A Cervical Spine Osteophyte Fracture in Patient with Diffuse Idiopathic Skeletal Hyperostosis Behaves Like a Long Bong Fracture

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Background context:

An interesting case of a 55 year old male patient suffering epilepsy with Diffuse Idiopathic Skeletal Hyperostosis (DISH) effecting the cervical spine, who initially presented with a minor osteophyte fracture at the C4/C5 junction following a seizure. This evolved into a significantly displaced fracture with neurological sequel despite appropriate management in a hard collar. Here we highlight the important learning points for future management of patients presenting with seemingly innocuous injuries.

Purpose:

Patients with DISH suffering from apparently innocuous injuries (such as osteophyte fracture) require caution and close continued surveillance or subtle cervical injuries can evolve into significant fracture configurations. This case reports aims to highlight this point.

Study Design/Setting (Patient Sample and Outcome Measures must be used in all Clinical Studies):

This is case report with important learning points for the future management of patients presenting with seemingly innocuous injuries to the neck where they are suffering from DISH.

Methods:

The methodology includes a review of the case notes, patient radiographs and the active involvement in the management of the patient. This has all been compiled retrospectively into a report of the patients' treatment.

Results:

- 1. Osteophyte cervical spine fracture in DISH behaves like a long bone
- 2. even minor injury may cause significant fracture
- 3. fracture management in hard collar needs close monitoring due to risk of displacement
- 4. early internal fixation should be considered

Conclusions:

Patients with DISH suffering from apparently innocuous injuries (such as osteophyte fracture) require caution and close continued surveillance or

subtle cervical injuries can evolve into significant fracture configurations. This may occur some time after the initial injury and even in the absence of further insult. A low threshold is required for repeat imaging in these patients.

STRUCTURED ABSTRACT - Here we present an interesting case of a 55 year old male epileptic patient with Diffuse Idiopathic Skeletal Hyperostosis affecting the cervical spine, who initially presented with a minor osteophyte fracture at the C4/C5 junction following a seizure. This evolved into a significantly displaced fracture with neurological sequel despite appropriate management in a hard collar. Here we highlight the important learning points for future management of patients presenting with seemingly innocuous injuries.

KEY POINTS – 1. Osteophyte cervical spine fracture in DISH behaves like a long bone

- 2. even minor injury may cause significant fracture
- 3. fracture management in hard collar needs close monitoring due to risk of displacement
- 4. early internal fixation should be considered

MINI ABSTRACT - An interesting case of a 55 year old male patient suffering epilepsy with Diffuse Idiopathic Skeletal Hyperostosis effecting the cervical spine, who initially presented with a minor osteophyte fracture at the C4/C5 junction following a seizure. This evolved into a significantly displaced fracture with neurological sequel despite appropriate management.

MANUSCRIPT

SCENARIO - A 55-year-old man, with a history of complex-partial-seizure epilepsy; presented complaining of neck pain 1 day following a generalised tonic-clinic seizure. Immediately following the seizure he was ambulatory. On examination he was tender in the mid-cervical spine. There were no abnormal focal neurological signs. Radiographs showed diffuse ossification of the anterior longitudinal ligament and widespread osteophytes, consistent with Diffuse Idiopathic Skeletal Hyperostosis (DISH). There was also a subtle undisplaced fracture of the anterior cortex of the cervical osteophytes. His cervical spine was immediately immobilised. Computerised tomography confirmed an undisplaced osteophyte fracture at the

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C4/C5 junction (figure-1a). The patient was managed in a hard Aspen Cervical collar, immobilising the neck, then was discharged.

Nine days later, having been compliant with the collar, the patient returned to clinic experiencing left shoulder weakness. Examination showed 3/5 power on shoulder abduction with preservation of sensation, reflexes and tone. There were no other focal neurological signs. Since discharge there had been no further injury. Magnetic resonance imaging was contra-indicated as the patient had an implanted vagal nerve stimulator for his epilepsy. Repeat CT showed slight displacement of C4 vertebra on C5 (figure-1b). His symptoms were considered to be secondary to a neuropraxia following the initial fall. He remained in the Aspen collar until his symptoms improved and he was discharged.

Over the following days, he experienced increasing neck pain and progressive neck deformity. He felt his neck becoming increasingly flexed in the collar. Examination he had kyphosis of the cervical spin with on-going reduced power 4/5 on the left arm with no other focal neurology. Subsequent plain radiographs showed a significant displacement of C4 on C5 (figure-1c). The patient underwent urgent surgical fixation with an anterior C4/C5 cervical disc replacement combined with posterior fusion of the C2-C6 vertebrae (figure-1d). The patient suffered a post-operative left recurrent laryngeal nerve palsy resulting in dysphonia and aspiration. Videofluoroscopy identified incomplete epiglottic closure on deglutition. After a period of nasogastric feeding and failed swallow assessments, a percutaneous enterogastrostomy feed (PEG) was placed. Subsequent DEXA scanning confirmed that the patient was not osteoporotic, with a spinal T score of 1.9. HLA-B27 was negative. Radiographs confirmed that the sacro-iliac joints were not involved. Both findings pointed to a diagnosis of DISH rather than ankylosing spondylitis. When last reviewed at 5 months the patient remained PEG-fed. He was well with no neck complaints and no focal neurological symptoms

DISCUSSION - DISH or Forestier's disease is a feature of senescence and is common in the elderly. It was initially described by Forestier's and Rotes-Querol in 9 men aged over 50 in 19501. The term DISH was coined by Resnick in 1975². The estimated prevalence is 25% and 18% in men and women respectively, over 50 years of age³. DISH is characterised by osteophytes arising from vertebral bodies, which connect at least 4 contiguous vertebrae (non-marginal osteophytes)4. It is distinguished from ankylosing spondylitis due its later age of onset and sparing of the sacro-iliac joints. The pathogenesis remains unclear3. It is associated with diabetes mellitus and hyperuricaemia³. DISH is frequently asymptomatic but may present with back or neck pain3. Dysphagia and laryngeal aspiration surprisingly may occur in DISH due to osteophytes

causing obstruction or the epiglottis impinging against these structures 3,5

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The physiological spine is plastic and deformable due to the ligaments, discs and facet joint movements. During trauma, energy is dissipated along the spine. However in DISH there is ossification of the whole osseo-ligamentous-disc nexus. This makes the spine brittle^{3,4}. In traumatic injury the energy is focussed at a single point and the spine fractures similarly to a long bone³. Ankylosis of the spinal column creates a long lever-arm with the skull at one pole. Even minor falls from a standing position can result in significant injury, and may cause neurological compromise^{3,4}. Previous ambulation does not exclude a fracture. Surgical correction has a significant risk including recurrent laryngeal nerve palsy⁴.

LEARNING POINTS - Patients with DISH suffering from apparently innocuous injuries (such as osteophyte fracture) require caution and close continued surveillance or subtle cervical injuries can evolve into significant fracture configurations. This may occur some time after the initial injury and even in the absence of further insult. A low threshold is required for repeat imaging in these patients. Similarly expeditious referral for consideration for cervical fixation is mandatory. Patient education is important; such that they are made aware of the hazards and the need to seek medical advice urgently should they develop new symptoms.

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