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# Rigid spine injuries – A comprehensive review on diagnostic and therapeutic challenges

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ABSTRACT

Injuries to the rigid spine have a distinguished position in the broad spectrum of spinal injuries due to altered biomechanical properties. The rigid spine is more prone to fractures. Two ossification bone disorders that are of particular interest are Ankylosing Spondylitis (AS) and Diffuse Idiopathic Skeletal Hyperostosis (DISH).

DISH is a non-inflammatory condition that leads to an anterolateral ossification of the spine.

AS on the other hand is a chronic inflammatory disease that leads to cortical bone erosions and spinal ossifications. Both diseases gradually induce stiffening of the spine.

The prevalence of DISH is age-related and is therefore higher in the older population. Although the prevalence of AS is not age-related the occurrence of spinal ossification is higher with increasing age. This association with age and the aging demographics in industrialized nations illustrate the need for medical professionals to be adequately informed and prepared.

The aim of this narrating review is to give an overview on the diagnostic and therapeutic measures of the ankylosed spine.

Because of highly unstable fracture configurations, injuries to the rigid spine are highly susceptible to neurological deficits. Diagnosing a fracture of the ankylosed spine on plain radiographs can be challenging. Moreover, since 8% of patients with ankylosing spine disorders (ASD) have multiple non-contagious fractures, a CT scan of the entire spine is highly recommended as the primary diagnostic tool.

There are no consensus-based guidelines for the treatment of spinal fractures in ASD. The presence of neurological deficit or unstable fractures are absolute indications for surgical intervention. If conservative therapy is chosen, patients should be monitored closely to ensure that secondary neurologic deterioration does not occur. For the fractures that have to be treated surgically, stabilization of at least three segments above and below the fracture zone is recommended. These fractures mostly are treated via the posterior approach.

Patients with AS or DISH share a significant risk for complications after a traumatic spine injury. The most frequent complications for patients with thoracolumbar burst fractures are respiratory failure, pseudoarthrosis, pneumonia, and implant failure.

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## 1. Introduction

Injuries to the rigid spine hold a distinctive position within the broad spectrum of spinal injuries, particularly given the aging demographics in industrialized nations. The prevalence of certain disorders that affect spinal rigidity increases with the age of the population (Fujimori et al., 2016). This illustrates the need for medical professionals to be adequately informed and prepared. Two ossification bone disorders, Ankylosing Spondylitis (AS) – also known as Morbus Bechterew – and Diffuse Idiopathic Skeletal Hyperostosis (DISH) – commonly referred to as Morbus Forrestier – are of particular concern. They both induce stiffening of the spine, subsequently altering its biomechanics (Mata et al., 1997; Braun et al., 2007).

AS is an inflammatory disorder from the rheumatologic spectrum and often has a symptomatic presentation (Koivikko et al., 2008). DISH on the other hand is a condition with unspecific symptoms or can be completely asymptomatic eluding detection until an incidental discovery (Rustagi et al., 2017; Schlapbach et al., 1989).

The divergent morphological spinal fusions exhibited by these conditions suggest varied clinical outcomes post fracture. It is well known that a rigid spine, irrespective of the underlying cause, is more prone to fractures, even with minimal trauma (Westerveld et al., 2009). This vulnerability is heightened in the context of multi-level injuries, emphasizing the intricate challenges in diagnostics and therapeutic intervention (Rustagi et al., 2017).

The objective of this review is not only to present an update of current knowledge but also to increase the understanding of both diseases. By shedding light on the latest diagnostic recommendations, therapeutic modalities, and a systematic account of documented complications, we aim to provide a comprehensive resource for healthcare professionals navigating this complex terrain.

### *1.1. What you need to know about – DISH*

DISH is a non-inflammatory systemic condition that is characterized by the ossification of entheses, both peripheral and paravertebral (Mata et al., 1997).

Different criteria exist trying to consider both the peripheral and paravertebral lesions. The most commonly used criteria are the radiographic criteria of Resnick that require the "flowing" anterolateral ossification of at least four contiguous vertebral bodies. Further obligatory requirements are the absence of a degenerative disc disease, apophyseal joint body ankylosis and signs of sacroiliac joint arthritis (Resnick et al., 1976).

The exact prevalence of DISH varies in the literature and ranges from 2.6% to 22.4% in adults over 40 years of age depending on the region (Finland, United States, Israel), type of criteria used and type of study conducted (population-based, hospital-based, autopsy). Regardless, most studies show an increase in prevalence with increasing age and significantly higher rates for male patients (Bloom, 1984; Boachie-Adjei et al., 1987; Julkunen et al., 1975).

Although studies have shown a higher prevalence of diabetes and obesity with DISH patients, the exact pathogenesis that leads to the formation of new bone remains largely unclear. Mechanical, metabolic and endocrine factors have been examined in an attempt to explain the etiology of the disease; no study has shown a significant result linking one of these factors to DISH so far (Schlapbach et al., 1989; Daragon et al., 1995; Forestier et al., 1971; Julkunen et al., 1971; Littlejohn et al., 1981).

Schlapbach et al. showed in a study that patients with radiographic evidence of DISH did not complain of back pain significantly more often than patients without DISH (Schlapbach et al., 1989). The lack of clinical symptoms such as pain might explain why the prevalence of the disease might be underestimated (Schlapbach et al., 1989). Furthermore, the strict Resnick criteria probably only include patients with advanced disease (Kuperus et al., 2020).

The spinal involvement is mostly described as either an anterolateral (Fujimori et al., 2016; Resnick et al., 1976; Bregeon et al., 1973) or a right-sided (Harris et al., 1974) hyperostosis and can affect the entire spine (Resnick et al., 1976).

Involvement of the anterior cervical spine can cause enthesophytes that can lead to compression of the esophagus or trachea leading to dysphagia or intubation difficulties (Mazieres, 2013; Rai, 2009).

The involvement of the entire spine leads to a stiffening of the spine similar to ankylosing spondylitis (AS). As resorption of energy by the intervertebral disc is no longer possible, minor trauma then is sufficient to cause spinal fractures (Kuperus et al., 2020). The most common trauma mechanism described in a systematic review by Westerveld et al. was hyperextension trauma (Westerveld et al., 2009). Most fractures in ankylosing disorders, both DISH and AS, involve the cervical spine (55%), followed by the thoracic (21%), thoracolumbar (16%) and lumbar spine (8%) (Caron et al., 2010).

Westerveld et al. showed that thoracic fractures in patients with DISH (34.5%) were more frequent than thoracic fractures in patients with AS (10.7%) (Westerveld et al., 2009). The same authors compared spinal fractures in patients with DISH and ankylosing spondylosis (AS) and showed that in DISH the fracture line predominantly involved the vertebral bodies, as opposed to the fracture line involving both the disc and the vertebral bodies in patients with AS (Westerveld et al., 2009). A study by Mazieres et al. could, however, not find a significant difference in fracture lines when comparing both diseases (Mazieres, 2013).

#### *1.1.1. Ankylosing spondylitis*

AS on the other hand is a chronic inflammatory arthritis and the most common entity in the group of spondyloarthritis (SpA). The term SpA refers to a group of immune-mediated diseases characterized by inflammation of the axial skeleton, peripheral joints, and entheses (Garcia-Montoya et al., 2018). SpA includes diseases such as reactive arthritis, psoriatic spondyloarthritis, spondyloarthritis of inflammatory bowel disease and undifferentiated spondyloarthritis (Khan, 2002). An extraarticular feature of AS can be anterior uveitis, occurring in about 40% of patients (Golder et al., 2013).

The prevalence of AS is related to the frequency of HLA-B27 in a population with an estimated prevalence of 0.4% in Togo (Belachew et al., 2009), 0.9% in the United States (Reveille et al., 2013) and 1.4% in Norway (Gran et al., 1985). AS is about twice as common in men than in women and patients typically complain of back pain and stiffness (Khan, 2002). Disease onset usually is in the adolescence or early adulthood; with about 80% of patients developing the first symptoms before the age of 30. Only less than 5% of patients are over the age of 45 when first symptoms occur (Braun et al., 2007).

Although the exact cellular and molecular mechanisms are not yet fully understood, the pathogenesis is deemed immune-mediated. The number of T-cells, macrophages and inflammatory cytokines are increased at the site of inflammation. This inflammation gradually leads to cortical bone erosions and new bone formation predominantly in the spine (Khan, 2002).

Early diagnosis of AS is difficult as the typical clinical characterization of inflammatory back pain with insidious onset before the age of 45, worsening with inactivity and improvement with exercise on its own is not very specific for AS (Khan, 2002). Due to its inflammatory origin, one would expect a huge role for CRP and ESR in the diagnostics. Yet theses markers are elevated in only 50–70% of patients (Golder et al., 2013), more so in patients with peripheral arthritis, than in those with only axial disease (Elyan et al., 2006). Although HLA-B27 is associated with AS, routine testing of HLA-B27 is not reasonable because of the relatively high false negative rates (about 10% of AS cases in absence of HLA-B27) (Feldtkeller et al., 2003) and the false positive rates. The test is more useful when the pretest probability is about 50% (Elyan et al., 2006).

Diagnosis of AS typically is delayed by 8–10 years (Poddubnyy et al., 2012), by which time, patients are in a more advanced stage of the disease. This is mostly because AS is diagnosed using the modified New York criteria which lack sensitivity to early stages of the disease. The criteria include the radiographic evidence of sacroiliitis grade ≥2 bilaterally, or grade 3 to 4 unilaterally and 2 of 3 clinical criteria (low back pain of at least 3 months not relieved by rest but improved by exercise, stiffness of the lumbar spine and limitation of chest expansion) (Poddubnyy et al., 2012).

In its extreme form in the spine, bridging syndesmophyte formation between the vertebrae with involvement of the intervertebral disc leads to the characteristic "bamboo spine". Similar to DISH patients with AS are more prone to spinal injury. This is due to osteopenia and the rigidity of the spine. A minor trauma then can cause a fracture in the AS patient. In contrast to DISH some patients cannot even recall a trauma occurring leading to a delay in diagnosis (Westerveld et al., 2009). Fractures of the ankylosed spine are often unstable with frequent secondary neurological deficits (Finkelstein et al., 1999).

Data on mortality and life expectancy regarding patients with AS is scarce. A nationwide population-based Swedish study showed an increased mortality rate of May 9, 1000 in the AS cohort compared to June 5, 1000 in the control cohort (Exarchou et al., 2016). The leading cause of death in the AS cohort were cardiovascular diseases (CVD). CVD was more frequent in the AS than in the control cohort (34.7% vs 30.6%). Due to the large size of the study they also could show that, the mortality risk in women with AS relative to controls were as high as in men (Exarchou et al., 2016). Interestingly, although spinal trauma was never registered as the leading cause of death, it was a more common intermediate cause of death in the AS than in the control cohort (2.2% vs 0.3%) (Exarchou et al., 2016).

According to the American College of Rheumatology, first-line treatment for symptomatic AS patients is NASIDs. Second-line treatment is DMARDs, such as tumor necrosis factor inhibitors (TNFi) (Infliximab, Etanercept) and IL-17 inhibitors (e.g. Secukinumab, ixekizumab) (Garcia-Montoya et al., 2018; Golder et al., 2013). Evidence suggesting that continuous use of NSAID results in less spinal fusion compared to on demand use is inconsistent (Ward et al., 2019). Other DMARDs such as methotrexate or sulfasalazine have little effect in the treatment of spinal disease and are recommended primarily for patients with prominent peripheral arthritis and few or no axial symptoms (Golder et al., 2013; Ward et al., 2019). Sulfasalazine is recommended in patients who have contraindications to TNFi, such as tuberculosis and chronic infection (Ward et al., 2019).

## 2. Imaging in the ankylosed spine in case of suspected fracture

Because of highly unstable fracture configurations, injuries to the rigid spine are highly susceptible to neurological deficits, both at the time of presentation and later on (Westerveld et al., 2009). Therefore, a high level of suspicion is recommended.

Diagnosing a fracture of the ankylosed spine on plain radiographs can be challenging. Compared to CT, MRI has a low sensitivity for detecting posterior element fractures of 11.5% and 36% (Klein et al., 1999; Izzo et al., 2019).

*Koivikko* et al. showed that only 48% of fractures in the cervical spine could be determined on plain radiographs (Koivikko et al., 2008). In the same study, the authors compared the accuracy of fracture detection with MRI and CT in the ankylosed spine of 20 patients. They showed that both imaging techniques were of comparable results – 94% and 84% detection rate by CT and MRI respectively. CT revealed six fractures not visible on MRI, while MRI revealed two fractures not visible on CT (Koivikko et al., 2008).

Since 8% of patients with ankylosing spine disorders (ASD) have multiple non-contagious fractures (Altenbernd et al., 2009) whole spine cross-sectional imaging is highly recommended. MRI has the advantage of detecting soft tissue damage, such as ligamentous injuries, epidural hematoma and cord signal abnormalities.

The utility of MRI is limited by respiratory compromise and severe thoracic kyphosis. These factors make it difficult for the patient to stay still, thus, increasing the occurrence of motion artefacts (Koivikko et al., 2008; Rustagi et al., 2017).

Therefore, a CT scan of the entire spine should always be used as the primary diagnostic tool. The CT is cheaper, more available, faster and shows better bone detail (Tavolaro et al., 2019). In case of neurological deficit, an additional MRI is recommended.

A multi-center study would help to clarify whether an additional MRI to the CT scan should be included as part of the diagnostic measures of the rigid spine. Some authors perform both in their respective hospitals and recommend the two measures (von der Hoh et al., 2020; Shah et al., 2021).

In AS patients, the CT usually is sufficient for fracture diagnostic and therapeutic screening as the absolute majority of fractures indicates a surgical approach. In DISH patients, we recommend additional standing radiographs, to determine the stability of spinal fractures as a significant proportion of these patients with stable fractures may receive a non-surgical treatment (Fig. 1).

## 3. Therapy

There are no consensus-based guidelines for the treatment of spinal fractures in ASD (Leone et al., 2016). Although, most patients should be treated surgically. A meta-analysis showed that 46% of patients with both DISH and AS were treated non-surgically due to high surgical risk and patient refusal (Westerveld et al., 2009).

The presence of neurological deficit or unstable fractures are absolute indications for surgical intervention. Otherwise, nonsurgical treatment would also be a possibility.

Once the conservative route is chosen, patients should be monitored closely to ensure that secondary neurologic deterioration does not occur (Whang et al., 2009). Options for immobilization include a cervical collar or halo vest for cervical spine fractures. The halo vest should only be used as a last resort due to higher mortality rates compared to a cervical collar (Majercik et al., 2005). Use of a halo vest is problematic for patients with ASD due to their advanced age, cervicothoracic

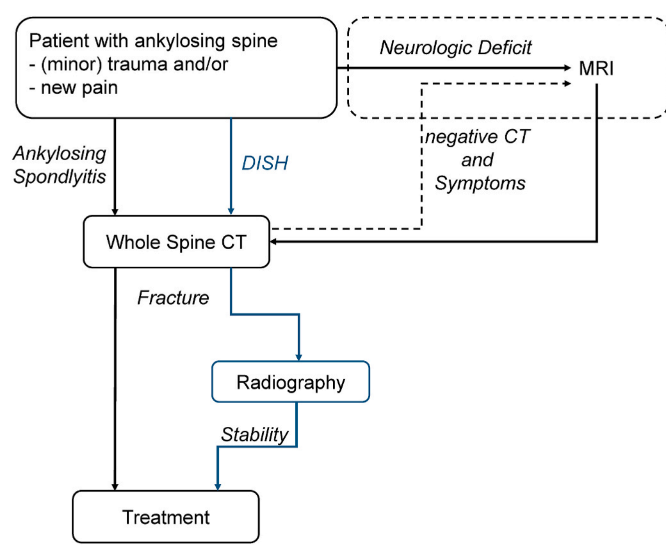


**Fig. 1.** Diagnostic algorithm. The primary diagnostic tool for evaluating spinal fractures in patients with ankylosing spine disorders is the whole spine CT. In patients with neurological deficits, an additional MRI is recommended. After identification of a fracture in the CT, DISH patients may benefit from a standing plain X-ray to determine spinal stability because this has relevant implications on indication for a surgical treatment strategy. In patients with AS, the whole spine CT is usually sufficient, as fracture severity in these patients excludes a non-surgical approach. In case of a negative CT, a total spine MRI is recommended (Ren et al., 2021; Vazan et al., 2019; Tavolaro et al., 2019).

kyphosis, and absent compliance of the chest wall. All of these things lead to an increased risk of aspiration and pulmonary deterioration (Rustagi et al., 2017). A thoracolumbrosacral orthosis can be used for thoracic, thoracolumbar, or lumbar spine fractures; and a cervicothoracolumbrosacral orthosis for high thoracic or low cervical fractures (Werner et al., 2016). Neurologic status typically has a stable course in fractures that qualify for conservative treatment (Westerveld et al., 2009; Whang et al., 2009).

For the fractures that have to be treated surgically, stabilization of at least three segments above and below the fracture zone is recommended (Rustagi et al., 2017). Several studies have shown a preference to percutaneous fixation if feasible. Clinical results are similar to open fixation but percutaneous fixation is faster and associated with lower blood loss and decreased bed rest duration (Buxbaum et al., 2021; Ye et al., 2022). If an open approach is chosen, anterior, posterior and combined anterior/posterior approaches are possible when treating fractures of the ankylosed spine (Westerveld et al., 2009). During surgery, patients should be positioned on a Wilson frame since the frame itself induces kyphosis. This corresponds more to the pre-existing kyphotic alignment. This technique also optimizes exposure of the disc space and minimizes neural-structure retraction for intervertebral work (Cardoso et al., 2010).

Patients with ASD usually present a fixed sagittal imbalance. In case of a fracture, it might seem reasonable to try to correct the preinjury sagittal alignment instead of just restoring the original sagittal axis. One of the procedures used to correct sagittal alignment is the pedicle subtraction osteotomy (PSO). PSO enables a correction of up to 30° (Fanous et al., 2018). Nevertheless, PSO and other techniques are open procedures that is subject to complications associated with open spinal surgery, such as neurological deficits pseudoarthrosis, rod breakage, substantial breakdown and kyphosis at segments proximal to the instrumented fusion (Bridwell et al., 2003).

The primary goal in surgical management of ASD, however, is to restore the preinjury alignment of the spine to achieve an adequate and stable reduction of the fracture and to address any compressive lesions (Werner et al., 2016). Since open surgery has a higher rate of complications such as pseudoarthrosis, nerve injury, increased blood loss and length of hospitalization compared to minimally invasive surgery, the indication for corrective surgery should be handled restrictively. Patients must be well informed about the alternative techniques and their advantages and disadvantages. (Whang et al., 2009).

Most fractures of the ankylosed spine affect the cervical spine (Westerveld et al., 2009). These fractures mostly are treated via the posterior approach (50% of cases) (Westerveld et al., 2009). This is because of an easy approach to the fracture zone in case of concomitant deformities and access to multiple additional fixation points. Furthermore, a multilevel decompression in case of a neurologic deficit is possible through the posterior approach (Rustagi et al., 2017). Reasons for not using the anterior approach as regularly (only in about 15% of cases (Westerveld et al., 2009)) most likely is because of an adverse constellation of present osteoporosis with limited screw purchase, long lever arms caused by the underlying ankylosing disorders, and frequently present kyphosis (Rustagi et al., 2017). Failure rates of initial anterior fixation is up to 50% (Werner et al., 2016). A combined anterior-posterior approach mostly is used when correction of fixed deformity and fracture fixation are to occur simultaneously (Werner et al., 2016).

Although thoracic and lumbar fractures are not as common as cervical fractures, these fractures still occur more frequently in patients with ASD than in the general population. Treatment of thoracic and lumbar fractures follows the same principles as cervical fractures: Posterior stabilization with long constructs to prevent adjacent segment fractures should be pursued (Werner et al., 2016).

## 4. Complications

Patients with AS or DISH share a significant risk for complications after a traumatic spine injury (Westerveld et al., 2009; Kohler et al., 2022). In a scoping literature review, we identified 47 studies providing complication measures in 10791 patients with spine fractures and ASD (also including the c-spine). We then focussed on studies (n = 37) presenting data on complications in specifically ankylosing spine or DISH and thoracolumbar spine injuries (Supplement S1 PRISMA Chart). We then pooled this cohort of 1520 patients with AS (91%) or DISH (9%). We extracted a pooled estimate across all patients with rigid spine (AS or DISH) and thoracolumbar injuries to summarize the most common complications (Table 1). The estimated in-hospital mortality during the first admission is 5.2 % (SD 15%). Overall, severe treatment complications are frequent in this group of patients. The most frequent complications for patients with thoracolumbar burst fractures are respiratory failure, pseudoarthrosis, pneumonia, and implant failure (Table 1). Our non-systematic results suggest lower rates of complications than previous systematic reviews (Westerveld et al., 2009). This and the potential differences within treatment groups and diseases (AS vs. DISH) warrant a thorough meta-analysis. Moreover, we also assessed complications within the treatment of c-spine fractures, which were also frequent in this cohort (Supplement S2).

## 5. Conclusion

Injuries to the rigid spine, typified by Ankylosing Spondylitis (AS) and Diffuse Idiopathic Skeletal Hyperostosis (DISH), present intricate challenges in spinal trauma management. With aging populations in industrialized nations, the prevalence of these conditions is on the rise, necessitating heightened awareness among healthcare providers. Diagnostic dilemmas arise due to the complexities in accurately assessing fractures, where plain radiographs may fall short, and advanced imaging modalities like CT scans and MRI play pivotal roles, albeit with their own limitations. Especially the role of a whole spine MRI should be elucidated further.

Therapeutic strategies vary based on fracture characteristics and underlying conditions, with surgical intervention often necessary for neurological deficits or unstable fractures. Percutaneous fixation techniques are favored where feasible, while conservative approaches are considered in select cases, emphasizing close monitoring to prevent secondary neurological deterioration. Patient-specific factors must be carefully weighed in treatment decisions to achieve the primary goal of restoring pre-injury alignment and stabilizing the fracture.

Complications following traumatic spine injuries in AS or DISH patients pose significant risks, including respiratory failure, pseudoarthrosis, pneumonia, and implant failure. Our findings suggest lower complication rates than previously reported, yet further investigation through thorough meta-analysis is warranted to clarify potential disparities within treatment groups and diseases.

Moving forward, continued research efforts are crucial to refine

**Table 1**
Complications in the management of thoracolumbar fractures of patients with ankylosing spondylitis or DISH.

| | Ankylosing Spondylitis (n = 627) | DISH (n = 114) |
|---|---|---|
| Complication | Mean % (min - max; SD %) | |
| Pseudoarthrosis | 6.6 (0.0–100.0; 21.5) | 7.2 (0.0–25.6; 14.3) |
| Respiratory failure | 6.6 (0.0–71.4; 16.2) | 15.2 (0.0–33.3; 16.8) |
| Pneumonia | 5.6 (0.0–100.0; 18.1) | 13.2 (0.0–33.3; 15.0) |
| Implant Failure | 4.2 (0.0–37.5; 10.4) | 0.8 (0.0–4.8; 1.9) |
| Infection | 3.5 (0.0–25.0; 6.3) | 1.6 (0.0–9.5; 3.9) |
| Conversion to surgery | 2.3 (0.0–72.7; 12.7) | 0 (0) |
| Sepsis | 2.3 (0.0–50.0; 9.2) | 1.6 (0.0–9.5; 3.9) |
| Cardiac | 1.2 (0.0–25.0; 4.6) | 0 (0) |
| Mal Union | 0 (0) | 1.2 (0.0–7.1; 2.9) |

diagnostic algorithms, therapeutic guidelines, and targeted interventions aimed at mitigating complications in this vulnerable population. By advancing our understanding of pathophysiology, diagnostic nuances, and therapeutic modalities associated with fractures in the ankylosed spine, healthcare professionals can better navigate complexities in spinal trauma management, ultimately improving outcomes and quality of life for affected individuals.

## Declaration of competing interest

The authors declare that there are no conflicts of interest regarding the publication of this paper. No financial or personal relationships with other people or organizations have influenced the work reported in this manuscript. This statement is made in the best interest of and truthfully by all authors.

## Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.bas.2024.102811.

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