Synovitis, Acne, Pustulosis, Hyperostosis, and Osteitis (SAPHO) Syndrome with Destructive Spondylitis: A Case Report

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Abstract:

\textbf{Introduction:} Spinal lesions in synovitis, acne, pustulosis, hyperostosis, and osteitis (SAPHO) syndrome generally have a good prognosis and rarely cause structural destruction or neurological deterioration. We described a surgical case of posterior instrumented surgery without anterior reconstruction and bone graft in a patient with SAPHO syndrome with destructive spondylitis and reviewed the literature on surgical treatment for this entity.

\textbf{Case Report:} We describe the case of a 73-year-old male who presented with palmoplantar pustulosis. He experienced progressive low back and leg pain for the past 3 months. Destructive spondylitis and lumbar canal stenosis were detected with magnetic resonance imaging (MRI), and aspiration biopsy was used to exclude pyogenic spondylitis and spinal tumors. He underwent posterior decompression and fixation surgery without anterior reconstruction and bone grafting. Low back and leg pain improved after surgery. Postoperative radiography and computed tomography showed boney bridge between vertebral bodies, and MRI showed the decrease of bone marrow edema.

\textbf{Conclusions:} Posterior fusion without anterior reconstruction produced a boney bridge between the vertebral bodies. Taking the pathophysiology of SAPHO syndrome into consideration, anterior reconstructed fusion for patients with SAPHO syndrome might not be needed.

\textbf{Keywords:}

synovitis, acne, pustulosis, hyperostosis, and osteitis (SAPHO), spine, spondylitis, surgery

Introduction

Synovitis, acne, pustulosis, hyperostosis, and osteitis (SAPHO) syndrome is considered a type of seronegative spondyloarthritis and is commonly recognized when skin and osteoarticular lesions are associated\textsuperscript{1-3)}. Spinal lesions in SAPHO syndrome generally show a good prognosis and rarely cause neurological deterioration\textsuperscript{4,5)}. For patients with spinal lesions in SAPHO syndrome with destructive spondylitis, anterior reconstruction with or without posterior fusion surgery using bone graft has been reported. We describe a case of SAPHO syndrome with rapidly progressing destructive spondylitis, which was treated with posterior percutaneous instrumentation without anterior reconstruction.

Case Report

A 73-year-old male has been treated for palmoplantar pustulosis for 10 years and has been using cyclosporine. He had experienced progressive lower back and leg pain for the past 3 months without any history of trauma. Plain radiography showed vertebral collapse at L3 with intravertebral instability and vertebral osteophyte (Fig. 1a). Computed tomography (CT) showed a severe erosion and destruction of vertebral endplate and vertebral body with marginal sclerosis on L3 (Fig. 1b, c). Magnetic resonance imaging revealed destructive and edematous change of L3 vertebral body and spinal canal stenosis at L2/3 (Fig. 1d, e). \textsuperscript{99m}Tc-HMDP bony scintigraphy showed abnormally high uptakes in the lumbar, cervical, and thoracic regions of the spine and sternocosto-
Figure 1. Plain radiogram (a: lateral plain radiogram) showing vertebral collapse at L3. Computed tomogram showing vertebral erosion at L3 and vertebral osteophyte formation together with destructive change, indicating the tendency of bony bridge (b: sagittal, c: coronal). Magnetic resonance image showing destructive and edematous change of L3 vertebral body (d: T1-weighted sagittal image, e: T2-weighted fat saturated sagittal image).

Figure 2. 99mTc-HMDP bony scintigraphy showing an abnormally high uptake of lumbar, cervical and thoracic spine and sternocostoclavicular joint.

clavicular joint (Fig. 2). Whole-body CT with contrast medium showed no evidence of infection or malignant tumor of organs. Laboratory examination demonstrated only a slight inflammatory reaction. C-reactive protein level was 0.871 mg/dL, white blood cell count was 7,500 cells/μl, and erythrocyte sedimentation rate was 14 mm/h. Blood culture, tumor markers, and polymerase chain reactions of tubercle bacillus, procalcitonin, and β-D-D-glucan were all negative. In order to make a diagnosis, an aspiration biopsy of the L3 vertebral body was performed. Histopathology demonstrated bone and bony marrow replaced by the immature fibrous tissue, and bacterium and tubercular culture of this specimen were negative. We diagnosed the patient with SAPHO syndrome with destructive spondylitis. Reconstructive surgery is usually performed4,5; however, taking into consideration the tendency of bony bridge and the pathophysiology of SAPHO syndrome, we planned to perform posterior decompression and fusion using percutaneous instrumentation without anterior reconstruction and bone graft. His lower back and leg pain improved immediately after surgery. One year after surgery, plain radiography and CT showed a bony bridge between L2-3 and L3-4 and sclerotic change of L3 vertebrae (Fig. 3a, b, c, d). Written informed consent was obtained from the patient for the publication of this case report and any accompanying images.

Discussion

This report is a retrospective review of a case of SAPHO syndrome with rapidly progressing destructive spondylitis treated with posterior percutaneous instrumentation without anterior reconstruction and bone graft. We successfully performed the surgery using posterior percutaneous instrumentation.

For patients with spinal lesions in SAPHO syndrome with destructive spondylitis, anterior fusion surgery (with or without posterior) using bone graft has been reported4,5. Prior to our patient’s surgery, CT showed vertebral osteophyte formation together with destructive change, indicating the tendency of bony bridge formation. We believed vertebral osteophyte would extend and form a bony bridge after stabilization using a percutaneous pedicle screw and rod system. Indeed, symptoms of back pain disappeared immediately after the surgery, and bony bridge and sclerotic change were evident. Regarding limitation, 1-year follow-up may be short considering the possibility of screw loosening or rod/screw breakage in the later period and there is a lack of evidence based on the long-term follow-up data.

To the best of our knowledge, this is the first report of posterior percutaneous instrumentation without anterior re-
construction and bone graft for destructive spondylitis in SAPHO syndrome. Although patients with a severe destruction of the spinal column and without the tendency of bone bridge formation generally require spinal reconstruction surgery, for patients with the tendency of the bony bridge of spondylitis in SAPHO syndrome, posterior fusion without anterior reconstruction may be a useful and less invasive surgical technique.

Conflicts of Interest: The authors declare that there are no relevant conflicts of interest.

Author Contributions: Toshio Nakamae wrote and prepared the manuscript, and all of the authors participated in the study design. All authors have read, reviewed, and approved the article.

References