Anterior dural ectasia mimicking a lytic lesion in the posterior vertebral body in ankylosing spondylitis

Case report

Keerthiraj Bele, M.D., D.M.,1 Hima Shrinivas Pendharkar, D.M.R.D., D.N.B., D.M.,1 Easwer Venkat, M.S., M.Ch.,2 and Arun Kumar Gupta, M.D., P.D.C.C.1

Departments of 1Imaging Sciences and Interventional Radiology and 2Neurosurgery, Sree Chitra Tirunal Institute for Medical Sciences and Technology, Trivandrum, India

Anterior dural ectasia is an extremely rare finding in ankylosing spondylitis (AS). The authors describe a unique case of AS in which the patient presented with cauda equina syndrome as well as an unusual imaging finding of erosion of the posterior aspect of the L-1 (predominantly) and L-2 vertebral bodies due to anterior dural ectasia. Symptomatic patients with long-standing AS should be monitored for the presence of dural ectasia, which can be anterior in location, as is demonstrated in the present case. (DOI: 10.3171/2011.8.SPINE1142)

Key words • ankylosing spondylitis • anterior dural ectasia • magnetic resonance imaging • cauda equina syndrome • lumbar spine

A nkylosing spondylitis is a seronegative spondyloarthropathy predominantly affecting the axial skeleton.17 Neurological symptoms are uncommon and any associated neurological deficit, pain, or spinal instability are usually caused by spinal fractures, rotary instability, progressive spinal deformity, and spinal canal stenosis.11 Lumbar radiculopathy in AS usually manifests as cauda equina syndrome.17 Posterior dural ectasia is an unusual but well-described manifestation of AS.2,4,11,21,26 On the other hand, anterior dural ectasia causing scalloping of the vertebral body posteriorly is very rare.10,21,26

Case Report

This 50-year-old man had back pain 14 years ago that was diagnosed as AS based on established diagnostic criteria. Currently, he presented to us with weakness of the right lower limb for the past 6 months, difficulty in getting up from a squatting position, and difficulty in climbing stairs for the past 5 months. There was no bowel or bladder dysfunction. On physical examination there were no neurocutaneous markers or hyperextensible joints. Postural changes due to AS were noted. He could walk with support. A neurological examination revealed subtle wasting of the muscles of the lower one-third of the right leg. Tone was normal in the right lower limb. Power in the right lower limb was 3/5 proximally, 0/5 in the dorsiflexors, and 1/5 in the plantar flexors of the right ankle. Knee jerk and ankle jerk reflexes were +1 on the right. A Babinski response was not obtained. Sensation to touch and pinprick was impaired in the right L5–S1 dermatomal distribution. These findings are consistent with right-sided L5–S1 (predominantly) radiculopathy with some proximal extension.

Plain radiographs of the lumbosacral spine revealed features of AS (Fig. 1). Computed tomography scanning of the lumbar spine revealed a well-defined lytic lesion causing scalloping of the posterior margin of the L-1 (predominantly; Fig. 2A) and the L-2 (minimally) vertebral bodies (not shown). Magnetic resonance imaging

Abbreviations used in this paper: AS = ankylosing spondylitis; CES = cauda equina syndrome; LPS = lumboperitoneal shunting.
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Fig. 1. Plain radiographs of the dorsolumbar spine, anteroposterior (left) and lateral (right) views, showing the classic features of AS with fusion of all spinal ligaments, giving the appearance of a “bamboo spine.” Mild lumbar scoliosis with convexity to the right is apparent.

Fig. 2. A: Bone window CT scan obtained at the L-1 level, showing a smoothly margined lytic lesion eroding the posterior vertebral border with extension into the body. Posterior spinal elements appear normal. B: Axial T2-weighted MR image corresponding to the image in panel A and showing the lesion to be hyperintense, that is, cystic. Note the fine septae extending anteriorly from the posterior margin of the lesion. Also note the forward displacement of nerve roots in the thecal sac and adherence to the dural diverticulum. C: Axial T2-weighted MR image obtained at a lower level, showing the septated cystic lesion in the dorsal part of the vertebral body with displacement of nerve roots into it.

Discussion

Ankylosing spondylitis is a seronegative spondyloarthropathy that characteristically affects the spine and sacroiliac joints. It often presents with low-back pain and progressive limitation of spinal movements. Neurological complications are uncommon and usually caused by atlantoaxial subluxation and compression of the spinal cord. Cauda equina syndrome is a late and poorly understood sequela of AS. The time interval between the onset of AS and the clinical manifestation of CES averages 35 years (range 17–53 years). Spinal instability,
The presence of dural ectasia and neurological symptoms in association with AS is an infrequent feature of spondylitis. Nerve roots are damaged in AS due to arachnoiditis and/or compression by expansive arachnoid diverticula, with the spread of inflammation from involved ligaments (local enthesitis) to meninges resulting in the local inflammation of arachnoid membranes with subsequent nerve root inflammation, degeneration, fibrosis, adhesion, and tethering. Inflammation and arachnoid diverticula establish a slowly progressive process resulting in bony erosion. Hauge described fibrous tissue adhering the cauda equina nerve roots to the dura mater intraoperatively in 1 patient. There is a window of opportunity for surgical treatment, but once the atrophy and fibrosis are established, any treatment would be ineffective. Another theory suggests that reduced compliance of the caudal sac and transmission of the CSF pulsations to surrounding structures result in diverticula formation and bony erosion.

Unlike other conditions that result in dural ectasia, erosions in AS are unique in that they occur in the posterior elements rather than the vertebral bodies. The structural changes primarily affect the lumbar region but can also appear in the thoracic region. When seen in the cervical spine, the pressure effects will be less compared with those in the lumbar region, and hence diverticula formation and bony pressure erosion are unlikely to occur.

Radiographs of the spine and sacrum are usually the initial images that reveal the lesion. Computed tomography and MR imaging are complementary imaging techniques in the characterization of these lesions. Myelography—conventional or CT guided—should be avoided as lumbar puncture in this condition can be technically difficult and hazardous because of bony ankylosis and ossification of the spinal ligaments.

Our case showed unique imaging features: anterior dural ectasia—the lytic lesion involving the L-1 (predominantly) and L-2 posterior vertebral bodies—in contrast to the posterior element involvement described in the literature. Magnetic resonance imaging and MR myelography confirmed the abnormal course of the nerve roots accounting for the cauda equina symptoms. While the radiographic findings of anterior dural ectasia in this patient are striking, it is not possible to be absolutely certain that these changes are a direct cause of the radicular symptoms. However, there is no other evidence of nerve root compression on MR imaging. Note that bladder and bowel symptoms, which are usually present in CES, are unusually absent in the featured case.

Dural ectasia is also seen in Marfan syndrome, Ehlers-Danlos syndrome, and neurofibromatosis. The dural ectasia in Marfan syndrome leads to generalized expansion of the thecal sac and nerve root sleeves, whereas that in Ehlers-Danlos syndrome and neurofibromatosis results in generalized thecal sac enlargement with or without root sleeve expansion. These features contrast with the multiple outpouchings or diverticula of the dura that are seen in AS. Similarly, the presence of posterior dural diverticula, as described in earlier case studies, is characteristic of AS. The MR imaging studies obtained in our patient 10 years ago showed no evidence of vertebral body erosion, thus ruling out congenital anterior meningocele and pointing more toward an acquired pathology. The possibility of a benign bony lesion, such as a giant cell tumor or an aneurysmal bone cyst, is unlikely because they would probably displace the nerve roots away from the bony lesion given their expansive nature.

Interestingly, in our case, erosions involved the vertebral bodies and not the posterior elements. To the best of our knowledge, to date only 1 case report by Ginsburg et al. documents findings similar to those in our case.

Various authors have reported different treatment options for posterior dural ectasia thought to be an end result of chronic inflammation. In their meta-analysis, Ahn et al. suggested that steroids are not effective in the chronic stage, whereas nonsteroidal antiinflammatory drugs decrease back pain but do not improve neurological deficits, probably because no active inflammation occurs in the chronic stage of CES–AS syndrome. Infliximab is a monoclonal antibody to tumor necrosis factor that is used for the treatment of active AS. Treatment with infliximab

**Fig. 3.** Left: Sagittal T2-weighted MR image of the lumbar spine showing the cystic ectatic lesion occupying the posterior third of the L-1 body. Another similar lesion is visible at the posterior superior corner of L-2. Note again the forward displacement of the cauda equina nerve roots into the lesion and adherence of the nerve roots to the wall of the diverticulum (black arrow). Also note a Romanus lesion at the T10–11 level. The lumbar intervertebral discs show degenerative changes. Right: Magnetic resonance myelogram showing continuity of the lesion with the thecal sac, suggesting anterior dural ectasia.
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has reportedly resulted in significant clinical improvement. Surgical options for CES include decompressive laminectomy, detethering of the cord, and LPS. Decompressive laminectomy provides more space for the neural elements at the cauda equina. In our case, however, this procedure was not favored because there was no radiological evidence of stenosis or compression. The conus medullaris was at the L-1 level but was tethered to the anterior aspect of the spinal canal and to the arachnoid sac. Hence, we considered releasing the arachnoid adhesions and fibrosis around the cauda equina roots; however, such release might cause neurological deterioration due to handling of the neural elements. Liu et al. performed detethering in their case, but the patient had no significant clinical improvement. Lumboperitoneal shunting helps in relieving the intradural pressure and has been hitherto offered for the more common posterior diverticulum in AS. Dinichert et al. documented the benefit of LPS in patients with AS presenting with progressive CES. The series by Ea et al. further emphasized the benefit of LPS in patients with AS and CES, with significant clinical improvement that persisted at follow-up. Although our case had an anterior dural diverticulum unlike previously reported surgically treated patients, we offered to perform LPS, as the pathogenesis for an anterior and posterior dural diverticulum remains the same. The patient requested some time for his decision but was eventually lost to follow-up. The best surgical option for an anterior dural diverticulum remains a matter of speculation.

Conclusions

Anterior dural ectasia is an unusual manifestation of AS presenting as CES due to extension of the nerve roots into the lesion with adherence of the nerve roots to the dura mater. Thus, anterior dural ectasia should be considered in any patient with AS and focal erosive lesions of the posterior aspect of the vertebral bodies, who presents with cauda equina symptoms. Treatment of this unusual entity remains speculative, however.

Disclosure

The authors report no conflict of interest concerning the materials or methods used in this study or the findings specified in this paper.

Author contributions to the study and manuscript preparation include the following. Conception and design: Pendharkar. Acquisition of data: Bele. Analysis and interpretation of data: Pendharkar. Drafting the article: Bele. Critically revising the article: all authors. Reviewed submitted version of manuscript: all authors. Approved the final version of the manuscript on behalf of all authors: Pendharkar. Study supervision: Gupta. Case management: Venkat.

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