Case Report

The First Case Report of Pulmonary Hypertension in Leprosy

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ABSTRACT

Leprosy is a chronic infectious disease caused by *Mycobacterium leprae*, an acid-fast, rod-shaped bacillus. The disease predominantly affects the skin, the peripheral nerves, the mucosa of the upper respiratory tract, and the eyes. [1] The diagnosis of leprosy is based on the presence of at least 1 of 3 cardinal signs: definite loss of sensation in a pale or reddish skin patch, a thickened or enlarged peripheral nerve with loss of sensation, and/or weakness of the muscles supplied by the nerve, and the presence of acid-fast bacilli in a slit-skin smear. However, there are no reports that describe the link between leprosy and pulmonary hypertension (PH). Here, we describe a 22-year-old man who was first diagnosed with leprosy and subsequently developed PH, confirmed by echocardiography. To our knowledge, this is the first case of leprosy associated with PH with a detailed clinical description. We recommend that physicians be aware of this rare comorbidity in patients with leprosy. Early echocardiographic screening is necessary for symptomatic patients. ***(Iranian Heart Journal 2024; 25(4): 100-104)***

KEYWORDS: Leprosy, Pulmonary hypertension, Echocardiography, Rare manifestation.

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Morbus Hansen, better known as leprosy, is a chronic infectious disease caused by *Mycobacterium leprae*. In 2021, Globally, the registered prevalence of leprosy was 133,802, and the prevalence rate was 16.9 per million population. Indonesia is one of the top 3 countries with the highest new leprosy cases. [2] Leprosy principally affects the skin, the peripheral nerves, the mucosa of the upper respiratory tract, and the eyes. The diagnosis of leprosy is based on the presence of at least 1 of 3 cardinal signs: definite loss of sensation in a pale or reddish skin patch, a thickened or enlarged peripheral nerve with loss of sensation, and/or weakness of the muscles supplied by the nerve, and the presence of acid-fast bacilli in a slit-skin smear.

Although leprosy predominantly involves peripheral nerves and skin, visceral involvement was recognized as early as 1894 by Hansen and Looft. There are also studies showing the effects of leprosy on the cardiac autonomic system, ECG changes, and cardiac amyloidosis. We also found no case report or special literature linking leprosy with pulmonary hypertension (PH).

Case Presentation

A 22-year-old man presented to the emergency department with progressively worsening shortness of breath during exertion and bilateral lower extremity edema

lasting for 3 months. He also reported experiencing fatigue during this period but denied any fevers, chills, orthopnea, joint pain, myalgias, or arthralgias. Additionally, he reported experiencing occasional chest pain during exertion for a similar duration, along with pruritus, swelling, and pain in the distal extremities and ears that had begun a year earlier. His past medical history was unremarkable.

The physical examination revealed mild bilateral pitting edema in the extremities, no jugular venous distension, a regular heart rhythm with a systolic murmur noted at the lower left sternal border, and bilateral air entry in the lungs. No evidence of peripheral cyanosis, arthritis, rash, jaundice, or skin telangiectasias was detected. There was also swelling of the earlobes, bilateral loss of eyelashes, and reddish skin patches accompanied by sensory loss.

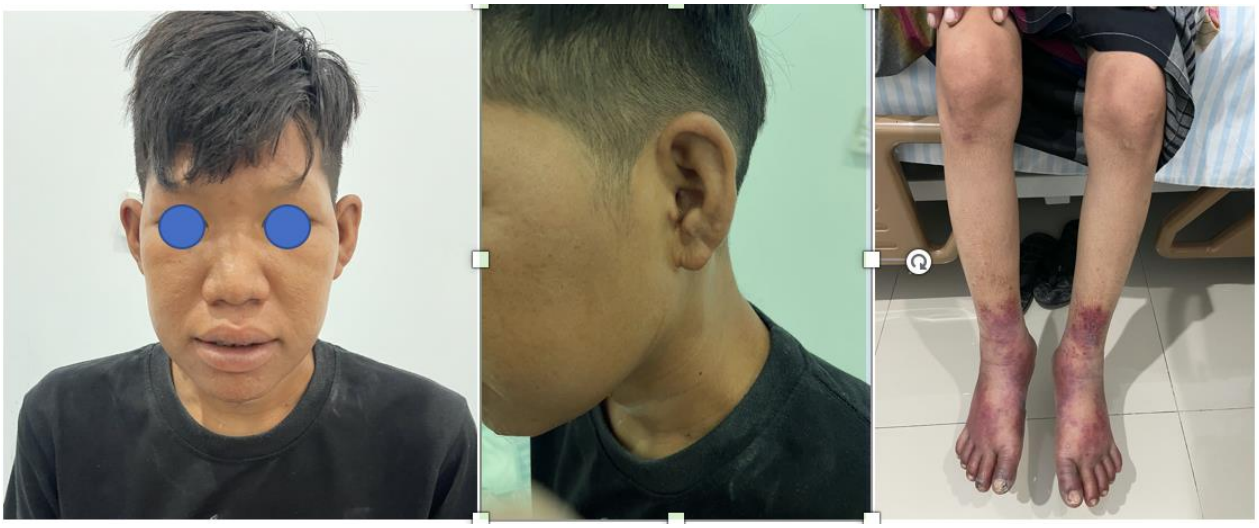

Figure 1: The images showcase the patient's clinical features, showing bilateral loss of eyelashes, swelling on the earlobes, and reddish skin patches with sensory loss.

The initial workup revealed a hemoglobin level of 11.6 g/dL, hematocrit of 38.8%, white blood cell count of 19.2 K/mL, platelet count of 171 K/mL, SGOT of 246 U/L, SGPT of 230 U/L, total protein of 6.1 g/dL, albumin of 3.2 g/dL, and globulin of 2.9 g/dL. The patient was diagnosed with multibacillary leprosy, as evidenced by a positive acid-fast bacilli test result. The chest X-ray was unremarkable. The 12-lead ECG showed sinus tachycardia, right-axis deviation, and inverted T waves in leads V_2–V_6, II, III, and aVF.

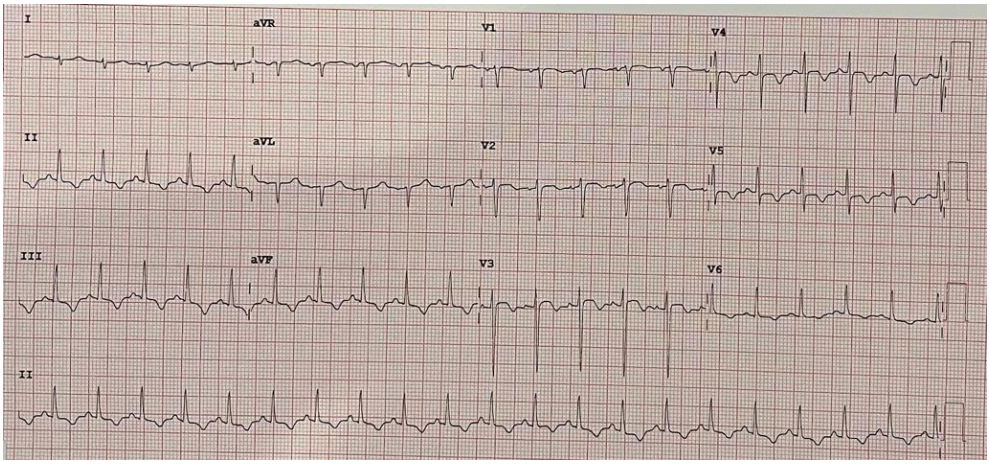

Figure 2: The patient's 12-lead ECG shows sinus tachycardia, right-axis deviation, and the T wave inverted in leads II, III, aVF, and V_3–V_6.

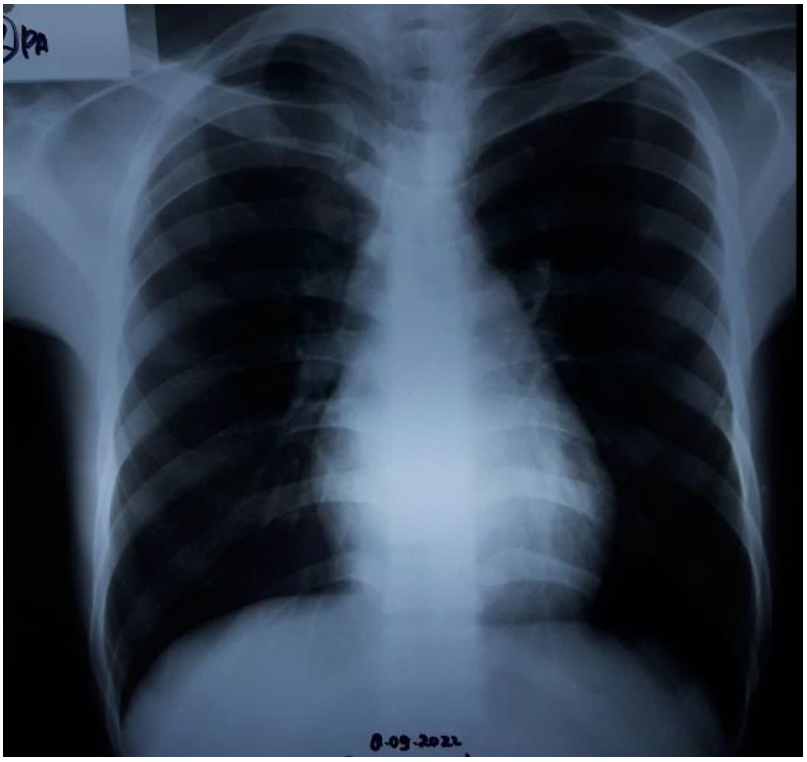

Figure 3: The patient's chest X-ray demonstrates unremarkable findings.

On admission, an echocardiogram revealed preserved left ventricular systolic function with a left ventricular ejection fraction of 68%. The study also showed right atrial and right ventricular dilatation, a D-shaped left ventricle with intraventricular septal flattening, and tricuspid regurgitation with a peak velocity of 3.43 m/s and a maximum pressure gradient of 47 mm Hg, indicating a high probability of PH. No intracardiac shunt was detected, and no definitive causes for the development of PH were identified in this case. Consequently, the patient was diagnosed with leprosy associated with PH.

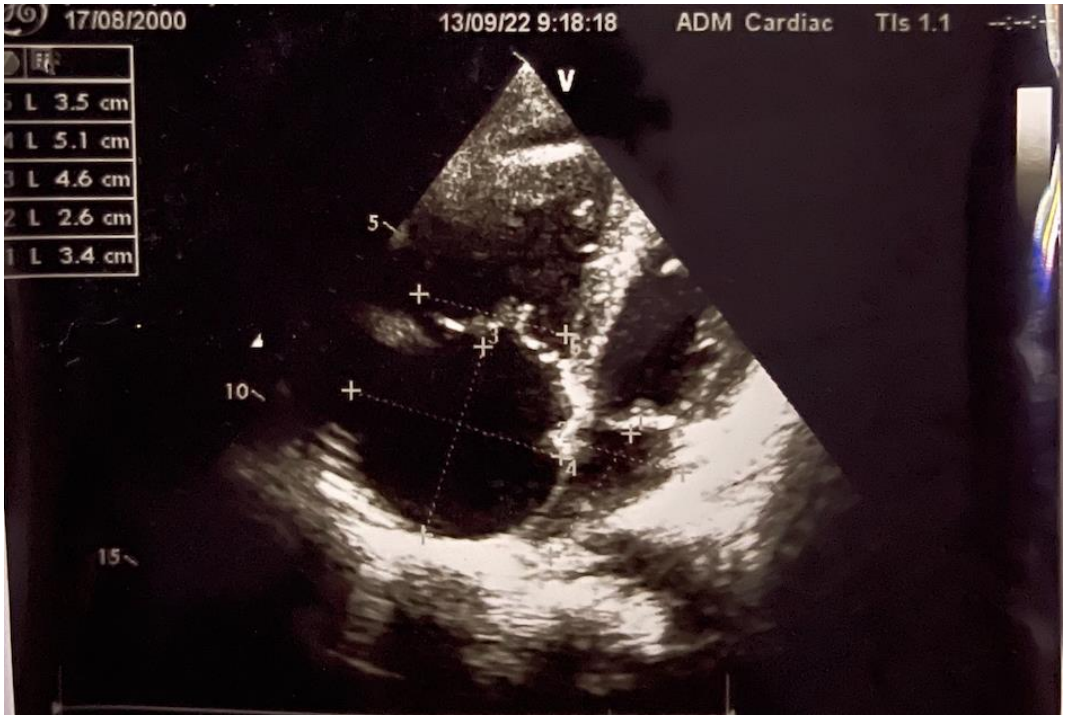

Figure 4: The patient's 2D echocardiography in the apical 4-chamber view shows right atrial and right ventricular dilatation.

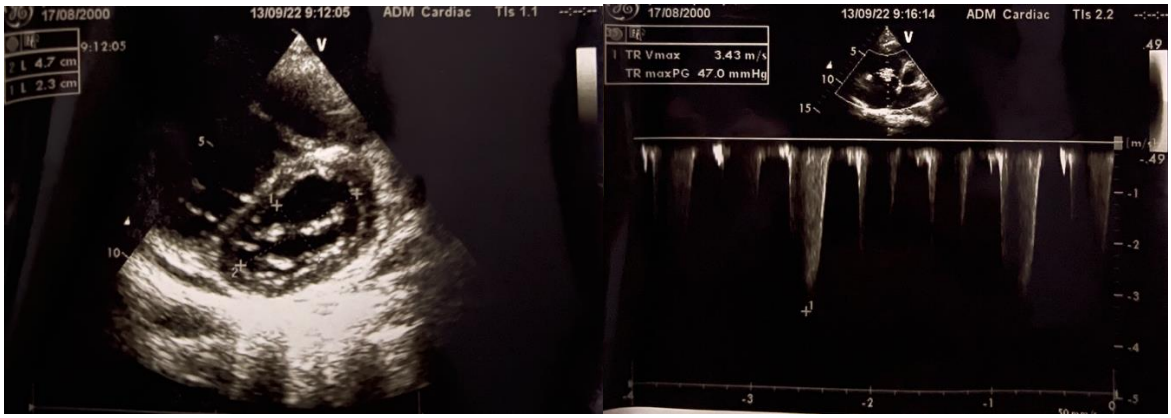

Figure 5: The patient's 2D echocardiography in the parasternal short-axis view shows a D-shaped left ventricle (left). The continuous wave Doppler study reveals tricuspid insufficiency with an estimated peak pressure gradient of 47 mm Hg.

During hospitalization, the patient was treated with diuretics and discharged with symptomatic improvement. The Department of Dermatology and Venereology was consulted for leprosy management. The patient was followed up in the outpatient department in stable condition and scheduled for echocardiographic monitoring during and after leprosy treatment

DISCUSSION

This article reports the diagnosis of PH in a patient newly diagnosed with leprosy. To our knowledge, this is the first case of leprosy associated with PH. The patient had no risk factors, including smoking, alcohol consumption, or sexually transmitted diseases. The diagnosis of PH was confirmed by transthoracic

echocardiography. It is challenging to definitively describe the pathogenesis of PH in this patient with leprosy, but a possible pathomechanism can be proposed. Pathomechanisms that may occur in these patients include metabolic alterations resulting from leprosy itself or the presence of other coexisting conditions. Anitha and Priyadharshini [3] assessed respiratory function in 23 treated leprosy patients and concluded that patients with leprosy suffered from impaired lung function. Significantly, 69.6% of their studied patients manifested the impairment. In addition, lepromatous patients had restrictive (66.7%) and obstructive (33.3%) patterns, while tuberculoid had a 36.4% restrictive pattern.

Structural changes in the lung parenchyma and functional abnormalities in gas exchange can lead to the development of PH in patients with leprosy, which, in turn, can result in right ventricular remodeling and hypertrophy.

In this patient, we did not evaluate for other infectious diseases, as there were no relevant risk factors present. Nevertheless, several infectious diseases, such as HIV and hepatitis, are known to be associated with PH.

CONCLUSIONS

The existing literature offers several case reports of dilated cardiomyopathy associated with leprosy; however, there are no documented cases of PH in patients with leprosy. Impairment of lung function is the primary pathomechanism underlying PH related to leprosy. This condition can be classified within the group of PH caused by lung disease.

Conflict of Interest: None declared.

REFERENCES

1. World Health Organization. Guidelines for the diagnosis, treatment and prevention of leprosy. New Delhi: WHO, Regional Office for South East Asia; 2018

2. World Health Organization. Weekly epidemiological record: global leprosy update, 2021: moving towards interruption of transmission, WHO. Geneva; 2022, 36: 429-50

3. Anitha A et al. Pulmonary dysfunction-an overt leprosy sequel: study done in rehabilitation centre. Int J Res Med Sci. 2016; 4(7): 2843-2849