Thoracic Vertebral Body Fracture After Total Hip Replacement in Diffuse Idiopathic Skeletal Hyperostosis

MATTHIAS KÖNIGSHAUSEN, MD; MARCEL DUDDA, MD; CHRISTIAN MERLE, MD; THOMAS ARMIN SCHILDHAUER, MD, PHD; TOBIAS FEHMER, MD

abstract

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This article describes the critical clinical sequelae of a patient with diffuse idiopathic skeletal hyperostosis who sustained an acute iatrogenic thoracic vertebral body fracture with subsequent spinal cord injury after a total hip replacement, with a final lethal course.

A 57-year-old woman was referred to the authors’ institution after undergoing a total hip replacement in the supine position for secondary osteoarthritis. Postoperatively, the patient had symptoms of an incomplete paraplegia. Computed tomography scan and magnetic resonance imaging revealed diffuse idiopathic skeletal hyperostosis and an acute unstable fracture of T11 with spinal contusion. A posterior spinal fusion of T10-L1 with laminectomy of T11 was performed immediately on admission. Postoperatively, no improvement of the neurological deficit was observed. After developing multiorgan failure while in intensive care, the patient died 2 months after the total hip replacement.

The morphological and functional symptoms of diffuse idiopathic skeletal hyperostosis with the typical ossification of the longitudinal ligaments and the associated loss of bending forces of the spine were detected postoperatively. This severe case demonstrates that the surgeon must be alert to possible complications due to intraoperative maneuvers in patients with stiffened spinal disorders undergoing total hip replacement.

Drs Königshausen, Dudda, Schildhauer, and Fehmer are from the Department of General and Trauma Surgery, BG Universitätssklinikum Bergmannsheil, Ruhr-Universität Bochum, Bochum, Germany; and Dr Merle is from the Nuffield Department of Orthopaedics, Rheumatology & Musculoskeletal Sciences, Botnar Research Centre, University of Oxford, Oxford, United Kingdom.

Drs Königshausen, Dudda, Merle, Schildhauer, and Fehmer have no relevant financial relationships to disclose.

Correspondence should be addressed to: Matthias Königshausen, MD, Department of General and Trauma Surgery, BG Universitätssklinikum Bergmannsheil, Ruhr-Universität Bochum, Bürkle-de-la-Camp-Platz 1, 44789, Bochum, Germany (m.koenigshausen@web.de).

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Figure: Postoperative sagittal (A) and transverse (B) computed tomography scans of the thoracic spine revealing the unstable hyperextension fracture through the vertebral body of T11. Note the extensive ossification of the spinous processes and the anterior longitudinal ligament in the thoracic spine with the right-sided predominance characteristic for diffuse idiopathic skeletal hyperostosis.
Diffuse idiopathic skeletal hyperostosis is a chronic disease with typical calcification and ossification of spinal ligaments and peripheral entheses, first described in detail in 1950. It affects middle-aged and elderly individuals and is often asymptomatic or associated with mild symptoms. Generally, susceptibility to fractures in stiffened spine disorders such as diffuse idiopathic skeletal hyperostosis or ankylosing spondylitis is a known but often underestimated problem. Most fractures in ankylosing spondylitis are a known but often underestimated problem. Generally, susceptibility to fractures in stiffened spine disorders such as diffuse idiopathic skeletal hyperostosis or ankylosing spondylitis is a known but often underestimated problem.1-3 Most fractures in ankylosing spondylitis are a known but often underestimated problem.4,5 Generally, susceptibility to fractures in stiffened spine disorders such as diffuse idiopathic skeletal hyperostosis or ankylosing spondylitis is a known but often underestimated problem.6,7

This article describes a middle-aged patient with diffuse idiopathic skeletal hyperostosis who sustained an iatrogenic unstable thoracic vertebral body fracture with subsequent paraplegia during total hip replacement. This case demonstrates a severe complication during an elective orthopedic surgery.

**Case Report**

A 57-year-old woman was referred to the authors’ institution after undergoing total hip replacement for secondary osteoarthritis the day before. Because of a varus malformation secondary to a healed proximal femoral fracture that had occurred several years before, a 2-dimensional corrective osteotomy of the femur using a plate osteosynthesis was performed simultaneously with the total hip replacement using a lateral approach (Figure 1). The patient’s medical history revealed mild developmental dysplasia of the affected hip, phosphate diabetes, asymptomatic spinal stenoses of the lumbar spine, arterial hypertension, bronchial asthma, euthyroid struma, and obesity. Immediately postoperatively, a lack of motor function of the lower extremities with intact sensitivity was observed, initially attributed to an anesthesia-related opiate overhang.

The symptoms persisted throughout the following day, presenting an incomplete paraplegia with preserved sensibility in the legs and anus area. Computed tomography revealed unspecified multisegmental changes in the lumbar spine with spinal stenosis of the L4/L5 segment. Due to the mismatched clinical findings, the computed tomography diagnosis was extended to the thoracic spine. Here, multisegmental hyperostotic degenerative changes of the thoracic spine with ossification of the anterior longitudinal ligament typical for diffuse idiopathic skeletal hyperostosis were diagnosed together with an acute, unstable T11 fracture (Figure 2). Subsequent magnetic resonance imaging demonstrated the spinal contusion at the level of the T11 fracture (Figure 3).

After referral to the authors’ institution, neurological reevaluation confirmed a complete motoric and incomplete sensitive spinal cord injury below the fracture level. A posterior spinal fusion of T10 to L1 with laminectomy of T11 was performed immediately after admission (Figure 4). Intraoperatively, extensive ossification of the ligamentum spinosum and ligamentum flavum was evident. The dural tube was found to be hourglass-shaped in the intervertebral space between T11 and T12; however, a direct lesion of the dura could not be visualized. Postoperatively, no neurological deficit improvement was observed.

The postoperative course was complicated by a pseudomembranous colitis confirmed by sigmoidoscopy, probably secondary to antibiotic therapy for urinary tract infection, a frequent accompanying problem in paraplegic patients. Simultaneously, the patient developed sepsis on the basis of peritonitis due to a perforated coecum, which was found in an immediately performed laparotomy. Intraoperatively, coecal suturing and protective colostomy were performed. Ten days later, while in intensive care, the patient suffered a tension pneumothorax of unknown cause and was resuscitated successfully. Despite thoracic...

**Figure 1:** Postoperative radiograph of the right hip after total hip replacement with simultaneous osteosynthesis for correction of a previous posttraumatic varus deformity of the proximal femur.

**Figure 2:** Postoperative sagittal (A) and transverse (B) computed tomography scans of the thoracic spine revealing the unstable hyperextension fracture through T11. Note the extensive ossification of the spinous processes and the anterior longitudinal ligament in the thoracic spine with the right-sided predominance characteristic for diffuse idiopathic skeletal hyperostosis.
drainage and further intensive therapy, the patient developed multiorgan failure and died 2 months after total hip replacement.

**Discussion**

This article describes a severe case of a patient with diffuse idiopathic skeletal hyperostosis and an acute thoracic vertebral body fracture with subsequent paraplegia secondary to a forced hyperextension during total hip replacement. To the authors’ knowledge, such a case has not been previously described in the literature. The known preexisting stenosis of the lumbar spine had not been symptomatic in the patient’s recent medical history. The morphological and functional symptoms of diffuse idiopathic skeletal hyperostosis with the typical ossification of the longitudinal ligaments and the associated loss of bending forces of the spine were detected postoperatively.

Diffuse idiopathic skeletal hyperostosis, also known as Forestier’s disease, is associated with ossification of paravertebral ligaments and peripheral entheses and occurs more frequently in men. Radiological alterations are mainly observed in the thoracic spine in the form of ossification of the longitudinal ligaments and anterolateral extensions of the vertebral bodies that bridge the intervertebral space, which can be less pronounced in the cervical and lumbar spine. The posterior longitudinal ligament is often seen to be less calcified than the prevertebral ligament, whereas the ossification of the anterior longitudinal ligament typically occurs predominantly on the right side.

As a key criterion for diagnosis, Resnick and Niwayama proposed this flowing ossification extending over 4 contiguous vertebrae, preservation of the integrity of the intervertebral disks without diminution of disk space height or other degenerative changes, and, in contrast to ankylosing spondylitis, absence of sclerosis or ankylosis in the sacroiliac joint. However, Olivieri et al recently reported that radiological changes also occur in the sacroiliac joint, such as sclerosis associated with diffuse idiopathic skeletal hyperostosis. In addition to spinal lesions, other pathological abnormalities are found in the peripheral skeleton. Often, heterotopic ossification in the pelvis and the hip are diagnosed, but bony attachments and ossified ligaments of the patella, shoulder, and hand may also occur. Rarely, dysphagia is noted in an extensively affected cervical spine.

The causes for the development of diffuse idiopathic skeletal hyperostosis are still largely unclear. Excessive osteoblast proliferation caused by an unknown genetic disposition and various external factors, such as environmental, dietary (obesity), and metabolic (diabetes mellitus), are thought to be responsible for this pathological condition. In addition to the noted changes of the peripheral skeleton, diffuse idiopathic skeletal hyperostosis is often asymptomatic or clinically associated with morning stiffness and mild dorsolumbar pain.

The prevalence of ankylosing disorders has been reported to be between 0.1% and 1.4% for ankylosing spondylitis and between 2.9% and 25% for diffuse idiopathic skeletal hyperostosis, with European-born Caucasians more likely to be affected. Due to the previously mentioned suspected association of diffuse idiopathic skeletal hyperostosis with metabolic disorders, dietary habits, and an aging society in Western industrial nations, an increase in its incidence is expected.

In the majority of cases, fractures in ankylosing diseases arise secondary to low-
energy impacts, compared with healthy individuals sustaining traumatic spine injuries.\textsuperscript{4,6,7} Only 2 cases of vertebral body fracture after a total hip replacement have been reported in the literature.\textsuperscript{28} In these cases, previously diagnosed ankylosing spondylitis was described as the underlying disease. Interestingly, both patients also sustained a T11 fracture, as in the current case. Because anterior longitudinal ligament ossifications in diffuse idiopathic skeletal hyperostosis are particularly found between T7 and T11,\textsuperscript{29} this area is connected with the highest rigidity and, thus, is least capable of compensating bending forces. Other reports of diffuse idiopathic skeletal hyperostosis also describe fractures of the thoracic spine, with caudal segments between T7 and T12 mainly involved.\textsuperscript{3,5,30,31} Even in ankylosing spondylitis, involvement of the thoracic region with fractures between T9 and L2 is predominant. These fractures are frequently associated with neurological deficits, which rarely regress postoperatively.\textsuperscript{32}

The increased incidence of fractures in the lower thoracic spine is probably due to physiological biomechanics, with the main motion for extension and flexion found in the caudal segments, particularly between T11 and T12, which is comparable with the reported range of motion in the lumbar spine.\textsuperscript{33} Therefore, it can be concluded that the highest biomechanical strain for extension and flexion, in addition to the lower and upper cervical spine, is at the junction between the thoracic and lumbar spines, as well as on the lower lumbar area. In Forestier’s disease, with its stiff-ossified column along the physiological curvatures (kyphosis/lordosis), an extension force puts its main load in the transition area from lumbar lordosis to thoracic kyphosis. This explains why this area is the second most common area for fractures after excessive extension in stiffening spinal disorders; the cervical spine is the foremost location.\textsuperscript{3,5}

In the current case, fractures extended through the vertebral body with a ventral-ly open angle, showing pathomechanism of a hyperextension injury. In a comprehensive literature search, Westerveld et al\textsuperscript{4} found the hyperextension fracture pattern to be the most common in stiffened spine disorders. In diffuse idiopathic skeletal hyperostosis, 70\% of all fractures were thoracic hyperextension fractures. Compared with ankylosing spondylitis, flexion fracture patterns were not found with diffuse idiopathic skeletal hyperostosis. In patients with diffuse idiopathic skeletal hyperostosis, as in the current case, the majority of fractures extended through the vertebral body, whereas in ankylosing spondylitis, the fracture line extend through the intervertebral disk space in 50\% of the cases.\textsuperscript{4} This is explained by the fact that the ossification of the anterior longitudinal ligament is especially pronounced in the intervertebral area and, thus, fractures can more easily extend through the vertebral bodies.\textsuperscript{3,9,34-36}

In this context, the current patient’s obesity represented an additional risk factor. A retrospective study of 965 patients with hyperextension injuries of the thoracic spine showed that all patients were obese, with an average body mass index of 32.75 (range, 30.9-36.4).\textsuperscript{37} This illustrates that, even in the absence of a stiffening spinal disease, obesity seems to be a major risk factor for the occurrence of thoracolumbar spine injuries. In combination with a stiffening spinal disorder, such as diffuse idiopathic skeletal hyperostosis, the bending forces of the spine are almost completely abolished with the increased fat content overstressing the supporting structures.

Whether the current patient’s vertebral body fracture occurred during intraoperative manipulation of the hip joint, perioperative positioning, or transfer of the patient between the operating table and the bed can not be fully clarified retrospectively. The intraoperative maneuver to expose the femoral shaft with external rotation and maximum adduction of the leg with a simultaneous lowering of the opposite leg is presumably responsible for the hyperextension fracture in the current patient. However, any movement that may result in hyperextension of the spine perioperatively is a potential cause for such a serious injury.

Although numerous criteria exist to differentiate between diffuse idiopathic skeletal hyperostosis and ankylosing spondylitis,\textsuperscript{14} stiffened spine disorders have functional similarities regarding susceptibility to fracture.\textsuperscript{3,5} Because hyperextension fractures in diffuse idiopathic skeletal hyperostosis mostly occur due to low-energy impacts, hyperextension of the spine in peri- and intraoperative maneuvers must be avoided. The current patient underwent a routine hip replacement with a final lethal course due to a severe perioperative complication. Stiffening spine disorders, with their concomitant pathobiomechanics, must be considered in any operative procedure, with special regard to exposing maneuvers in total hip replacement. The positioning of the lower extremities and the choice of the surgical approach must be adjusted accordingly to avoid any hyperextension forces on the thoracolumbar spine during operative intervention.

\textbf{REFERENCES}


