

SPINAL CORD STIMULATOR IMPLANTATION IN A PATIENT WITH DIFFUSE IDIOPATHIC SKELETAL HYPEROSTOSIS: CASE REPORT

Shabaaz M. Baig, MD¹, Adejuyigbe Adaralegbe, MD¹, Sangel Gomez, BA¹, Tabhata Paulet, BA¹, Carol Apai, MD¹, Mia Castiglione DO¹, Adejimi Adaralegbe, BA², and Akwasi Amponsah, MD¹

Background: Diffuse idiopathic skeletal hyperostosis (DISH) is a noninflammatory skeletal disorder causing calcifications and ossification of ligaments and tendons. We present a patient with undiagnosed DISH in whom a spinal cord stimulator (SCS) provided adequate pain relief while the disease progressed to become debilitating.

Case Report: The patient is a 66-year-old woman with an SCS placed for lower back pain due to degenerative changes. The patient presented to the emergency department with progressive right-sided lower extremity weakness and a 3-day history of urinary incontinence. Computed tomography showed new osteophytes consistent with DISH throughout the thoracic spine to the level of T10-T11.

Conclusions: Our aim with this case report is to demonstrate the importance of preoperative imaging as well as follow-up imaging to evaluate the progression of DISH and having an extensive conversation with patients about whether an SCS is the right choice to manage their pain.

Key words: Spinal cord stimulator, lumbar spine, pain management, DISH

BACKGROUND

Diffuse idiopathic skeletal hyperostosis (DISH), otherwise known as Forestier's disease or ankylosing hyperostosis, is a noninflammatory skeletal disorder causing calcifications and ossification of ligaments and tendons (1). We present a patient with undiagnosed DISH in whom a spinal cord stimulator (SCS) initially provided adequate pain relief until her symptoms recurred and progressed to become debilitating. We aim to demonstrate the importance of obtaining preoperative imaging prior to accessing the spine for permanent lead placement and how the presence of DISH can affect the outcome and safety profile of an SCS implant. Informed consent was obtained for the publication of this case report.

The term DISH was coined, in 1975, by radiologists

who noticed the extraspinal manifestations of the known degenerative spine diseases recognized at the time. The prevalence of DISH is unknown, with various studies (2-4) giving broad estimates. However, it is generally accepted that DISH typically presents more commonly in men over the age of 50 and that there is an increase in prevalence with increasing age (2,3).

DISH most commonly develops on the right side of the thoracic spine. While typically an indolent disorder, symptoms can occur due to spinal ossifications causing pain, stiffness, and decreased range of motion (1-3). Some other more serious complications include unstable spinal fractures and cord compression (3). Radiographical findings are typically used to confirm the diagnosis. The current criteria used to establish a diagnosis include the formation of new vertebral ossifications in at least

From: ¹Department of Anesthesia and Perioperative Care, Rutgers New Jersey Medical School, Newark, NJ; ²Ross University School of Medicine, Miramar, FL

Corresponding Author: Shabaaz M. Baig, MD, E-mail: smbaign94@gmail.com

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4 contiguous vertebrae and enlarged bony bridges present in either the cervical, thoracic, or lumbar spine (1). One other key feature of DISH that may be seen is peripheral joint involvement. The major lower body joints, such as the hip and knee, tend to be less likely to be affected by DISH; however, the foot and ankle are more likely to show hypertrophic changes (3).

DISH is generally considered to be a relatively mild disease, especially when compared to other degenerative spine diseases, such as ankylosing spondylitis. Treatment is primarily noninvasive and involves physical therapy and nonsteroidal anti-inflammatory drugs (3,5). However, when DISH continues to result in significant back pain despite maximizing noninvasive therapy, patients are often referred to interventional pain specialists for more invasive options. For those cases, which present with severe symptoms or complications, a surgical evaluation is indicated (4).

CASE

The patient is a 66-year-old woman with a longstanding history of lower back pain radiating down her right leg due to degenerative changes in the lumbar spine. She had a prior L2-L4 right partial laminectomy, in 2017, with significant relief of her symptoms and another lumbar laminectomy, in 2020, due to symptom recurrence without any relief. The patient was evaluated by neurosurgery, in January 2022, and was told definitive treatment of her pain would entail significant invasive spine surgery. Imaging of the lumbar spine, from both 2020 and 2022, showed slowly progressive degeneration from L1-S1. No thoracic spine imaging was performed at this time. The patient declined surgery and was instead referred to pain management for an SCS trial. She acknowledged significant improvement of her pain with a 7-day trial.

In April 2022, the patient was scheduled for an SCS implantation in the operating room. Using loss-of-resistance-to-air technique, the epidural space was accessed. Two stimulator leads were inserted through Tuohy needles. Lateral fluoroscopic views confirmed the wires to be in the posterior epidural space. Under live fluoroscopy, the wires were easily driven to the T7 and T8 superior vertebral end plate. Lateral films were obtained again to ensure the electrodes were in the posterior epidural space. After creating a pocket for the generator and connecting the leads, the SCS was tested and confirmed to be functioning. The patient did well after the procedure. She followed-up with the

pain clinic, in November 2022, where she had substantial overall improvement of her pain but reported muscle cramping in the right lower extremity. Her oral pain medications were increased at that time.

In January 2023, she reported a one-week history of weakness in her right lower extremity. No intervention was done at that time as the patient did not feel it caused her significant discomfort. However, 3 weeks later, the patient presented to the emergency department with progressively worsening right-sided lower extremity weakness and a 3-day history of urinary incontinence. Pain management evaluated her and noted that the leads of the SCS were in place and that there was no change in her symptoms after it was turned off. Computed tomography showed confluent anterolateral osteophytes consistent with DISH extending throughout the thoracic spine to the level of T10-T11. Additionally, a magnetic resonance imaging of the thoracic and lumbar spine noted widening of the anterior aspect of the disc space at T10-T11 and degenerative changes at T10-T11 with some mass effect on the ventral and dorsal aspects of the thoracic spinal cord. There was also underlying signal abnormality on the cord, which was highly suspicious for edema with cord contusion. Neurosurgery recommended urgent decompressive surgery to prevent disease progression and possible paraplegia, but the patient refused and preferred to follow-up in outpatient care. She later underwent a T10-T11 posterior instrumented fusion with reduction of extension/distraction fracture, SCS removal, and posterior T10-T11 laminectomy. The surgical team reported that when removing the SCS, they noted that the leads of the SCS became ossified, preventing them from being easily removed. The patient did well after the procedure and reported an improvement in her symptoms with acute inpatient rehabilitation.

DISCUSSION

This case illustrates how DISH can be another cause of severe stenosis that must be evaluated with imaging prior to placing an SCS. In this case, the diagnosis of DISH had not been established due to a lack of thoracic spine imaging, with the diagnosis at the time being postlaminectomy syndrome from the patient's prior lumbar laminectomies. Thus, it was unknown if the patient's spine had already undergone any changes characteristic of DISH, though given the indolent nature of the disease, she likely already had bony changes that were contributing to her pain symptoms. Even though

implantation of an SCS is generally considered to be safe with recognized beneficial effects, absence of imaging to assess the anatomy beforehand can lead to complications further down the line (6).

A review of the literature does not show any previous cases where SCS had been used to manage patients affected by DISH. However, in recent years, DISH has become more well known to clinicians. Several studies and reviews have been published on the pathophysiology and etiology of the disease, but further advancements in treatment modalities have not been documented. Conservative management remains the mainstay of treatment. However, as SCSs have become more popular, it is also likely that patients with mild symptomatic DISH have had adequate pain control with an SCS, even though they may have been undiagnosed or misdiagnosed with a similar presenting condition. Unfortunately, given the risk of developing more serious symptoms from the progression of DISH, it is crucial for the interventional pain specialist to perform a thorough evaluation with imaging and discuss the risks and benefits of the procedure with the patient.

The utility of obtaining further diagnostic imaging before pain management physicians implant an SCS cannot be understated, particularly if the leads of the SCS will extend into the thoracic spine. Though this patient's pain was initially very well controlled via SCS, if preoperative imaging had demonstrated changes in the thoracic spine, this would have alerted the clinician to the increased likelihood of complications following the SCS implant. In addition, while in our case DISH may have been the cause of stenosis from the formation of new osteophytes, the need for preoperative imaging

is imperative to evaluate for any cause of stenosis that could lead to cord compression. If imaging shows the presence of a disease process that causes stenosis, it must be considered that implantation of any device may result in a higher likelihood of complications. Follow-up imaging is also imperative, as it may identify disease progression from any cause of spinal stenosis before symptoms advance and can lead to earlier surgical intervention. It is also key for the pain management specialist to discuss lead ossification as a potential complication of SCSs, and that if there is disease progression, surgical removal of the leads and decompressive spine surgery may be the only possible methods of symptom control. Given these factors, the only alternative form of pain control aside from surgery currently recommended in these high-risk patients would be steroid injections in conjunction with nonsteroidal anti-inflammatory drugs and physical therapy (5).

CONCLUSIONS

While the SCS was beneficial for the patient's pain, it is not possible to tell whether the patient previously had DISH of the thoracic spine without imaging prior to the SCS implant. Evaluating patients with imaging for any disease process that can lead to spinal stenosis should be the standard of care to prevent further progression to cord compression and surgical intervention. Our aim with this case report is to demonstrate the absolute necessity of preoperative imaging as well as follow-up imaging to evaluate the thoracic spine and having an extensive conversation with patients about whether an SCS is the right choice to manage their pain.

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