

Authors' contributions

Camino Prada-García: The study concept and design; data collection, analysis, and interpretation of data; writing of the manuscript or critical review of important intellectual content; effective participation in the research guidance; intellectual participation in the propaedeutic and/or therapeutic conduct of the studied cases; critical review of the literature; final approval of the final version of the manuscript.

Asunción González-Morán: The study concept and design, analysis and interpretation of data; writing of the manuscript or critical review of important intellectual content; intellectual participation in the propaedeutic and/or therapeutic conduct of the studied cases; critical review of the literature; final approval of the final version of the manuscript.

Xenia Pérez-González: The study concept and design; data collection, analysis, and interpretation of data; intellectual participation in the propaedeutic and/or therapeutic conduct of the studied cases; final approval of the final version of the manuscript.

Conflicts of interest

None declared.

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Löfgren's syndrome manifestation of acute sarcoidosis: short-term resolution with association of anti-inflammatory drugs*

Dear Editor,

A 43-year-old woman, with no previous comorbidities, came to the outpatient clinic after being referred by the Infectious Diseases Division. Twenty-one days before, she had developed a painful lesion on her left calf, dry cough and persistent fever (38 °C). After five days, lesions characterized as violet nodules appeared on the legs (Figure 1) and upper limbs (Figure 2). A thorough physical examination was performed, with palpation of the joints of the hands, wrists, elbows, knees, and ankles.



There was joint edema and erythema on the knees and left metacarpophalangeal joints. She denied previous use of medication.

A chest tomography was requested (Figure 3), which showed perihilar lymph node disease and peripheral centrilobular micronodules. Recent serology for syphilis, HIV, hepatitis B and C, and blood cultures were negative. There was an increase in erythrocyte sedimentation rate (66 mm/h) and C-reactive protein level (8.8 mg/dL). The tuberculin skin test (PPD) and antinuclear factor were negative. The 24-h urinary calcium and angiotensin-converting enzyme levels were unchanged. Subcutaneous adipose tissue histopathology showed changes compatible with erythema nodosum – septal lymphocytic panniculitis, without vasculitis, with granuloma formation and presence of multinucleated giant cells (Figure 4). The patient ocular fundus examination showed no alterations. No adeno/visceromegaly was identified on clinical examination.

The signs and symptoms allowed the diagnosis of Löfgren's syndrome (LS), considered an acute manifestation of sarcoidosis and occurring in 5% to 10% of cases of the disease.^{1,2} The patient was treated with prednisone

* Study conducted at the Department of Infectious Diseases, Dermatology, Imaging Diagnosis and Radiotherapy, Faculty of Medicine, Universidade Estadual Paulista, Botucatu, SP, Brazil.

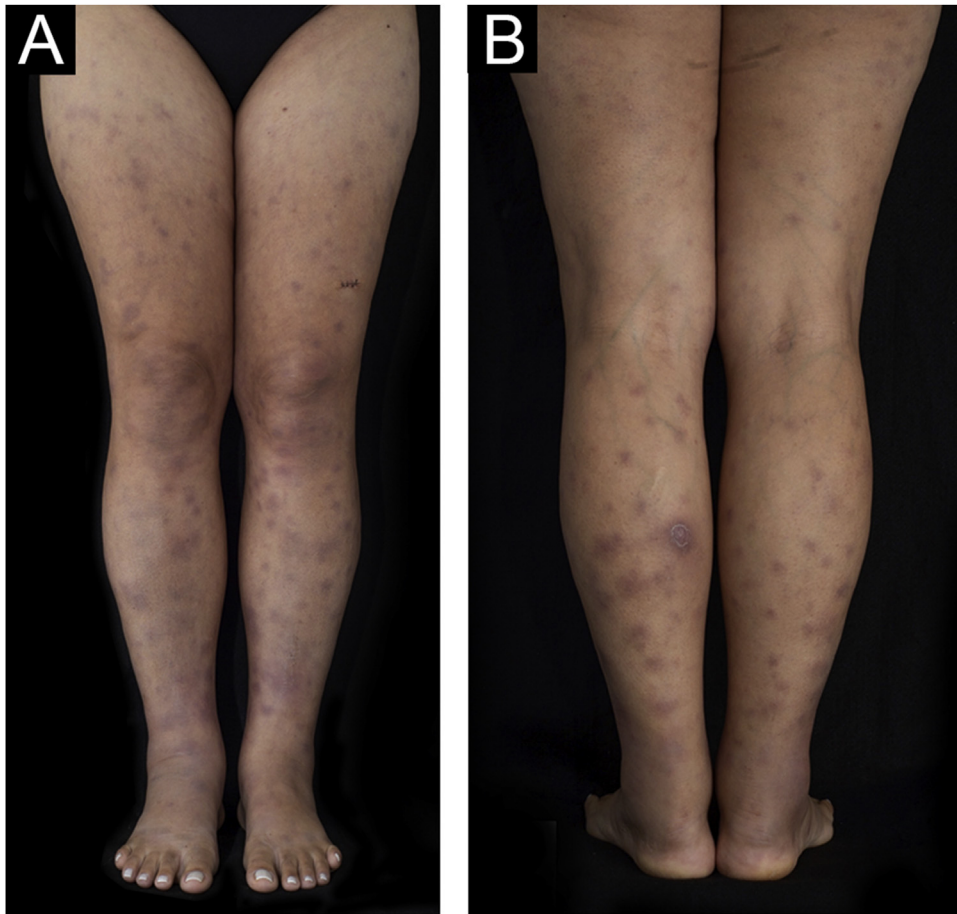


Figure 1 Multiple erythematous and edematous nodules on the lower limbs. (A) Anterior view. (B) Posterior view.

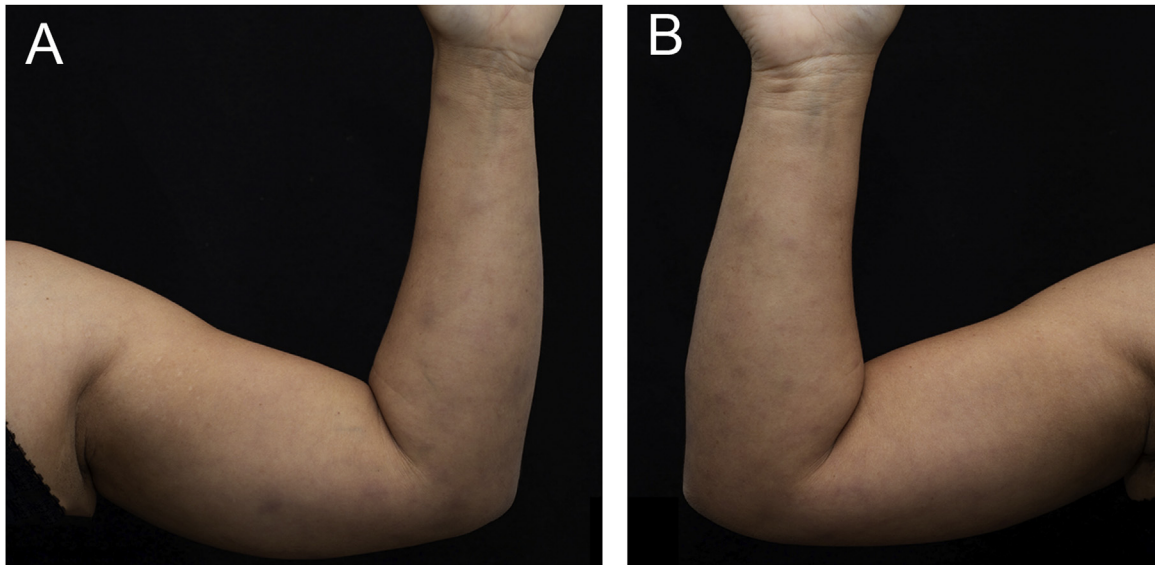


Figure 2 Multiple erythematous and edematous nodules on the upper limbs. (A) Medial aspect of the left upper limb. (B) Medial aspect of the right upper limb.

60 mg/day, hydroxychloroquine 400 mg/day, methotrexate 15 mg/week, and folic acid 5 mg/week. After five days, she no longer had a fever, and the pain and lesions had improved. After 30 days, she had a complete resolution of the com-

plaints and the erythema nodosum. The corticosteroid was completely discontinued in 90 days, the antimalarial in 120 days, and methotrexate in 12 months. The control CT carried out ten months later, showed minimal micronodu-

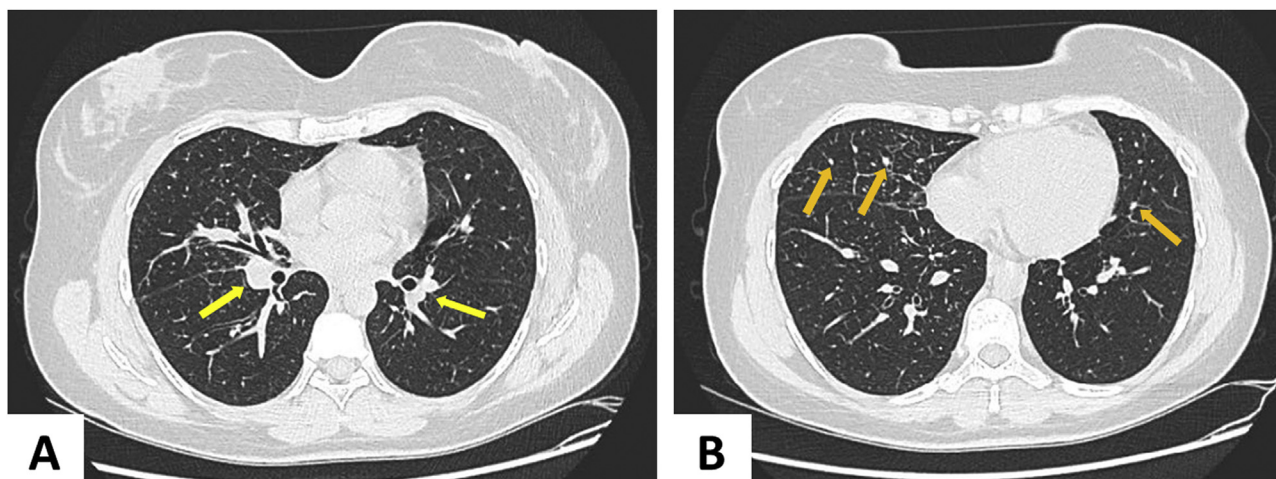


Figure 3 Computed tomography of the chest showing parabranchial nodules – yellow arrows (A) and multiple peripheral centrilobular micronodules – orange arrows (B).

lar findings, leading to discharge from the Pulmonology Division. At the 15-month follow-up, the patient remains asymptomatic.

LS mainly affects people between 25 and 30 to 40 years old, with a similar incidence between genders. There is, however, a second peak between 45 and 65 years of age, when 70% of cases are in women.^{1,2} The presence of the syndrome suggests a good prognosis, with 85% of patients showing spontaneous resolution of the condition within two years of symptom onset. The classic triad of symptoms includes erythema nodosum (60%), bilateral hilar lymphadenopathy (100%), and arthritis/arthralgias (10% to 30%). The diagnosis requires two of the three cardinal symptoms – when all are present the specificity of the clinical diagnosis is 95%.²⁻⁴

Most patients with LS do not have respiratory symptoms; nevertheless, parenchymal radiographic changes are observed in 90% of the cases. Stage I disease (hilar adenopathy without pulmonary infiltrates) resolves in 60% to 80% of the patients.^{3,4}

A biopsy of the affected organs should be performed to exclude infection, rheumatological disease, or malignancy. The best location depends on the accessibility, safety and feasibility of the procedure. Biopsies of superficial lesions – non-erythema nodosum skin lesions or palpable peripheral lymph nodes – are preferable. The pathognomonic finding of sarcoidosis is a well-formed, noncaseating epithelioid granuloma. Erythema nodosum-like lesions present as septal panniculitis that may have a granulomatous outline (Fig. 4).⁵

PPD is necessary as a screening tool for tuberculosis. The test is negative in patients with sarcoidosis; when positive, the diagnosis of tuberculosis should be considered.⁵⁻⁷

Treatment is symptomatic, in most cases with non-steroidal anti-inflammatory drugs and rest. Glucocorticoids are associated with clinical and pulmonary function improvement, without changing the course of the disease. The use of chloroquine has been reported in patients with cutaneous sarcoidosis, hypercalcemia, hypercalciuria and in neurosarcoidosis refractory to corticosteroids. Methotrex-

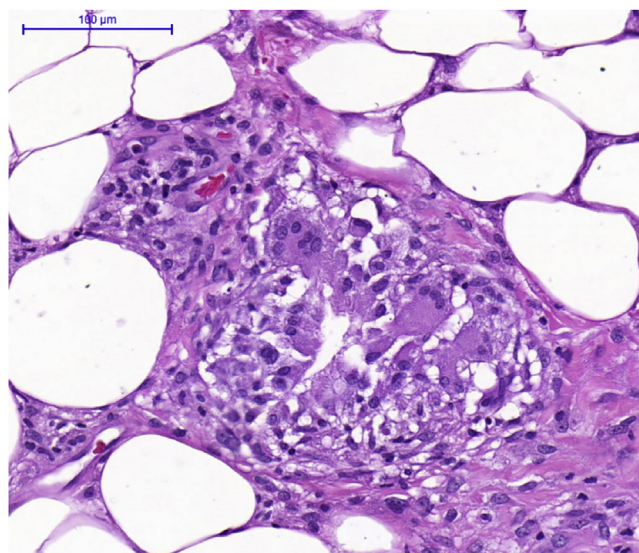


Figure 4 Subcutaneous adipose tissue histopathology showing septal lymphocytic panniculitis, without vasculitis, granuloma formation and presence of multinucleated giant cells.

ate administered at a dose between 7.5-15 mg/week is well tolerated and accepted in cases of lung, muscle and skin changes.^{2,5-7}

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Conflicts of interest

None declared.

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Lupus miliaris disseminatus faciei with extra-facial involvement in a 6-year-old Japanese girl*



Dear Editor,

Lupus miliaris disseminatus faciei (LMDF) predominantly occurs at 20–30 years of age and is rarely seen in children. We report a pediatric case of LMDF affecting the face and the labia majora.

A 6-year-old Japanese girl was referred to our department, complaining of a 5-month history of pruritic papular eruptions on the face. She had been treated with oral antiallergic drugs and topical corticosteroids, but without effects. Physical examination showed numerous 1–2 mm dome-shaped small reddish papulonodules around the mouth and lower eyelids (Fig. 1 A and B). In addition, reddish papules were scattered in the labia majora (Fig. 2). A skin biopsy was carried out from papular eruptions on the right jaw. Histological examination revealed dermal epithe-

lioid cell granulomas without caseous necrosis (Fig. 3A). Magnified epithelioid cell granuloma images showed multinucleated giant cells in the dermis (Fig. 3B), as well as inflammatory lymphocytic and histiocytic infiltration around the granulomas and hair follicles area. Immunostaining was positive for CD68 and CD163 antigens (Fig. 3 C and D). A tuberculin test was negative. Treatment with oral administration of clarithromycin showed favorable effects on the vulvar lesions after 5 months and the facial lesions after 9 months.

The present case developed multiple palpebral and perioral papulonodular lesions, which are the frequently involved sites of LMDF. In addition, vulvar involvement was observed. The patient did not have any organ symptoms suggestive of juvenile-onset sarcoidosis. Granulomatous rosacea was excluded, because neither facial erythema nor telangiectasia was observed, and the patient denied flushing.

Clinical and pathological features of pediatric LMDF are slightly different from adult LMDF, such as i) Papules concentrated around the mouth, on the nasolabial fold, and on the lower eyelids, ii) Small papule size, iii) Few pustules and scarring, iv) Redness around the mouth, v) Few caseous necrosis within epithelioid granulomas, and vi) Short clinical course.¹ On the other hand, Childhood Granulomatous Periorificial Dermatitis (CGPD) was reported as a disease in which yellow-brown papular eruptions lim-

* Study conducted at the Fukushima Medical University, Fukushima, Japan.