International Journal of Surgery Case Reports 94 (2022) 107053

Contents lists available at ScienceDirect

International Journal of Surgery Case Reports

journal homepage: www.elsevier.com/locate/ijscr

Case report

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

Ahmad Faried [a,b,f,*], Sumartini Dewi [c], Herry Herman [d,f], Alif Noeriyanto Rahman [e,f]

[a] Department of Neurosurgery, Intan Medika KIM Hospital, Pangkalpinang, Bangka Island, Indonesia
[b] Department of Neurosurgery, Faculty of Medicine, Universitas Padjadjaran–Dr. Hasan Sadikin Hospital, Bandung, West Java, Indonesia
[c] Division of Rheumatology, Department of Internal Medicine, Faculty of Medicine, Universitas Padjadjaran–Dr. Hasan Sadikin Hospital, Bandung, West Java, Indonesia
[d] Department of Orthopaedic and Traumatology, Faculty of Medicine, Universitas Padjadjaran–Dr. Hasan Sadikin Hospital, Bandung, West Java, Indonesia
[e] Depok Orthopaedic and Pain Interventional Center, Sentra Medika Hospital, Depok, West Java, Indonesia
[f] Precursor - Comprehensive Course in Pain Intervention and Regeneration, Faculty of Medicine, Universitas Padjadjaran, Bandung, West Java, Indonesia

ARTICLE INFO

Keywords:
Sjogren's syndrome
Chronic pain
Lumbar interlaminar epidural steroid injections
Case report

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 Intan 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, hyperstesis 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 lymphocytic cells 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

* Corresponding author at: Department of Neurosurgery, Faculty of Medicine, Universitas Padjadjaran–Dr. Hasan Sadikin Hospital, Bandung, Indonesia.
E-mail addresses: ahmad.faried@unpad.ac.id (A. Faried), sumartini.dewi@unpad.ac.id (S. Dewi), herry.herman@unpad.ac.id (H. Herman).

https://doi.org/10.1016/j.ijscr.2022.107053
Received 8 January 2022; Received in revised form 5 April 2022; Accepted 5 April 2022
Available online 6 April 2022

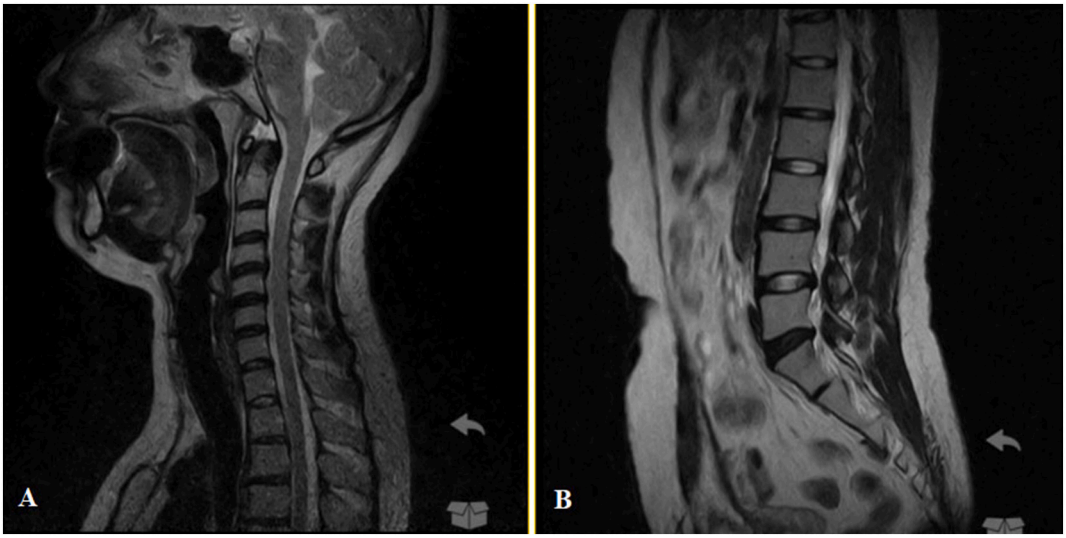

Fig. 1. Computed tomography (CT) scan images of the spinal cord. (A) Cervical, C-spine and (B) lumbal, L-spine, sagittal imaging demonstrated multiple protruded.

mass, as observed in several systemic diseases such 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, hyperestesis 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 successfully managed with the prescription of muscle relaxant and a 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.

Table 1
Laboratory test results of our case.

Laboratory test	Value	Reference
Erythrocyte sedimentation rate (mm/h)	13	0–20
Leukocyte		
Basophil (%)	0.2	0–1
Eosinophil (%)	**5.0**	2–4
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)	**1:1000**	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
Scl-70	Negative	Negative
PM-Scl	Negative	Negative
Jo-1	Negative	Negative
Centromere B	Negative	Negative
PCNA	Negative	Negative
Urinalysis		
Blood (/μL)	**10.0 (+1)**	Negative
Keton (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; Scl-70, scleroderma; PM-Scl, polymyositis-systemic scleroderma; Jo-1, for myopathies inflammatory idiopathic; PCNA, proliferating cell nuclear antigen.
Bold: Abnormal value(s).

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. Total 25-OH vitamin D level was significantly low and total IgE was significantly elevated. Anti-nuclear, anti-Ro, ant-La and dsDNA antibodies were positive; leakages of blood, ketone and bilirubin were found

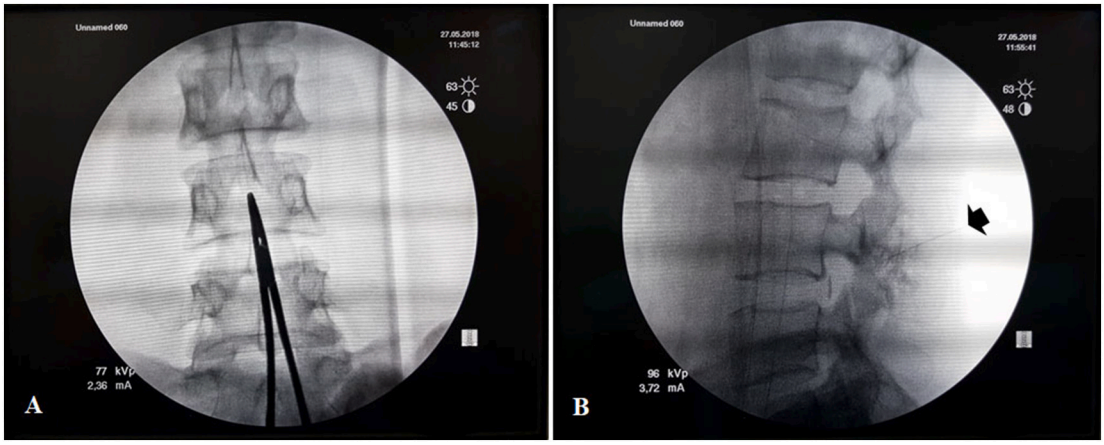

Fig. 2. Lumbar interlaminar epidural steroid injections. (A) Antero-posterior view of marking and (B) lateral view showing spinal needle insertion (arrow head).

in the urine (Table 1). Based on her recent and past-medical history and the auto-antibody test, the patient was diagnosed as SS in accordance with the American-European diagnostic criteria [15]. Both the anti-Ro (SSA) and anti-La (SSB) antibody were also positive in our patient. Hematological investigations disclosed an abnormal values of antinuclear antibodies [granularneoplasm/speckled (+) pattern with titer ≥1:1000 and cytoplasm cytoplasmic/granular (+) pattern with titer ≥1:100]. Ribonucleo-protein/Smith (RNP/Sm), Smith (Sm), scleroderma (Scl-70); polymyositis-systemic scleroderma (PM-Scl), myopathies inflammatory idiopathic (Jo-1), proliferating cell nuclear antigen (PCNA) were negative.

Total 25-OH vitamin D level was significantly low and total IgE was significantly elevated in this case. The relationship between low levels of vitamin D and SS is still controversial. The lack of exposure to UV rays as part of the treatment for the skin manifestations of the disease has been postulated as a risk factor for vitamin D deficiency [12]. Vitamin D deficiency is relatively frequent in patients with primary SS. Few studies have investigated the role of vitamin D in patients with SS. The link between vitamin D and neuropathy was reported in other conditions and data suggests that vitamin D may play a role in the SS pathogenesis. Low levels of vitamin D have been found in SS patients, which are associated with extra-glandular manifestations, such as lymphoma or neuropathy [12]. Vitamin D deficiency in this case associated with poly neuropathy severity. Plausible beneficial effect for vitamin-D supplementation may thus be suggested.

Lumbar interlaminar epidural steroid injections was planned for her treatment, considering the patient's autoimmune states and associated deterioration of bone density, we argued that the patient was not a good candidate for laminectomy and posterior stabilization, the gold standard for the management multiple levels of protruded disc as the consequences of instrumentation on an osteoporotic spine may outweigh its benefit. Interlaminar epidural was performed by injecting local anesthetic (2% lidocaine HCl) and steroid cocktail (80 mg triamcinolone plus 0.5% bupivacaine) under guidance of C-arm (Fig. 2), 5–10 min following the procedures, the pain was actually reduced to VAS 0–1.

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 [16]. 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 [17]. 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 [12]. 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 life-style 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 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.

Provenance and peer review

Not commissioned, externally peer-reviewed.

Ethical approval

Obtained.

Funding

A.F. supported by the Grants-in-Aid from Universitas Padjadjaran, Bandung, West Java, Indonesia.

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.

References

[1] Y. Peri, N. Agmon-Levin, E. Theodor, Y. Shoenfeld, Sjögren's syndrome, the old and the new, Best Pract. Res. Clin. Rheumatol. 26 (1) (2012) 105–117, https://doi.org/10.1016/j.berh.2012.01.012.

[2] F.B. Vivino, Sjogren's syndrome: clinical aspects, Clin. Immunol. 182 (2017) 48–54, https://doi.org/10.1016/j.clim.2017.04.005.

[3] M.U. Çevik, S. Varol, E. Akıl, A. Arıkanoğlu, I. Batmaz, U. Alabalık, Primary Sjögren's syndrome with sensory ganglionopathy and painful legs and moving toes syndrome, Turk. J. Neurol. 20 (2) (2014) 54–56.

[4] R.K. Olney, AAEM minimonograph 38: neuropathies in connective tissue disease, Muscle Nerve 15 (1992) 531–542.

[5] K.N. Bıçak, F.E. Bayam, Z. Çolakoğlu, Ganglionopati nedeni: sjögren sendromlu bir olgu, Ege Tıp Dergisi 2 (2007) 115–117.

[6] F. Tennant, L. Hermann, Intractable or chronic pain: there is a difference, West J. Med. 173 (2000) 306.

[7] S. Delalande, J. de Seze, A.L. Fauchais, E. Hachulla, T. Stojkovic, D. Ferriby, et al., Neurologic manifestations in primary sjogren syndrome: a study of 82 patients, Medicine (Baltimore) 83 (2004) 280–291.

[8] T.C. Salman-Monte, C. Sanchez-Piedra, M.F. Castro, et al., Prevalence and factors associated with osteoporosis and fragility fractures in patients with primary Sjögren syndrome, Rheumatol. Int. 40 (8) (2020) 1259–1265.

[9] C. Roux, Osteoporosis in inflammatory joint disease, Osteoporos. Int. 22 (2011) 421–433.

[10] U. Lange, J. Teichmann, J. Strunk, U. Müller-Ladner, K.L. Schmidt, Association of 1.25 vitamin D3 deficiency, disease activity and low bone mass in ankylosing spondylitis, Osteoporos. Int. 16 (2005) 1999–2004.

[11] J. Jacobs, L.A. Korswagen, A.M. Schilder, et al., Six-year follow-up study of bone mineral density in patients with systemic lupus erythematosus, Osteoporos. Int. 24 (2013) 1827–1833.

[12] M.G. Carrasco, E.A.J. Herrera, J.L.G. Romero, L. Vázquez de Lara, C.M. Pinto, I. E. Morales, et al., Vitamin D and Sjögren syndrome, Autoimmun. Rev. 16 (6) (2012) 587–593.

[13] B.S. Goodman, L.W. Posecion, S. Mallempati, M. Bayazitoglu, Complications and pitfalls of lumbar interlaminar and transforaminal epidural injections, Curr. Rev. Musculoskelet. Med. 1 (3–4) (2008) 212–222, https://doi.org/10.1007/s12178-008-9035-2.

[14] R.A. Agha, T. Franchi, C. Sohrabi, G. Mathew, for the SCARE Group, The SCARE 2020 guideline: updating consensus Surgical CAse REport (SCARE) guidelines, International Journal of Surgery 84 (2020) 226–230.

[15] C. Vitali, S. Bombardieri, R. Jonsson, H.M. Moutsopoulos, E.L. Alexander, S. E. Carsons, et al., European study group on classification criteria for Sjögren's syndrome. Classification criteria for Sjögren's syndrome: a revised version of the european criteria proposed by the american-european consensus group, Ann. Rheum. Dis. 61 (6) (2002 Jun) 554–558, https://doi.org/10.1136/ard.61.6.554.

[16] A.F. Freeman, S.M. Holland, Clinical manifestations of hyper IgE syndromes, Dis. Markers 29 (2010) 123–130.

[17] R. Mondal, S. Sarkar, V. Aggarwal, T. Sabui, Hyperimmunoglobulin E syndrome with Sjogren's syndrome in a child. Case report, SA J of childHealth 6 (1) (2012) 21–22.