Letters to the Editor

Disseminated Cystic Echinococcosis Cured With Lengthy Albendazole and Praziquantel Oral Therapy

To the Editors:

A 12-year-old boy with ill-defined chest pain was referred to us due to a chest radiograph (Fig. A, A1) demonstrating several round lung lesions. The initial hemogram demonstrated eosinophils 1,060/μL. A serum test for antibodies against *Echinococcus granulosus* was positive. A computed tomography (CT) scan of the chest and abdomen demonstrated 5 cysts in the lungs (3 on the left), 10 smaller hepatic cysts, along with a single 22-mm cyst in the splenic hilum. Whole-body magnetic resonance imaging (MRI) confirmed the above lesions and showed an additional single 15-mm cyst on the right sacral bone. No cysts were detected inside the peritoneal cavity or brain. He was started on albendazole 400 mg per os bid with the goal of completing 3 monthly cycles with drug-free intervals of 14 days. During the first drug-free interval, he was readmitted because of right-sided thoracic pain. One month later, that is, again soon after albendazole was discontinued due to the prescribed treatment cycle, he returned with right-sided thoracic pain. Therefore, we received informed consent from the family and started daily oral therapy with albendazole 400 mg per os bid and praziquantel 600 mg per os 3 times per week for 2 months because the regional supply of praziquantel was limited. After the first 2 months of therapy, a generous supply of praziquantel was imported from Belgium by the family, and he completed 16 months of uninterrupted daily oral therapy with albendazole 400 mg bid along with praziquantel 600 mg bid. Both drugs were taken sequentially with fatty foods. Therapy was extremely well-tolerated, with absence of hematologic, hepatic, or other toxicity. Although ultrasonography is the method of choice for diagnosis, staging, and follow-up of cystic echinococcosis (CE) cysts, we used whole-body MRI and thoracic CT because of the disseminated nature of his disease with multiorgan involvement. Imaging studies confirmed the impressive response to medical therapy (Fig. A, A2, A3, B and D). The patient is now more than 3.5 years off therapy and continues to be in excellent health.

In Europe, albendazole is licensed for interrupted treatment of CE, as described above, usually for a maximum of 3 cycles. However, the French Agency for the safety of medical and health products stresses that continuous treatment may be recommended in disseminated disease, since it achieves increased cyst nonviability with comparable toxicity with the interrupted regimen.1 Praziquantel, an effective drug against the intestinal stages of *E. granulosus* in carnivores, has been added to albendazole for combined treatment of CE and was found to substantially increase the serum concentrations of its active metabolite, albendazole sulfoxide.2 Praziquantel side effects are temporary and dose-related.3 Alvela-Suarez et al administered combination therapy in 57 patients with CE.4 Only 8 patients reported mild adverse effects, mostly gastrointestinal, followed by headaches and dysgeusia. Although cysts of CE which become inactive through treatment need close monitoring due to potential reactivation,5 the extended follow-up in our patient makes us feel confident that his disease will not relapse. Praziquantel deserves a randomized clinical trial of combination therapy versus albendazole monotherapy in patients with similar responsive cyst stages. However, steps must be taken to make praziquantel easily available and affordable in endemic countries.

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The Issue of Body Mass Index Increase in Adolescents Living With HIV on ART

To the Editors:

We have read with interest the recently published letter by Yeoh et al1 describing an increase in body mass index (BMI) in children living with HIV, switched to tenofovir alafenamide fumarate (TAF) or dolutegravir containing regimens. The integrase inhibitor dolutegravir is, since 2018, the first-line therapy for people living with HIV, including children and adolescents.2 Recently, data from clinical trials including adults raised concern as a notorious weight gain was described among study participants receiving dolutegravir combined with TAF.3 Whether this effect is related to immune reconstitution or should be considered a detrimental effect

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of the antiretroviral regimen has been object of intense debate. Without data from clinical trials in children and adolescents, a possible effect on BMI is extremely worrisome for pediatricians, especially for those managing adolescents. On one hand, because hormone changes could amplify these effects in body composition, contributing to obesity, metabolic and cardiovascular risk in this vulnerable population. On another hand, because the fear of weight gain could have a negative impact on treatment adherence, which is the main concern of all clinicians treating adolescents. We therefore agree with the authors that the possible effect on weight of first-line treatments should be addressed carefully. However, the analysis of body composition during adolescence is challenging, as natural growth leads to a BMI increase in this unique period of life.

In October 2020, Neil Thivalapill et al published the first retrospective cohort of virologically suppressed adolescents 10–19 years living with HIV in Eswatini and reported an increase in the rate of BMI, especially among women, after switch to dolutegravir. However, authors did not standardized BMI by gender and age according to reference growth charts and were unable to analyze the potential effect of TAF, which was not included in the regimen. However, these data are consistent with previous studies in adult populations, reporting a significant weight gain among sub-Saharan women. Some authors have suggested that these findings may be related to potential genetic factors and therefore the data presented by Yeoh et al are interesting, as they report data from another continent that are consistent. Unfortunately, ethnicity and country of origin are not reported in this series. The use of means and standard deviations to present data with such a reduced sample size can be treacherous, as the mean could be easily influenced by extreme values, that is, a significant change in BMIZ in only one or a few particular patients. In fact, the proportion mean/standard deviation of the variable BMIZ suggest that the values are widely distributed. Together with the fact that body composition changes during puberty include certain increase in BMIZ, we understand these data should be interpreted cautiously. Finally, authors underline the fact that a significant proportion of patients did not maintain viral suppression after switch, which they interpret as probably related to adherence issues. We understand adherence issues should also make us question the potential deleterious effect of treatment on weight gain.

We agree with the authors that larger and prospective studies are urgently needed to address the specific question of body composition changes in relation to dolutegravir- and TAF-containing regimens in children and adolescents.

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