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Progressive diffuse idiopathic spinal hyperostosis: a case report

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Abstract

Diffuse idiopathic spinal hyperostosis (DISH) causes various problems, such as adjacent disc dysfunction, pseudarthrosis, or reossification, when spinal surgery is performed or spinal fracture occurs. The authors herein describe a patient with DISH in whom ossification of the anterior longitudinal ligament progressively advanced from the thoracic to sacral vertebrae during a 14-year period. Surgery was performed three times to treat the characteristic problems associated with DISH: abnormal sagittal alignment of the spine, ossification of the posterior longitudinal ligament, reossification of the laminectomy-treated regions and accompanying spinal canal stenosis, pseudarthrosis after spinal fracture, and delayed palsy. DISH-associated problems after spinal fusion are not rare, but this patient developed a particularly large number of problems often seen in the long term after spinal fusion in patients with DISH. Clinicians must be aware of problems being likely to occur in ossification-discontinuous and fragile regions during the postoperative follow-up.

Keywords

Diffuse idiopathic hyperostosis, lumbar operation, complication, ossification of ligament, sagittal vertical axis, thoracic kyphosis, lumbar lordosis, fracture, pseudarthrosis, delayed palsy

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Introduction

Diffuse idiopathic spinal hyperostosis (DISH), previously also termed Forestier disease and ankylosing spinal hyperostosis,[1] is characterized by advancement of ossification of the anterior longitudinal ligament

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until continuity of the spine and reduced spinal mobility occur. Although the disease slowly progresses, mobility of the sacroiliac joint and major joints of the extremities are retained and daily life is generally not markedly affected.[2] However, DISH causes various problems when spinal surgery is performed[3,4] or spinal fracture occurs.[1,5–7] We herein describe a patient in whom ossification of the anterior longitudinal ligament advanced and became continuous from the thoracic to sacral vertebrae during a 14-year period, causing various problems, and three surgeries were required during this period.

DISH-associated problems after spinal fusion are not rare and have been extensively analyzed.[2–7] However, few case reports have described the long-term follow-up. The present case illustrates the development of a large number of problems during the long-term follow-up after spinal fusion in a patient with DISH.

Case report

This study was conducted in conformity with the ethical guidelines for clinical application of human subjects established by the Japanese Ministry of Health, Labour and Welfare. Nihon University Itabashi Hospital does not require institutional review board approval for case reports. Written informed consent was obtained from the patient.

A 48-year-old woman presented to our hospital for evaluation of lower back pain, 15-minute intermittent claudication, and numbness of the bilateral lower limbs (Figure 1(a)–(c)). She was diagnosed with lumbar degenerative spondylolisthesis, and surgery was performed. Laminectomy of L4/L5 and posterior lumbar interbody fusion were applied, and her symptoms were resolved after surgery. No ossification or bone bridge formation was noted in the anterior longitudinal ligament at the

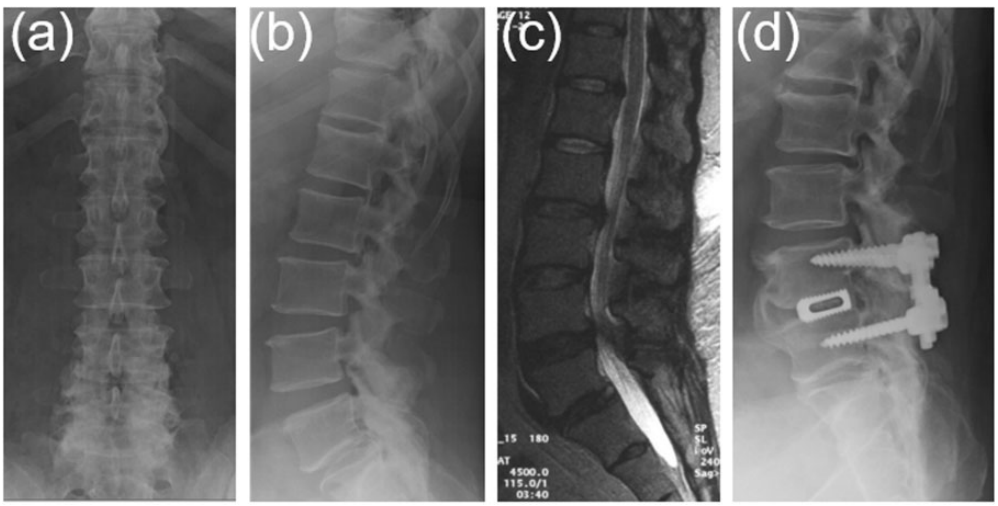

Figure 1. Imaging before and after the initial surgery. (a) Anteroposterior view of the lumbar spine on a plain radiograph. (b) Lateral view of the lumbar spine on a plain radiograph. No ossification or bone bridge formation was noted in the lumbar anterior longitudinal ligament. (c) Sagittal view of the lumbar spine on plain T2-weighted magnetic resonance imaging. Spinal canal stenosis was shown at the L4/L5 level. (d) Lateral view of the lumbar spine on a plain radiograph 2 years after surgery. Bone bridge formation was noted at L4/L5 after posterior lumbar interbody fusion of L4/L5.

lumbar level before surgery (Figure 1(b)). Bone bridge formation at L4/L5 was noted 2 years after surgery (Figure 1(d)).

Numbness of the bilateral lower limbs and 2-minute intermittent claudication recurred about 6 years postoperatively. Progression of spinal canal stenosis was noted at L2/L3 and L3/L4 (Figure 2(b)–(e)), for which decompression and fusion of L2–L4 was additionally performed (Figure 2(f)). At this time point, ossification and bone bridge formation of the anterior longitudinal ligament had extended from the thoracic vertebrae to L1 in the lateral view of a plain radiograph of the lumbar spine, and DISH was diagnosed based on the Resnick diagnostic criteria[1] (Figure 2 (a)). Incomplete bone bridge formation was also noted in the anterior longitudinal ligament at L1/L2. After the reoperation, the symptoms remitted and the patient returned to work.

She fell on her backside 7 years after the reoperation and presented for evaluation of persistent lower back pain. The L5 vertebral body, which was the lowest end of the fused region, was fractured. Surgical treatment was recommended, but conservative treatment was performed at the patient's request. The lower back pain remained 6 months after the injury, and numbness developed in the bilateral lower limbs. Bone fusion of the fractured region could not be achieved, and pseudarthrosis developed (Figure 3(a)). Continuous ossification and bone bridge formation of the anterior longitudinal ligament were noted on the cranial side of the L5 vertebral body and at L5/S1. On computed tomography, continuous ossification and bone bridge formation of the anterior longitudinal ligament were noted from the thoracic vertebrae to the cranial and caudal sides of the L5 fracture region, and L5 exhibited

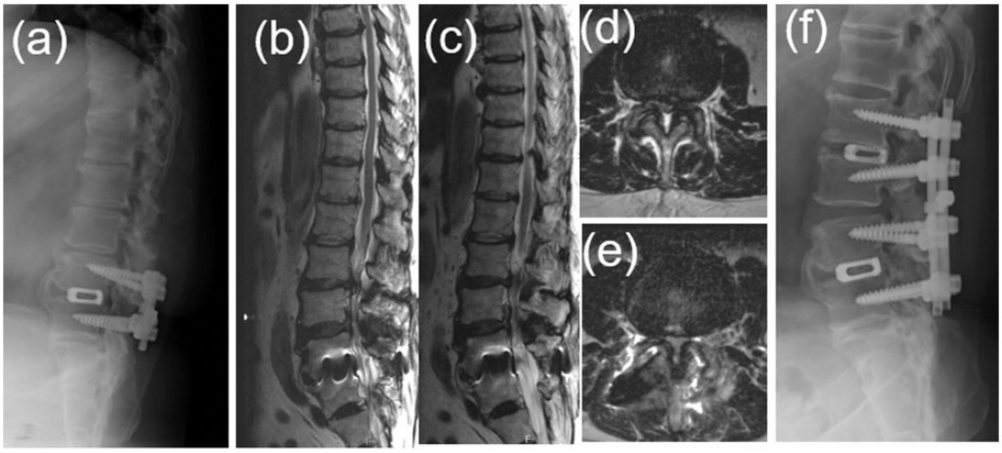

Figure 2. Imaging before and after the second surgery. (a) Lateral view of the lumbar spine on a plain radiograph 6 years after the initial surgery. Ossification and bone bridge formation of the anterior longitudinal ligament from the thoracic vertebra to L1 were observed. (b, c) Sagittal view of the lumbar spine on plain T2-weighted magnetic resonance imaging (MRI) 6 years after the initial surgery. Progression of spinal canal stenosis was noted at L2/L3 and L3/L4. (d) Axial view of the lumbar spine on plain T2-weighted MRI 6 years after the initial surgery. Circumferential stenosis was noted at the L2/L3 level. (e) Axial view of the lumbar spine on plain T2-weighted MRI 6 years after the initial surgery. Circumferential stenosis was noted at the L3/L4 level. (f) Lateral view of the lumbar spine on a plain radiograph after reoperation. Decompression and fusion of L2–L4 were applied.

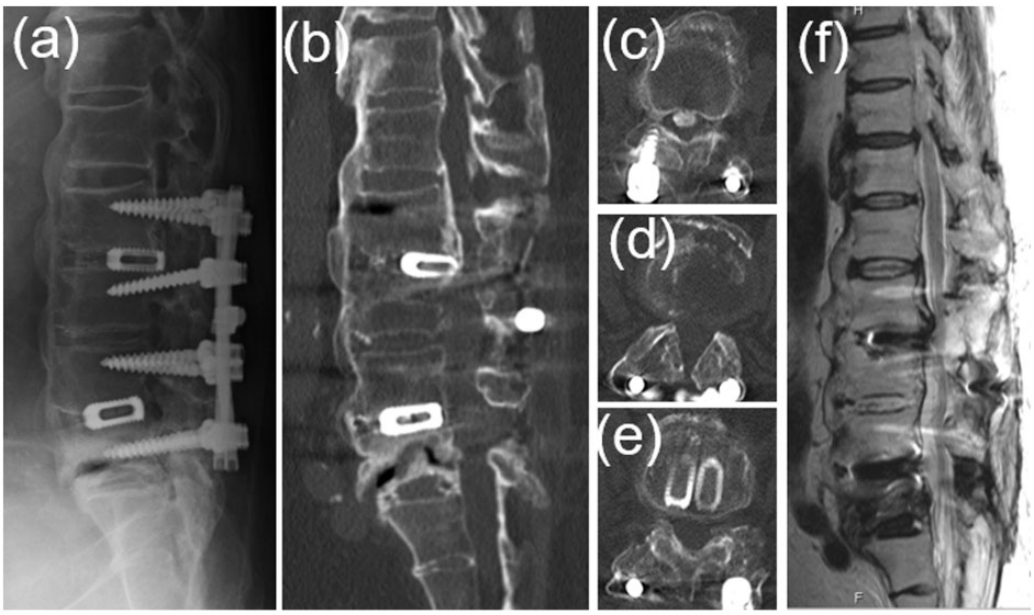

Figure 3. Imaging before the third surgery. (a) Vertebral body fracture of L5 became a pseudarthrosis 7.5 years after reoperation (6 months after the patient fell on her backside). Continuous ossification and bone bridge formation of the anterior longitudinal ligament were noted at L5/S1 (cranial and caudal sides of the L5 fracture region, respectively). (b) Sagittal view on computed tomography (CT). Continuous ossification and bone bridge formation of the anterior longitudinal ligament from the thoracic vertebrae to the cranial and caudal sides of the L5 fracture region were observed, and the L5 vertebral body exhibited pseudarthrosis. Ossification of the posterior longitudinal ligament (OPLL) was noted from L1/L2 to L2/L3, and the laminectomy-treated regions were ossified posterior to the spinal canal. (c) Transverse view on CT. OPLL was observed at the L2/L3 level. (d) Transverse view on CT. Reossification of the laminectomy-treated region was noted at the L3/L4 level. (e) Transverse view on CT. Reossification of the laminectomy-treated region was noted at the L4/L5 level. (f) Sagittal view on T2-weighted magnetic resonance imaging. Spinal canal stenosis caused by the ossified lesions anterior and posterior to the spinal canal was observed at L2/L3.

pseudarthrosis (Figure 3(b)). Moreover, ossification of the posterior longitudinal ligament was noted from L1/L2 to L2/L3 (Figure 3(b), (c)). Reossification of the laminectomy-treated regions posterior to the spinal canal was also noted, and the largest ossification was observed at L2/L3 (Figure 3(b)–(e)). Magnetic resonance imaging showed spinal canal stenosis by the ossified lesions anterior and posterior to the spinal canal at L2/L3.

Resection of the ossified laminectomy-treated regions at L2/L3 to L4/L5 and additional posterior fusion from L4 to the iliac bones were performed (Figure 4). The patient's lower back pain and gait disturbance improved after surgery. Retrospectively, ossification and bone bridge formation of the anterior longitudinal ligament at T5–T11 could be confirmed from the initial examination (Figure 5(a)) to 1 year after the initial surgery (Figure 5(b)), but these changes were absent in the lumbar and sacral vertebrae. Six years after the initial surgery, ossification on the cranial side had advanced to T5–L1, and that on the caudal side had advanced to L4–S1 (Figure 5(c)). Thirteen years after the initial surgery (7 years after the reoperation), ossification and bone bridge formation of the

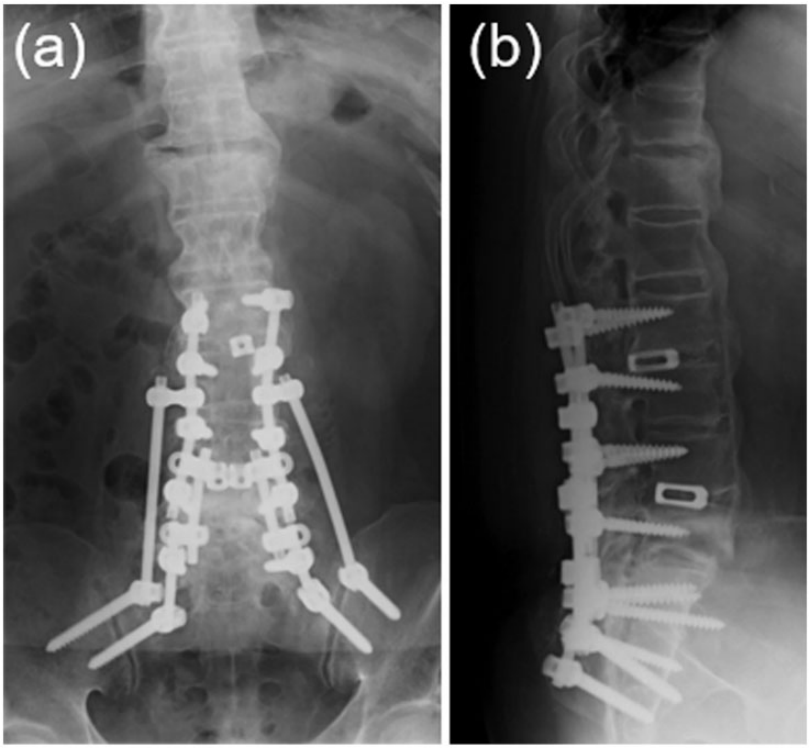

Figure 4. Postoperative plain radiograph after the third operation. (a) Anteroposterior view of the lumbar spine after the third surgery. Resection of the ossified laminectomy-treated regions at L2/L3 to L4/L5 and additional posterior fixation of L4–iliac bone were performed. (b) Lateral view of the lumbar spine on a plain radiograph after the third surgery.

thoracic and lumbar anterior longitudinal ligament had become continuous from the thoracic to sacral vertebrae (Figure 5(d)). During this period, the sagittal vertical axis[8] had become aggravated from 65.0 mm at the first examination to 120.0, 168.9, and 180.8 mm at 1, 6, and 13 years after the initial surgery, respectively. The thoracic kyphosis angle (angle between the superior endplate of T5 and inferior endplate of T12)[8] also increased from 23.0° to 37.1° at 6 years and to 37.4° at 13 years. The lumbar lordosis (angle between the superior endplate of L1 and superior endplate of S1) also changed after surgery, with angles of 42.0° before surgery and 27.0°, 0.6°, and 10.2° at 1, 6, and 13 years after the initial operation, respectively (Table 1).

Discussion

In patients with DISH, characteristic bone growth occurs in the spine and regions other than the spine. DISH has also been termed ankylosing spinal hyperostosis or Forestier disease, and its pathology differs from that of ossification of the posterior longitudinal ligament and ligamentum flavum and ankylosing spondylitis. The diagnostic criteria of this disease are based only on imaging findings, and normally, the following three Resnick diagnostic criteria must be met: (1) the presence of flowing calcification and ossification along the anterolateral aspects of at least four contiguous vertebral bodies with or without associated localized pointed excrescences at the

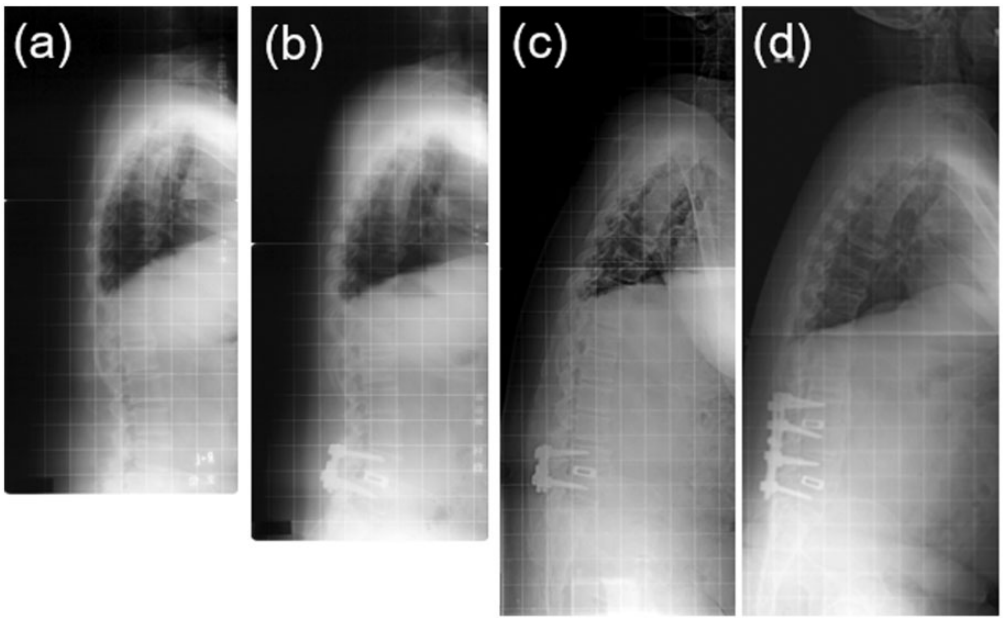

Figure 5. The course of the lateral view of the lumbar spine on plain radiographs in the standing position. (a) Before the initial surgery. (b) One year after the initial surgery. (c) Six years after the initial surgery. (d) Thirteen years after the initial surgery (7 years after the second operation).

Table 1. Ossification of the anterior longitudinal ligament, bony bridge formation, and spinopelvic parameters.

	Before operation	1 year after operation	6 years after initial operation	7 years after reoperation
Bridge of OALL	T5–T11	T5–T11	T9–L1 L4–S1	T5–S1
SVA (mm)	65.0	120.0	168.9	180.8
Thoracic kyphosis (°)	23.0	20.0	37.1	37.4
Lumbar lordosis (°)	42.0	27.0	0.6	10.2
Pelvic incidence (°)	N/A	N/A	53.3	54.8
Pelvic tilt (°)	N/A	N/A	36.3	36.3
Sacral slope (°)	N/A	N/A	17.1	18.5

OALL, ossification of the anterior longitudinal ligament; SVA, sagittal vertical axis; Thoracic kyphosis, angle between superior endplate of T5 and inferior endplate of T12; Lumbar lordosis, angle between superior endplate of L1 and superior endplate of S1; Pelvic incidence, angle between the line perpendicular to the sacral plate at its midpoint and the line connecting this point to the axis of the femoral heads; Pelvic tilt, angle between the vertical line and the line through the midpoint of the sacral plate to the axis of the femoral heads; Sacral slope, angle between a horizontal line and the sacral plate.

intervening vertebral body–disc junctions; (2) relative preservation of the intervertebral disc height in the involved vertebral segments and the absence of extensive radiographic changes of "degenerative" disc disease, including vacuum phenomena and vertebral body marginal sclerosis; and (3) absence of apophyseal joint bony

ankylosis and sacroiliac joint erosion, sclerosis, or bony fusion.

DISH does not clearly cause clinical symptoms other than reduction of spinal mobility in many cases, and most patients have a normal daily life. When fixation of the spine is necessary because of degenerative disease or spinal injury, investigation of a surgical procedure in consideration of the specific morphology and mechanical environment is necessary.[3,4,9] Because the pathology is associated with a poor outcome of surgical spinal fusion in many cases, the disease has recently been attracting attention and has been investigated from various aspects. Metabolic abnormalities and genetic and environmental factors have been proposed as contributors to DISH, but its cause remains unclear. Changes in the overall spinal alignment due to an increase in the thoracic kyphosis angle and a decrease in the lumbar lordosis angle,[4,5] ossification of the other ligaments around the spinal canal and spinal canal stenosis,[10] fracture and pseudarthrosis formation in discontinuous and fragile regions,[11–13] and delayed palsy after fracture[6] have been shown to influence the advancement of ossification and bone bridge formation of the anterior longitudinal ligament.[14]

We performed surgery three times during a 14-year period in this patient. Anterior longitudinal ligament ossification rapidly progressed during this period, and ossification and bone bridge formation were noted in almost the entire anterior longitudinal ligament from the thoracic to lumbar and sacral vertebrae. Additionally, the thoracic kyphosis angle increased and the lumbar lordosis decreased, promoting abnormal spinal alignment. Ossification of the ligaments around the spinal canal other than the anterior longitudinal ligament was also noted: ossification of the posterior longitudinal ligament and reossification of the laminectomy-treated regions and accompanying spinal canal stenosis developed. Moreover, the fragile region between continuous vertebrae was fractured and pseudarthrosis was formed. The patient experienced many DISH-induced spinal problems throughout the 14 years. This case illustrates that in patients with DISH, ossification and bone bridge formation of the anterior longitudinal ligament may rapidly progress within 14 years, which is a relatively short period. Although abnormal spinal alignment does not further progress in regions with complete bone bridge formed by ossification, clinicians should be aware that the lack of due consideration for fixation without focal lumbar lordosis has a greater effect on global alignment than does the progression of DISH and that problems readily occur in discontinuous and fragile regions. Additionally, treatment should be performed with consideration of alignment of the entire spine.

Conclusions

DISH-associated problems after spinal fusion are not rare, but the present case report illustrates the possibility of long-term development of multiple problems in such patients. In the present case, a patient with DISH developed ossification and bone bridge formation of the thoracic and lumbar anterior longitudinal ligament that advanced and became continuous from the thoracic region to sacrum during a 14-year period. She developed characteristic DISH-associated problems that required three surgeries throughout this time period: abnormal sagittal alignment of the spine, ossification of the posterior longitudinal ligament, reossification of the laminectomy-treated regions, and fragile fracture and pseudarthrosis. During follow-up, clinicians must be aware that a lack of due consideration for fixation without focal lumbar lordosis has a greater effect on global alignment than does the progression of

DISH and that problems are likely to occur in ossification-discontinuous or fragile regions.

Declaration of conflicting interest

The authors declare that there is no conflict of interest.

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References

1. Sarzi-Puttini P and Atzeni F. New developments in our understanding of DISH (diffuse idiopathic skeletal hyperostosis). *Curr Opin Rheumatol* 2004; 16: 287–292.
2. Chi D, Miyamoto K, Hosoe H, et al. Symptomatic lumbar mobile segment with spinal canal stenosis in a fused spine associated with diffused idiopathic skeletal hyperostosis. *Spine J* 2008; 8: 1019–1023. DOI: 10.1016/j.spinee.2007.09.007.
3. Etebar S and Cahill DW. Risk factors for adjacent-segment failure following lumbar fixation with rigid instrumentation for degenerative instability. *J Neurosurg* 1999; 90: 163–169.
4. Otsuki B, Fujibayashi S, Takemoto M, et al. Diffuse idiopathic skeletal hyperostosis (DISH) is a risk factor for further surgery in short-segment lumbar interbody fusion. *Eur Spine J* 2015; 24: 2514–2519. DOI: 10.1007/s00586-014-3603-5.
5. Olivieri I, D'Angelo S, Cutro MS, et al. Diffuse idiopathic skeletal hyperostosis may give the typical postural abnormalities of advanced ankylosing spondylitis. *Rheumatology (Oxford)* 2007; 46: 1709–1711. DOI: 10.1093/rheumatology/kem227.
6. Westerveld LA, Verlaan JJ and 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* 2009; 18: 145–156. DOI: 10.1007/s00586-008-0764-0.
7. Caron T, Bransford R, Nguyen Q, et al. Spine fractures in patients with ankylosing spinal disorders. *Spine* 2010; 35: E458–E464. DOI: 10.1097/BRS.0b013e3181cc764f.
8. Schwab F, Lafage V, Patel A, et al. Sagittal plane considerations and the pelvis in the adult patient. *Spine* 2009; 34: 1828–1833. DOI: 10.1097/BRS.0b013e3181a13c08.
9. Greiner-Perth R, Boehm H, Allam Y, et al. Reoperation rate after instrumented posterior lumbar interbody fusion: a report on 1680 cases. *Spine* 2004; 29: 2516–2520.
10. Sade R, Ulusoy OL, Sirvanci M, et al. Cord compression and myelopathy due to stress fracture in a patient with diffuse idiopathic skeletal hyperostosis (DISH). *Spine J* 2016; 16: e503–e504. DOI: 10.1016/j.spinee.2016.01.022.
11. Hunter T, Forster B and Dvorak M. Ankylosed spines are prone to fracture. *Can Fam Physician* 1995; 41: 1213–1216.
12. Paley D, Schwartz M, Cooper P, et al. Fractures of the spine in diffuse idiopathic skeletal hyperostosis. *Clin Orthop Relat Res* 1991; (267): 22–32.
13. Miyamoto K, Shimizu K, Arimoto R, et al. Spontaneous symptomatic pseudoarthrosis at the T11-T12 intervertebral space with diffuse idiopathic skeletal hyperostosis: a case report. *Spine* 2003; 28: E320–E322. DOI: 10.1097/01.brs.0000083321.62608.ee.
14. Yaniv G, Bader S, Lidar M, et al. The natural course of bridging osteophyte formation in diffuse idiopathic skeletal hyperostosis: retrospective analysis of consecutive CT examinations over 10 years. *Rheumatology (Oxford)* 2014; 53: 1951–1957. DOI: 10.1093/rheumatology/ket335.