Bilateral isolated C5 paralysis of the shoulder: Atypical presentation of a transdiscal C4-C5 cervical spine fracture

Elisa Mareddu , Aurélien Traverso , Pietro Laudato, Stefan Bauer

Orthopaedics and Trauma Surgery, EHC Morges, Morges, Switzerland

Correspondence to
Dr Elisa Mareddu;
elisa.mareddu@gmail.com

Accepted 30 December 2020

SUMMARY

After a low-energy fall, an 83-year-old man presented with bilateral weakness of the upper arms without loss of sensation associated with a rigid cervical spine (ankylosing spinal disorder, ASD). Because of an atypical presentation during history, examination and initial imaging, a late diagnosis of a transdiscal C4-C5 fracture was made by dynamic radiographs. Anterior cervical discectomy and fusion were performed with delay. Strength improved from grade C to D (American Spinal Injury Association classification) after surgery. To our knowledge, this is the first description of a bilateral, isolated upper limb C5 paralysis without any loss of sensation caused by a transdiscal C4-C5 fracture. A high clinical and diagnostic index of suspicion is mandatory to make the diagnosis. We present three clinical 'Awareness Criteria' (1: recognition of ASD; 2: high index of fracture suspicion; 3: necessary imaging) helping clinicians to safely and promptly diagnose occult spinal fractures in ASD.

BACKGROUND

Ankylosing spinal disorders (ASDs) are responsible for a higher susceptibility to lesions even after low-energy impacts.[1]

Improvements in the medical treatment available for spine diseases will increase the life expectancy of affected individuals, and thus the total number of people at risk of unstable fractures. This is why the incidence of complicated spinal fractures can be expected to rise during the coming decades. Moreover, the diagnosis is often delayed because of subtle trauma and pre-existing, long-standing back pain that both decrease the alertness of patients and caregivers. In addition, these fractures can be easily missed on standard plain films, CT and even MRI scans. Delay in diagnosis can lead to catastrophic consequences.

We describe a case of a transdiscal fracture at the C4-C5 level that led to bilateral isolated C5 paralysis of the upper limbs. To our knowledge, this is the first report of a transdiscal fracture associated solely with a motor deficit of the root of C5. Current guidelines for the assessment of cervical spine trauma might not be sufficient to make the diagnosis in the context of a rigid spine.

The aim of this report was to highlight the existing difficulties with respect to initial management and diagnosis of these injuries and to rise the index of suspicion among clinicians during the assessment of patients at risk.

CASE PRESENTATION

An 83-year-old man presented to the emergency department after a fall backwards from standing height, striking his head (occipital impact) against the ground. At arrival, he could not give any details concerning his previous medical history and comorbidities. Immediately after the accident, he experienced important posterior neck pain. The physical examination revealed some tenderness in the cervical region but no neurological deficit was initially detected and recorded. Advanced Trauma Life Support assessment was unremarkable. A high-resolution three-dimensional spinal and cerebral CT scan in the emergency department was considered to be normal (figure 1) and plain radiographs were omitted since a CT scan was indicated. No fractures or dislocation of the spine was detected, although the typical radiographic features of a degenerative, partially ankylosed spine were apparent. The patient was subsequently discharged with pain medication and no immobilisation.

Two weeks later, he sought medical attention from his general practitioner for an aggravation of neck pain, neck stiffness and development of bilateral upper limb weakness, predominantly on the right side.

A right shoulder lesion was suspected and plain radiographs and a shoulder ultrasound were ordered which demonstrated a mild omarthrosis and no rotator cuff lesion. These investigation findings were inconclusive and insufficient to explain the clinical picture. Because of persisting disabling neck pain and aggravation of upper limb weakness, the patient reattended the emergency department.

Based on the standard classification of spinal cord injuries (American Spinal Injury Association), deltoid abduction and flexion strength, external rotation of the shoulders and flexion at the elbows ranged from grade M2 to grade M3 (standard strength grading)

Figure 1 CT images of the cervical spine obtained during first presentation. Cervical spine considered aligned. C6-C7 discarthrosis. (A) Arrow showing increased space between anterior portions of vertebral bodies C4-C5. (B) Arrow showing anterior osteophytes.

To cite: Mareddu E, Traverso A, Laudato P, *et al*. *BMJ Case Rep* 2021;**14**:e236323. doi:10.1136/bcr-2020-236323

Figure 2 Dynamic radiograph of cervical spine showing rupture of posterior and anterior cervical line at C4-C5 with augmentation of interspinous distance (red arrow) and grade 1 anterolisthesis of C4 on C5 corresponding to a ligamentous and discal lesion.

predominantly on the right side. No loss of sensation was detected. The patient was admitted for further investigations.

INVESTIGATIONS

The attempt of an MRI scan failed since cervical pain, stiffness and flexion of the cervical spine were too important to allow for positioning in the MRI scanner. We want to point out that it is crucial not to passively extend the cervical spine of patients with kyphotic posture, neither for imaging nor for immobilisation in rigid collars or braces. Finally, dynamic cervical spine radiographs in an alert patient (active flexion and extension as tolerated) were the only option to assess the stability and integrity of the cervical spine. They showed a transdiscal flexion-distraction type fracture at C4-C5 with posterior opening between the spinous processes C4-C5 and grade 1 anterolisthesis of C4 on C5 (figure 2). The diagnosis of degenerative ankylosing spondylosis of the cervical spine was made since there were degenerative changes in the facet joints and disc space with narrowing and sclerosis and adjacent degenerative spondylophytes with anterior vertebral bony contact and therefore rigidity between C5, C6 and C7. No characteristic syndesmophytes were seen.

After further pain control, preoperative CT and MRI scans were performed which demonstrated compression of the C5 roots bilaterally (figure 3).

Figure 3 MRI scans were performed which demonstrated compression of the C5 roots. MR examination was often interrupted because of the poor condition and discomfort of the patient and produced ever-decreasing image quality with each succeeding sequence. (A) Sagittal T1-weighted MRI of cervical spine demonstrating stenosis of the intervertebral foramen C4-C5 level. (B) Sagittal CT-scan reconstruction of the cervical spine showing the fracture. (C) Axial T1-weighted MRI of cervical spine demonstrating compression of the C5 root.

DIFFERENTIAL DIAGNOSIS

The clinical differential diagnosis of the patient's neck pain needs to be seen in the context of his pre-existing spinal disease. Clinical pain and stiffness of the cervical spine should raise the clinicians index of suspicion for an ASD.

The most important diagnostic performance of the treating clinician are suspicion and awareness of a vulnerable patient with ASD which is radiologically distinguishable by several features (table 1).

TREATMENT

As the neurological deficits were slowly progressive, emergency surgical decompression of the spinal cord with internal fixation of the fracture was considered to be necessary to prevent further deterioration. The integrity of the posterior longitudinal ligament allowed us to perform the intervention safely under general anaesthesia. A standard anterior approach was chosen to perform anterior cervical discectomy at C4-C5 with fusion (figure 4).

OUTCOME AND FOLLOW-UP

The muscle strength of key muscles responsible for abduction, external rotation and flexion of the shoulders recovered from grade C to grade D by postoperative week 1. The patient was transferred for rehabilitation on day 11. At 2-month follow-up, the patient was already independent in performing activities of daily living.

DISCUSSION

ASD presents a fivefold higher risk of vertebral fracture and a 35% increased risk of non-vertebral fracture.[2] ASD-related fractures are atypical. The intervertebral disc undergoes degenerative changes, such as chondroid metaplasia and calcifications, and becomes the weakest point of the spine, most prone to fracture.[3] Moreover, they tend to be highly unstable as they involve all three vertebral columns and ossified ligaments. Hence, spinal cord injury complicates up to one-third of these fractures.[4] A history of minor trauma with only moderate symptoms, described as the 'fatal pause' before neurological deterioration by Einsiedel *et al*,[5] inappropriate radio-diagnostic examinations and a substantially altered anatomy are some of the reasons why it can be challenging to identify the fracture. Most of the complications would be preventable by a prompt treatment; however, in almost half of the cases, the diagnosis is delayed.[6] Late diagnosis is the result of misinterpretation of the clinical symptoms by physicians (doctor's delay) as well as by patients (patient's delay).[7] If the patient's delay is understandable, and could be prevented through better education once ASD is diagnosed,[8] doctor's delay is alarming, considering that it is accountable for two-thirds of the overall deferment. There is a lack of definite guidelines. The German Society for Orthopaedics and Trauma is the only group that attempted to establish an assessment protocol.[9] They consider simple plain radiography insufficient and advocate for the use of advanced imaging techniques such

Table 1 Differences among ankylosing spinal disorders

	Spondylosis	DISH (Diffuse Idiopathic Skeletal Hyperostosis)	AS (Ankylosing Spondylitis)	Surgical fusion
Age	Older	Older or middle	Younger	Older
Hallmark	Interrupted osteophytes	Flowing candle wax	Bamboo spine, squaring, kyphosis, sagittal balance lost	No disc space
Osteophytes/syndesmophytes	Spondylophytes (osteophytes) interrupted	Non-marginal syndesmophytes, at least four vertebrae	Marginal syndesmophytes	None or few
Disc space	Narrowing	Preserved	Ossification of the disc	None
Osteopenia	Present	Absent	Present	Absent
Sclerosis	Present	Absent	Absent	Absent
Facet joint involvement	Sclerosis and narrowing	Spared	Involved	Spared or fused

as CT. Concomitant use of MRI is still debated[9 10] and recommended according to the circumstances, mandatory in the case of neurological involvement. Dynamic flexion–extension radiographs in awake and alert patients with muscle control are in some cases necessary to make a diagnosis. Advanced Trauma and Life Support guidelines recommend immobilisation on a spinal board with a hard cervical collar in patients with a suspected spine injury. Unfortunately, vertebral alignment is problematic when the column is rigid. Moreover, most of the cervical spine injuries are the result of a hyperextension mechanism and the recommended immobilisation equipment forces the neck into extension, opening up the column anteriorly, potentially displacing the fracture and causing further risks of deterioration.[3 11 12]

It is preferable to allow the conscious patient to maintain a comfortable position, most often a mild flexion supported by pillows, in line of the pre-existing kyphosis. Disruption of this setting should be avoided until definitive treatment, even during imaging studies,[3 11 12] and the personnel involved in patient manipulation have to be informed of the applied immobilisation protocol.

Classical teaching recommends an anterior or posterior approach with multiple level fixation for this type of injury. Unfortunately, our patient was extremely frail, so we opted for the least invasive option. Nevertheless, the patient was immobilised in a Minerva Brace for 3 months after surgery and an MRI scan was performed on day 7 to evaluate the stability and the necessity of additional fixation with a posterior approach.

In summary, patients with ASD represent a unique cohort in which the standard use of high-resolution imaging techniques is justified to avoid missing fractures with initially mild symptoms. Diagnosis of transdiscal cervical spine fractures can be particularly challenging, as shown in this case. Statistics show that doctors are accountable for most of the delays in the diagnosis. The medical community needs to develop increased awareness since the personal and socioeconomic burden in terms of morbidity (~50%)[4 6] and mortality (~10%)[4 6] for these patients are unacceptably high. We therefore describe three Awareness Criteria for emergency doctors, as well as treating junior and senior clinicians to reduce the delay of diagnosing occult fractures in ASD:

Awareness criterion 1: Recognition of ASD (ankylosing spondylitis, diffuse idiopathic skeletal hyperostosis, spinal spondylosis/rigid degenerative spine, status after adjacent surgical or idiopathic fusion) on initial imaging is a mandatory first step of awareness.

Awareness criterion 2: A high index of clinical suspicion of an occult spine fracture in ASD after trivial mechanisms is a mandatory second step of awareness.

Awareness criterion 3: A CT scan does not rule out an unstable fracture in ASD and an MRI scan and/or dynamic flexion–extension films are often needed.

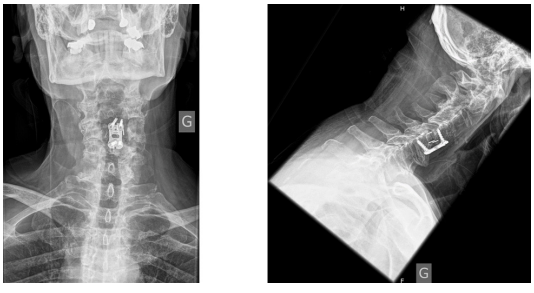

Figure 4 Plain radiographs showing reduction of the fracture and appropriate position of plate and screws.

Learning points

- Failure to diagnose fractures after trivial trauma in ankylosing spinal disorders (ASDs) is common. Doctor and patient awareness could decrease devastating complications.
- Recognition of ASD on initial imaging: Mandatory first step of awareness (Awareness criterion 1).
- High index of clinical suspicion of occult spine fracture after trivial mechanisms in ASD: Mandatory second step of awareness (Awareness criterion 2).
- A CT does not rule out unstable fractures. MRI, radiographs or active flexion–extension films might be indicated: Mandatory third step of Awareness (Awareness criterion 3).
- Passive neck extension for positioning, imaging or immobilisation should be avoided.

Contributors EM wrote the paper and follow the patient. AT supervised CR writing. PL operated the patient. SB: Concept of manuscript, keypoints ans corrections

Funding The authors have not declared a specific grant for this research from any funding agency in the public, commercial or not-for-profit sectors.

Competing interests None declared.

Patient consent for publication Obtained.

Provenance and peer review Not commissioned; externally peer reviewed.

ORCID iDs
Elisa Mareddu http://orcid.org/0000-0001-6203-1134
Aurélien Traverso http://orcid.org/0000-0002-2059-8746

REFERENCES

1 Fox MW, Onofrio BM, Kilgore JE. Neurological complications of ankylosing spondylitis. *J Neurosurg* 1993;78:871–8.

2 Prieto-Alhambra D, Muñoz-Ortego J, De Vries F, *et al*. Ankylosing spondylitis confers substantially increased risk of clinical spine fractures: a nationwide case-control study. *Osteoporos Int* 2015;26:85–91.
3 Leone A, Marino M, Dell'Atti C, *et al*. Spinal fractures in patients with ankylosing spondylitis. *Rheumatol Int* 2016;36:1335–46.
4 Teunissen FR, Verbeek BM, Cha TD, *et al*. Spinal cord injury after traumatic spine fracture in patients with ankylosing spinal disorders. *J Neurosurg Spine* 2017;27:709–16.
5 Einsiedel T, Kleimann M, Nothofer W. Therapie von Verletzungen der Halswirbelsäule bei Spondylitis ankylosans (M. Bechterew) [Special considerations in therapy of injuries of the cervical spine in ankylosing spondylitis (Bechterew disease. *Unfallchirurg* 2001;104.
6 Katoh H, Okada E, Yoshii T, *et al*. A comparison of cervical and thoracolumbar fractures associated with diffuse idiopathic skeletal Hyperostosis-A nationwide multicenter study. *J Clin Med* 2020;9. doi:10.3390/jcm9010208. [Epub ahead of print: 12 01 2020].
7 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* 2009;18:145–56.
8 Nugent M, Berney MJ, Morris S. Clinical outcomes following spinal fracture in patients with ankylosing spondylitis. *Ir J Med Sci* 2017;186:677–81.
9 Reinhold M, Knop C, Kneitz C, *et al*. Spine fractures in ankylosing diseases: recommendations of the spine section of the German Society for orthopaedics and trauma (DGOU). *Global Spine J* 2018;8:56S–68.
10 Tavolaro C, Ghaffar S, Zhou H, *et al*. Is routine MRI of the spine necessary in trauma patients with ankylosing spinal disorders or is a CT scan sufficient? *Spine J* 2019;19:1331–9.
11 Kurucan E, Bernstein DN, Mesfin A. Surgical management of spinal fractures in ankylosing spondylitis. *J Spine Surg* 2018;4:501–8.
12 Werner BC, Samartzis D, Shen FH. Spinal fractures in patients with ankylosing spondylitis: etiology, diagnosis, and management. *J Am Acad Orthop Surg* 2016;24:241–9.