

Case report

Contents lists available at ScienceDirect

International Journal of Surgery Case Reports



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

Complete bone fusion in failed back surgery syndrome using teriparatide in neurosyphilis: A case report

Dong-Ju Lim

Department of Orthopaedic Surgery, Seoul Spine Institute, Sanggye Paik Hospital, College of Medicine, Inje University, Republic of Korea

ARTICLE INFO	A B S T R A C T
<i>Keywords:</i> Teriparatide acetate Failed back surgery syndrome Neurosyphilis Cauda equina syndrome Spinal fusion	Introduction and importance: Syphilis is a sexually transmitted disease that appears in various organs. Neuro- syphilis in the spine is infrequent, and report of failed spinal arthrodesis surgery is rare. We report the first presentation of complete bone fusion in failed back surgery syndrome with teriparatide. <i>Case presentation:</i> A 65-year-old man presented to the outpatient clinic after being admitted to the spine department. The patient visited the neurology department 30 years prior for syphilitic myelitis and had been walking with a cane. He underwent an L5–S1 stenosis operation earlier. Severe proximal adjacent L4–L5 level stenosis was observed due to syphilitic gumma with cauda equina syndrome. A posterior decompression and posterolateral fusion to S1 were performed. Four weeks post-surgery, bilateral lower extremity muscle weakness recurred, and a radiographic examination revealed bilateral posterior screw loosening and fracture of the fourth lumbar vertebrae body. After the stagnant fluid and metal removal, for the augmentation of bone union, ter- iparatide was used for six months, and a complete bone union was confirmed by radiography without pain. <i>Clinical discussion:</i> Spinal syphilitic gumma has been rarely reported, most of which undergo surgical treatment. Surgical decompression and fixation with a pedicle screw are usually needed. There are complications after spinal surgery using a pedicle screw fixation, and parathyroid hormone (PTH) might be suitable for application in the prevention of nonunions or to augment bone fusion. <i>Conclusion:</i> This case report is the first description of complete spinal bone fusion in failed back surgery syndrome using teriparatide in tertiary syphilis.

1. Introduction

Syphilis is a sexually transmitted disease that presents multiple symptoms in different organs. The incidence of infectious syphilis has been increasing globally. The World Health Organization (WHO) reported that 5.6 million new cases of syphilis occurred globally in 2012 [1]. From 2017 to 2021, it showed an increase in the prevalence of 68.4 % in the US [2]. In addition, neurosyphilis can cause various neurological symptoms. Spinal neurosyphilis is less common than that with central involvement, which can be caused by myelitis, tabes dorsalis, and gummas. Studies report that failed back surgery syndrome (FBSS) affects approximately 10–40 % of patients who undergo spinal surgery [3], but there is no report about surgery failure after neurosyphilis gummas. Teriparatide hormone improves fracture union and nonunion fracture [4,5]. We report the first case of complete bone fusion in FBSS using teriparatide in tertiary phase syphilis. This article has been written according to the SCARE criteria described by Agha et al. for the SCARE group [6]. This case study was approved by the Institutional Review Board of Sanggye Paik Hospital.

2. Presentation of case

A 65-year-old man with syphilitic myelitis, known to our neurology department 30 years ago and to be using a cane, presented to our department in a wheelchair. The patient was reluctant to describe his illness in detail. A laboratory test performed at the hospital showed positive rapid plasma regain and *Treponema pallidum* hemagglutination assay findings for syphilis. A fluorescent treponemal antibody absorption test (FTA-ABS) also showed a positive result. However, the patient did not have HIV or immunosuppressed disease. Before admission, the remote onset of symptoms five years prior was due to bilateral lower extremity pain accompanied by worsening lower back pain. Severe

E-mail address: scd25@paik.ac.kr.

https://doi.org/10.1016/j.ijscr.2022.107816

Received 17 September 2022; Received in revised form 17 November 2022; Accepted 23 November 2022 Available online 29 November 2022

^{*} Department of Orthopaedic Surgery, Seoul Spine Institute, Sanggye paik Hospital, College of Medicine, Inje University, Dongil-ro 1342, Nowon-gu, Seoul 139-707, Republic of Korea.

^{2210-2612/© 2022} The Author. Published by Elsevier Ltd on behalf of LJS Publishing Group Ltd. This is an open access article under the CC BY-NC-ND license (http://creativecommons.org/licenses/by-nc-nd/4.0/).

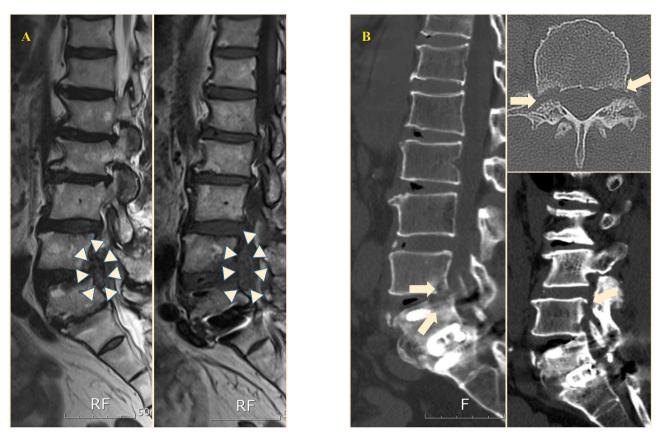


Fig. 1. A: Spinal syphilitic gumma located at L4–5 level. (arrowhead) B: An osteolytic lesion was detected in the L4 and L5 endplates. Both pedicles of L4 were absorbed in osteolytic lesions, as seen by 3D CT. (arrow).

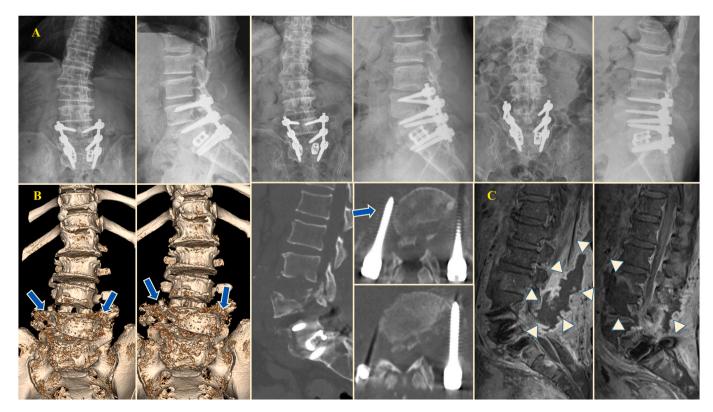


Fig. 2. A: Immediately after surgery, the pedicle screw was adequately fixed. The screw position changed over time, as seen in the radiograph. B: The proximal screws are dislocated, and a vertebral body fracture was found in the 3D CT scan. (arrow) C: Fluid stagnation is shown in the surgery site six weeks after the operation by MRI. (arrowhead).

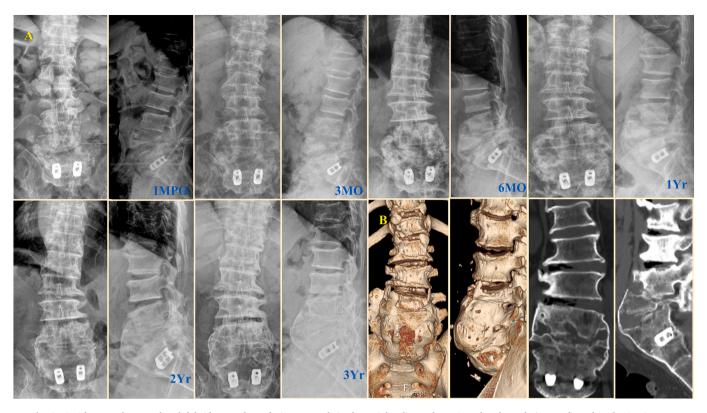


Fig. 3. A: After metal removal and debridement, bone fusion proceeds in the serial radiograph. B: Complete bone fusion confirmed in the 3D CT scan.

central spinal stenosis was detected, and posterior decompression and posterolateral fusion of L5–S1 were performed.

One year and five months later, at a second admission, the patient suffered a loss of sensation below the L4 dermatome level. Sphincter tone was decreased, and hypoesthesia of the scrotum and the perianal area was observed. Back pain, bilateral lower extremity pain, and motor weakness worsened, and the patient revisited the hospital. Magnetic resonance imaging (MRI) observed a proximal adjacent segmental disease at the L4–5 level with spinal syphilitic gummas. An osteolytic lesion was detected in the L4 and L5 endplates. Both pedicles of L4 were absorbed in osteolytic lesions, as seen by 3D CT (Fig. 1). A posterior decompression at L4–5 and a posterolateral fusion to S1 were performed. After the operation, the motor weakness of the lower extremities improved; however, after the removal of the catheter, urinary disturbance persisted, and the patient was transferred to the urology department with a diagnosis of a neurogenic bladder.

During urology treatment, at four weeks post-surgery, bilateral lower extremity motor weakness was observed again (Gr 2). A general radiographic examination revealed bilateral posterior screw loosening and a fracture of the fourth lumbar vertebra body. An MRI showed bilateral spinal stenosis of L3–4, and fluid retention was suspected of infection around the implant (Fig. 2). The fluid and metal were removed from the posterior spinal approach, and an additional L3–4 lumbar laminectomy was performed. There was no growth of bacteria in the culture.

After the operation, lower extremity motor strength showed improvement (Gr 3), and a body cast was put in place to secure the stability of the spine. On a bone density test, the T score revealed osteopenia. For the augmentation of bone union, teriparatide was used for six months, and the bone union was confirmed by radiography and CT imaging at a three-year postoperative follow-up (Fig. 3).

3. Discussion

Syphilis may present different symptoms in several stages (primary, secondary, latent, tertiary, or congenital). The manifestations of

neurosyphilis are rarely seen today, particularly due to the availability of antibiotics like penicillin [7]. A gumma lesion in the spine can cause symptoms of stenosis and occur in both the cervical and lumbar areas [8–10]. Hannuksela et al. and Nielsen et al. reported bone involvement was expected in approximately 1-5 % of untreated syphilis patients [11,12]. Consequently, long-term untreated syphilis can cause damage to the brain, heart, liver, nerves, eves, joints, and bones [13]. The gummatous spine disease may be sporadic, and it is difficult to differentiate between spinal and tumors [8,10,14]. The lesion can occur in the intradural and extradural areas [14], and in our case, there were no superficial skin, mucous, or chancre lesions during the admission period. However, given the history and the results of the syphilis serologic laboratory tests and the MRI scan, a spinal syphilitic gumma was strongly suspected. Due to the rarity of this disease, the diagnosis and treatment of spinal syphilitic gumma and the relevant prognosis have not been outlined. In previous studies, limited cases of spinal syphilitic gumma have been reported, most of which underwent surgical treatment. Surgical decompression and fixation with a pedicle screw were usually required [14,15].

There may be complications after spinal surgery using pedicle screw fixation. Halos around the screws or osteolysis, which may usually cause loosening of the pedicle screw, fracture, and screw pullout [16]. Some postoperative fractures are also caused by osteoporosis, inflammation, and trauma [17]. We hypothesized that neurosyphilis gumma invades the vertebral body and pedicle area, and abnormal tissue lessens the purchasing power of the pedicle screw fixation. So, after the operation, during rehabilitation, screw loosening occurs, and wound fluid stagnates and flows from the wound. Nonunions occur at a rate of 2.6–16 % in fractures of the long bones and 26 % in posterolateral lumbar fusions in degenerative lumbar surgery [18].

Surgery aims to provide symptom relief through decompression and complete fusion. Several studies report satisfactory radiographic outcomes. However, there is no report about postoperative complications like loosening and fixation site fracture in neurosyphilis patients. Parathyroid hormone (PTH) plays a central role in the coupling of osteogenesis and osteolysis and is an essential regulator in the process of remodeling. Therefore, PTH might be suitable for application in the prevention of nonunions. A possible positive effect of PTH on the healing of nonunions has already been observed in a number of case reports [4,5,19]. After spinal decompression and fusion surgery in neurosyphilis patients, there have been reports about unsuccessful cases of surgery [7,13] but no success after FBSS in neurosyphilis patients, and this is the first report.

4. Conclusion

As the number of syphilitic patients increases, we can predict the increasing number of neurosyphilis in the spinal column that needs surgery. Unlike spinal stenosis or disc disease, cautious treatment for spinal neurosyphilis gumma is required. In failed back surgery syndrome, especially bone fusion failure, treatment using PTH can be the strategic option to overcome bone fusion failure.

Ethics approval

This case study was approved by the Institutional Review Board of Sanggye Paik Hospital (SGPAIK 2020-10-001). Written informed consent was obtained from the patient to publish this case report and the accompanying images. The approval date is October 7, 2020.

Sources of funding

This research did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors.

Author contribution

Dong-Ju, Lim – study concept, design, data collection, data analysis, writing the paper, and data interpretation.

Provenance and peer review

Not commissioned, externally peer-reviewed.

Guarantor

Dong-Ju, Lim.

Research registration

Registered.

Declaration of competing interest

None.

Acknowledgements

We would like to thank Editage (www.editage.co.kr) for English

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.

References

language editing.

Consent

- [1] L. Newman, J. Rowley, S. Vander Hoorn, N.S. Wijesooriya, M. Unemo, N. Low, G. Stevens, S. Gottlieb, J. Kiarie, M. Temmerman, Global estimates of the prevalence and incidence of four curable sexually transmitted infections in 2012 based on systematic review and global reporting, PLoS One 10 (2015), e0143304.
- [2] Centers for Disease Control and Prevention, Preliminary 2021 Sexually Transmitted Disease Surveillance Data, Dept of Health and Human Services, 2022.
- [3] S. Inoue, M. Kamiya, M. Nishihara, Y.P. Arai, T. Ikemoto, T. Ushida, Prevalence, characteristics, and burden of failed back surgery syndrome: the influence of various residual symptoms on patient satisfaction and quality of life as assessed by a nationwide internet survey in Japan, J. Pain Res. 10 (2017) 811–823.
- [4] Y.U. Yolcu, J. Zreik, M.A. Alvi, N.R. Wanderman, B.C. Carlson, A. Nassr, et al., Use of teriparatide prior to lumbar fusion surgery lower two-year complications for patients with poor bone health, Clin. Neurol. Neurosurg, 198 (2020), 106244.
- [5] I. Kastirr, M. Reichardt, R. Andresen, S. Radmer, G. Schröder, T. Westphal, et al., Therapy of aseptic nonunions with parathyroid hormone, Eur. J. Orthop. Surg. Traumatol. 29 (2019) 169–173.
- [6] R.A. Agha, T. Franchi, C. Sohrabi, G. Mathew, A. Kerwan, SCARE Group, The SCARE 2020 guideline: updating consensus surgical CAse REport (SCARE) guidelines, Int. J. Surg. 84 (2020) 226–230.
- [7] Centers for Disease Control and Prevention, Sexually Transmitted Disease Surveillance, 2020, Department of Health and Human Services, Atlanta, GA, 2022.
- [8] R. Yin, L. Wang, T. Zhang, B. Zhao, Syphilis of the lumbar spine: a case report and review of the literature, Medicine 96 (2017), e9098.
- [9] A. Mejdoubi, M. Khoulali, N. Raouzi, S. Nasri, Y. Mebrouk, N. Oulali, et al., Neurosyphilis revealed by compressive cervical spine syphilitic gumma: a case report, Spinal Cord Ser. Cases 6 (2020) 56.
- [10] C. Yang, G. Li, J. Fang, H. Liu, B. Yang, Y. Xu, Spinal intramedullary syphilitic gumma: an unusual presentation of neurosyphilis, World Neurosurg. 95 (622) (2016) e17–e23.
- [11] M. Hannuksela, E.O. Karaharju, Syphilis of the spine, Br. J. Vener.Dis. 48 (1972) 397–399.
- [12] J.P. Nielsen, Follow-up of syphilitics; late manifestations in 467 male patients with early syphilis followed for 29–36 years, Acta Derm. Venereol. 30 (1950) 507–512.
- [13] S. Tuddenham, S.S. Katz, K.G. Ghanem, Syphilis laboratory guidelines: performance characteristics of nontreponemal antibody tests, Clin. Infect. Dis. 71 (Suppl 1) (2020) S21–S42.
- [14] Y. Bai, F. Niu, L. Liu, H. Sha, Y. Wang, S. Zhao, Tertiary syphilis in the lumbar spine: a case report, BMC Infect. Dis. 17 (2017) 513.
- [15] R. Jameson, C. Garreau de Loubresse, A. Maqdes, Spinal neuroarthropathy associated with Guillain-Barre syndrome, Eur. Spine J. 19 (Suppl 2) (2010) S108–S113.
- [16] X. Wu, J. Shi, J. Wu, Y. Cheng, K. Peng, J. Chen, et al., Pedicle screw loosening: the value of radiological imagings and the identification of risk factors assessed by extraction torque during screw removal surgery, J. Orthop. Surg. Res. 14 (2019) 6.
- [17] R.F. Mclain, Functional outcomes after surgery for spinal fractures: return to work and activity, Spine 29 (2004) 470–477.
- [18] T. Tsutsumimoto, M. Shimogata, Y. Yoshimura, H. Misawa, Union versus nonunion after posterolateral lumbar fusion: a comparison of long-term surgical outcomes in patients with degenerative lumbar spondylolisthesis, Eur. Spine J. 17 (2008) 1107–1112.
- [19] J.W. Kim, S.W. Park, Y.B. Kim, M.J. Ko, The effect of postoperative use of teriparatide reducing screw loosening in osteoporotic patients, J. Korean Neurosurg. Soc. 61 (2018) 494–502.