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Case Report

Trichuris dysentery syndrome, the neglected tropical disease: a case series

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Almost 2 billion people, about a quarter of the world's population, are infected with soil-transmitted helminths (STH) worldwide. Approximately 270 million preschool children and more than 550 million school-age children live in areas of extensive parasite transmission.[1,2] Indonesia is a moderate-to-high-risk area of STH, with an overall mean prevalence of 28.12%. However, the prevalence in Papua is higher.[3] A study reported that 50% of school-aged children in Jayapura, Papua, a high-risk area, suffered from STH, with distributions of 48.5% Ascaris lumbricoides, 28.6% Trichuris trichiura, 14.3% hookworm, and 8.6% mixed infection.[4] **[Paediatr Indones. 2022;62:430-4 ; DOI: 10.14238/pi62.6.2022.430-4]**.

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In tropical and subtropical regions worldwide, trichuriasis is common, but often neglected.[1] Clinical manifestations differ according to degree of infection, with generally asymptomatic mild infection. When larger numbers of worms (>300) are present, chronic bloody diarrhea or symptoms of dysentery may develop in association with dehydration, anemia, and malnutrition, reaching a failure to thrive state that causes stunting.[5-7] Adult worms are found in the large intestine, especially the cecum, but in heavy infections, they can be found throughout the colon and rectum, such that rectal prolapse may occur.[5,6] Trichuris dysentery syndrome (TDS) is chronic bloody diarrhea caused by heavy trichuriasis, often characterized by mucus in diarrhea, prolapsed rectum, iron deficiency anemia (IDA), and clubbed fingers.[6-8]

Trichuriasis can be diagnosed by finding typical eggs through direct fecal smears or with concentration techniques. The eggs are barrel-shaped, with refractile plugs at both ends and usually measure 50 to 55 μm long by 22 to 24 μm wide.[9] Egg quantitation techniques occasionally may be requested to assess infection intensity, therapeutic efficacy, and reacquisition rate of parasites. The eggs are passed in the stool, mature in warm, moist soil, and require approximately 3 weeks to become infective. Under ideal conditions, the eggs survive for 1 year. After the embryonated egg is ingested, the first-stage larvae begin maturing in the distal small bowel. The larvae then migrate to the cecum, where they mature into adult worms. Males and females measure up to 50 mm in length; they remain attached to and penetrate the intestinal mucosa by the long, slender anterior end, while the thicker posterior end hangs free in the lumen. Female worms are elongated, whereas the tails on males are coiled.[2]

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The heavy infection called TDS correlates with poor sanitation and is often misdiagnosed as dysentery diarrhea by health workers, thus the helminth infection does not resolve since it is not managed promptly. That misdiagnosed issue is very important to discuss to improve child health, especially in remote areas of Papua.

The cases

This case series was conducted in the Pediatric Ward at Scholoo Keyen Hospital, Sorong Selatan, Papua Barat from September 2018-February 2019. Three boys aged 4, 7, and 11 years were hospitalized with the same chief complaints of severe anemia and heart failure. They came from a poor, rural family, with low socioeconomic background, poor hygiene, and lack of access to good medical services. They also had a history of long-term, ongoing pica, due to neglect by parents and health providers. All three patients complained of recurrent bloody diarrhea and worm contamination in vomit and feces. Stool consistency was not always watery, frequency was 2-6 times per day often accompanied by mucus, and recurring 2-3 days per week. The three patients had no other bleeding manifestations except bloody diarrhea. Furthermore, their nutritional intake was also insufficient in quality and quantity. They had been diagnosed with bacterial dysentery diarrhea in the past and took antibiotics for several days, but the complaint persisted.

The boys' clinical manifestations were similar: they all looked very thin, stunted, weak, and pale, in otherwise stable vital signs. Their nutritional status was categorized as marasmus severe malnutrition with stunting. Cardiac examination revealed systolic murmur in 3-4th left parasternal border and cardiomegaly. The liver was palpable about 2-4cm below the costal arch, and clubbed fingers were noted in all of their extremities. Clubbing indicates prolonged hypoxia as a result of chronic anemia.[5] Routine blood examination result revealed microcytic hypochromic anemia. Fecal smear examinations also showed multiple Trichuris eggs. At 3-4 days prior to treatment, multiple worms were also visible in the feces, but they decreased after therapy. The children received malnutrition management and were screened for possible tuberculosis infection.

The first patient was a boy aged 7 years, diagnosed with severe anemia, heart failure, marasmus severe malnutrition, history of intestinal obstruction due to heavy ascariasis several years ago, trichuriasis, bloody diarrhea with rectal prolapse, and hypoalbuminemia. The duration of TDS was 4 years. We started him on a program of malnutrition management, PRC transfusion, albendazole 400 mg/day for 3 consecutive days, and rehydration solution for malnutrition after every episode of diarrhea. After 1 week, his condition improved, no tuberculosis infection was found, the bloody diarrhea and rectal prolapse resolved, and malnutrition management continued at home. At a follow-up visit to the outpatient clinic three weeks later, fecal examination showed no Trichuris eggs and the child had gained weight.

The second patient was a 3.5-year-old boy, with a TDS duration of 2 years, and diagnosed with severe anemia (Hb 1.8 g/dL), heart failure, marasmus severe malnutrition, ascariasis, trichuriasis, recurrent bloody diarrhea, and lung tuberculosis. Peripheral blood smear showed aniso-poichilo cytosis, microcytic hypocromic erythrocytes (**Figure 1**). The child underwent PRC transfusion, malnutrition management, anthelmintic with 400mg/day albendazole for 3 consecutive days, and a lung tuberculosis regimen. The patient was discharged after one week of hospitalization with an improved condition and body weight gain. Two weeks later, the patient visited the outpatient clinic; no Trichuris eggs were found in the fecal examination at that time.

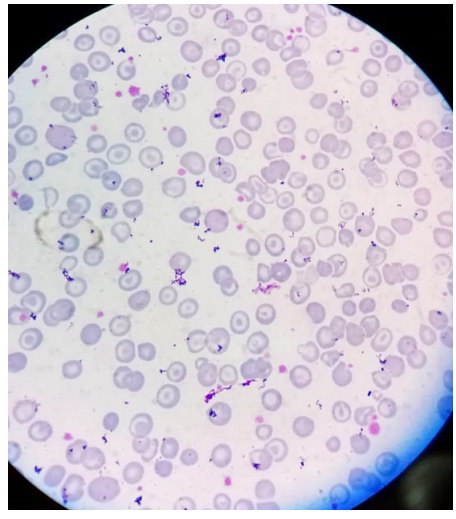

Figure 1. Peripheral blood smear of the second patient with severe anemia (Hb 1.8 g/dL)

The third patient was an 11-year-old boy, already diagnosed with severe anemia, stunting, severe marasmus malnutrition, bloody diarrhea with recurrent rectal prolapse, and trichuriasis. This patient had already been hospitalized 4 times with anemia. He was the eldest, and had the longest period of TDS. The patient received PRC transfusion, malnutrition management, and anthelmintic albendazole 400 mg/day for 3 consecutive days. During his 8-day hospitalization, the rectal prolapse resolved. One week after discharge, the patient visited the outpatient clinic. Fecal examination showed multiple Trichuris eggs (**Figure 2**), and his parents said that sometimes worms still appeared in stool, but decreased in number as compared to before. So, he received 200mg/day mebendazole for 5 more consecutive days at home. When we evaluated him again 3 weeks after, no Trichuris eggs were observed in the fecal exam.

All three patients were monitored monthly in the pediatric outpatient clinic to continue malnutrition management and oral iron therapy. The characteristics of patients are shown in **Table 1**.

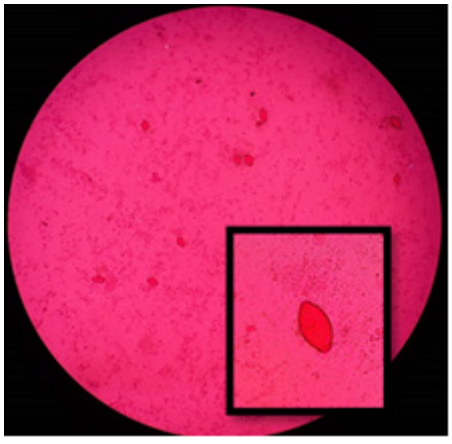

Figure 2. Microscopic fecal smear of the third patient showed multiple Trichuris eggs

Discussion

Trichuris dysentery syndrome endangers the growth and development of children because of its chronic and severe nature.[5] Studies from Honduras and India noted that TDS was related to IDA, infection, severe malnutrition, and stunting,[5] therefore, education on nutrition and hygiene should be included in the management. A study noted that adult TDS patients suffered from IDA by occult blood loss from colonic lesions.[7] Such lesions are presumably caused by petechial lesions and blotchy mucosal hemorrhages from the burrowing anterior end of the worm into the mucosa. during severe infection, they found mucosal edema, luminal narrowing, and lack of distensibility, with the histopathological findings of a slight increase in lymphocytic infiltrates in the lamina propria and the presence of eosinophils and plasma cells.[5,7] As such, TDS should be considered in the differential diagnosis of IDA accompanied by occult blood loss (bloody diarrhea) in endemic areas.[7]

Trichuris dysentery syndrome and related conditions are treatable (except stunting before 2 years of age, which affects cognitive function11) and, from our experience, show good response with comprehensive management and prompt medication. Routine fecal examination, anthelminthic therapy, and hygiene education are recommended for all family members of TDS patients to prevent recurrent infection. In addition to long-term monitoring, intense parental counseling is essential to enhance compliance.

To date, preventive chemotherapy (PC), i.e., periodic administration of anthelminthic drugs to at-risk populations without a prior diagnosis, is the cornerstone of helminth control put forth by the *World Health Organization* (WHO) to reduce the burden of STH. Preventive chemotherapy (PC)is implemented in the form of annual mass drug administration (MDA) campaigns. The recommended target populations have expanded from only school-aged children to include

Table 1. Characteristics of the three patients

Patient number	Sex	Age at diagnosis	Duration of TDS symptoms	Hemoglobin level, g/dL	Rectal prolapse
1	Male	7 years	4 years	5.7	Yes
2	Male	4 years	2 years	1.8	No
3	Male	11 years	6 years	5.5 At previous hospitalizations: 2.8, 5.8, 3.8	Yes

younger children (1-5 years of age), adolescent girls (10-19 years), women of reproductive age (15-49 years), and pregnant women after the first trimester in areas with an STH prevalence of ≥20%.[1] Mass drug administration program in Indonesia, especially in Papua, needs to be evaluated in terms of coverage and efficacy.

Anthelmintic treatment efficacy data against Trichuriasis are limited and varied. A previous study showed that the efficacy of albendazole vs. mebendazole in Trichuriasis was not consistent; one study revealed that albendazole alone had an 88.3% cure rate (CR),[12] but another study reported only a 10% CR.[13] The CR after combination ivermectin treatment was 38%, and after combination with levamisole was 95.8%. Moreover, egg reduction (ER) by albendazole was 40%.[12] Mebendazole showed better results with 19% CR, increased to 55% if combined with ivermectin, and 83.3% if combined with levamisole; the ER was 67%.[13] The efficacy of albendazole and mebendazole was better if combined with levamisole or ivermectin,[12,13] but those drugs are not available in Papua, so we used monotherapy in our cases.

Trichuriasis can be comorbid with another STH infection, so a study into types of STH in the area will be a key to eradication of STH, especially trichuriasis. The three cases diagnosed in 6 months in our series may represent the tip of the iceberg, because of limited medical services due to difficulty of the geographic area and limited medical providers in primary health care. We also recommend routine fecal examination for every patient with anemia, and malnutrition screening for those with STH especially *Ascaris, Trichuris,* and hookworm infection.

Scholoo Keyen District Hospital is the only hospital in Sorong Selatan that provides basic and specialist medical services, but special diagnostic tools for colonoscopy and histopathological biopsy were not available, so we did not evaluate our TDS patients by those modalities. Anthelminthic availability was also limited. We used albendazole alone as a single regimen and mebendazole as a second-line regimen for the third patient whose condition did not resolve with albendazole. The cure rate still needs to be monitored in that patient.

Further investigation of trichuriasis and other STH prevalence in Indonesia, especially Papua, should be conducted, followed by integrated eradication and prevention programs or policies. Development of such programs is challenging because low socioeconomic and education levels, difficult geography, and lack of primary health care. Soil transmitted helminths management requires not only empowerment of medical providers, but also engagement of local government, education providers, and prominent figures. Further study on the prevalence of STH and good program support is needed to improve child health in Papua.

References

1. WHO. Guideline: Preventive chemotherapy to control soil-transmitted helminth infections in at-risk population groups. Geneva: World Health Organization; 2017.
2. Schulz JD, Moser W, Hürlimann E, Keiser J. Preventive chemotherapy in the fight against soil-transmitted helminthiasis: achievements and limitations. Trends Parasitol. 2018;34:590-602. DOI: 10.1016/j.pt.2018.04.008.
3. Badan Litbangkes Kemenkes RI. Laporan Nasional RISKESDAS 2017. Jakarta: Kemenkes RI; 2017.
4. Martila M, Sandy S, Paembonan N. Hubungan higiene perorangan dengan kejadian kecacingan pada murid SD Negeri Abe Pantai Jayapura. J Plasma. 2016;1:87-96. DOI : 10.22435/plasma.v1i2.4538.87-96.
5. Kaminsky RG, Castillo RV, Flores CA. Growth retardation and severe anemia in children with Trichuris dysenteric syndrome. Asian Pac J Trop Biomed. 2015;5:591-7. DOI: 10.1016/j.apjtb.2015.05.005.
6. Noorizan AM, Raj SM. Trichuris dysentery syndrome : evidence that it may be underdiagnosed in Kelantan. Med J Malaysia. 2001;56:53-7. PMID: 11503297.
7. Khuroo MS, Khuroo MS, Khuroo NS. Trichuris dysentery syndrome : a common cause of chronic iron deficiency anemia in adults in an endemic area (with videos). Gastrointest Endosc. 2010;71:200-4. DOI: 10.1016/j.gie.2009.08.002.
8. Brooker SJ. Soil-transmitted helminth treatment: multiple drug regimens. Lancet Infect Dis. 2018;18:698-9. DOI: 10.1016/S1473-3099(18)30268-8.
9. WHO Working group on Soil-transmitted helminthiasis. Monitoring anthelmintic efficacy for soil transmitted helminths (STH). 2008. (cited 2020 February 20) Available from: https://www.who.int/docs/default-source/ntds/soil-transmitted-helminthiases/monitoring-anthelmintic-efficacy-for-sth-march-2008.pdf.
10. Agarwal A. Pica- an enigma of malnutrition. J Nutr Disord Ther. 2017;07:7-8. DOI: 10.4172/2161-0509.1000e132.
11. WHO. WHO Global Nutrition Targets 2025: Stunting Policy

Brief [Internet]. Genewa, Swiss; 2014 [cited 2019 Jun 27]. Available from: https://www.who.int/nutrition/topics/globaltargets_stunting_policybrief.pdf

12. Anto EJ, Nugraha SE. Efficacy of albendazole and mebendazole with or without levamisole for ascariasis and trichuriasis. Open Access Maced J Med Sci. 2019;7:1299-302. DOI: 10.3889/oamjms.2019.299.
13. Knopp S, Mohammed KA, Speich B, Hattendorf J, Khamis IS, Khamis AN, *et al.* Albendazole and mebendazole administered alone or in combination with ivermectin against Trichuris trichiura: a randomized controlled trial. Clin Infect Dis. 2010;51:1420-8. DOI: 10.1086/657310.