



# Epidural Hematoma Associated with Spinal Fracture in Diffuse Idiopathic Skeletal Hyperostosis

## *Difüz İdiyopatik İskelet Hiperostozunda Spinal Kırıkla İlişkili Epidural Hematom*

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### ABSTRACT

The authors describe the clinical findings, radiographic appearance and surgical treatment of a spinal epidural hematoma (SEH) associated with spinal fracture in diffuse idiopathic skeletal hyperostosis (DISH). An 81-year-old male patient injured after falling from a 2.5 m tree presented with severe back pain. On plain radiographs and CT images, DISH with anterolateral osteophytes and fused thoracolumbar vertebrae was found along with a T12 fracture. Patient was initially treated with bed rest followed by placement into a body cast. Three weeks later, he presented with incomplete paraplegia of his lower limbs. CT images did not reveal any fracture displacement, but MRI images showed an epidural hematoma compressing the dura mater. The patient was successfully treated by posterior fixation surgery using pedicle screws and rod with vertebroplasty. We aimed to report a unique case of a SEH complicating a spinal fracture in DISH.

**KEYWORDS:** Spinal epidural hematoma, Spinal fracture, Diffuse idiopathic skeletal hyperostosis

### ÖZ

Yazarlar difüz idiyopatik iskelet hiperostozunda spinal kırıkla ilişkili spinal epidural hematomun klinik bulguları, radyografik görünümü ve cerrahi tedavisini tanımlamaktadır. 81 yaşında bir erkek hasta 2,5 metre yüksekliğinde bir ağaçtan düştükten sonra şiddetli bel ağrısıyla geldi. Röntgen filmleri ve BT görüntülerde T12 kırığıyla birlikte anterolateral osteofitler ve torakolumbar vertebra füzyonuyla difüz idiyopatik iskelet hiperostozu görüldü. Hasta başlangıçta bir yatak istirahatıyla tedavi edildi ve bir vücut alçısına kondu. Üç hafta sonra alt uzuvların tam olmayan paralejisi ile geldi. BT görüntüleri herhangi bir kırık displasmanı göstermedi ama MRG görüntülerinde dura materi sıkıştıran bir epidural hematom vardı. Hasta vertebroplastiyle pedikül vidaları ve çubuk kullanılarak posterior fiksasyon cerrahisiyle başarılı şekilde tedavi edildi. Difüz idiyopatik iskelet hiperostozunda spinal kırık komplikasyonu olarak spinal epidural hematom şeklinde benzersiz bir olguyu bildiriyoruz.

**ANAHTAR SÖZCÜKLER:** Spinal epidural hematom, Spinal kırık, Difüz idiyopatik iskelet hiperostozu

### INTRODUCTION

Diffuse idiopathic skeletal hyperostosis (DISH), also called ankylosing spinal hyperostosis, has been well recognized as a cause of spinal ankylosis (6,11). Spinal fractures that occur with DISH are unstable: the ankylosed spine, with its osteophytes and ossification of ligaments and discs, acts as a long lever arm which concentrates stress and deformity at the site of injury. So when fractures with DISH occur, this mechanical instability increases the incidence of neurological deficit (2,7). Although spinal epidural hematoma (SEH) has been associated with ankylosing spondylitis (AS) (5,12-14), there have been only a few reports of SEH associated with a spinal fracture in DISH (3,4,9). We report on a patient who developed incomplete paraplegia due to the SEH with spinal fracture in DISH.

### CASE REPORT

An 81-year-old male patient presented with severe back pain

after falling from a 2.5 m tree. Plain radiographs revealed DISH with anterolateral osteophytes and fused thoracolumbar vertebrae and a T12 fracture (Figure 1). CT images also showed a fracture line in the ossified portion of the supraspinous ligament (Figure 2). On the MRI images, the fracture line through the vertebral body was horizontal and traveled from the anterior to the posterior margin of the vertebral body, and also to the ossification of the supraspinous ligament (Figure 3).

The initial management was bed rest for eight days. The patient was then allowed to mobilize with body cast and rehabilitation was started. Three weeks later, he presented with incomplete paraplegia of his lower limbs, and he could not walk. Although the fracture was not displaced on CT images, MRI images showed a SEH compressing the dura mater and causing the paraplegia (Figure 4A,B). Decision to proceed to surgery was made to prevent further neurological deterioration. Under general anesthesia, the patient was

placed in the prone position with a Hall frame. A posterior midline incision was made from the level of Th10 to L2. We performed a laminectomy from T11 to T12 and found a dorsal epidural hematoma at these levels compressing the spinal cord. The hematoma was strongly adherent to the dura mater, making it difficult to remove completely. We were able to partially resect it and confirm decompression by ultrasonography. We performed posterior spinal fixation using instrumentation by placing pedicle screws bilaterally into T10, T11, L1 and L2. In addition, we performed vertebroplasty using HA blocks in the T12 vertebra. We fixed the pedicle screws and rod with posterolateral fusion using autogenous iliac crest and local bone graft (Figure 5A,B). Postoperatively,

the patient was immobilized for three months with a hard corset which was changed to a soft corset for another three months. One year postoperatively, bony union had been achieved and muscle strength of his lower limbs recovered. The patient could walk with a cane.

### DISCUSSION

Forestier et al. first introduced the term “senile ankylosing hyperostosis of the spine” and described the characteristic roentgen features in the spine: involvement of the anterior and right lateral aspect of the thoracolumbar region, undulating calcification and ossification and cortical hyperostosis (6). However, since these patients also have extraspinal manifestations with ossification of other joints, Resnick et al. proposed the name “diffuse idiopathic skeletal hyperostosis (DISH)” for this skeletal disorder that produces characteristic alterations in both spinal and extraspinal structures (11).

Several studies reported that minor trauma could cause a fracture of the ankylosed spine, which easily becomes unstable due to the inflexibility of the anteriorly- and posteriorly-fused spine. These lengths of fused spine simulate long, rigid lever arms that cause a concentration of force at the site of injury (1,2,7). Hyperextension leads to an anterior distraction of the vertebral body that fractures transversely to posterior compression resulting in posterior element injuries (2,8).

SEH is an uncommon entity, which may be caused by traumatic or nontraumatic conditions that include coagulopathy, vascular lesions and iatrogenic or idiopathic causes. Although



**Figure 1:** Initial plain radiographs showing the ankylosed spine and the fracture line of T12.



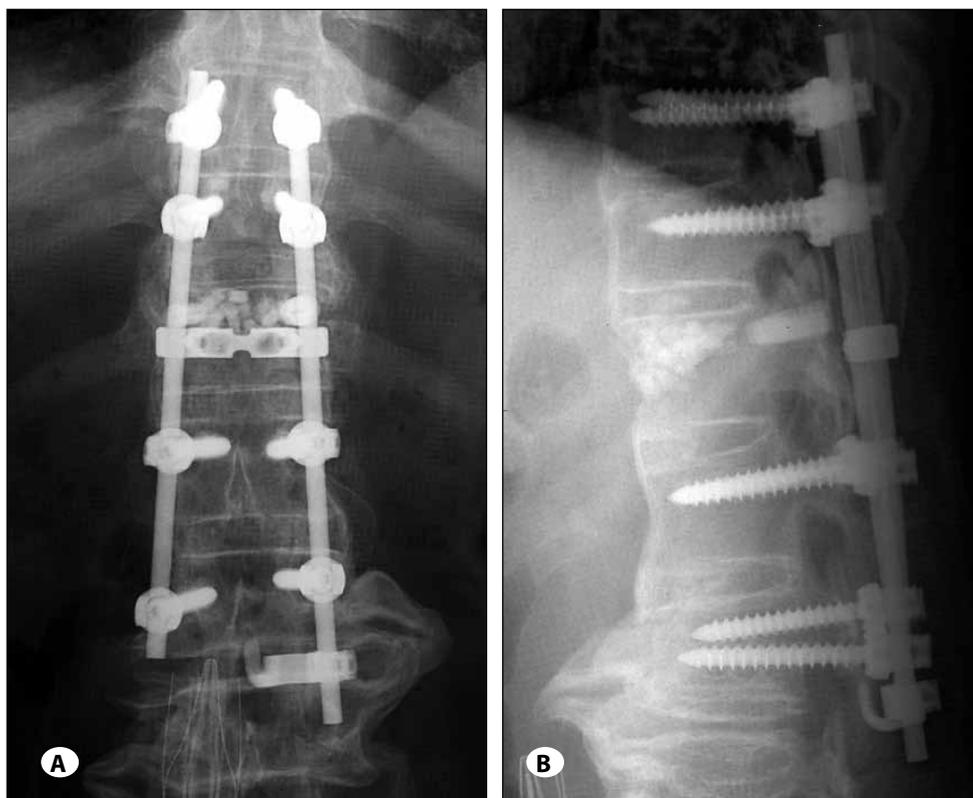
**Figure 2:** Sagittal image on CT scan also showing a fracture line in the ossified part of the supraspinous ligament.



**Figure 3:** MRI T2-weighted image showing the fracture line traveling from the anterior part to the ossified part of the supraspinous ligament.



**Figure 4:** MRI T2-weighted images (**A:** sagittal image. **B:** axial image) showing the epidural hematoma compressing the dura mater 3 weeks after placing the cast.



**Figure 5:** Postoperative anteroposterior (**A**) and lateral (**B**) plain radiographs. We performed posterior spinal fixation using instrumentation with vertebroplasty.

the risks of SEH associated with AS are well established in the literature (5,12-14), only a few cases of SEH associated with DISH have been reported (3,4,9). Caron et al. mentioned that SEH was induced by the unstable nature of the fracture and the hyperemic tissues in the area (3). The causes of our DISH patient's SEH were likely the injured epidural soft tissues by the unstable fractured bone or the fractured bone itself.

For fractures occurring with DISH, surgical treatment is recommended when it is significantly displaced and unstable. On the other hand, conservative treatment such as bed rest, plaster or halo vests can be used for the more stable, minimally displaced fractures (10). Whang et al. reported that most surgical repairs for fractures occurring with DISH have been performed through a posterior approach. However, when there is significantly comminuted fracture of the vertebral column or where there is a characteristic "fish-mouth" defect indicating pseudoarthrosis, an anterior procedure may be necessary<sup>13</sup>. We performed posterior spinal fixation with vertebroplasty to achieve anterior support because this patient had a "fish-mouth" defect; consequently, the patient made a near complete recovery after surgery and could walk. We employed the vertebroplasty along with posterior spinal fixation to achieve anterior support to avoid an anterior approach in elderly patients with multiple comorbidities.

### CONCLUSION

We report a case of delayed presentation of SEH with subsequent surgical treatment in a patient with DISH. Although SEH rarely occurs with a spinal fracture in DISH, we should have a clear understanding of this disease entity; an immediate MRI upon any change in neurological status to confirm the diagnosis with prompt surgery to avoid permanent neurological deficit.

### REFERENCES

- Bernini PM, Floman Y, Marvel JP Jr, Rothman RH: Multiple thoracic spine fractures complicating ankylosing hyperostosis of the spine. *J Trauma* 21:811-814, 1981
- Burkus JK, Denis F: Hyperextension injuries of the thoracic spine in diffuse idiopathic skeletal hyperostosis. Report of four cases. *J Bone Joint Surg Am* 76:237-243, 1994
- Caron T, Bransford R, Nguyen Q, Agel J, Chapman J, Bellabarba C: Spine fractures in patients with ankylosing spinal disorders. *Spine (Phila Pa 1976)* 35:E458-464, 2010
- de Peretti F, Sane JC, Dran G, Razafindratsiva C, Argenson C: Ankylosed spine fractures with spondylitis or diffuse idiopathic skeletal hyperostosis: Diagnosis and complications. *Rev Chir Orthop Reparatrice Appar Mot* 90:456-465, 2004
- Elgafy H, Bransford RJ, Chapman JR: Epidural hematoma associated with occult fracture in ankylosing spondylitis patient: A case report and review of the literature. *J Spinal Disord Tech* 24:469-473, 2011
- Forestier J, Lagier R: Ankylosing hyperostosis of the spine. *Clin Orthop Relat Res* 74:65-83, 1971
- Hendrix RW, Melany M, Miller F, Rogers LF: Fracture of the spine in patients with ankylosis due to diffuse skeletal hyperostosis: Clinical and imaging findings. *AJR Am J Roentgenol* 162: 899-904, 1994
- Le Hir PX, Sautet A, Le Gars L, Zeitoun F, Tubiana JM, Arrive L, Laredo JD: Hyperextension vertebral body fractures in diffuse idiopathic skeletal hyperostosis: A cause of intravertebral fluidlike collections on MR imaging. *AJR Am J Roentgenol* 173:1679-1683, 1999
- Michel JL, Souteyrand AC, Kabre M, Dubost JJ, Soubrier M, Ristori JM: Fractures of the ankylosed spine: MRI features. *J Radiol* 88:1703-1706, 2007
- Paley D, Schwartz M, Cooper P, Harris WR, Levine AM: Fractures of the spine in diffuse idiopathic skeletal hyperostosis. *Clin Orthop Relat Res*:22-32, 1991
- Resnick D, Niwayama G: Radiographic and pathologic features of spinal involvement in diffuse idiopathic skeletal hyperostosis (DISH). *Radiology* 119:559-568, 1976
- Westerveld LA, Verlaan JJ, Oner FC: Spinal fractures in patients with ankylosing spinal disorders: A systematic review of the literature on treatment, neurological status and complications. *Eur Spine J* 18:145-156, 2009
- Whang PG, Goldberg G, Lawrence JP, Hong J, Harrop JS, Anderson DG, Albert TJ, Vaccaro AR: The management of spinal injuries in patients with ankylosing spondylitis or diffuse idiopathic skeletal hyperostosis: A comparison of treatment methods and clinical outcomes. *J Spinal Disord Tech* 22:77-85, 2009
- Wu CT, Lee ST: Spinal epidural hematoma and ankylosing spondylitis: Case report and review of the literature. *J Trauma* 44:558-561, 1998