SERVIZIO SANITARIO REGIONALE
EMILIA-ROMAGNA
Azienda Unità Sanitaria Locale di Modena

UNIMORE
UNIVERSITÀ DEGLI STUDI DI MODENA E REGGIO EMILIA

IS THERE AN ALIEN IN MY BELLY? AN ATYPICAL CASE OF STIFF-PERSON SYNDROME

E. Menozzi[1], S.R.M. De Martino[1], F. Vitetta[1], N. Fini[1], P. Sola[1], F. Valzania[1], S. Meletti[1]

1: Department of Neuroscience, S. Agostino-Estense Hospital and University of Modena and Reggio Emilia, Modena, Italy.

Introduction: Stiff-person syndrome (SPS) is a rare autoimmune disorder characterized by progressive muscle rigidity and episodic muscle spasms that may be triggered by emotional upset, startle or sudden movements[1,2].

Case report: A 59 year-old Caucasian woman, with an anxiety-depressive disorder in SSRI treatment and a 2 years-history of insulin-dependent diabetes mellitus (IDDM), was admitted for a 2 month-history of low back pain and a recent onset of sudden and involuntary abdominal movements dyskinesia-like, hyperthermia, diaphoresis and psychomotor agitation. Neurological examination was normal. Brain MRI and electroencephalography were negative. A non-contrast computed tomographic (CT) scan of lumbar spine showed lumbarization of S1. No history of abdominal surgery or trauma was referred, but the abdomen CT scan revealed a bilateral iliopsoas bursitis. Firstly supposing iliopsoas bursitis as a local painful trigger for this Belly Dancer's Dyskinesia[3], a treatment with Clonazepam and Valproic Acid was successfully completed. One week after therapy suppression, the same symptomatology reappeared. An electromyography study showed a continuous involuntary motor-unit activity at Rectus Abdominis Superior muscle with a dystonic-myoclonic pattern. High titers (2000 UI/ml) of anti-GAD in the serum were found, and a diagnosis of SPS was made. Eight cycles of plasmapheresis were completed, and a long-term immunosuppressive treatment with Mycophenolate Mofetil was started with good tolerance. Iliopsoas bursitis completely recovered after the treatment.

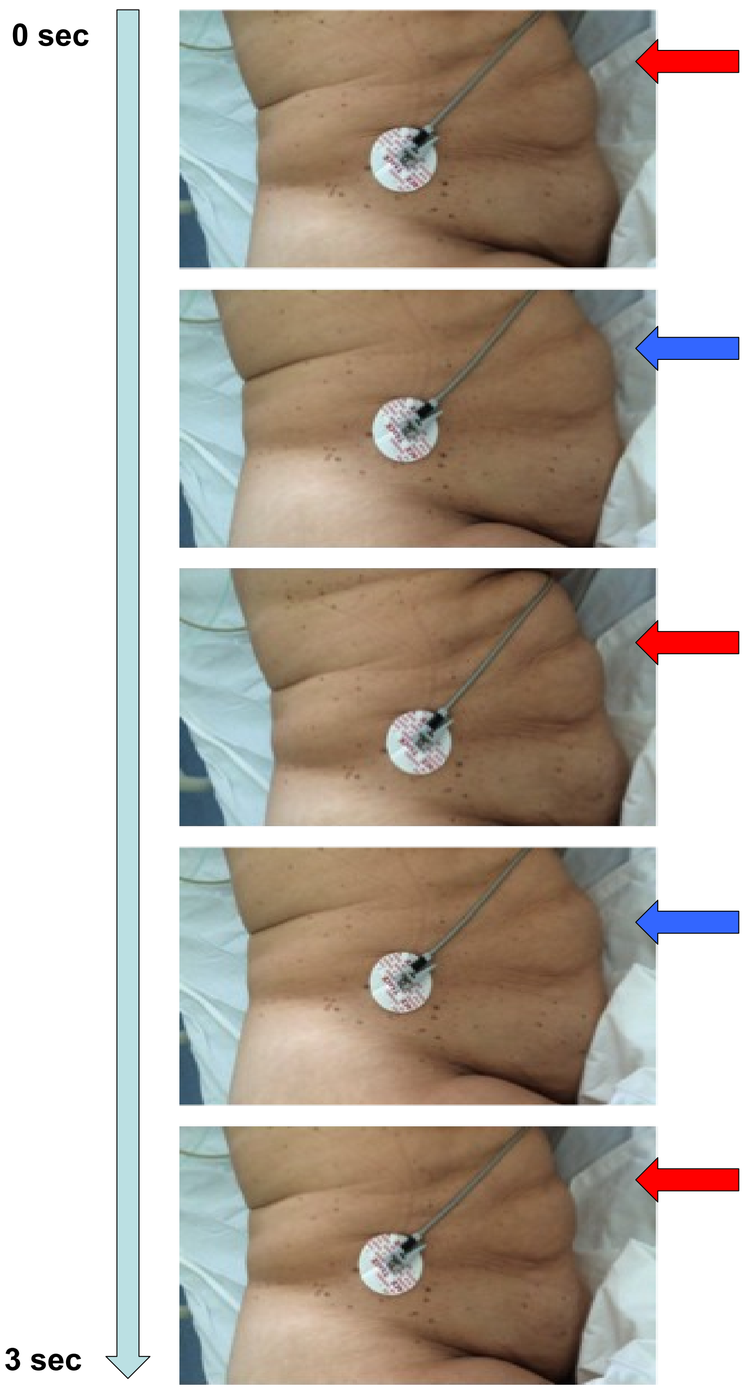

Fig. 1: Snapshots of a video recording, representing a sequence of three abdominal spasms. Each phase of contraction of Rectus Abdominis Superior muscle (red arrow) is followed by a phase of muscle release (blue arrow). The combination of muscle contraction-release lasts approximately 1 second.

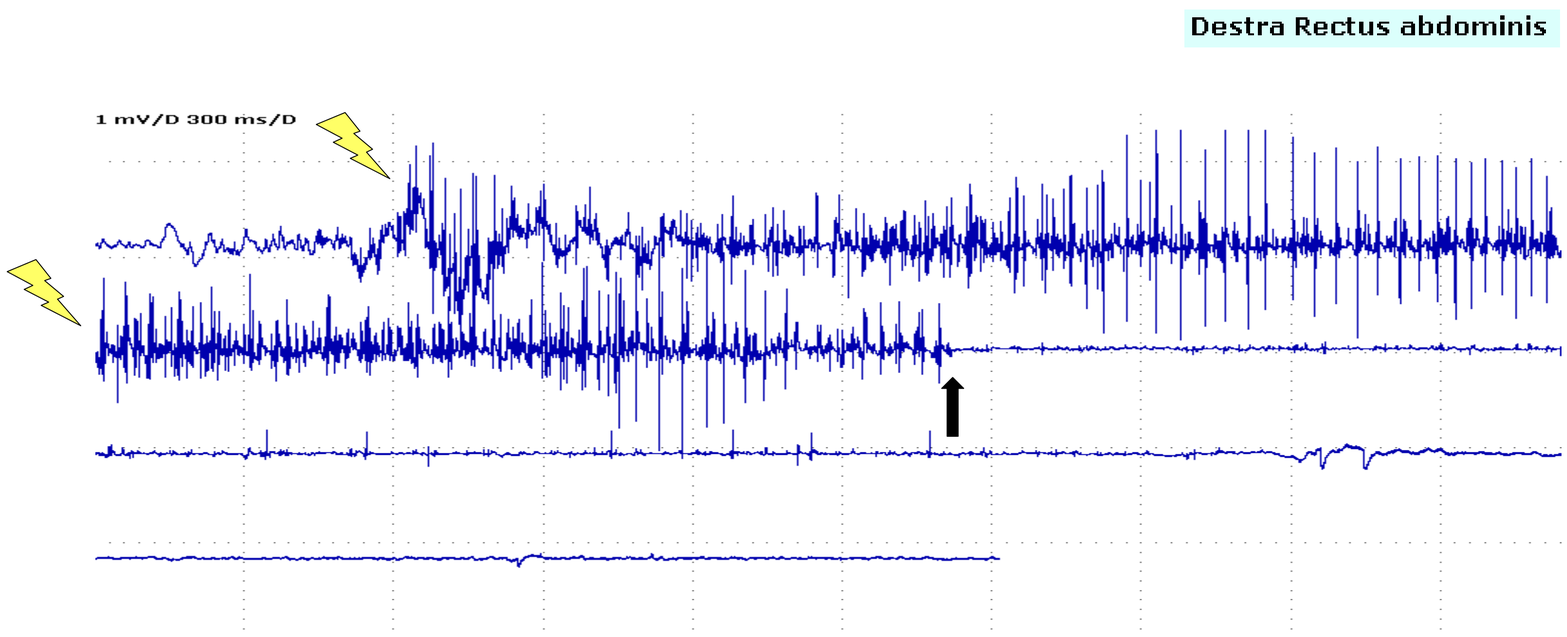

Fig. 2: Involuntary and sudden motor-unit activity at Rectus Abdominis Superior muscle recorded by EMG, correlated to abdominal spasms as those showed in figure 1. This recording shows the onset of the involuntary muscle contractions (yellow lightning), suddenly interrupted (black arrow).

Discussion: Abdominal painful dyskinesias without significant rigidity and legs impairment, represent an atypical presentation of SPS. A history of late-onset IDDM, the presence of lumbar column abnormalities and of subjective emotional involvement precipitant symptoms, might alert to the possibility of SPS diagnosis. In our case, iliopsoas bursitis, a rare and often undetermined condition, was finally considered as a consequence of abdominal spasms.

1. Rakocevic G, Floeter MK. Autoimmune stiff person syndrome and related myelopathies: Understanding of electrophysiological and immunological processes. *Muscle and Nerve*. 2012;45(5):623-634. doi:10.1002/mus.23234.
2. Baizabal-Carvallo JF, Jankovic J. Stiff-person syndrome: insights into a complex autoimmune disorder. *J Neurol Neurosurg Psychiatry*. 2015;86(8):840-848. doi:10.1136/jnnp-2014-309201.
3. Linazasoro G, Van Blercom N, Lasa A, Fernández JM, Aranzábal I. Etiological and Therapeutical Observations in a Case of Belly Dancer's Dyskinesia. *Mov Disord*. 2005;20(2):251-253. doi:10.1002/mds.20312.

Sin

XLVII CONGRESSO NAZIONALE
VENEZIA, 22 – 25 OTTOBRE

WebPoster