Journal of the Neurological Sciences 267 (2008) 182–186

Journal of the Neurological Sciences

www.elsevier.com/locate/jns

Short communication

Sjogren's syndrome-associated meningoencephalomyelitis: Cerebrospinal fluid cytokine levels and therapeutic utility of tacrolimus

Takayuki Hoshina, Yui Yamaguchi, Shouichi Ohga *, Ryutaro Kira, Masataka Ishimura, Hidetoshi Takada, Tamami Tanaka, Toshiro Hara

Department of Pediatrics, Graduate School of Medical Sciences, Kyushu University, 3-1-1 Maidashi, Higashi-ku, Fukuoka 812-8582, Japan

Received 20 July 2007; received in revised form 5 October 2007; accepted 8 October 2007

Abstract

Serial changes in the circulating and cerebrospinal fluid (CSF) cytokine levels were assessed in a patient with Sjogren's syndrome (SS)-associated meningoencephalomyelitis. A 16-yr-old girl diagnosed as having primary SS at 8 yr of age presented headache and vomiting. CSF studies revealed lymphocyte-dominant pleocytosis and high IgM index, but no evidence of infection. Disturbed consciousness and diffuse slow waves on electroencephalogram led to the diagnosis of SS-meningoencephalitis. The clinical condition subsided after a cycle of dexamethasone therapy, however, 2 months later urinary retention and paresthesia of the lower body developed. Craniospinal magnetic resonance imaging (MRI) showed extensive intraparenchymal lesions with high T2-weighted signal intensity adjacent to the posterior left horn of lateral ventricle of the brain and the longitudinal lesion from C5 to T10 of the spinal cord. High-dose methyl-prednisolone and subsequent tacrolimus therapy has effectively controlled the activity of SS-meningoencephalomyelitis. Monitoring of systemic and CSF cytokine levels during the course of illness revealed that CSF interleukin-6, but not interferon-γ or tumor necrosis factor-α levels were the sensitive indicator of disease activity. The unique cytokine profile, differing from those of infectious meningitis may be useful for predicting the central nervous system involvement in autoimmune disease.
© 2007 Published by Elsevier B.V.

Keywords: Sjogren's syndrome; Meningoencephalitis; Encephalomyelitis; Tacrolimus therapy; Cytokine profile; Interleukin-6

1. Introduction

Sjogren's syndrome (SS) is a chronic autoimmune disease of exocrine glands, involving in particular the salivary and lacrymal glands. This leads to diminished secretion of saliva and lacrima, resulting in xerostomia and keratoconjunctivitis sicca, respectively. SS can be observed as a clinically isolated syndrome (primary SS) or associated with other autoimmune diseases (secondary SS) including rheumatoid arthritis and systemic lupus erythematosus. Extraglandular manifestations occur in one-third of the affected patients with primary SS, represented by arthritis, Raynaud's phenomenon, lymphadenopathy, vasculitis, interstitial pneumonia, renal and neurological disorders, and malignant lymphoma [1]. Central nervous system (CNS) involvement as neurological manifestation of SS includes myelopathy, neuropathy, multiple sclerosis-like syndrome, aseptic meningitis or meningoencephalitis [2–4]. The incidence of CNS disease in patients with primary SS has reportedly varied ranging from 6% to 25% [2,5]. On the other hand, the acute myelitis has been estimated to occupy only 1% of primary SS patients [6]. Pleocytosis and/or the detection of oligoclonal bands of immunoglobulins (Ig) in the cerebrospinal fluid (CSF) might be helpful to assess the neurological complication in SS patients [7,8]. However, the early diagnosis is challenging in terms of the differentiation from CNS infection. There is little

Abbreviations: SS, Sjogren's syndrome; CSF, Cerebrospinal fluid; CNS, central nervous system; PSL, prednisolone.

* Corresponding author. Tel.: +81 92 642 5421; fax: +81 92 642 5435.
E-mail address: ohgas@pediatr.med.kyushu-u.ac.jp (S. Ohga).

0022-510X/$ - see front matter © 2007 Published by Elsevier B.V.
doi:10.1016/j.jns.2007.10.009

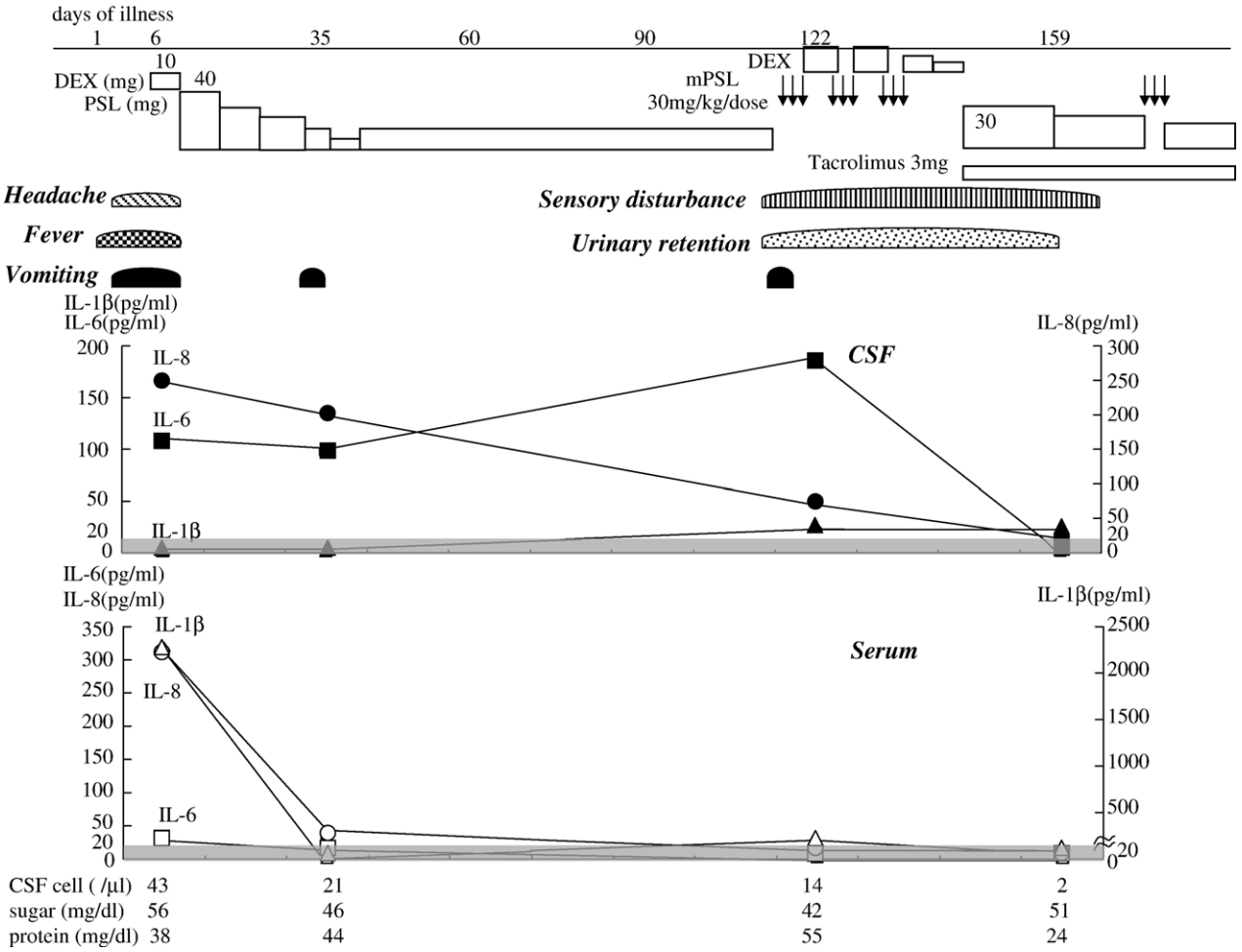

Fig. 1. Clinical course and cytokine profiles. Serum IL-8 (open circle) dropped 10-fold over the first month, as did IL-1β (open triangle), while IL-6 (open square) remained unchanged (IL-8: 314.8→31.5→<20→<20 pg/ml, IL-1β: 2286.5 pg/ml→<20→135.4→<20 pg/ml, IL-6: 29.9→<20→<20→<20 pg/ml). CSF IL-8 (closed circle) dropped 10-fold over months 1–5 while CSF IL-6 (closed square) dropped 5-fold over only the 5th month (IL-8: 251.1→202.2→67.4→21.4 pg/ml, IL-6: 105.1→92.9→186.1→<20 pg/ml). CSF IL-1β (closed triangle) levels were slightly elevated on the exacerbation. Circulating and CSF levels of TNF-α, IL-10 and IFN-γ were lower than 20 pg/ml during the course of CNS disease. The gray column represents the levels below 20 ng/ml.

information about the cytokine profile and effective treatment in SS-associated CNS diseases.

In this report, we analyzed sequential changes in the circulating and CSF cytokine levels in a patient during the progression of SS-meningoencephalomyelitis, and discussed the unique cytokine profile in intrathecal inflammation.

2. Material and methods

2.1. Cytokine assays

Serum and CSF samples were collected and stored at −30 °C for cytokine assays, after the informed consent was obtained. Interleukin (IL)-1β, IL-2, IL-4, IL-6, IL-8, IL-10, interferon (IFN)-γ, tumor necrosis factor (TNF)-α concentrations were measured by cytometoric bead array kit (BD Biosciences Pharmingen San Diego, CA) according to the manufacturer's protocol. The detectable range of each cytokine was 20–5000 pg/ml.

3. Case report

A 16-yr-old Japanese girl was admitted to our hospital at the 6th day of illness because of fever, headache and vomiting. This patient had been diagnosed as having primary SS at 8 yr of age, presenting recurrent parotitis, dryness of the eyes and mouth, the positivity of Schirmer's test, and autoantibodies (anti-Ro/SS-A, anti-La/SS-B, and antinuclear antibodies). Immune-associated thrombocytopenia occurred at 15 yr of age, and then resolved spontaneously. No prednisolone (PSL) therapy had been required for 8 yr because of only stable SS symptoms. There was no preceding virus infection to this admission. Her grandmother and aunt had a history of rheumatoid arthritis.

Download English Version:

https://daneshyari.com/en/article/1915850

Download Persian Version:

https://daneshyari.com/article/1915850

Daneshyari.com