depending on clinical subtypes.² The clinical presentations of leprosy include skin rash, peripheral neuropathy, and organ deformities.

The global prevalence rate of leprosy is 0.2 cases per 10,000 population as of 2018,³ while its prevalence rate in Thailand is 10-fold lower than the global prevalence in the year 2019 (0.02 cases per 10,000 persons).⁴ In Thailand, leprosy as a public health problem is eliminated according to the definition of the World Health Organization (WHO).³ In 2021,⁵ the WHO launched the campaign for "Toward zero leprosy" for the decade. The WHO guidelines for leprosy diagnosis are based on the presence of at least one of the following three cardinal signs: 1. definite loss of sensation in a pale (hypopigmented) or reddish skin patch/plaque; 2. thickened or enlarged peripheral nerves with loss of sensation and/or weakness of the muscles supplied by the nerve, and 3. presence of acid-fast bacilli in a slit-skin smear.⁶ The WHO guidelines for leprosy treatment recommend a 3-drug regimen (multidrug treatment: MDT) of rifampicin, dapsone, and clofazimine for all leprosy patients, with a treatment duration of 6 months for paucibacillary leprosy and 12 months for multibacillary leprosy.

In this report, we present a case of lepromatous leprosy diagnosed using all three cardinal signs at a community hospital in Thailand. A written informed consent for the publication was obtained from patient.

Case report

Clinical findings

A 48-year-old woman with no underlying disease presented with hyperpigmented lesions on her back for 2 years. She felt numbness at the tip of her fingers and toes for 4 days, but denied contact with any chemicals. Thirty years ago, she spent a week at her father-in-law's house after he was diagnosed with leprosy. No other family members or relatives showed symptom. Skin examination revealed asymmetrical, multiple, discrete, poorly defined borders and hyperpigmented plaques with decreased sensation on her back, nose, and both cheeks, loss of the outer two-thirds of both eyebrows, and infiltrated plaques on both earlobes (Figure 1). Neurological examination revealed a decrease in sensation of pain and temperature at the tips of her fingers and toes. Other physical examinations and peripheral nerve findings were unremarkable. At this stage, the provisional diagnosis of the patient's illness was leprosy.

We then obtained slit skin smears of both earlobes and plaques of the skin on her back. Acid-fast staining of the samples revealed the presence of acid-fast stained bacilli (Figure 2) in all slit skin samples.

The patient was diagnosed with leprosy as all three cardinal signs of the WHO diagnostic criteria were met. The



Figure 1: Skin manifestation of the patient. Multiple, discrete, poorly-defined borders, irregular and hyperpigmented patches were seen on the back of the patient. Also, infiltrated plaques were observed on both earlobes.

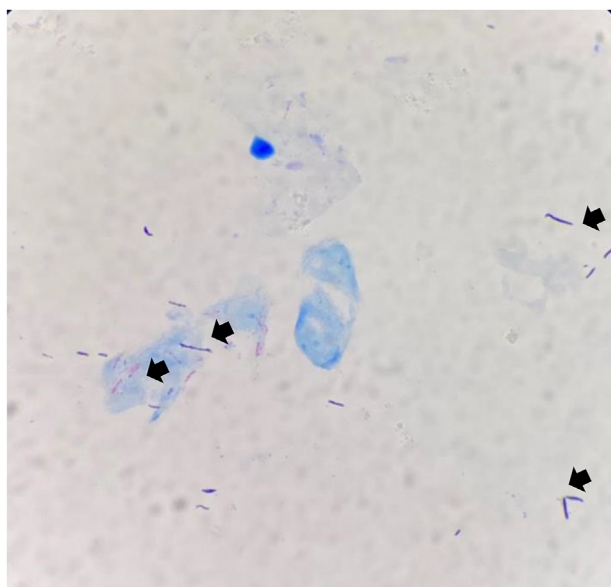


Figure 2: The acid-fast staining of the slit skin smear from the patient. Acid fast stained bacilli (arrow) are seen in all slit skin samples. The image was taken by high-power field (x400 magnification) microscopic examination.

generalised distribution of plaques on the face, trunk, and eyebrows was compatible with lepromatous leprosy. According to the WHO guideline for leprosy treatment, she was treated with oral Rifampicin 600 mg plus Clofazimine 300 mg once per month, and Dapsone 100 mg plus Clofazimine 50 mg, daily, for two years. At the six-month follow-up, the patient's skin lesions had improved and numbness had decreased. She will have to go for annual follow ups for five years.

Discussion

Leprosy remains a public health problem in many developing countries, especially, in tropical regions. Since the causative *Mycobacterium* spp. are slow-growing, people may have subclinical leprosy for years.² The international migration and importation of workers from one country to another could be a cause of expansion of leprosy from endemic areas to uncommon areas.⁷ The long incubation period of this disease makes it difficult to estimate its real incidence and eradication. According to the WHO, leprosy as a public health problem can be considered eradicated in regions where the prevalence rate has become <1 case per 10,000 persons.^{1,3} Since 1994, Thailand has achieved its goal of eradication. From the records available at Lamplaimat Hospital, the prevalence rate in the study area was 0.06 cases per 10,000 persons in the year 2020. Therefore, the patient's area of residence has already achieved the goal of eradicating leprosy as a public health problem.

The transmission mode of leprosy occurs via droplets from untreated cases, with substantially long and frequent contact.¹ However, the patient in the present report lives on her own in a rural area of the Lamplaimat District, Buriram Province, where the prevalence rate of leprosy is very low

and no known case is currently reported. The most reasonable cause of her diagnosis is a history of close contact with her father-in-law who was diagnosed with leprosy 30 years ago. This incubation period was particularly long and has not been usually reported in human leprosy cases. However, this long incubation period for leprosy has been reported in chimpanzees.⁸ This reminds healthcare providers that proper surveillance of leprosy should not be neglected. Patients may undergo a subclinical stage for decades and present symptoms in unusual regions after their international migration.⁷ Moreover, since leprosy is one of the greatest imitators of other skin diseases, a complete history taking could be helpful for an appropriate differential diagnosis.⁹

This report, however, has some limitations; 1) the real incubation period of the patient could be less than 30 years since the unobvious symptoms might not be recognised. 2) In the community hospital setting, molecular tests and skin biopsies to confirm *M. leprae* species are not available. Nevertheless, the clinical manifestation and microscopic examination of this patient met the diagnostic criteria of the WHO, and the patient's symptoms improved after treatment with the MDT regimen.

In summary, we reported a case of lepromatous leprosy with an unusually long incubation period of approximately 30 years in an eradicated area. This reminds healthcare providers that leprosy is not a disease of the past despite its rare incidence. Good surveillance and early detection are crucial for the prevention of long-term complications and disabilities in patients.

Source of funding

This work did not receive any specific grants from funding agencies in the public, commercial, or not-for-profit sectors.

Conflict of interest

The authors have no conflict of interest to declare.

Ethical approval

This report was reviewed and approved by the Ethics Committee for Human Research, Buriram Provincial Health Office (approval no. BRO-E 2021-004, approval date: August 31, 2021).

Authors contributions

NiJ and CS conceptualised the study protocol, which was critically curated for its scientific content and relevance by NiJ; formal analysis by NaJ and CS; a preliminary draft of the manuscript was prepared by NiJ and CS. All authors have critically reviewed and approved the final draft and are responsible for the content and similarity index of the manuscript.

Acknowledgment

We would like to thank Mrs. Bussakorn Tiamwong, a medical technologist at Lamplaimat Hospital, for her

assistance with slit skin smear preparation and microscopic examination. We are grateful to Professor Yukifumi Nawa, M.D., Ph.D., for editing this manuscript at the KKU Publication Clinic, Khon Kaen University. CS was supported by the Long-term Pre-clinical Staff Development Program of the Faculty of Medicine, Khon Kaen University, and the Research Grant for Talented Young Researcher (N41A640108), National Research Council of Thailand.

References

1. Hambridge T, Nanjan Chandran SL, Geluk A, Saunderson P, Richardus JH. Mycobacterium leprae transmission characteristics during the declining stages of leprosy incidence: a systematic review. *PLoS Neglected Trop Dis* 2021; 15:e0009436.
2. White C, Franco-Paredes C. Leprosy in the 21st century. *Clin Microbiol Rev* 2015; 28: 80–94.
3. World Health Organization. Global leprosy (Hansen disease) update, 2019: time to step-up prevention initiatives. *Wkly Epidemiol Rec* 2020; 417–440.
4. Nawalerspanya S, Sangmala S, Aiempanakit K. A case report of prurigo nodularis-like lesions in a patient with lepromatous leprosy. *Case Rep Dermatol* 2020; 12: 236–240.
5. World Health Organization. *Towards zero leprosy. Global leprosy (Hansen's Disease) Strategy 2021–2030*. New Delhi: World Health Organization Regional Office for South-East Asia; 2021. p. 30.
6. World Health Organization. *Guidelines for the diagnosis, treatment and prevention of leprosy*. New Delhi: World Health Organization, Regional Office for South-East Asia; 2017. p. 106.
7. Tan YE, Yeo YW, Ang DJQ, Chan MMF, Pang SM, Sng LH. Report of a Leprosy case in Singapore: an age-old disease not to be forgotten in developed countries with low-prevalence settings. *Access Microbiol* 2019; 1:e000014.
8. Suzuki K, Udono T, Fujisawa M, Tanigawa K, Idani G, Ishii N. Infection during infancy and long incubation period of leprosy suggested in a case of a chimpanzee used for medical research. *J Clin Microbiol* 2010; 48: 3432–3434.
9. Kundakci N, Erdem C. Leprosy: a great imitator. *Clin Dermatol* 2019; 37: 200–212.

How to cite this article: Jariyakulwong N, Julanon N, Saengboonmee C. Lepromatous leprosy with a suspected 30-year incubation period: A case report of a practically eradicated area. *J Taibah Univ Med Sc* 2022;17(4):602–605.