Case report

Lumbar interlaminar epidural steroid injections for chronic low back- and lower extremity-pain in Sjogren's syndrome: A case report

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ABSTRACT

Introduction and importance: Peripheral nervous system involvement is very common in Sjogren's syndrome (SS); however, polyradiculopathy has been reported rarely in association with SS, and predominantly chronic forms have been described. Here, we reported a case from our Neurosurgery Department in Inam Medika KIM Hospital, Bangka Island, Pangkalpinang, Indonesia; as Academic Health System of Universitas Padjadjaran.

Case presentation: A 32-year-old woman, diagnosed with Sjogren's syndrome that was characterized by anti-nuclear, anti-Ro, anti-La and anti dsDNA-antibodies positives since 3 years ago; consulted to our department for a chronic low back with a radicular pain in both lower limbs from the gluteal area to both feet together with numbness, hyperesthesia and allodynia. The pain was evaluated by the visual analogue scale (VAS) score of 8; we then performed cervico-lumbal computed tomography (CT) scan that demonstrated multiple protruded discs of the cervical- and lumbar-spine.

Clinical discussion: Pain was treated with lumbar interlaminar epidural steroid injections as a safe technique that allows relieving patient symptoms; after 10 min, the patient experienced an improvement in her pain with reduced scores to 0-1 in VAS, as well as a significant improvement on her quality of life later on.

Conclusion: The use of lumbar interlaminar epidural steroid injections for an alternative therapeutic for neuropathic pain in SS gives a satisfactory result in terms of improvement of pain as well as a significant improvement on patients' quality of life.

1. Introduction

Sjogren's syndrome (SS) is a systemic autoimmune rheumatic disease, characterized by immune-mediated injury of exocrine glands, mainly affecting salivary and lacrimal glands, and a diverse array of extraglandular manifestations [1]. Sjogren's syndrome is marked by lymphocytic infiltrations of the exocrine glands and other organs in association with the production of various autoantibodies in the blood, typically develops insidiously over a period of months or even years [2]. In addition to dry eyes (e.g., a lack of tears), the most common symptoms include dry mouth, fatigue, musculoskeletal pain, and swelling of the major salivary glands [2]. A sensory ganglionopathy (neuropathy) can take place as a result of the infiltration of lymphocytes into dorsal root ganglia. Ten to 15% of the SS patients present with polyneuropathy [3], Sjögren syndrome (SS) is the only connective tissue disease that presents with a pure sensory neuropathy [4,5]. This polyneuropathy can involve motor and sensory tracts or remains a purely sensory involvement [4,5]. In primary SS, pure sensory neuropathy has a subacute onset. The bouts of pain are often accompanied by limitations in patient's daily activity and living patterns, such as sleep impairment and depression [6]. Anti-SSA (Ro) and anti-SSB (La) antibodies were found in 46% and 19% of the patients with the peripheral nervous system involvement, respectively [7].

Recently, in 2020, a study by Salman-Monte et al., evaluated factors associated with osteoporosis in patients with SS; the author suggested that patients with SS can develop osteoporosis and fragility fractures over the course of the disease [8]. The persistence of inflammation and vitamin D deficiency may contribute to the presence of decreased bone
mass, as observed in several systemic diseases such as rheumatoid arthritis [9], ankylosing spondylitis [10] and systemic lupus erythematosus [11]. Further, a significant association was found between the presence of anti-La antibody and osteoporosis [12]. Clinicians should be aware of these findings, which underline the importance of prevention and treatment of osteoporosis and fragility fractures in SS patients. Interlaminar epidural is the injection of local anesthetic and steroid to the epidural space, inside the spinal canal but outside spinal fluid containing dural space [13]. This pain management alternative led to noticeable improvements of chronic pain, for example, in patient with SS involving the low back pain and lower extremity pain. Herewith, we present a case report with a literature review.

2. Materials and methods

This case report has been reported in line with the SCARE 2020 criteria [14] and has been approved by our ethics committee No LB.02.01/X.6.5/193/2020. Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

2.1. Case presentation

A 32 years old female, married with 8-years-old twin daughters, working as a nurse while completing a master degree was consulted from Department of Internal Medicine to our Department of Neurosurgery, Intan Medika KIM Hospital, Bangka Island, Pangkalpinang, Indonesia; as Academic Health System of Universitas Padjadjaran, Bandung; with a history of 3 years chronic low back and lower extremity pain that had worsened during the time. The chronic low back pain was accompanied by pain that radiated to both lower limbs from the gluteal area to both feet with numbness, hyperesthesia and allodynia for several years, the pain however had significantly worsened over the previous 1–2 months. The pain used to associate with a flare-up of SS and accompanied by pain that radiated to both lower limbs from the gluteal area to both feet with numbness, hyperesthesia and allodynia for several years, the pain however had significantly worsened over the previous 1–2 months. The patient used to associate with a flare-up of SS and successfully managed with the prescription of muscle relaxant and physical therapy referral. Both interventions provided a slight relief of the pain up to 1 day prior to the visit where by, she reported an acute worsening of the back pain that had prevented her from completing her physical therapy session earlier that day. The use of heat therapy and muscle relaxants, although somewhat effective previously, did not provide any sort of pain relief. The pain caused limitations in her daily activity and living patterns, as well as her quality of sleep. On her history taking, the patient underlined that she had been depressed since she struggled with her chronic pain.

On physical examination we found dry mouth, dry eyes and musculoskeletal pain; with VAS score 8, that radiated to both lower limbs from the gluteal area to both feet with numbness and weakness in legs (foot and toe dorsiflexion) 4/5 according to the Medical Research Council (MRC) scale. Cervical and lumbosacral computed tomography (CT) scan studies showed multiple protruded disc of the C-spine and L-spine level (Fig. 1). Blood profiles were normal; except for slight increase of eosinophil, lymphocyte and monocyte along with low neutrophil. Laboratory test results of our case.

Table 1

| Laboratory test                                      | Value   | Reference |
|-----------------------------------------------------|---------|-----------|
| Erythrocyte sedimentation rate (mm/h)               | 13      | 0–20      |
| Leukocyte                                          | 9.2     | 4.0–11.0  |
| Basophil (%)                                        | 0.2     | 0–1       |
| Eosinophil (%)                                      | 5.0     | 1.0–1.8   |
| Neutrophil (%)                                      | 43.8    | 50–70     |
| Lymphocyte (%)                                      | 41      | 25–40     |
| Monocyte (%)                                        | 11.2    | 2–8       |
| Vit D 25-OH total (ng/mL)                           | 7.5     | 30–100    |
| IgE total (IU/mL)                                   | 267     | <87       |
| Anti-nuclear antibody (ANA)                         | Positive| Negative  |
| Anti-Ro (SSA) antibody                              | Positive| Negative  |
| Anti-La (SSB) antibody                              | Positive| Negative  |
| dsDNA antibody (IU/mL)                              | <10     | <7.0 (negative) |
| RNP/Sm                                              | Negative| Negative  |
| Sm                                                  | Negative| Negative  |
| ScI-70                                              | Negative| Negative  |
| PM-Sc                                               | Negative| Negative  |
| Jo-1                                                | Negative| Negative  |
| Centromere B                                        | Negative| Negative  |
| PCNA                                                | Negative| Negative  |
| Urinalysis                                          |         |           |
| Blood (µL)                                          | 10.0 (+1)| Negative  |
| Ketone (mg/dL)                                      | 5.0 (+1)| Negative  |
| Bilirubin (mg/dL)                                   | 3 (+2)  | Negative  |

Note: Vit D, vitamin D; IgE, immunoglobulin E; dsDNA, double-stranded DNA; RNP/Sm, ribonucleo-protein/Smith; Sm, Smith; ScI-70, scleroderma; PM-ScI, polymyositis-systemic scleroderma; Jo-1, for myopathies inflammatory idiopathic; PCNA, proliferating cell nuclear antigen. Bold: Abnormal value(s).
3. Discussion

Hyperimmunoglobulin E syndrome (HIES) is a primary immunodeficiency disorder that characterized by recurrent sino-pulmonary infections, cutaneous abscesses and chronic eczematous dermatitis. The syndrome is also associated with coarse facies, growth restriction, osteoporosis, eosinophilia and autoimmune disorders. Two distinct genetic variants of HIES have been described, namely autosomal recessive HIES (AR-HIES) and HIES with STAT3 mutation. The immunopathogenesis of STAT3 deficiency is still a matter of debate, and T-helper 1 (Th-1/Th2) cytokine imbalance has been suggested as a causative factor.

In one report of a case in an adult, where primary SS diagnosis was subsequently followed by HIES diagnosis, the authors proposed the Th2 cytokine as the predominant mechanism. In our case no symptoms and the onset of HIES was identified.

Definitive procedures for multiple levels of protruded disc, such as laminectomy and posterior stabilization were ruled out considering the patient’s autoimmune status and associated deterioration of bone density; SS patients tend to develop osteoporosis and fragility fractures over the course of the disease. We chose instead to go with interlaminar epidural steroid injections; satisfactory pain relief was achieved with the injection of local anesthetic and steroid under C-arm guidance along with post-procedures education which includes lifestyle modification such as water-exercise to strengthen her extremity muscle, the use of lumbar-support for prolonged activity and body weight reduction. No side effects were observed during 2 years follow up. To the best of our knowledge, until now, no report or data regarding the SS case with related chronic low back and lower extremity pain in Indonesia have ever been internationally published. Hence, this report would also add a reported case of SS with chronic low back and lower extremity pain worldwide. Regarding the limitations of this case report, it should be pointed out, we should improvise our methods, in the case that a definitive treatment couldn’t be done, alternative management plan tailored individually to patient’s condition ought to be attempted.

4. Conclusion

The use of lumbar interlaminar epidural steroid injections for an alternative therapeutic for neuropathic pain in SS gives a satisfactory result in terms of improvement of pain as well as a significant improvement on patients’ quality of life. This is the first SS with chronic low back and lower extremity pain case reported from Indonesia.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

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Guarantor

Ahmad Faried, MD., PhD.

Research registration number

1. Name of the registry: -
2. Unique identifying number or registration ID: -
3. Hyperlink to your specific registration (must be publicly accessible and will be checked): -.

CRediT authorship contribution statement

First author as study concept, design, data collector, operator, analysis, and editing; second author as study concept, data collector, analysis, and editing; other authors as study design, interpretation, analysis, and editing.

Declaration of competing interest

The authors report no conflicts of interest. The authors alone are responsible for the content and writing of this article.

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