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# Spinal Arachnoiditis Ossificans: Report of Quadruple-Triggered Case

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**Key words**
- Ankylosing spondylitis
- Arachnoid cysts
- Arachnoiditis ossificans
- Spinal surgery
- Spinal trauma

**Abbreviations and Acronyms**
3D: Three-dimensional
AO: Arachnoiditis ossificans
AS: Ankylosing spondylitis
CT: Computed tomography
MRI: Magnetic resonance imaging

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BACKGROUND: **Arachnoiditis ossificans (AO) is a rare condition often associated with previous spine surgery. Here we describe a unique case of a patient affected by ankylosing spondylitis (AS), presenting with progressive neurologic deterioration due to AO. We also review the literature on evaluation and management of patients suffering from AO.**

CASE DESCRIPTION: **The 65-year-old patient had a history of previous spinal trauma and related thoracolumbar surgery. Magnetic resonance imaging revealed multiloculated intradural/extramedullary cysts on the posterior surface of the spinal cord at Th9-L1, with clustered nerve roots. Computed tomography, with 3-dimensional reconstruction, demonstrated a likely ossification of both the dura and arachnoid from Th9 to S1. Microsurgical debridement of scar tissue from previous surgery, drilling of posterior ossified plaques at Th11-Th12-L1, and marsupialization and drainage of arachnoid cysts at Th11-Th12 were performed.**

CONCLUSIONS: **We submit that AS, spinal trauma, epidural hematoma, and related surgery may be synergistic and independent factors in the etiopathogenesis of AO. This should be considered in patients with AS and/or a history of spinal surgery who present neurologic worsening.**

## INTRODUCTION

Spinal arachnoiditis ossificans (AO) is an uncommon chronic disorder characterized by ossification of the arachnoid layer; however, its true incidence is still unknown.[1] Calcified arachnoid and/or dural plaques can be found in surgical and autoptical specimens with an incidence of 43%–76%.[2] Rudolf Virchow first described both symptomatic and asymptomatic calcifications, and in 1931 Puusepp reported the first 4 cases of the disease.[3] Intradural ossifications with chronic arachnoiditis are often associated with progressive neurologic deficits that can require surgical treatment.[4] AO can lead to a wide range of neurologic symptoms and signs with regard to the size and location of the ossification inside the thecal sac. Only 3 cases of AO associated with ankylosing spondylitis (AS) have been reported.[5-7] Although AO etiology is not clearly understood yet, specific factors such as spinal trauma, previous spinal surgery, subarachnoid hemorrhage, thecal puncture during myelography, and infections are involved in its etiopathogenesis. We report the first case of a patient with AS, history of previous spinal trauma, and related thoracolumbar spinal surgery and AO.

## CASE REPORT

### History

A 68-year-old man was admitted to our unit because of low-back pain radiating to both legs, a tingling sensation from Th10 dermatome down both proximal lower extremities, progressive lower limb weakness, and intermittent claudication. Urinary dysfunction was not reported. Five years before he had been diagnosed with AS, a spine computed tomography (CT) scan showed a typical "bamboo appearance" with marginal syndesmophytes between adjacent vertebral bodies and thoracic kyphosis. When he was 64 years old, he underwent a Th11-Th12 decompressive laminectomy and Th10-Th11, L1-L2 posterior percutaneous vertebral fixation for an unstable Th12 vertebral body fracture. An associated posttraumatic spinal epidural hematoma was evacuated during the same surgical procedure.

### Neurologic Examination

On admission, his neurologic examination showed a right brisk patellar reflex and right lower limb sustained clonus. Anal sphincter tonus was normal.

### Preoperative Imaging

Preoperative spine CT scan with 3-dimensional (3D) reconstruction showed ossification of both the dura and arachnoid from Th9 to S1, around the spinal cord and nerve roots (Figures 1 and 2). Magnetic resonance imaging (MRI) also revealed multiloculated intradural/extramedullary cysts on the posterior surface of the spinal cord at Th9-L1, with clustering of nerve roots (Figure 3).

### Surgical Intervention

A microsurgical debridement of scar tissue of previous surgery, drilling and dissection of posterior ossified thecal sac and arachnoid at Th11-Th12-L1, and marsupialization and drainage of arachnoid cysts at Th11-Th12, with decompression of neural structures, were performed (Figure 4). A complete removal of the ossified plaques was not performed

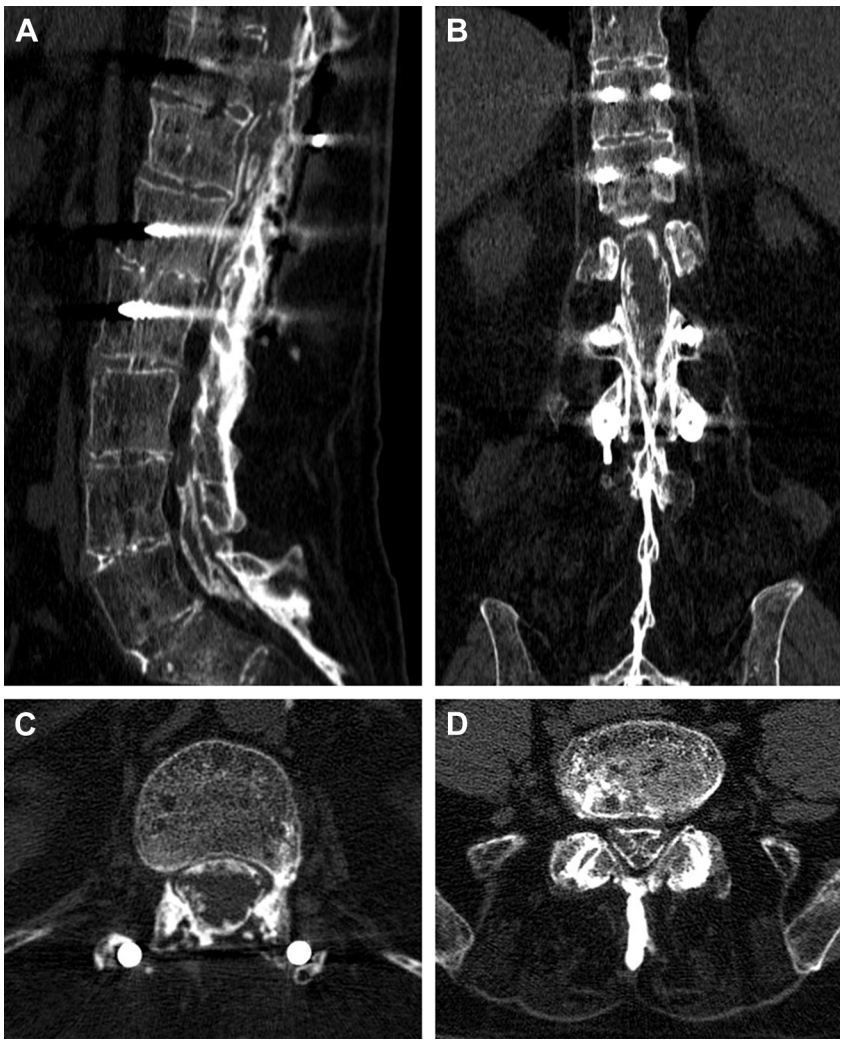


**Figure 1.** Preoperative sagittal, coronal, and axial computed tomography images showing thecal sac and arachnoid ossification (AO) (**A**–**D**) and 2 different ossification patterns in the same patient. In type II AO, concentric ossification surrounds the thecal sac (**C**). In type III AO there is a "honeycomb" appearance in which nerve roots may be encased in arachnoid ossification (**D**).

**Figure 2.** Computed tomography scan with 3-dimensional reconstruction showing the extent of dura and arachnoid ossification along the thoracolumbosacral spinal canal (*white asterisks*), providing a detailed study of contiguity with previous thoracolumbar fixation system.

because of tight adherences to the spinal cord and its blood vessels.

### Postoperative Course

Postoperatively, the patient improved neurologically despite a partial resection of the ossified plaques. On the third postoperative day, after transient urinary incontinence, he progressively started walking with assistance and reduced back pain, as well as increased leg strength, were observed.

### Postoperative Imaging

Early and 3-month postoperative MRI showed proper decompression of the neural structures (Figure 5). Postoperative CT scan confirmed partial resection of ossified arachnoid and dura.

## DISCUSSION

AO is ossification of the spinal arachnoid associated with chronic arachnoiditis, with or without dural ossification, leading to neurologic impairment.[4]

Incidental dural and leptomeningeal calcifications are commonly found during spinal surgery or in autopsy studies; however, they are composed of calcium crystals and do not usually have clinical relevance.[2] AO generally involves the thoracic spine in about 64% of all patients and the lumbar spine in 24%, with progressive neurologic worsening.[8] Both spine segments can be involved in the same patient. To date, only 1 case of cervical spinal AO has been reported.[9] The osseous plaques are usually found in the dorsal arachnoid layer of the middle-lower thoracic and lumbar spine and occur more frequently with aging. AO pathogenesis is not clearly understood yet. Predisposing factors such as trauma, previous spinal surgery, subarachnoid hemorrhage, spinal anesthesia, myelography procedures, meningitis, genetic factors, dural vascular anomalies, neoplastic conditions, and repeated mechanical injury to the spinal meninges are related to the etiopathogenesis of AO.[2]

**Figure 3.** Preoperative T2-weighted sagittal and axial magnetic resonance imaging demonstrating multiple arachnoid cysts between Th10 and L1. An intradural/extramedullary cyst is shown on the right posterolateral aspect of the spinal cord at level Th11 (*white arrow*).

Burton et al[10] considered ossification as the effect of chronic adhesive arachnoiditis. In this late stage of the chronic inflammatory process, metaplasia may cause the proliferation of osteoblasts leading to the development of ossification.[11]

A literature review performed through a Medline web search, using the key words "spinal arachnoiditis ossificans," "arachnoiditis ossificans," and "arachnoid ossification" provided 58 articles with 11 additional articles found in the reference lists. Particularly, our search revealed only 3 papers describing AO in association with AS.[5-7] Bilgen et al[5] suggested that AO is caused by dystrophic changes as a result of degenerative and/or chronic inflammatory process in patients with AS. Joo et al[6] reported a case of a patient with AO who underwent spinal surgery for AS. Moreover, Hasturk et al[7] described the case of a patient suffering from AS, history of previous related spinal surgery, and syringomyelia with AO. The development of syringomyelia and/or arachnoid cysts secondary to AO suggests possible altered cerebrospinal fluid dynamics due to subarachnoid space obstruction by arachnoid and/or dural ossified plaques.[12,13] We submit that in our case, arachnoid cysts significantly contributed to neurologic deterioration. Furthermore, our patient improved neurologically despite a partial resection of the ossified plaques, suggesting that the symptoms mentioned earlier were caused by not only direct compression of arachnoid ossification but also arachnoid cysts and their related altered cerebrospinal fluid dynamics. The combination of previous spinal surgery and AS is considered a possible risk factor for AO.[6,14] Our patient showed evidence of previous spinal trauma, epidural hematoma, related spinal surgery, and history of AS; thus these should be considered as triggers and synergistic factors for AO.

In case of nerve root involvement, associated syringomyelia, and/or arachnoid cysts, a good knowledge of AO MRI appearance has become mandatory. Spine MRI showed clustered nerve roots of the cauda equina, representative of arachnoiditis; associated ossification may have a wide-ranging appearance represented by overlying linear or masslike signal abnormalities, usually hyperintense on T1-weighted images and hypointense or hyperintense on T2-weighted sequences. These various findings on MRI depend on the stage of ossification and type of calcium macromolecular aggregates. A further investigation with spine CT is, in our opinion, fundamental for AO diagnosis because of its sensitivity and specificity to

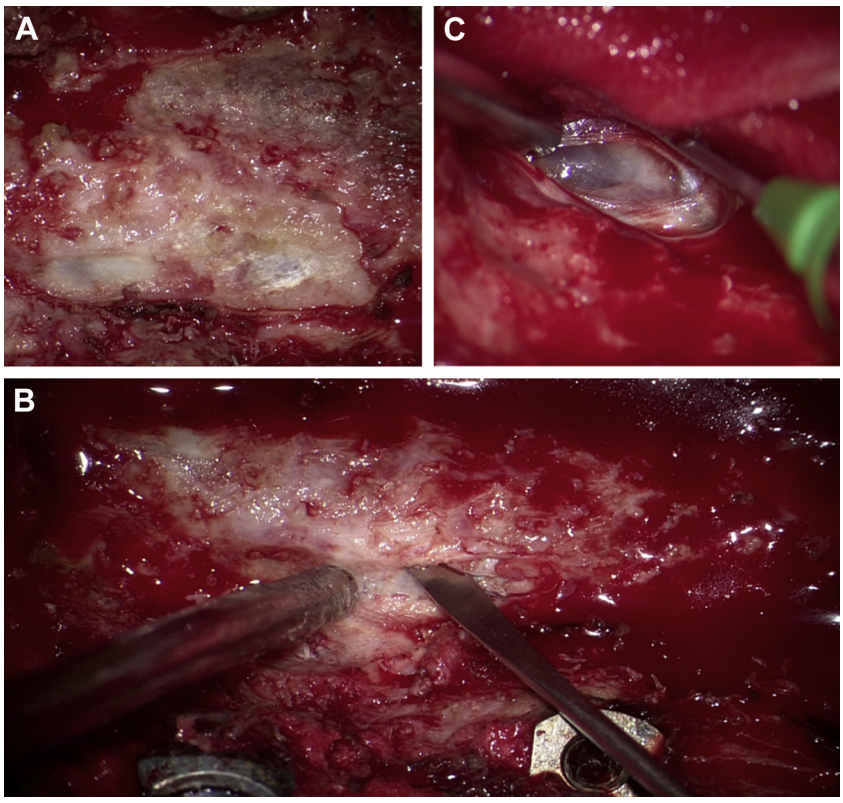


**Figure 4.** Intraoperative findings included extended ossified dural plaques (**A**), dissection of the dural plaques to expose underlying arachnoid ossification (**B**), and marsupialization of arachnoid cyst wall with a microsurgical cutter (**C**).

detect ossification plaques, recognize specific patterns, and make a differential diagnosis. Indeed, unlike arachnoid cysts, in cases of dermoid cysts, a CT scan may show intralesional calcifications. Unenhanced CT scan with 3D reconstruction is useful to realize adequate preoperative planning and recognize the extent of ossification, and none of the spinal arachnoiditis ossificans cases have described or reported its use (Figure 2).

AO has been classified into 3 types, according to Domenicucci et al[15]: In type I there is a semicircular appearance of ossification on CT, often located in the dorsal area of the spinal cord; type II AO is more circular and uniformly surrounds the thecal sac; and type III is characterized by a "honeycomb" appearance, in which nerve roots are bundled and encased in arachnoid and dural sac ossifications. Exceptionally, in our case, sagittal, axial, and 3D CT reconstruction images showed both types II and III calcification patterns in the same patient (Figure 1). Although only in type II AO, which mainly contributes to the onset of symptoms, is surgery feasible, we removed part of the ossified structures, drained cysts, and decompressed the neural structures. Often, it is not possible to radically treat type III AO because removal of ossified plaques from the nerve roots can lead to neural and vascular injuries, particularly in the lumbar spine.[16] A plain radiography examination may demonstrate arachnoid and/or dural calcification with nonspecific findings. The optimal treatment, as well as its timing, for AO patients has not yet been established. In patients with mild symptoms and/or small calcified plaques, conservative management with clinical and radiologic follow-up is recommended. Surgical options may be problematic because surgery itself is known to be risky for AO. However, in patients with severe and progressive symptoms, surgery is often performed to remove calcified plaques, decompress the spinal cord and nerve roots, marsupialize arachnoid cysts, and restore physiologic subarachnoid cerebrospinal fluid circulation. In the cases reported, only 50% of surgically treated patients improved. Prognosis of AO patients is variable. Some patients develop progressive neurologic worsening, but others have only mild symptoms despite the ossification rate. We add to the scant literature with our peculiar case of a patient harboring combined types II and III AO, who was treated surgically and sustained a significant clinical improvement after surgery.

## CONCLUSION

AS may be a synergistic and independent factor, along with previous surgery, for a spinal trauma and epidural hematoma, triggering the development of AO. To our knowledge, this is the first case of AO in a patient with AS and previous spinal

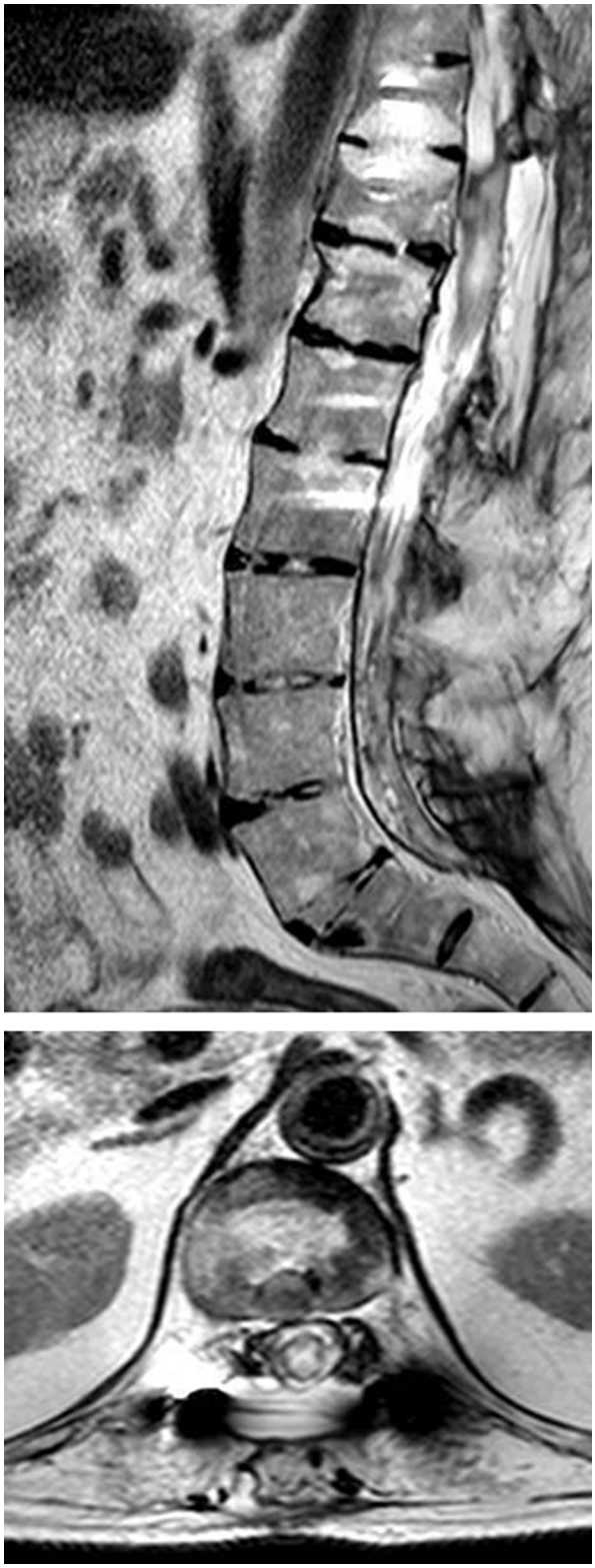

**Figure 5.** Three-month postoperative T2-weighted sagittal and axial magnetic resonance imaging showing a proper decompression of the spinal cord at the thoracolumbar junction.

surgery for a vertebral fracture associated with posttraumatic epidural hematoma that showed both types II and III ossification patterns. We submit that AO should be considered in the differential diagnosis of patients with AS or a history of spinal surgery who present with progressive neurologic deficits.

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