New cases of childhood leprosy in two male siblings in low endemic province: an iceberg phenomenon?

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Abstract

Children are the leprosy population most at risk; multibacillary infections are common. We reported pediatric cases with WHO-multibacillary lepromatous type morbus Hansen. We discovered several hypopigmented patches on the right upper arm, anesthesia-filled claw hands, and infiltrates in the ears in one 17-year-old child. In the other case (13-year-old), we discovered reduced sensory sensitivity in nearly every area of the body, infiltrates on the ear, and numerous irregular hypopigmented plaques on the belly, lumbar, and gluteus. Both patients had positive acid-fast bacteria. After three months of treatment, multibacillary leprosy patients began to show improvement. Low socioeconomic and environmental conditions, as well as a history of intimate contact with leprosy patients, have been identified as risk factors.

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Introduction

Leprosy remains a significant public health problem globally.[1] The World Health Organization (WHO) reports that there were 127,558 cases worldwide in 2020, with 8629 of those cases being children under the age of 15 and 7198 cases involving people with disabilities.[2,3] Indonesia ranking third worldwide with new cases detection rate (NCDR) of leprosy were found at 4.03 cases per 100,000 population.[4] According to WHO-AFRO Leprosy burden scale, West Nusa Tenggara Children are the leprosy population most at risk; multibacillary infections are common. We reported pediatric cases with WHO-multibacillary lepromatous type morbus Hansen. We discovered several hypopigmented patches on the right upper arm, anesthesia-filled claw hands, and infiltrates in the ears in one 17-year-old child. In one more (13-year-old), we discovered reduced sensory sensitivity in nearly every area of the body, infiltrates on the ear, and numerous irregular hypopigmented plaques on the belly, lumbar, and gluteus. Both patients had positive acid-fast bacteria. After three months of treatment, multibacillary leprosy patients began to show improvement. Province is categories of low endemic areas with NCDR 4.9 cases per 100,000 population with incidence rate in children (0-14 years) is 4.8%.[5,6] New cases of child leprosy in Mataram City were found to be 6.45% in 2018-2019 with cases that often found in children are the multibacillary type.[7] This report presents a case of Morbus Hansen Lepromatous type in two male siblings.

Case Report

Two male siblings with initials of child W (17-year-old) and child A (13-year-old) were diagnosed with Morbus Hansen's WHO-MB Lepromatous type. On child W (Figure 1), redness accompanied by itch on the left arm was reported for 4 years which then spread to other body parts including the face and body with tingling and numbness in the fingers of his left hand. Patient was in good nutritional status with weight 47.2 kg, height 161 cm, and body mass index 18.22 kg/m^2. Infiltrates were found on both ears and contracture were visible of the left hand (claw hand) accompanied by anesthesia. Dermatologic examination revealed multiple hypopigmented patches in the right upper extremity.

His brother, child A (Figure 2) complained of redness and itching on the back which then spread to the face and almost the whole

body in the past 1 year. Currently child A was in good nutritional status with weight 26.2 kg, height 125 cm and body mass index 16.97 kg/m^2. In the right and left ears, infiltrates were found, and dermatology examination revealed multiple irregular hypopigmented plaques with distribution in the abdomen, lumbar and gluteal regions followed by decreased sensibility in almost all parts of the body except the soles of the feet. No peripheral nerve thickening was found. The result of acid-fast staining and laboratory on child W was positive with Bacterial Index (BI) +4 and Morphology Index (MI) is 89% and child A's BI was +4 with MI 94%. Therapy regimen given to child W was rifampicin 600 mg/month, dapsone 100 mg/day, clofazimine 300 mg/month continued with 50 mg/day and neurotropic vitamins (B1, B6 and B12). Child A was given rifampicin 450 gr/month, dapsone 50 gr/month continued with 5 gr/day, clofazimine 150 gr/month continued with 50 gr/2 days. History of close interactions with their cousin living 100 meters away having the same symptoms and recently diagnosed with Morbus Hansen was reported. Family history was also found in late grandfather experiencing the same symptoms but never been examined in health facilities. Observation of patients' environment found the humidity was 82% with temperature 28°C as well as limitation of sun exposure and ventilation (Figure 3).

Follow up after three months of medication showed some skin lesions improved with decreased of inflammation. The acid-fast staining result on child W decreased with BI +3 and MI 64%, and child A also decreased with BI +2 and MI 79%. Both patients reported no side effects while taking the medication.

Discussion

Leprosy cases are often found in children aged 10-14 years or under 15 years old due to long incubation period of approximately 3-5 years.[8] *M. Leprae* can be transmitted by direct contact and through droplets during prolonged close contact. One case that has been reported in Mexico is a male suffering from leprosy with a disability, then after 5 years his sister experienced the same symptoms and was diagnosed with leprosy.[9]

Genetic factors are involved in one's predisposition to leprosy, one of which is the influence of a specific variant of genes from human leukocyte antigen (HLA) and the non-HLA.[10] The development of leprosy also has been associated with genes that control the macrophage and Schwann cells response to bacillus, such as variations in the *PRAK2* and *PACRG* genes.[11] For genetics in this case need deeper assessment on the associated genes, for the reason of under advance technology, we are not able to execute this factor.

Several studies reported a higher risk of leprosy in individuals with poor environment condition. A humid environment is a breeding ground for *M. Leprae* bacteria. These bacteria are able to survive in the environment for 7 days with a temperature of 20.6°C and humidity of 43.7%.[12] The poor ventilation and not exposed to direct sunlight may have contribute to increase the survival of the leprosy bacteria in the droplets. Another risk factors we found contributing to this case were living together, low socioeconomic and environmental condition.

Multi Drug Therapy (MDT) is a regimen used to treat a lep-

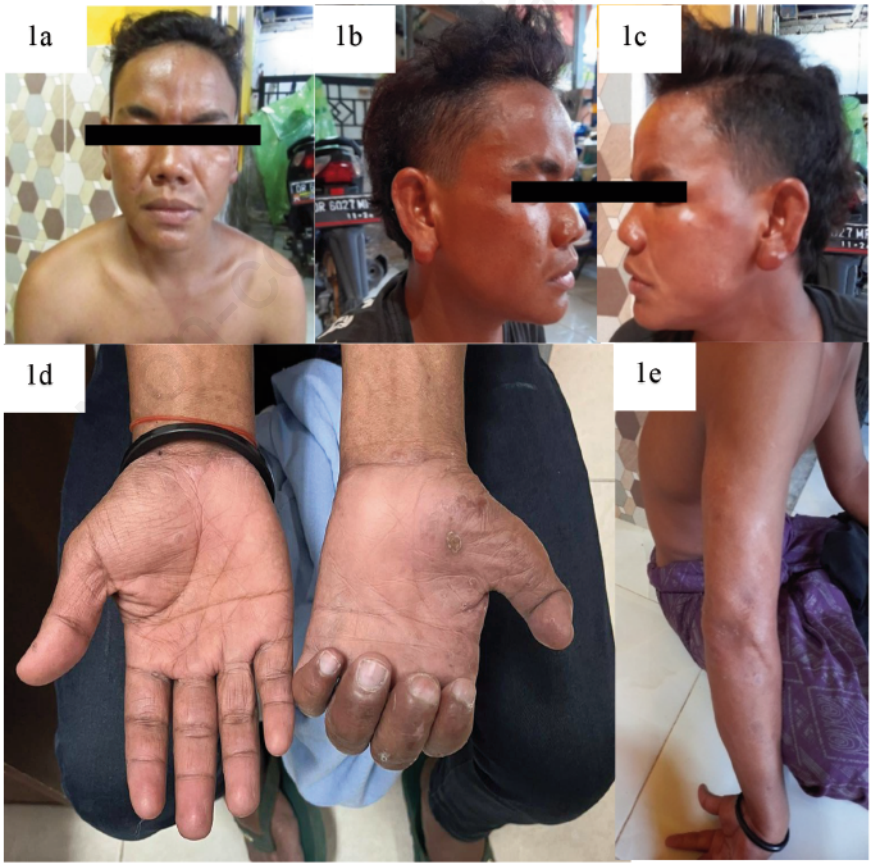

Figure 1. **a**) Child W's face front view; **b**) child W's face from right side with right ear infiltrate; **c**) child W's face from left side with left ear infiltrate; **d**) an image of a claw hand on the finger of child W's left hand; **e**) hypopigmented patch on child W's right hand.

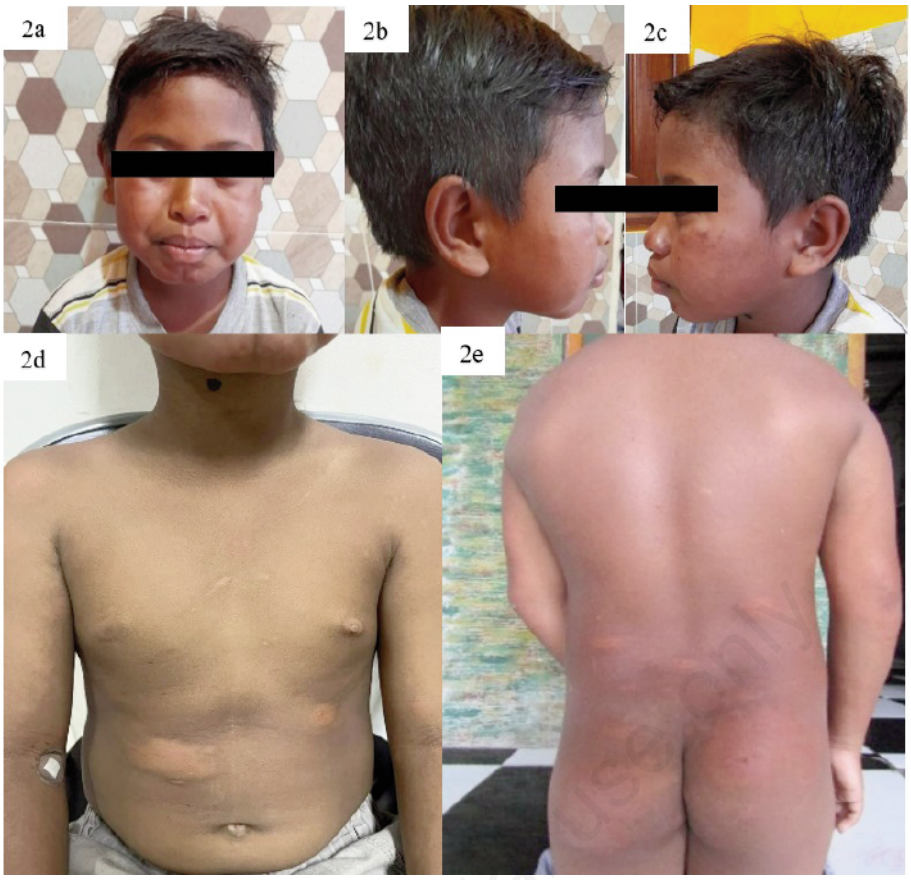

Figure 2. **a**) Child A's front view face; **b**) child A's face from right side with right ear infiltrate; **c**) child A's face from left side with left ear infiltrate; **d**) hypopigmented plaques in child A's body, front view; **e**) hypopigmented plaques in child A's body, rear view.

Figure 3. **a**) Front view of the patient's house; **b**) view of the patients' living room; **c**) look at the patient's warehouse; **d**) patients kitchen; **e**) child W's bedroom which is located outside the house; **f**) patient's bathroom.

rosy consists of several antibiotics adjusted to the type of leprosy (PB or MB). In children aged 10-15 years with MB type as in this case there is a regimen package with a duration of administration of 12 months.[13] Few cases reported improvement after taking MDT for several months which in this case showed an improvement after three months medication.[14,15]

Frequent interactions with the community's sufferers also increased the likelihood that, in a few more years, leprae would strike. Contact tracing of the family member and community should be considered as a way to find more active cases and stop the spreading of the disease.[16] In low endemic areas, the lack of leprosy medications in primary health facilities necessitates increased coordination in leprosy prevention.

Conclusions

This case report made clear that, despite the rarity of pediatric cases of the Morbus Hansen lepromatous type – particularly in low-endemic areas – there is still a chance of the "iceberg" phenomenon when it comes to the discovery and tracking of new cases.

References

1. Sangupta U. Recent laboratory advances in diagnostics and monitoring response to treatment in leprosy. Indian Dermatol Online J 2019;10:106-14.
2. White C, Franco-Paredes C. Leprosy in the 21st century. Clin Microbiol Rev 2015;28:80-94.
3. World Health Organizations. Leprosy, 2022. Available from: https://www.who.int/news-room/fact-sheets/detail/leprosy
4. Kementrian Kesehatan RI. Profil Kesehatan Indonesia Tahun 2021. Kementrian Kesehatan RI, 2022.
5. Ogunsumi DO. Measuring endemicity and burden of leprosy across countries and regions: a systematic review and Delphi survey. PloS Negl Trop Dis 2021;15:1-19.
6. Dinas Kesehatan Provinsi NTB. Profil Kesehatan Provinsi NTB Tahun 2021. Kementrian Kesehatan RI, 2022.
7. Affarah WS, Primayanti I, Sari LS. Gambaran Epidemiologi Kusta Pada Anak Dan Pelaksanaan Kemoprofilaksis Kusta Di Kota Mataram. J Kedokt Unram 2021;10:413-21.
8. Ruiz-Fuentes JL. , Rumbaut Castillo R, Hurtado Gascón LdlC, Pastrana F. Leprosy in children: a Cuban experience on leprosy control. BMJ Paediatrics Open 2019;3:1-7.
9. Sotiriou MC, Stryjewska BM, Hill C. . Case report : two cases of leprosy in siblings caused by Mycobacterium lepromatosis and review of the literature. Am J Trop Med Hyg 2016;95:522-7.
10. Gracino ME, Mendes B, Garbelin MV, et al. Leprosy : a systematic review. Int J Dermatol Sci 2020;2:8-21.
11. Mira MT, Alcais A, Nguyen VT, et al. Susceptibility to leprosy is associated with PARK2 and PACRG. Nature 2004;427:636-40.
12. Cendaki QA. The findings of Mycobacterium leprae DNA Existence in the air as an indication of leprosy transmission from respiratory system. J Kesehat Lingkung 2018;10:181-90.
13. Kementrian Republik Indonesia. Peraturan Menteri Kesehatan Republik Indonesia Nomor 11 Tahun 2019 Tentang Penanggulangan Kusta. 2019.
14. Pratama N, Mas-Rusyati LM, Sudarsa PS, et al. Borderline lepromatous leprosy with severe erythema nodusum leprosum: a case report. Available from: https://e-journal.unair.ac.id/BIKK
15. 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 Sci 2022;17:602-5.
16. Richardus JH, Tiwari A, Barth-Jaeggi T, et al. Leprosy post-exposure prophylaxis with single-dose rifampicin (LPEP): an international feasibility programme. Lancet Glob Health 2021;9:e81-90.