Cauda equina syndrome is an uncommon complication of ankylosing spondylitis characterized by the slow and insidious development of severe neurologic impairment related to dural ectasia. This report describes a unique case of cauda equina syndrome in a patient with ankylosing spondylitis after hip revision surgery. A 70-year-old man with long-standing ankylosing spondylitis underwent standard hip revision surgery; combined spinal and general anesthesia was administered. Pain was controlled with intravenous opioids postoperatively (patient-controlled analgesia). As per routine protocol, on the first postoperative day, the patient remained supine on a hip abduction pillow; mobilization was initiated on the second postoperative day. On postoperative day 1, the patient had severe low back pain that was controlled with patient-controlled analgesia. On postoperative day 2, the Foley catheter was removed and the patient sat and dangled. Back pain persisted while supine; in addition, the patient noticed involuntary loss of urine. On postoperative day 3, the patient had below-the-knee numbness that progressed to saddle anesthesia and foot flexor and extensor weakness. An epidural hematoma was suspected and urgent magnetic resonance imaging was performed, which showed severe degenerative stenosis at the L4-L5 level (mainly by dense ligamentum flavum). An L4-L5 decompression and instrumented fusion was performed; intraoperatively, L4-L5 was found to be the sole mobile segment. The extension of the spine in the supine position that completely obliterated the spinal canal was considered the mechanism of cauda equina syndrome. The intensity of back pain is a good indicator of a severe spinal lesion; however, pain can be dampened by intravenous opioids. High suspicion is required in patients with preexisting spinal pathology, such as ankylosing spondylitis.
Cauda equina syndrome is considered a severe neurologic condition with progressive loss of function of the neurologic elements of the spinal canal below the termination of the spinal cord. Common causes include intervertebral disk prolapse, metastatic disease, direct trauma as a result of lumbar puncture, and spinal anesthesia. Degenerative spinal stenosis, developmental defects, and inflammatory diseases, including ankylosing spondylitis, may also be associated with the development of cauda equina syndrome.1,2

Bowie and Glasgow3 first described ankylosing spondylitis-related cauda equina syndrome in 1961. The incidence of ankylosing spondylitis-related cauda equina syndrome is very low, accounting for approximately 2% of patients with ankylosing spondylitis.4 The exact pathogenesis of the syndrome is unclear. The hypothesis that initial inflammation in the ligaments may lead to adjacent meningeal inflammation and arachnoiditis, with subsequent nerve root inflammation, degeneration, fibrosis, adhesion, and tethering, all resulting in cauda equina syndrome, was described in 1968 by Matthews.5 The characteristic clinical presentation of ankylosing spondylitis-related cauda equina syndrome usually involves slowly progressive neurologic symptoms consisting of motor and sensory deficits of the lower limbs and loss of voluntary control of the bowel and bladder.1 This article reports a unique case of ankylosing spondylitis-related cauda equina syndrome with acute onset of neurologic symptoms after hip revision surgery that was probably related to postoperative stay in the supine position that caused tethering and subsequent ischemia of nerve elements. The goals of this report are to increase awareness among hip surgeons about this possible postoperative complication after total hip replacement surgery, to note the severity of this condition, and to emphasize the need for immediate surgical treatment.

CASE REPORT

A 70-year-old man was admitted to a tertiary hospital for revision surgery of a mechanically loose left total hip arthroplasty (Figure 1). He underwent bilateral primary total hip replacement 20 years ago. This patient had ankylosing spondylitis diagnosed at the age of 27 years, and he was treated with 75 mg of indomethacin daily. The treatment provided relief from symptoms for many years, with the exception of minor, occasional low back pain. The preoperative range of left hip motion was 95° flexion, -10° extension, 20° abduction, 15° adduction, and 20° internal and external rotation. The hip was in slight flexion contracture (10°). The range of motion for the lumbar spine included 20° flexion, 5° extension, 20° right and left lateral flexion, and 10° rotation. The rest of the medical history was unremarkable.

Revision total hip replacement surgery (Figure 2) was performed under combined regional combined anesthesia. Postoperatively, pain was controlled with intravenous opioids (patient-controlled analgesia). According to the routine protocol, on the first postoperative day, the patient remained supine on a hip abduction pillow and mobilization was initiated on the second postoperative day.

On postoperative day 1, the patient had severe low back pain that was controlled with patient-controlled analgesia. On postoperative day 2, the Foley catheter was removed and the patient sat and dangled his legs. Back pain persisted while the patient was supine; additionally, the patient noticed involuntary loss of urine. On postoperative day 3, he had below-the-knee numbness that progressed to saddle anesthesia and weakness of the foot flexors and extensors. Repeated sensory examination showed decreased urethral sensation, saddle and perianal anesthesia (S2, S3, and S4 dermatomes prominent on the left side), and loss of anal sphincter tone and sensation. Lower-extremity deep tendon reflexes and the bulbocavernous reflex were absent. Motor examination showed lower-extremity weakness and slight left sciatica (S1 distribution).

Epidural hematoma was suspected, and an urgent magnetic resonance imaging (MRI) scan was performed and showed severe degenerative stenosis at L4-L5 (mainly by dense ligamentum flavum and arachnoid mater) with nearly complete obliteration of the spinal canal (Figure 3). A L4-L5 decompression with instrumented fusion was performed. Intraoperatively, L4-5 was found to be the sole mobile segment.

On postoperative day 1, the patient showed immediate improvement of neurologic status, partially regaining motor strength and experiencing complete resolution of the lancinating back pain with accompanying left sciatica. By postoperative day 3, he could walk with the aid of a walker. The patient had moderate consti-

---

Figure 1: Preoperative anteroposterior radiograph of the pelvis showing cemented total hip replacement of both hips with excessive polyethylene wear of the acetabular components and loosening at the cement-bone interface of the left hip 20 years postimplantation.

Figure 2: Postoperative anteroposterior radiograph of the pelvis showing the revised acetabular component on the left side using a jumbo cluster hole cup (Trabecular Metal Revision Shell; Zimmer Inc, Warsaw, Indiana) and 2 screws.
pation and urinary retention, necessitating the use of a temporary urinary catheter. One month later, an urodynamic study was performed that showed persistent autonomous neuropathic bladder.

At the latest follow-up, 1 year postoperatively, the patient had no symptoms as a result of the fused lumbar spine. He could walk independently, including ascending stairs. There was mild motor weakness (4 of 5 according to the Frankel scale) on dorsiflexion of the left toes and ipsilateral hip extension and abduction and diminished left foot plantar flexion. The patient’s bowel incontinence improved. However, urinary retention persisted and the use of intermittent self-catheterization for bladder drainage and laxatives for emptying of the large intestine was necessary. Medical therapy included a selective alpha-1-blocker (tamsulosin) and distigmine bromide that improved bladder contraction, relieving urine retention. On bladder distention, the patient had minimal voiding sensation.

**DISCUSSION**

Cauda equina syndrome is a rare complication of advanced ankylosing spondylitis that presents with slowly progressive neurologic symptoms that involve the bowel, bladder, and lower limbs. This report describes a unique case of acute onset of neurologic symptoms of ankylosing spondylitis-related cauda equina syndrome in a patient with chronic ankylosing spondylitis who underwent revision total hip replacement. This case vividly highlights the effect of acute tethering of nerve elements to the fibrotic arachnoid mater that was probably induced by the postoperative positioning of the patient.

Cauda equina syndrome is usually a late and uncommon or underdiagnosed complication of advanced chronic ankylosing spondylitis. The prevalence of neurologic deficits in patients with ankylosing spondylitis is very low. Edgar showed that cauda equina syndrome was found in 2.1% of 290 patients with ankylosing spondylitis. Ahn et al. showed that the prevalence of neurologic findings in ankylosing spondylitis-related cauda equina syndrome is very high, even when loss of bowel or bladder function is not included. This case report is the only case in the literature of ankylosing spondylitis-related cauda equina syndrome occurring suddenly after a routine surgical procedure. Most of the other cases in the literature developed slowly with insidious onset. Mahesh et al. presented a case of ankylosing spondylitis-related cauda equina syndrome with acute neurologic symptoms. This patient had asymptomatic early, incomplete Anderson lesion of the spine that progressed rapidly to fracture dislocation and resulted in complete paraplegia after total hip arthroplasty. The authors hypothesized that stress localized to a potentially weak area in the anterior aspect of the otherwise ankylosed spine induced the 3-column defect. This could cause a fracture dislocation, even in the absence of significant trauma.

The etiopathogenesis of ankylosing spondylitis-related cauda equina syndrome is poorly understood. Hypotheses include adhesive arachnoiditis, small vessel angiitis of the nerve roots, and decreased elasticity of the dural sac, leading to increased cerebrospinal fluid pressure from arterial pulsations. Pathologic specimens have shown posterior arachnoid di-verticula with laminar and spinous process erosions as well as atrophy and demyelination of several nerve roots, without inflammatory infiltrates. Studies of surgical specimens in cases of advanced chronic ankylosing spondylitis have shown obliteration of the epidural space and adhesion of the dura mater to the vertebrae but no evidence of inflammation. The dura mater is usually thin and friable. There are several foci of fibrosis and thickening of the arachnoid with hemosiderin deposition, whereas lymphocytes are found infiltrating the dura mater.

The reported pathophysiologic mechanism involves fibrosis and adhesion of the arachnoid and dura mater to the vertebrae, which may in turn impair cerebrospinal fluid resorption. Furthermore, fibrotic meninges may be unable to dampen brief fluctuations in cerebrospinal fluid pressure secondary to breathing and arterial pulsations. According to another hypothesis, the pathogenesis of ankylosing spondylitis-related cauda equina syndrome may involve lesions of the apophyseal joints and entheses, such as the ligamentum flavum or interspinous ligaments and joint capsules. These lesions may lead to epidural tissue inflammation, arachnoiditis, thickening of the arachnoid, meningeal fibrosis, and adhesion of the arachnoid to the dura mater and periosteum.
The intraoperative findings of the current case were consistent with those reported in the literature, including mainly dense fibrotic tissue of the ligamentum flavum and arachnoid mater at the level of L4-L5 that was the only mobile segment of the lumbar spine.

Nerve root tethering and adhesion to the dural sac may cause obliteration of the epidural space with loss of the blood supply to the nerve roots that subsequently induces ischemic neurologic manifestations. In the absence of other mechanisms, ischemia is usually believed to account for acute spinal cord deficits, although toxicity from local anesthetics and epidural hematomas have been also reported. In the current case, epidural hematoma after regional anesthesia was excluded by MRI scan and the intraoperative findings. Toxicity from local anesthetics was excluded because there was a symptom-free interval between regional anesthesia and the development of cauda equina syndrome.

Ankylosing spondylitis-related cauda equina syndrome is associated with a poor prognosis. The physical history of the disease consists of neurologic impairments that worsen steadily over time. In the largest reported case series, all 14 patients had worsening of neurologic manifestations over the mean follow-up period of 8 years (range, 2-23 years). A meta-analysis has also shown that 80% of untreated patients experienced worsening of neurologic status.

Treatment is controversial, and options include cerebrospinal fluid peritoneal shunting, surgical decompression, and use of tumor necrosis factor inhibitors. A meta-analysis of outcomes after medical and surgical treatment found no evidence that nonsteroidal anti-inflammatory drugs or local or systemic glucocorticoids might be effective. Cornec et al recently reported the efficacy of infliximab. In the same meta-analysis, surgical treatment (laminctomy or lumboperitoneal shunting) appeared superior to pharmacotherapy for improving pain, sensory and motor deficits, urinary symptoms, and anal sphincter dysfunction.

In the current literature there are reports on 8 patients treated by laminectomy. Three patients had stabilization of neurologic features, and 1 had transient worsening of motor deficits and improvements in urinary symptoms, and 1 had improvement of radicular pain. No information on postoperative outcomes was available for 2 of these patients, whereas 1 died of postoperative meningitis.

CONCLUSION

Cauda equina syndrome is a rare complication of advanced ankylosing spondylitis, and acute onset of neurologic symptoms after total hip arthroplasty is uncommon. However, hip surgeons should have increased suspicion of this entity, especially in patients with preexisting spinal pathology, such as ankylosing spondylitis.

The intensity of back pain is a good indicator of a severe spinal lesion; however, pain can be dampened by intravenous opioids. The authors believe that extension of the spine in the supine position that completely obliterates the spinal canal might be the mechanism of cauda equina syndrome.

Preoperative imaging in patients with ankylosing spondylitis who undergo total hip arthroplasty should probably include MRI scan of the lumbar spine to assess for epidural tissue inflammation, arachnoiditis, thickening of the arachnoid, meningeal fibrosis, and adhesion of the arachnoid to the dura mater and periosteum. Dynamic radiographic views of the lumbar spine, including flexion, extension, and lateral flexion, as well as sagittal views in the sitting and standing positions, can be useful for preoperative evaluation of motion, the number of moving segments, and the radiographic range of motion of the lumbar spine.

There is some question about appropriate postoperative positioning of patients. Alterations in the routine postoperative rehabilitation protocol should be considered, possibly by adopting a slightly bent position of the lumbar spine during bed rest or a flexed position of the hips that would diminish traction on the nerves at the single mobile spine segment.

REFERENCES


