ABSTRACT
Systemic lupus erythematosus (SLE) is a chronic autoimmune systemic disease of the connectivitis group. This condition particularly affects young women between the ages of 20 and 40. Several systems are affected during the disease, including the nervous system, where central damage is more described than peripheral damage. We report the case of a 15-year-old male teenager with systemic lupus erythematosus whose initial clinical manifestation was acute inflammatory axonal polyneuropathy. This diagnosis was made based on allodynia predominantly in gloves and socks with clinical examination of bilateral peripheral neurogenic syndrome predominant in both lower limbs, all associated with fever. The electroneuromyogram showed signs of sensory and motor impairment (more marked in sensory) of the axonal type in both lower limbs, compatible with polyneuropathy. In addition, the patient developed several antibodies tested in the blood that returned positive. The skin biopsy described the proliferation of vascular capillaries with a fibrous and myxoid wall, dissociated by inflammatory cells, suggesting inflammatory involvement. Under treatment with hydroxychloroquine and corticosteroids, the patient presented a marked improvement in the general condition as well as on the functional level with regression of sensory and motor disorders.

KEYWORDS
polyneuropathy, lupus, teenager, case report

INTRODUCTION
Systemic lupus erythematosus (SLE) is a chronic autoimmune systemic disease of the connectivitis group, which has several manifestations which vary among individuals. This condition particularly affects young women between the ages of 20 and 40 [1]. Several systems are affected during the disease (cardiovascular, renal, haematological, respiratory, locomotor, integumentary, and immune), including the nervous system, where central damage is more described than peripheral damage [2, 3]. We report the case of a patient with systemic lupus erythematosus whose initial clinical manifestation was acute inflammatory axonal polyneuropathy.
muscle strength was normal proximally and rated at 4/5 for distal muscles symmetrically; ulno-pronator and radial reflexes were normal and symmetrical. Tactile sensitivity was reduced (coarse tact) on the extremities, and deep sensitivity (kinesthesia, pallesthesia) was retained. Trophic disorders such as dryness of the skin more marked on the elbows and ankles were found, but there was no muscular atrophy or oedema. The examination of the cardiovascular and respiratory systems was unremarkable.

**Diagnostic Assessment**

With these signs of peripheral neurogenic damage to the lower and upper limbs, we performed an electroneuromyogram (ENMG) which showed bilateral axonal-type damage to the lower limbs with a decrease in sensory potentials: amplitudes of 4.3µV (normal > 10µV) and 1.6µV (normal > 5µV) respectively on the sural and musculocutaneous right nerves. A decrease in motor potentials was also noted with an amplitude of 1.8 mV for external popliteal sciatic (normal > 3 mV), but the distal motor latencies were preserved. The internal popliteal sciatic was not stimulable. Ultimately, we could note a collapse of conduction velocities (almost zero on the sensitive and motor trunks of the lower limbs), symmetrical, predominantly on sensitivity and only on the lower limbs, distal motor latencies, amplitudes and conduction velocities were preserved in the upper limbs (See figures 1 and 2).

**Figure 1** Decrease in the sensory amplitude of the musculocutaneous nerve with the collapse of sensory conduction velocity. Sensory amplitude and speed retained on the median nerve.

The cerebrospinal fluid examination was normal (clear appearance, cytology with two cells per mm3, protein 0.40g / l, glucose 0.62g / l, chloride 121.60 mEq / l). The serologies for HIV, hepatitis B and C came back negative. C Reactive Protein was positive and very high at 122 mg / l. The complete blood count showed anaemia at 10.4 g/dl haemoglobin, a hematocrit level of 30.2, a mean corpuscular volume of 97fl, and a mean corpuscular haemoglobin concentration of 34 with 9,700 leukocytes and 319,000 platelets. We found no kidney damage (urea 0.39 g / l. Creatinine 9.6 mg / l).

One week after the onset of symptoms, the patient developed several skin lesions such as erythematous and scaly patches on the extremities and purpuric macules of the palms of hands (See figures 3 and 4).

**Figure 2** Decreased motor amplitude in the external popliteal sciatic nerve, with the collapse of motor conduction velocity.

**Figure 3** Erythematous and scaly plaques of the face.

**Figure 4** Purpuric macules of the palms of hands.
We observed a clear improvement in the general condition, in particular polyneuropathy, according to the American Academy of Neurology [8]. The diagnosis of lupus was polyneuropathy. This diagnosis was made based on allodynia predominantly in gloves and socks with clinical examination of bilateral peripheral neurogenic syndrome predominant in both lower limbs. The electroneuromyogram showed signs of sensory and motor impairment (more marked in sensory) of the axonal type in both lower limbs, compatible with polyneuropathy, according to the American Academy of Neurology [8].

The diagnosis of SLE was made based on clinical criteria: fever, skin lesions (maculopapular rash), a non-scarring alopecia, peripheral neuropathy in the absence of other causes, and biological: hemolytic anaemia, antinuclear antibody titre higher than the laboratory standard, Anti-native DNA antibodies higher than the laboratory standard, presence of an antibody to the Sm antigen. However, according to the American College of Rheumatology [9], the definite diagnosis of SLE has been retained.

**Therapeutic Intervention**

As management, the patient received prednisone at 1 mg/kg/day for 2 months with a current reduction to 0.5 mg/kg/day and hydroxychloroquine at 400 mg/day for 4 months (treatment in progress).

**Follow-up and outcomes**

We observed a clear improvement in the general condition, in sensitivity with a regression of neuropathic pain (DN4 score from 7/10 to 3/10), a regression of allodynia, evolution from tactile anaesthesia in socks to tactile hypoesthesia, kinesthesia of the toes has normalized, all this after 2 weeks of treatment. After 1 month of treatment, we observed an improvement on the motor level (the muscle strength in the distal region went from 4/5 to 5/5 in the upper limbs and 2/5 to 4/5 in the lower limbs), the Achilles reflexes initially abolished evolved to a rating of 2+. The patient is also currently doing motor physiotherapy sessions.

**Discussion**

Peripheral nervous system damage (polyneuropathy, mononeuropathy, myasthenia gravis, cranial nerve palsy, acute inflammatory demyelinating polyradiculoneuropathy) can be found in lupus. In the literature, they are found in 1.5% to 15% of cases [4, 5]. Polyneuropathies are the most frequent damage to the peripheral nervous system, according to several authors [6], and are more common in women [7]. These damages to the peripheral nervous system very often occur several years after the diagnosis of lupus has been made [4] and most often affects people over 30 years of age [4, 7]; they are frequently found when the patient also has damage to the central nervous system [6]. So what makes the particularity of this case, where our patient was a young boy of 15 years whose initial manifestation of lupus was polyneuropathy. This diagnosis was made based on allodynia predominantly in gloves and socks with clinical examination of bilateral peripheral neurogenic syndrome predominant in both lower limbs. The electroneuromyogram showed signs of sensory and motor impairment (more marked in sensory) of the axonal type in both lower limbs, compatible with polyneuropathy, according to the American Academy of Neurology [8].

Wang et al. in a study conducted on 4924 patients with systemic lupus erythematosus, found polyneuropathies, particularly in patients with an advanced form of lupus. In 0.1% of cases, polyneuropathy appeared before lupus [5]. Fever is one of the clinical signs most frequently associated with polyneuropathies in lupus. Biology often finds the positivity of anti-Sm antibodies, as described in our patient [4]. The analysis of cerebrospinal fluid returns abnormal most often in acute inflammatory demyelinating lesions (Guillain-Barré like syndrome), according to several authors [10]. Electroneuromyographic examination most often finds a sensory-motor axonal polyneuropathy, with a predominance of sensitivity and impaired conduction velocities most often linked to axonal damage [4] as found in our patient.

Several molecules are used in the management of lupus: hydroxychloroquine, corticosteroids (cortisone, prednisone, methylprednisone), high doses of aspirin, non-steroidal anti-inflammatory drugs, immunosuppressants and immunomodulators, particularly in refractory cases. In addition, several studies are still underway concerning biological therapies for lupus [10].

Our patient’s progress was very satisfactory under treatment with hydroxychloroquine combined with prednisone (which are first-line molecules and more accessible in our context); the literature describes good functional recovery under treatment [7, 10].

**Conclusion**

Systemic lupus erythematosus is an inflammatory disease most commonly affecting the female sex and having well-defined diagnostic criteria. Peripheral nervous system damage in systemic lupus erythematosus is not frequent, and little is described as an initial manifestation. This clinical case of a young teenage male with polyneuropathy as an initial manifestation shows the multiple forms of inflammatory diseases and should prompt us to perform an inflammatory assessment in front of any young subject with a clinical picture of polyneuropathy.

**Competing Interests**

Authors have no financial, political, personal, religious, ideological, academic, intellectual, commercial or any other conflicts of interest to declare in relation to this manuscript.

**Authors’ Contribution**

PCM and GN examined the patient in the hospital; PCM did all his follow-ups and wrote the first draft of this case report. PCM, GN, DGM, YFF and CTK revised subsequent versions and approved the final article.

**Patient perspective**

According to the patient and his family, the treatment (still in progress) is really effective in view of its improvement but has side effects such as weight gain.

**Informed consent**

The authors had the informed consent of the patient’s parents.

**References**


