Complex regional pain syndrome of the foot in a girl with post-SARS-CoV-2 chilblains

Sirs,

We have found of great interest the case of a 16-year-old-girl diagnosed with complex regional pain syndrome (CRPS) after SARS-CoV-2-chilblain-like lesions (CLL). CLL or 'Covid-toes' are one of the most interesting para-viral eruptions triggered by SARS-CoV-2 presenting as indolent or itchy erythematous-purpuric macules and papules and as blisters of toes and fingers. Several case series have reasonably ensured that only young adults and especially healthy adolescents show these skin findings probably due to an exuberant response of the innate immune system to the virus, with enhanced type I interferon (IFN-I) response (1). Several adolescents have been referred to our clinics with this clinical picture, in clear temporal relationship to SARS-CoV-2 epidemic. Our girl presented with a two-month history of bilateral discoloration of the toes associated with progressive pain. No history of trauma, three-step pattern of skin colour changes typical of Raynaud's phenomenon, joint pain or fever was reported, and there was no family history of autoimmune or rheumatologic diseases. No one in her family was affected with Coronavirus disease 2019 (COVID-19), nor did she refer contacts with people likely to be affected with it. Nasopharyngeal swab and serology were negative. The toes were bilaterally cold and bluish, mainly on the left side with multiple small ulcers on the big toe. Apart from this, the physical examination was unremarkable. Blood tests were normal (blood count, erythrocyte sedimentation rate, anti-nuclear antibodies). Interferon score was negative. Capillaroscopy did not reveal any abnormality in capillary architecture. A diagnosis of SARS-CoV-2-CLL was made. She was discharged on a course of prednisone 1 mg/kg/day with mild benefit on pain. After two weeks, the left foot became swollen and the patient was unable to walk without crutches. At the evaluation, her left foot was extremely cold and sweaty with a pale-bluish colour and swelling from the ankle to the toes (Fig. 1). Pedal pulses were palpable. Passive and active mobilisation were possible, but extremely painful. A diagnosis of CRPS was made due to complete clinical criteria satisfaction (2). A single 100 milligram infusion of neridronate was administered and well-tolerated. Moreover, according to the protocol proposed by Dietz and Compton, a treatment plan based on a patient-directed programme of mobilisation and foot massage was explained to the patient and well accepted (3). As early as the following day, the family reported a prompt improvement with a reduction in pain intensity and the patient started to walk without crutches. At the follow-up visit a week after, the foot had no skin or colour changes. A magnetic resonance imaging (MRI) showed diffuse soft tissue oedema and bone marrow oedema limited to the toes of the left foot (Fig. 2), while the right foot did not show any abnormality. There was no further need for other bisphosphonate infusion as the girl continued self-physiotherapy and resumed dancing a month later. This case of CLL showed a more severe clinical course strongly impaired by pain as a key feature. To our knowledge, this is the first case of CRPS following a severe case of CLL in a previously healthy adolescent girl. Our hypothesis is that underlying vasculopathy (as shown at the MRI bone marrow oedema), persistent immobilisation and lockdown-related stress may have triggered a local neuro-inflammatory response leading to the described clinical picture. In our experience, a treatment plan clearly explained to the young patient based on a desensitisation-relaxation technique (3), without parents or other figures involved, has shown prompt clinical benefit while waiting for bisphosphonate maximal effect (4).

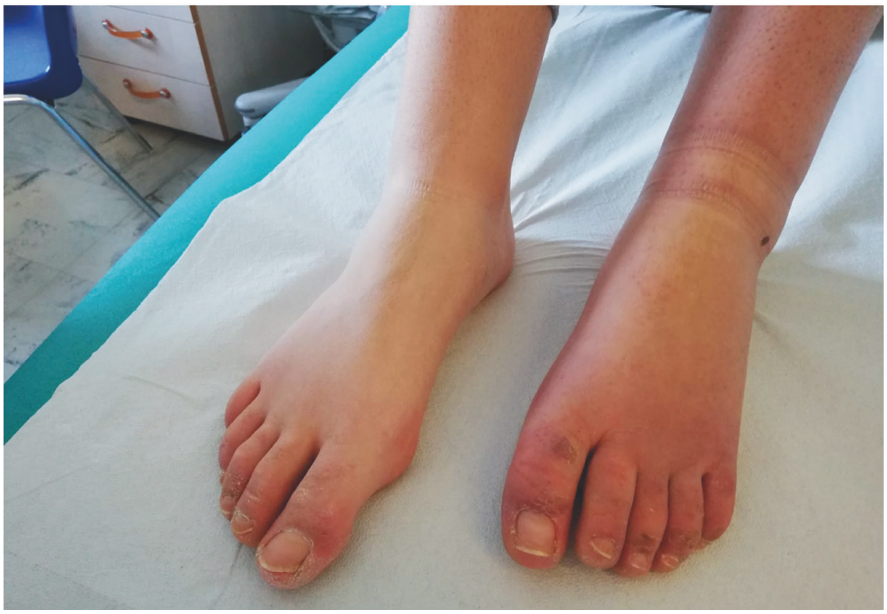

Fig. 1. CRPS and CLL of the left foot, before neridronate infusion.

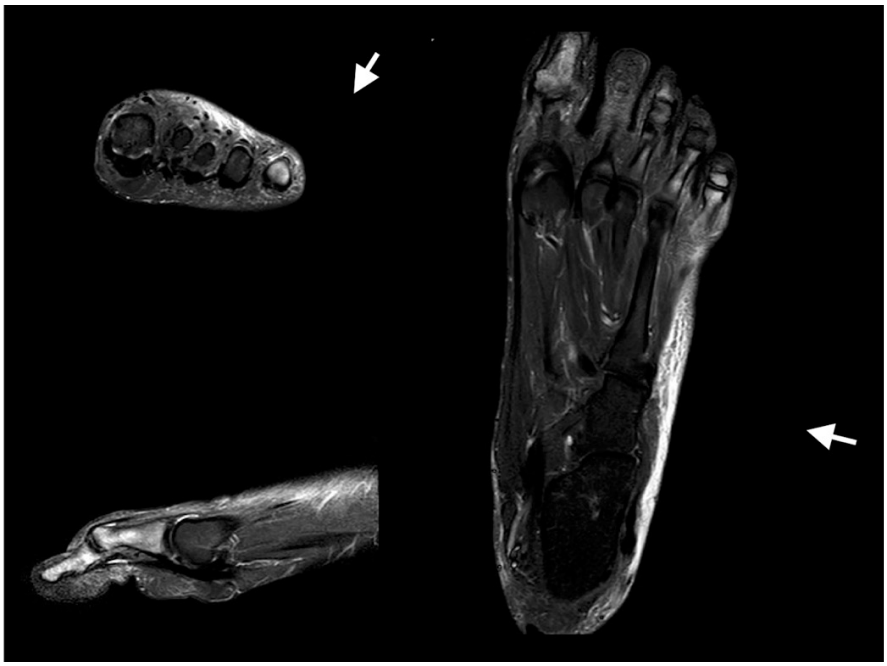

Fig. 2. MR STIR images show subcutaneous oedema of the dorsal and lateral aspect of the left foot and bone marrow oedema of the toes (arrows).

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