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Bilateral Symmetrical Eruption in Dermatomal Distribution in Amyotrophic Lateral Sclerosis

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Abstract

A 72-year-old woman with amyotrophic lateral sclerosis was in our treatment for 12 years. She was mechanically ventilated and late in the course the management was limited to life support and palliation. During the last two years we observed the unusual occurrence of bilateral, symmetrical patchy erythema and edema confined to dermatomes C5 to T1. Eruption and edema were fading and relapsing in the same territories, essentially unchanged and limited to these areas. The differential diagnosis with disorders of dermatomal mimicry is presented. Advanced investigations to explore the diagnosis were not performed. We call attention to the three features defining the syndrome: dermatomal distribution, bilateral symmetrical involvement by eruption and edema suggestive of a vasomotor mechanism. The clinical findings were suggestive of a spinal cord disorder. We could not find reports of a similar occurrence in the PubMed database. Thus, it is clinically important to raise awareness and hopefully more publications of similar cases in the future might widen our understanding.

Keywords: Dermatomal; Eruption; Myelitis; Complex regional pain syndrome

Introduction

We describe the unusual occurrence of dermatomal, bilateral, symmetrical patchy erythema and pitting edema in a patient with late phase amyotrophic lateral sclerosis (ALS). We could not find reports of similar cases in the survey of the PubMed database.

Case History

A 75-year-old woman was on long term mechanical ventilation for the last 12 years. At age 60 she had developed weakness, wrist drops and gait instability, muscle cramps, dysarthria and dysphagia. On neurologic examination there was evidence of lower motor neuron and upper motor neuron disease involving bulbar, thoracic, and lumbosacral levels, findings consistent with amyotrophic lateral sclerosis. Pneumonia was the trigger to severe respiratory failure. The patient was mechanically ventilated through tracheostomy. Trials failed weaning her from mechanical ventilation. Swallowing dysfunction was the indication for enteral feeding by gastrostomy. Several years later, self-decannulation followed by resuscitation resulted in anoxic brain damage and the resultant unresponsive wakefulness state. Thereupon, treatment focused on basic life support and palliative care. The later course was largely uneventful until 2024 when a cutaneous eruption was noticed, confined to dermatomes C5 to T1, bilaterally. The eruption consisted of confluent erythematous patches, blanching under compression, and was associated with grade 2 pitting edema on forearms and hands (Figure 1). During the upcoming two years there was intermittent waning and recurrence of the eruption and pitting edema in cycles of 4 - 8 days (Figures 2,3). There were no changes in blood pressure, cardiac and respiratory parameters, nor any new findings on physical examination and laboratory tests. During a febrile episode, when the neutrophile leukocyte count increased from 3810/mm3 to 19300/mm3 and the C reactive protein increased from 1.2 mg/dL to 19.3 mg/dL the eruption did not change. A further diagnostic workup (CT/MRI/Lumbar puncture) was not authorized by the patient's

legal guardian, in accordance with his prior decision to restrict investigations and focus on palliative care only. Hence, clinical findings nurtured a diagnostic suspicion that could not be verified and specified by imaging, neurophysiologic and laboratory tests. Nevertheless, the distinctive picture and the message it conveys may be clinically meaningful.

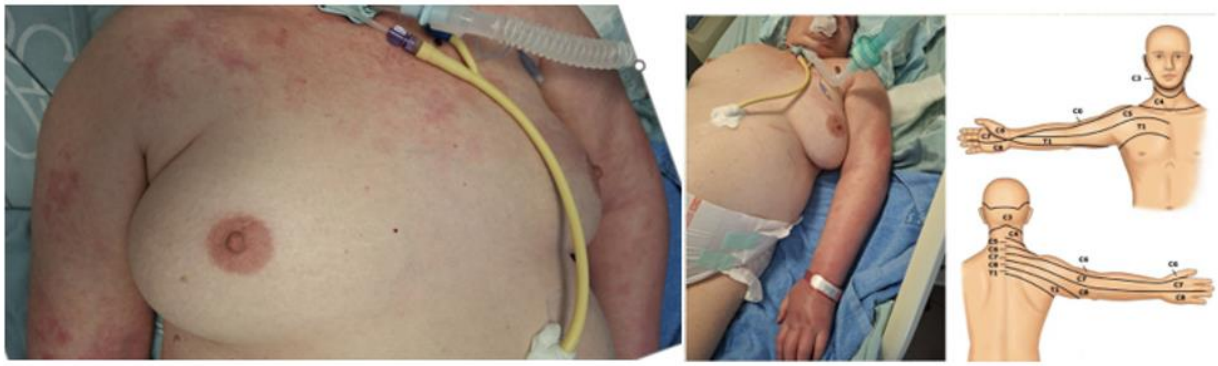

Figure 1: *Bilateral symmetrical erythematous eruption in the dermatomes C5-T1.*

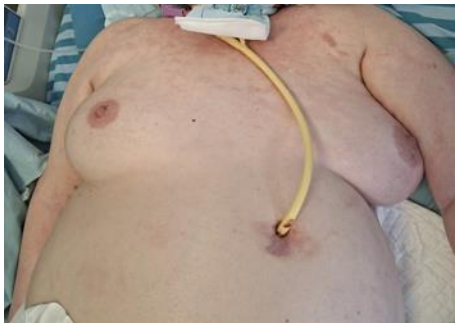

Figure 2: *Eruption in course of resolution.*

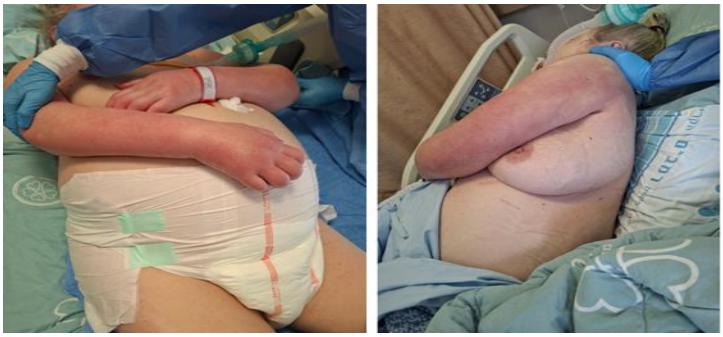

Figure 3: *Flair of the eruption and edema on forearms and dorsal aspect of the hands.*

Table 1: *Symptoms and signs of cervical-thoracic myelopathy versus mimickers, based on review PubMed data. CRPS: complex regional pain syndrome.*

Symptoms & signs	Myelopathy	Radiculopathy	CRPS
Dermatomal	+	+	-
Bilateral	+	+/-	-
Symmetrical	+	-	-
Pain	+	+	+
Dysesthesia	+	+	+
Eruption & edema	-	-	+/-
Autonomic	+/-	+/-	-

Discussion

Three features define the patient's evolving disorder: 1. dermatomal distribution, 2. bilateral symmetrical involvement, and 3. eruption and edema suggestive of a vasomotor mechanism. Clinical signs and symptoms confined to a dermatome indicate a disorder related to a specific nerve root (a radiculopathy) or to the corresponding spinal nerve ganglion. Radiculopathies typically manifest with pain radiating in a dermatomal distribution, that may be associated with numbness, weakness, paresthesia, sensory deficit, motor and trophic changes. Spinal nerves also contain autonomic fibers, but autonomic symptoms such as vasomotor changes are clinically not prominent under radiculopathy [1]. True dermatomal distribution of symptoms and signs should be differentiated from dermatomal mimicry. Such may occur in complex regional pain syndrome (CRPS), which develops as a complication after injury, surgery, myocardial infarction, or stroke. CRPS is characterized by continuous pain, motor and trophic changes and vasomotor symptoms, such as skin color changes, temperature asymmetry, edema and sweating asymmetry [2]. Though resembling a dermatomal distribution, complex regional pain syndrome typically involves a regional area rather than defined dermatomal distribution and usually unilateral [3]. Vasomotor symptoms in the present patient like those of CRPS differ from the classical functional vascular acrosyndromes: Raynaud phenomenon, acrocyanosis, erythromelalgia [4], and do not resemble idiopathic cyclic edema characterized by facial edema and swelling of the fingers in the morning and swelling of the legs at the end of the day [5]. Furthermore, there is no resemblance of the present case with vasomotor hot flushes and night sweats of menopause, carcinoid syndrome, and flushing associated with fever, emotions, medications, alcohol, food, hypersensitivity reactions, dumping syndrome, mast cell activation disorders [6,7].

Bilateral pain in dermatomal distribution, the presence of a sensory level of dysesthesia, bilateral extremity weakness, and autonomic dysfunction (bladder, bowel, cardiovascular or thermoregulatory dysfunction) suggest the presence of a lesion involving the spinal cord [8,9]. In PubMed database we could not find a mention of dermatomal eruption and edema, as in the present case. Herpes zoster has a genuine dermatomal distribution. In reviewing the PubMed database focused on atypical, bilateral symmetrical herpes zoster sine herpete [10,11] we could not find a case resembling this patient's coming and fading, long lasting dermatomal eruption and edema. Bilateral symmetrical dermatomal signs imply that the spinal cord, between left and right nerve roots, is at the origin of the syndrome, i.e. a myelopathy. A wide range of diseases may affect the spinal cord: nutritional deficiencies, infections, immune-mediated disorders, vascular and neurodegenerative diseases. The time course of myelopathies may be acute, subacute, or chronic. Neither patient history nor physical examination can distinguish between vascular, compressive causes or inflammatory causes. Therefore, patients with a suspected myelopathy are managed in emergency. The first is to exclude extrinsic cord compression that might warrant surgical intervention. MRI of the spine is the initial step in the diagnostic workup. When an inflammatory or infectious cause is suspected, a lumbar puncture is performed for cultures, immunoglobulin assessment and cytology. Initial blood tests also include C-reactive protein, thyroid stimulating hormone, syphilis serologies, HIV antibodies, and serum vitamin B12 levels [12].

In this patient with late-stage ALS, we reviewed the literature for a possible causality between ALS myelopathy and the patient's dermatomal eruption and edema. Vasomotor phenomena and autonomic dysfunction (sympathetic hyperactivity, sympatho-vagal imbalance, and central autonomic network impairment) are recognized recently in ALS. These autonomic dysfunctions in ALS, usually clinically occult, are detectable on electrophysiological autonomic function tests, e.g. as an abnormal electrical potential of the skin generated by activated sweat glands [13-15]. We could not find any report of findings resembling the present case. Altogether, the features observed in the present case differ from the well-known phenotypes, as summarized in (Table 1). In conclusion, the chronic dermatomal, bilateral, symmetrical patchy erythema and pitting edema in this patient suggests a pathology process involving the spinal cord in ALS patient. The patient being in a conscious unresponsive state, classical symptoms of myelopathy, namely dermatomal pain and sensitivity, could not be elicited. Standard investigations to advance the diagnosis were not considered respecting the guardian's request. Yet, the distinctive clinical picture featured above might increase interest and understanding of the extra-motor manifestations in ALS.

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