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Two-Stage Total Hip Arthroplasty for Severe Hip Joint Destruction in SAPHO Syndrome: A Case Report

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Abstract: The syndrome of synovitis, acne, pustulosis, hyperostosis, and osteitis (SAPHO) is becoming more well-known. The hip joint is a less commonly afflicted location in SAPHO syndrome, and there have been few reports of hip joint illnesses caused by SAPHO syndrome, thus effective treatment for this disease spectrum is yet unknown. The case of a 22-year-old man who was hospitalized for SAPHO syndrome and later diagnosed with advanced secondary hip arthritis and severe right hip pain is described. The identification of SAPHO syndrome was delayed; in a prior tertiary health center, the patient was diagnosed with osteomyelitis and treated with extensive courses of antibiotics and open surgical debridement. In our facility, the patient had two stages of joint replacement surgery. He is doing well a year after surgery, with minor right hip discomfort and a functional prosthesis. This case demonstrates the safety and efficacy of a two-stage joint replacement in the treatment of degenerative hip disease caused by SAPHO syndrome, which mimics infectious arthritis.

Keywords: SAPHO syndrome, total hip arthroplasty, two-stage joint replacement, hip joint destruction

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Introduction

Hip replacement surgery is one of medicine's most effective procedures. Total hip replacement has been much more successful since the early 1960s, thanks to advancements in joint replacement surgical methods and technologies. Whether you're just starting to look at treatment choices or have already made the decision to have a total hip replacement, this information will help you understand the advantages and disadvantages of the procedure. This article explains how a healthy hip functions, the reasons for hip discomfort, what to expect following hip replacement surgery, and what exercises and activities will help you regain mobility and strength so you can resume your usual activities.

SAPHO syndrome is now thought to be an uncommon illness with an underestimated true frequency. The diagnosis and management of SAPHO syndrome remain difficult due to the wide range of clinical manifestations. There are numerous established diagnostic criteria for SAPHO, and the presence of only one of them is enough to confirm the diagnosis. When considering all of them, it can be concluded that the criteria developed by Kahn and revised in 2003 appear to be the most precise when combined.

A 22-year-old man came to us with acute right hip pain that has been getting worse for the past year. He was generally in good health, with no previous medical or trauma history. He was diagnosed with chronic osteomyelitis at the prior health institution and treated with extensive rounds of antibiotics and open surgical debridement. Despite all these therapies, right hip discomfort persisted, as did limited range of motion in the right hip joint.

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When he arrived, he was given a full physical examination, which revealed severe motion discomfort and substantial loss of range of motion in his right hip joint, as well as plantar pustulosis in both feet. Except for substantial elevations in C-reactive protein (CRP) and erythrocyte sedimentation rate (ESR) (2.92 mg/dL and 57 mm/h, respectively), laboratory tests were normal. Radiographic exams of the entire body were performed. With numerous cystic lesions at the femoral head and acetabulum, the right hip showed uniformly reduced joint space and osteophyte development.

Increased signal intensity in the gluteus and iliacus muscles, as well as joint effusion, synovitis, and sacroiliitis, were all seen on MRI. Increased tracer uptake was seen on bone scintigraphy in the right hip joint, manubrium, and sternoclavicular joint. SAPHO syndrome was suspected based on these observations.

Method

We intended to use complete joint replacement surgery for this condition because of the extreme debilitating hip pain and end-stage joint deterioration caused by osteitis refractory to prior antibiotic therapy and open surgical debridement. However, due to the possibility of infectious osteitis involving the right hip joint, we proceeded with 2-stage THA as a definitive operation.

Periosteal new bone formation with multiple subcortical cysts in the right ilium, cartilage denudation and joint space narrowing of the right hip joint associated with osteophyte formation and subchondral bone cysts in the right iliac and acetabulum, indicating secondary osteoarthritis, was seen on MRI of the right hip.

All necrotic and devitalized tissues were removed in the first step, and the functional temporary prosthesis was implanted using cement containing vancomycin and streptomycin. A tissue sample (5-6 specimens from the acetabulum and femur) was taken and submitted for 2 weeks of prolonged culturing. The femoral head and acetabulum were extensively degraded at the whole right hip joint, where the cartilage had peeled off, according to intraoperative observations. There was a slight dark brown tint to the joint fluid, but no unpleasant pus-like fluid.

Frozen biopsy revealed 5 neutrophil counts/mm^3 in all intraoperative specimens, and femoral neck cutting and extensive synovectomy surrounding the right hip joint were done. For each bag (40 g) of antibiotic-impregnated polymethyl methacrylate (PMMA) that was applied in the doughy phase to the temporary functional hip prosthesis, vancomycin powder (3 g) and 1 g of streptomycin were combined. The prosthesis was placed late in the polymerization process to reduce osseous interdigitation and enable removal in the second stage.

Result and Discussion

During the first step, many intraoperative samples were collected for microbiological investigation. All intraoperative gram stain and culture results from the right hip joint were negative. Histopathology revealed extensive lymphoplasmacytic infiltration with synovial hyperplasia, indicating subsequent osteoarthritis. Intravenous antibiotics were given for only one day postoperatively, with no additional antibiotics given, and the patient was allowed to mobilize and ambulate partially weight-bearing with a frame the day following surgery.

Intraoperative results revealed that there was no sign of infection in the second stage. The decision was taken to convert to a permanent THA system. A complete hip prosthesis was successfully implanted during the second-stage operation, with no microorganisms growing on any culture samples collected from the operating site during the second-stage surgery. The patient was ambulating without the need of any assistive equipment six months after surgery, and his hip pain was well managed by low-dose oral nonsteroidal anti-inflammatory medications.

SAPHO is now thought to be an uncommon illness with an underestimated true frequency. The diagnosis and management of SAPHO syndrome remain difficult due to the wide range of clinical manifestations. Despite all attempts to accurately diagnose SAPHO syndrome, it may prove to be a difficult diagnosis. Al-Juhani and colleagues highlighted how individuals with SAPHO syndrome frequently have soft tissue involvement around inflammatory bone, leading to diagnostic challenges. It was also shown by Boutin and Resnick that distinguishing SAPHO syndrome from other illnesses with comparable radiographic features but distinct prognoses and therapies, such as osteomyelitis, Ewing sarcoma, metastasis, and Paget disease of bone, might be challenging.

There has recently been more proof of the medicine used to treat SAPHO syndrome, which shows a significant improvement in the clinical picture of this disease. As a result, delaying disease development or controlling pain produced by SAPHO syndrome with medicine or physical therapy in the early stages of the disease is a viable treatment strategy. Unfortunately, our patient's SAPHO condition was not appropriately identified in the early stages, resulting in ineffective treatment. Systemic examination, radiographic examination, and bone scintigraphy, which has been shown to be a sensitive and reliable early diagnostic test

for SAPHO syndrome bone lesions, provided hints for our patient's diagnosis.

According to reports, hip joints are not frequently implicated with SAPHO syndrome, according to reports. Our patient had significant right hip discomfort and restricted range of motion, and imaging investigations revealed joint obliteration, severe arthritic change, and myositis surrounding the joint. When we searched the literature for references on how to effectively manage our patient, we found that there was very little information to help us decide on the best course of action.

A young girl presented with right hip discomfort, which was first diagnosed as septic arthritis but did not respond to antibiotic therapy, according to O'Connor et al. SAPHO syndrome was discovered after further clinical and radiological research. Following that, the patient chose conservative treatment and made a full recovery. In contrast to this account, our patient's right hip discomfort lasted around three years, and imaging scans revealed that the hip joint was destroyed with numerous osteophyte and cyst forms, making him an excellent candidate for joint replacement surgery.

Currently, THA is regarded as a safe treatment option for advanced degenerative hip joint disease. Moreover, other inflammatory diseases affecting the hip joint have been well treated with THA in severe cases. For the treatment of pigmented villonodular synovitis involving the hip joint, synovectomy is generally accepted as the most appropriate surgical option; however, in cases demonstrating end-stage joint destruction, THA is commonly coupled with an open synovectomy.

Although the cause of SAPHO syndrome is uncertain, one theory suggests that it is linked to seronegative spondyloarthropathies, particularly psoriatic arthritis. THA is a widely utilised therapy for psoriatic arthritis patients who have severe and painful deformities. As a result, we chose THA for our patient, whose hip had been badly injured by SAPHO syndrome.

Despite a negative Gram stain and culture result from an open surgical debridement at a previous hospital and an imaging study indicating SAPHO syndrome, we couldn't rule out the possibility that his right hip pathology was caused by an infectious condition due to disabling motion-related hip pain and elevated inflammatory serological markers. It's also worth noting that Propionibacterium acnes, Staphylococcus aureus, and other germs have been isolated from osteoarticular lesions in the anterior chest wall, spine, pustules, and synovial fluid tissue. SAPHO syndrome is most likely primitive reactive osteitis in genetically susceptible people. Culture-negative joint fluid specimens, according to recent findings, cannot conclusively rule out septic arthritis. With culture-negative septic arthritis, Slinger and colleagues discovered Kingella kingae. During the research period, Berbari and colleagues found that 7 percent of 897 instances of periprosthetic joint infection were impaired by culture-negative periprosthetic joint infection.

When it comes to treating infectious hip joint illness, 2-stage THA has been hailed as the gold standard and therapy of choice, especially when used in conjunction with antibiotic-loaded PMMA spacers. This is the only paper that we are aware of that focuses on 2-stage THA for patients with severe hip degeneration caused by SAPHO syndrome. This study examines the safety and efficacy of a two-stage THA procedure for the treatment of destructive hip disease caused by SAPHO syndrome, which mimics infectious arthritis.

Conclusion

Because new medicines for treating this disease have just been introduced, it is critical to diagnose SAPHO syndrome early to avoid ineffective treatments and to prevent joint damage. Despite increased awareness of SAPHO disease and improved medical treatments, we believe this study is extremely useful because we have only discovered a few references to treatment of severely injured hip joints caused by SAPHO syndrome in any national or international professional literature.

To our knowledge, this is the first study describing 2-stage THA as a treatment for severely injured hip joints exacerbated by SAPHO syndrome's delayed diagnosis and poor treatment. Based on our findings, 2-stage THA was an effective and safe definitive management strategy for controlling severe hip pain associated with severe secondary osteoarthritis complicated by SAPHO syndrome, despite the difficulty in distinguishing non-infectious osteitis caused by SAPHO syndrome from other infectious osteitis involving hip joints.

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Author Contributions

Conceptualization, V.M. and P.S.; methodology, V.M.; validation, V.M. and P.S.; formal analysis, V.M.; investigation, V.M.; resources, V.M.; data curation, V.M.; writing—original draft preparation, V.M.; writing—review and editing, V.M. and P.S.; visualization, V.M.; supervision,

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Conflicts of Interest

The authors declare no conflict of interest. The funders had no role in the design of the study; in the collection, analyses, or interpretation of data; in the writing of the manuscript; or in the decision to publish the results.

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