Emerging Multiple Parasite Reinfection as Neglected Tropical Disease: A Case Series

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Abstract

土传寄生虫感染在印度尼西亚流行，尽管有单次治疗和大规模治疗程序，也会有再感染的病例。描述了三种多种寄生虫感染，可能与慢性便血和生长迟缓有关。这些病例系列警告儿科医生，未经治疗的寄生虫感染是儿童出现胃肠出血的可能原因。在高风险地区进行环境监测可以防止再感染和生长迟缓。在流行病区，出现便血就需要进行早期的粪便常规分析。正确的诊断、及时的治疗和源头控制对于防止长期并发症至关重要。

Keywords
► trichuriasis
► multiple helminth infections
► gastrointestinal bleeding
► growth faltering

Introduction

土壤传寄生虫（STH）再感染常见于治疗后，且在发展中国家仍然是一种主要的热带疾病。STH包括Ascaris lumbricoides，Trichuris trichiura，以及钩虫（Necator americanus和Ancylostoma duodenale）。T. trichiura，或鞭虫，是最常见的四种蠕虫之一，影响全球4.6亿人。1,2

印度尼西亚的寄生虫感染在农村地区达到25%以上，24.6%的学龄儿童在印度尼西亚都有涉猎。3

大多数STH共有症状，包括生长迟缓和认知限制。在治疗后，Trichuriasis可以表现为慢性腹泻或肠梗阻，腹痛、恶心、呕吐、严重缺铁性贫血，以及脱肛。4

再感染STH可发生于个体化治疗后及大规模治疗计划中。这些案例系列警告了儿科医生，未经治疗的寄生虫感染是儿童出现胃肠出血的可能原因。在高风险地区进行环境监测可以防止再感染和生长迟缓。在流行病区，出现便血就需要进行早期的粪便常规分析。正确的诊断、及时的治疗和源头控制对于防止长期并发症至关重要。

Case Presentation

Case 1

患者A，一名17岁的男性，从西爪哇转诊到Cipto Mangunkusumo医院，评估10年前开始的持续性便血。怀疑原因是直肠多发性息肉。患者诉粪便有恶臭味，排便时肛门排出白色虫体和肛门内有肿块。他因慢性贫血进行了多次输血。体格检查显示正常的生命体征，但头围（<2标准差）和严重的生长迟缓（体重和身高<3%）；然而，营养状况良好（体重/身高91%）。直肠检查显示没有polypl/mass.
Laboratory tests showed normal hemoglobin, platelet count, leucocyte count, and blood clotting function. Parasitological stool examination using light microscopy showed eggs of *T. trichiura*, *A. lumbricoides*, and hookworm and cysts of *Entamoeba hystolitica/dispar* and *Entamoeba coli* (► Fig. 1). Colonoscopy examination showed hundreds of worms covering the mucosa of the colon, and no polyp was identified (► Fig. 2A, B). The patient was diagnosed with severe trichuriasis and multiple intestinal parasites infections. Mebendazole was administered at 100 mg twice daily for 5 days, and metronidazole at 500 mg three times daily for 10 days. Clinical symptoms improved after 5 days treatment, supported by no parasites found on re-evaluation.

**Case 2**

Patient N, a 5-year-old male, was referred to Cipto Mangunkusumo Hospital due to suspicion of rectal polyposis. He suffered from bloody diarrhea for 2 years before admission and also had a serial blood transfusion. He also had white worms coming out from the anus and rectal lump while...


The patient also was diagnosed with severe trichuriasis and multiple intestinal parasites infections. We administered mebendazole at 100 mg two times daily and metronidazole at 240 mg three times daily for 5 days. Clinical symptoms also improved after five days of treatment, and no parasites were found on re-evaluation.

**Case 3**

Patient AL, a 13-year old male, was the sibling of patient A and N. Patient had no gastrointestinal complaint; however, we performed parasites stool examination for screening purpose. Interestingly, evaluation result showed positive eggs of *A. lumbricoides*, *T. trichiura*, hookworm, and vacular form of *Blastocystis spp.*\(^{(\text{Fig. 1E})}\). The patient was treated with the same medications as cases 1 and 2. The patient reported improved clinical symptoms, and no parasites were found on re-evaluation examination.

We identified a slight difference in all three cases. Case 1 and case 2 showed major long-term consequences of untreated chronic multiple helminths infections, whereas case 3 showed mild STH infection without any symptom present.

**Six Months of Follow-up after Treatment**

We performed a new parasitological stool examination 6 months after treatment on each patient. Case 1 had positive eggs of *T. trichiura*, *A. lumbricoides*, and *B. hominis*, while case 2 and 3 had both eggs and adult worms of *T. trichiura* and hookworm. Evaluation of additional family members (parents and one other sibling) showed positive multiple parasite infections (eggs of *T. trichiura*, *A. lumbricoides*, and hookworm and cysts of *Giardia lamblia* and *Blastocystis hominis*). Laboratory tests were not performed. All family members started their treatment with mebendazole with additional metronidazole as conjunctive treatment.

**Discussions**

Without adequate treatment, trichuriasis could cause severe infection resulting in chronic bloody diarrhea, rectal prolapse, malnutrition, short stature/stunting, and severe anemia due to chronic bowel mucosa infection.\(^{7}\) As shown in case 1, chronic malnutrition occurred due to 10 years of inadequate parasitic treatment, resulting in stunting and microcephaly.\(^{6}\) Anal lumps due to rectal prolapse are often caused by increased intra-abdominal pressure during diarrhea and irritation of bowel mucosa caused by severe trichuriasis infection. Trichuriasis infection could cause loss of red blood cells directly from the gut, followed by iron deficiency anemia and growth faltering. Thus, increased tumor necrosis factor-α as an inflammatory mediator is thought to inhibit patient’s appetite and interfere with metabolic processes.\(^{7}\)

The patients’ families came from low economic status, had low education, lived in an environment with poor sanitation (e.g., lack of hand washing before meal and walking barefoot), limited water supply, and grounded house, with defecation habits in inappropriate places (e.g., land or river) in consequence of unavailability of the private latrine. These lifestyles further caused multiple parasitic reinfections in all family members although effective parasitic treatment had been achieved. The availability of latrine, periodic health evaluation and education, and better hygiene promotions by health care providers will reduce infections.\(^{3}\)

Treatment of trichuriasis requires the combination of at least two anthelmintic regimens, as monotherapy was deemed unsatisfactory.\(^{8}\) Albendazole monotherapy resulted in the lower cure rate compared with mebendazole (2.6 vs. 11.8%) for trichuriasis.\(^{9}\) However, albendazole is a drug of choice for hookworms and *A. lumbricoides* infection. In triple infections cases, oxantel pamoate–albendazole regimen was reported to be superior to mebendazole, with a cure rate of 31.2 versus 11.8% and egg reduction of 96 versus 75%.\(^{9}\) Oxantel pamoate has low efficacy in eradicating hookworm and *A. lumbricoides*. Single-dose mebendazole had a cure rate of 92.6% for *A. lumbricoides*, 27.5% for *T. trichiura*, and 25.5% of hookworms, and high egg-reduction rate for all three cases.\(^{10}\)

In Cuba, mebendazole 500 mg twice a year was used for mass drug administration in the eradication of trichuriasis.\(^{11}\) This is in accordance with our serial cases showing 5 days of mebendazole monotherapy treatment effective in curing multiple helminths infections, as proved by parasite stool re-evaluation showing negative results in all cases.\(^{10}\) Moreover, albendazole availability in Indonesia is limited to primary health centers. Furthermore, combination regimens were not used in the cases above to prevent anthelmintic resistance. Metronidazole is only used in patients with *E. histolytica* co-infection who are also suspected of having bacterial overgrowth.

Reinfection can occur rapidly after treatment with standard regimen, particularly for *A. lumbricoides* and *T. trichiura*.\(^{12}\) Reinfection was higher in stunted children, with inadequate STH treatment, poor sanitation/hygiene, and scarce clean water supplies.\(^{13,14}\) Following the administration of albendazole–oxantel pamoate treatment, it was reported that post-treatment the reinfection rate of *A. lumbricoides* reached 34.6% and *T. trichiura* 37.2% in 18 weeks.\(^{15}\) Other strategies to control STH infections include regular deworming of school-aged children, which is considered to be more cost-effective in settings where resources are limited. In addition to reducing reinfection rate, long-term solutions needed include improvement in water quality, sanitation, hygiene, poverty reduction, health education, and attention to chronic malnutrition.\(^{13–15}\) The parents were educated for the prevention of reinfection before returning to their hometown.
Conclusions

*Trichuris trichiura* may result in severe infection such as chronic bloody diarrhea, recurrent abdominal pain, severe anemia, loss of appetite, weight loss, rectal prolapse, malnutrition, and severe stunting. Bloody diarrhea should be evaluated early by routine fecal analysis and colonoscopy if possible and treated promptly to prevent long-term complications. Recurrent infections of helminths and parasites, especially in low-educated and poor hygiene areas, should be prevented by continuous health evaluation and education by local health care providers. Health education should be incorporated in school curricula, which will inculcate awareness among the younger generation and eventually disseminating to families and larger communities. In addition, cooperation with the local government in regulating environmental household programs is also essential. Periodic helminths evaluation should be done by local health care providers in endemic areas for early diagnosis, and anthelmintic regimen should be given if needed. Treating worm infection without proper source controlling was impractical and not economical.

Consent for Publication

Written informed consent was obtained from the patient for the publication of this case report and accompanying images.

Authors’ Contributions

F.S.A. coordinated the writing of the case series and was the attending senior consultant who examined the patient and performed a colonoscopy examination. A.A.P. helped in the drafting of the manuscript and follow-up of the patient. A.K. helped with parasitology examination of the patients and writing of discussion, and A.H.P. helped to draft the manuscript and supervise the project. All authors have read and approved the final version of the manuscript.

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Conflict of Interest

None declared.

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