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Lymphatic Filariasis Caused by *Brugia timori* Presenting as Lower Limbs Edema in A Pediatric Patient in Malaka Regency, East Nusa Tenggara, Indonesia

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ABSTRACT

There were 12,677 cases of LF in Indonesia, 2,864 of them were found in East Nusa Tenggara. People who suffer from LF usually have no clinical manifestation until adults. This rare case report found a pediatric case caused by LF in a 14-year-old male with marasmus. This study adopts a case study design to investigate the chronic progressive swelling of both lower extremities in a 14-year-old boy, attributing the condition to lymphatic filariasis caused by *Brugia timori.* Anthropometric measurements revealed Length For Age (LFA) $<P_5$, Weight For Age (WFA) $<P_5$, Ideal Body Weight (IBW) (%) 55,64%, and Upper Arm Circumference (UAC) $<P_5$. The physical examination showed both lower extremities have non-pitting edema, and no hydrocele was found. Through night, a blood smear with Giemsa stain examination confirmed the character of Brugia timori's microfilaria. The patient got Diethylcarbamazine and Albendazole for two weeks. After two weeks of medical and nutritional intervention, we performed a blood smear evaluation, and no microfilaria was detected. He also gained weight of 2,8 kg after nutritional intervention. Pediatric patients rarely suffer from lower limb edema caused by LF infection. Traditional regimens 200 mg of Diethylcarbamazine (DEC) and also 400 mg of Albendazole still work for this patient. Furthermore, this new case of LF should become the government's matter of interest to apply good policy to reach elimination status.

Keywords: *Brugia timori*, children, lymphatic filariasis, and leg swelling

INTRODUCTION

Lymphatic filariasis is estimated affects 51,4 million people globally.[1] Even though the disease is endemic in 80 nations, India, Nigeria, Bangladesh, and also Indonesia reaching out for 70% of cases of LF worldwide.[2] It is estimated there are 12,677 cases of LF in Indonesia, and 2,864 of the cases are found in East Nusa Tenggara province.[3] In areas of high intensity of transmission, infection of LF is generally found during childhood. Lymphatic filariasis in children usually asymptomatic, but once it manifest rarely presenting as bileteral leg swelling. A meta-analysis in children covering a literature search from 1966-2000 identified there were 57 lymphedema case of a limb in individuals 20 years old or younger.[4] In a 2007 study in a *Brugia malayi* endemic area, there were 4 pediatric cases had swelling of a limb out of 100 children who reported having microfilaremia. [5] In a filariasis endemic area of Haiti, from 186 school-aged children with positive microfilariae underwent physical examinations, none of them had limb swelling as clinical manifestation.[6] Other prior studies in Indonesia, Spain, Brazil, Indonesia, Nigeria, and India that were published after 2008, reported pediatric cases with positive microfilaremia without lymphedema of extremities as clinical features. [7–11] Here a case of lymphedema found in a 14-year-old male that caused by LF.

Despite the prevalence of LF, there is a dearth of research specifically addressing pediatric cases with positive microfilaremia and lower limb lymphedema. Existing studies, conducted in various endemic regions such as Brugia malayi-endemic areas, Haiti, Indonesia, Spain, Brazil, Nigeria, and India, have reported cases of positive microfilaremia in children without concurrent lymphedema. However, a research gap persists, particularly regarding the occurrence and management of lower limb edema in pediatric LF patients, making the presented case of a 14-year-old male in Malaka Regency, East Nusa Tenggara, Indonesia, noteworthy.

This case report aims to contribute to the existing body of knowledge by providing insights into a rare pediatric case of LF-induced lower limb edema. By utilizing traditional treatment regimens involving Diethylcarbamazine (DEC) and Albendazole, the study explores the effectiveness of these interventions in the context of pediatric LF. This investigation not only addresses the current research gap but also offers valuable information for the formulation of public health policies aimed at LF elimination in endemic regions. The novelty and specificity of this research lie in its focus on the pediatric population, shedding light on an underexplored aspect of LF manifestation and treatment. The outcomes of this study hold implications for healthcare practices and underscore the importance of targeted interventions in the fight against LF, aligning with global efforts to eliminate this debilitating disease.

RESEARCH METHOD

This study adopts a case study design to investigate the chronic progressive swelling of both lower extremities in a 14-year-old boy, attributing the condition to lymphatic filariasis caused by *Brugia timori.* The case study approach allows for an in-depth exploration of the specific case, capturing the nuances and details relevant to the manifestation, treatment, and outcomes.

The primary subject of this research is a 14-year-old boy residing in the rural area of Malaka Regency, Timor Island, who has experienced chronic swelling in both lower extremities. The study aims to understand the progression of symptoms, associated complications, and the response to the implemented treatment.

Data collection involves a comprehensive approach, including clinical assessments, laboratory investigations, and interviews. Anthropometric measurements were conducted to assess the patient's nutritional status, and a detailed physical examination was performed to identify clinical manifestations. Laboratory investigations comprised thick blood film smears, conducted both at noon and night, utilizing Giemsa stain to confirm the presence of *Brugia timori* microfilaria.

The Case

A boy suffered from a chronic progressive swelling of both lower extremities. The problem had begun in his left ankle twelve months earlier and had extended to the left lower limb. The boy then developed episodes of fever. A few months after the swelling was first noted in his left ankle, the same symptoms arose in the boy’s right ankle and lower limb. The patient also complains of intermittent stabbing sensations in the lower extremities, especially at night. The patient said some wounds started to appear on the soles of his feet which made him walk painfully and prevented the patient from going to school for the last two months. This boy had already stayed and lived with the parents in the rural area of Malaka Regency, Timor Island since birth. Malaka Regency is an LF endemic area and already implemented preventive chemotherapy from 2015 to 2019. However, the boy and his mother said they did not remember having received this medication.

On the physical examination, vital signs were normal. A 14-year-old boy weighing 30.6 kg, 142.7 cm with an upper arm circumference of 17 cm. The anthropometric calculation revealed weight for age was <P5, length for age was <P5, upper arm circumferences were <P5 and percentage of ideal body weight was 55,64% classified as severe malnutrition. Non-pitting edema has been present on both lower extremities. Both lower extremities had dermatological abnormalities such as thick and hard grey-yellowish crust. Other systems were normal, including there were no *lymph scrotum* or hydrocele found in this patient.

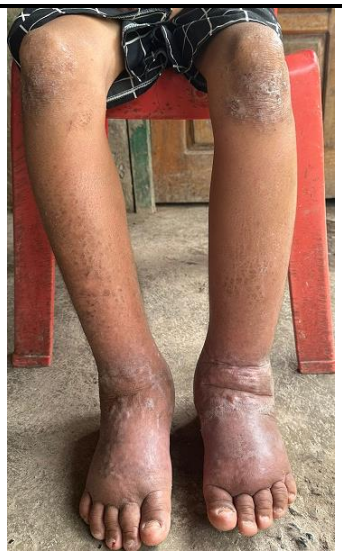

Figure 1. *Lower Legs Swelling with Non-Pitting Edema*

The laboratory result showed albumin 2,8 g/dL. Thick blood film smears were performed on this patient at noon (10.50 a.m.) and at night (11.35 p.m.). There were no parasites found in the noon sample. A night blood smear examination confirmed *Brugia timori*. The general feature of the parasite in blood smear appears to be stiff curved with a pointy end tail and no sheathed.

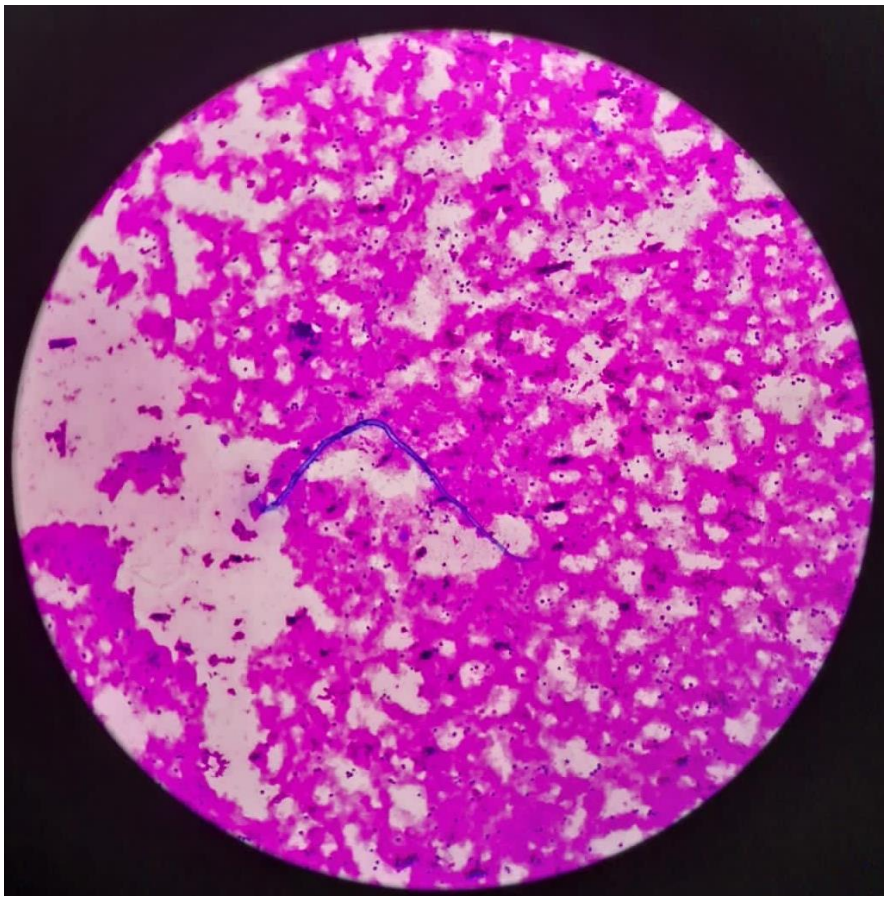

Figure 2. A Night Blood Smear Showing *Brugia timori* (Giemsa Stain with 10x Oil Magnification)

The patient was hospitalized and began treatment in daily dose of 6 mg/kgBW diethylcarbamazine (DEC) for 12 days and albendazole for 400 mg of dose to eradicate the circulating microfilaria and the adult worms. The patient

got a malnourished formula of F-100 a day and was prescribed a high-calorie, high-protein oral nutritional supplementation for his severe malnutrition. He was also prescribed multivitamins and trace elements. Intravenous albumin administration to solve hypoalbuminemia. After 12 days of DEC and albendazole treatment, he was discharged from the hospital with the noon and night blood smear tests showing no microfilaria. He gained 2.8 kgs and his albumin level was improved to 3.6 g/dL.

DISCUSSIONS

The universal burden of case infection is still found mostly in Asia, predominantly in Indonesia and Papua New Guinea.[12] The occurrence of LF among the pediatric population is not clearly elucidated. The filariasis prevalence in children aged 5-18 years in Brazil was 13,8% while in Samoa revealed prevalence in children ≥ 10 years (4,7%) was 3 times higher than in children 5-9 years (1,3%).[13,14]

The gold standard diagnosis of LF is to detect the microfilariae presence inside blood or lymphatic vessels. The specimens of the peripheral blood should be collected between 10.00 p.m. to 02.00 a.m. because the parasite actively circulates in the blood system at night. The type of LF infection usually correlates with the particular clinical manifestation. In *Wucheria bancrofti'*s infection will be found lymphedema in the scrotum and whole extremities, but the manifestation of lymphedema in *Brugia sp* was found below the knee or below the elbow.3,15 A night blood smear examination confirmed *Brugia timori* in this patient. The lymphedema in both lower extremities which were below the knee and there was no lymph enlargement in the scrotum as the clinical manifestation in this patient supports this type of infection. The fact that this boy had lived in Timor Island since birth also supports the evidence of LF which was caused by *Brugia timori.*

Traditionally, DEC alone, Ivermectin alone, or with one of these agents paired with albendazole. The WHO currently recommends triple therapy, which is a single dosage of ivermectin, albendazole, and single dosage or given as a 12-day course of DEC.[16,17] This patient got a 12-day course 200 mg of DEC and a single dosage 400 mg of albendazole, and there were clinical improvements, and the blood smear evaluation revealed no microfilaria.

Malnutrition increases vulnerability to infection and is commonly associated with recurring and variable infections.18 Severe malnutrition can become such a serious and also highly public health issue prevalent, particularly in poor or developing countries. Several factors give influence toward adequate status of nutrition, these are like socioeconomic poverty or deprivation. The socio-economic factor is the main reason for the severe malnutrition of this patient. [19] In this patient, besides socioeconomic factors, feeding practice also plays an important role. He had only rice and *Moringa oleifera'*s leaves for his daily diet which provided a high carbohydrate and also load of calorie but did not fulfil the protein requirement for his age. In this patient, we give prompt treatment to treat severe malnutrition. He gained weight of 2,8 kg after being discharged from the hospital. Furthermore, this new case of LF should become the government's matter of interest to apply good policy to reach elimination status.

CONCLUSIONS

This research highlights the presence of lymphedema in a 14-year-old male caused by lymphatic filariasis (LF) in the endemic area of Malaka Regency, Timor Island, Indonesia. The patient exhibited chronic progressive swelling in both lower extremities, intermittent stabbing sensations, and dermatological abnormalities on the soles of his feet. Laboratory results confirmed the presence of Brugia timori microfilariae in the night blood smear. The treatment approach involved a 12-day course 200 mg of diethylcarbamazine (DEC) and a single dose 400 mg of albendazole, along with nutritional supplementation to address severe malnutrition. The patient showed clinical improvement, with no microfilariae detected in blood smears after treatment. Additionally, the patient gained weight, and there was an improvement in albumin levels. This study underscores the ongoing burden of LF in Asia, particularly in Indonesia, and emphasizes the need for effective public health strategies to achieve elimination. The gold standard diagnosis of LF involves detecting microfilariae in peripheral blood, preferably collected between 10:00 p.m. and 02:00 a.m. The manifestation of lymphedema in this case, primarily below the knee, supports the diagnosis of Brugia timori infection. Socio-economic factors, including living conditions and feeding practices, contribute to individuals' vulnerability to severe malnutrition, exacerbating the impact of LF. Prompt treatment for severe malnutrition, including nutritional supplementation, is crucial for the overall well-

being of affected individuals. In conclusion, this case emphasizes the importance of integrated approaches, including diagnosis, treatment, and nutritional support, in addressing lymphatic filariasis and its associated complications. The findings call for continued efforts by the government to implement effective policies aimed at eliminating LF and improving the overall health and nutrition of affected populations.

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