

Case report

A Guillain-barre syndrome revealing cutaneous leishmaniasis in an immunocompetent adolescent



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Abstract

Guillain-Barre Syndrome (GBS) is an acute autoimmune polyradiculoneuropathy that often follows an antecedent infection. Leishmaniasis is a zoonosis due to a flagellate protozoan of the *Leishmania* genus and transmitted by the sandfly. Here we report a 15 years old teenager who presented with an ascending installation of the motor deficit in three weeks associated with dysphonia and swallowing disorders. The physical examination showed a flaccid proximo-distal quadriparesis, akinesthesia, apallesthesia and cutaneous lesions on the forehead, wrist and calf that appeared two months earlier. The HIV serology was negative and cerebrospinal fluid examination was normal. The electroneuromyography showed a reduction in motor and sensitive amplitudes with an increase in distal latencies and F waves in the upper limbs and segmental and focal conduction blocks. Parasitological examination of the dermal juice around the edges of the skin lesions revealed amastigote forms of leishmaniasis. The evolution was favorable with only symptomatic treatment.

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Introduction

Guillain-Barré Syndrome (GBS) is an acute monophasic autoimmune polyradiculoneuropathy that often follows an antecedent infection. Leishmaniasis is a zoonotic disease due to a flagellate protozoan of the leishmania genus and transmitted by the sting of sandfly. Neurological manifestations in cutaneous leishmaniasis were limited to focal neuropathies at the level of lesions reported in small case series in the 1990s [1,2]. We report a case of Guillain-Barré syndrome revealing cutaneous leishmaniasis in an apparently immunocompetent adolescent.

Patient and observation

A 15-year-old teenager was hospitalized in November 2017 with a motor deficit involving the four limbs. The signs began, 3 weeks before his admission, with the progressive and ascending installation of the motor deficit associated with paroxysmal electrical discharge-like lower limb pain and root irradiation, dysphonia and swallowing disorders. The patient reported a notion of a trip to the north of Senegal in an endemic zone of cutaneous leishmaniasis in August 2017 where he noticed the appearance of cutaneous lesions. He did not report diarrhoea episodes or flu-like symptoms occurring before neurological signs. On admission, neurological examination outlined flaccid proximo-distal quadriparesis rated at 2/5 for upper limb and at 1/5 for lower limb according to the Medical Research Council scale, tactile dysesthesia of the backs of the foot, plantar tactile anaesthesia, distal akinesthesia and apallesthesia. Dermatological examination revealed hypertrophic, ulcerous, painless and non-itching lesions on the forehead, wrist and calf. The rest of the clinical examination was unremarkable (Figure 1, Figure 2). Blood tests (Complete blood count, C-reactive protein, Fasting blood glucose, Aspartate aminotransferase, Alanine aminotransferase, Blood urea nitrogen and creatinine) were

normal. HIV serology was negative. The electroneuromyography (ENMG) showed a reduction in motor amplitudes on the nerve trunks in all four limbs, with an increase in distal latencies and F waves in the upper limbs, segmental and focal conduction blocks, and at the sensitive level, a reduction in amplitudes in all four limbs was noted, sparing the medians. Cytochemical and bacteriological examination of the cerebrospinal fluid was normal. Parasitological examination of the dermal juice around the edges of the skin lesions revealed amastigote forms of leishmaniasis (Figure 3).

Management was based on symptomatic treatment of pain with amitriptyline, motor rehabilitation sessions and clinical monitoring. A week later, the dysphonia and swallowing problems had disappeared. Muscle strength improved to 4/5 in the upper limbs and 3/5 in the lower limbs according to the same scale. Standing was still impossible, probably due to proprioceptive disorders. After a year of absence, the patient was seen again in consultation with a clinical examination carried out in December 2018 which was unremarkable apart from a decrease in osteotendinous reflexes and skin lesions that had completely healed. The control electroneuromyography, compared to that of November 2017, showed a lifting of conduction blocks, an overall improvement in motor and sensory parameters, but with ever decreasing conduction amplitudes and speeds Table 1.

Discussion

Leishmaniasis is a neglected disease, its cutaneous form is endemic in northeastern Senegal [3,4]. Beyond this region, cases originate from various regions of Senegal [5,6]. Studies reporting cases of cutaneous leishmaniasis with neurological manifestations are very rare in the literature. Kubba *et al.* (1987) reported for the first time in a series of 288 patients with cutaneous leishmaniasis, 14 cases of peripheral nerve damage

(5%) whose clinical manifestations were decreased tactile and thermo-algesic sensitivities in skin lesions. These nerve lesions were related to the skin lesions and nerve biopsy showed inflammatory infiltrates in the nerves or nerve sheaths with the presence of amastigotic forms of leishmania in some cases [1,2]. In our patient it is more like Guillain-Barré syndrome. The latter is an acute immune-mediated polyradiculoneuropathy often secondary to an infection. Certain germs are classically recognized such as *Campylobacter jejuni*, cytomegalovirus, *Epstein-Barr virus* and more recently the *Zika virus*, but the list of incriminated germs has not stopped growing [7,8].

One of the major mechanisms suggested by some studies, particularly in the case of *Campylobacter jejuni*, is a cross-immune reaction between bacterial antigens and gangliosides of the myelin sheath due to structural similarities between these molecules. The significant association between anti-GM1 and anti-GD1b antibodies and *Campylobacter jejuni* infection also supports this hypothesis [9]. We did not find any cases of Guillain-Barré syndrome in the literature during cutaneous leishmaniasis. However, some cases of Guillain-Barré syndrome occurring during visceral leishmaniasis were reported, revealing the visceral leishmaniasis. The case reported by Fasanaro *al.* had two episodes of acute polyradiculoneuropathy in less than 2 months, the first regressed in 3 weeks with plasma exchange and the second slowly improved with anti-leishmania chemotherapy. This evolutionary aspect suggests a dysimmune mechanism in which anti-leishmania antibodies also attack the constituents of the myelin sheath of peripheral nerves [10].

Conclusion

Our case raises the question of the accountability of cutaneous leishmaniasis (i.e. leishmaniasis species with cutaneous tropism) in the occurrence of Guillain-Barré syndrome. Clinical,

electrophysiological and serological surveillance in cohorts of patients followed for cutaneous leishmaniasis could possibly support this hypothesis.

Competing interests

The authors declares no competing interests.

Authors' contributions

All the authors have contributed to the conception, designing and writing of the manuscript. They all read and agreed to the final manuscript.

Table and figures

Table 1: comparison of motor potentials between 2017 and 2019

Figure 1: cutaneous leishmaniasis lesion on forehead

Figure 2: cutaneous leishmaniasis lesion on forearm

Figure 3: amastigotes of leishmanias seen under an optical microscope

References

1. Kubba R, El-Hassan AM, Al-Gindan Y, Omer AHS, Bushra M, Kutty MK. Peripheral nerve involvement in cutaneous leishmaniasis (Old World). *Int J Dermatol.* 1987;26(8):527-531. **PubMed | Google Scholar**

2. Satti MB, El-Hassan AM, Al-Gindan Y, Osman MA, Al-Sohaibani MO. Peripheral neural involvement in cutaneous leishmaniasis: a pathologic study of human and experimental animal lesions. *Int J Dermatol.* 1989;28(4):243-247. **PubMed | Google Scholar**
3. WHO. Global Health Observatory (GHO) data: Leishmaniasis. Accessed April 15, 2020.
4. Lariviere M, Abonnenc E, Kramer R. Studies on Skin Leishmaniasis in West Africa. The Problem of the Vector. *Bull Soc Pathol Exot Filiales.* 1961;54(5):1031-1046. **Google Scholar**
5. Ndongo S, Dieng MT, Dia D, Sy TN, Leye A, Diop MT *et al.* Cutaneous leishmaniasis in hospital area: epidemiological and clinical aspects, about 16 cases. *Dakar Med.* 2004;49(3):207-210. **PubMed | Google Scholar**
6. Diadie S, Diatta BA, Ndiaye M, Seck NB, Diallo S, Niang SO *et al.* La leishmaniose cutanée au Sénégal: une série de 38 cas à l'hôpital Aristide Le Dantec, de Dakar. *Médecine et Santé Tropicales.* 2018;28(1):106-108. **PubMed | Google Scholar**
7. Wijdicks EFM, Klein CJ. Guillain-Barré Syndrome. *Mayo Clin Proc.* 2017;92(3):467-479. **PubMed | Google Scholar**
8. Jacobs BC, Rothbarth PH, van der Meché FG, Herbrink P, Schmitz PI, de Klerk MA *et al.* The spectrum of antecedent infections in Guillain-Barré syndrome: a case-control study. *Neurology.* 1998;51(4):1110-1115. **PubMed | Google Scholar**
9. Loshaj-Shala A, Regazzoni L, Daci A, Orioli M, Brezovska K, Panovska AP *et al.* Guillain Barré syndrome (GBS): new insights in the molecular mimicry between *C jejuni* and human peripheral nerve (HPN) proteins. *J Neuroimmunol.* 2015;289:168-176. **PubMed | Google Scholar**
10. Fasanaro AM, Scoleri G, Pizza V, Gaeta GB, Fasanaro A. Guillain-Barré syndrome as presenting manifestation of visceral leishmaniasis. *Lancet.* 1991;338(8775):1142. **PubMed | Google Scholar**

Table 1: comparison of motor potentials between 2017 and 2019

Nerves / Sites	ENMG in 2017			ENMG in 2019		
	Lat. ms	Ampl. mV	Sp. m/s	Lat. ms	Ampl. mV	Sp m/s
R_Median						
1. WRIST	3,95	4,8		3,65	13,6	
2. ELBOW	12,60	2,2	37,0	8,75	12,8	52,9
L_Median						
1. WRIST	3,85	7,6		2,75	8,5	
2. ELBOW	11,35	1,8	49,3	7,75	8,6	56
R_Ulnar						
1. WRIST	3,05	6,0				
2. ELBOW	10,00	3,5	46,0			
L_Ulnar						
1. WRIST	3,25	5,0				
2. ELBOW	9,25	2,0	50,0			
R_Sciatic						
1. ANKLE	9,75	1,5		7,35	2,2	
2. PERONEAL HEAD	21,20	0,0		19,50	1,8	33,7
L_Sciatic						
1. ANKLE	10,15	1,3		5,15	1,6	
2. PERONEAL HEAD	20,50	0,0		18,80	1,9	30
R_Comm peronal						
1. MALLEOLUS	9,10	0,2		5,35	0,6	
2. POPLITEAL FOSSA	21,40	0,0	30,9	17,65	0,9	29,3
L_comm peronal						
1. MALLEOLUS	10,30	0,1		6,70	0,5	
2. POPLITEAL FOSSA	21,70	0,1	30,7	17,45	0,6	34,4

Ampl: amplitude; lat: distal latency, sp: speed mV: millivolt, ms: millisecond



Figure 1: cutaneous leishmaniasis lesion on forehead



Figure 2: cutaneous leishmaniasis lesion on forearm

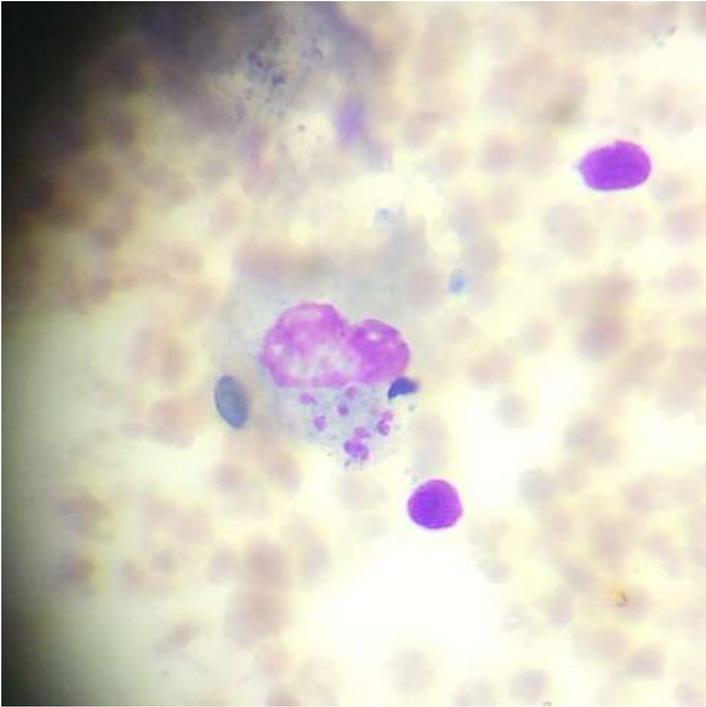


Figure 3: amastigotes of leishmaniasis seen under an optical microscope