Journal of Spine
ISSN: 2165-7939

Ghani, J Spine 2014, 3:6
DOI: 10.4172/2165-7939.1000191

Case Report Open Access

Functional Outcome in Dish Asssociated with Opll and Dens Hypertrophy Resulting in Rapidly Progressive Hemiplegia – A Case Report

Imtiaz Ghani*

Consultant Spine surgeon, Apollo Hospitals, India

Abstract

Study design: A case report of a patient with diffuse idiopathic skeletal hyperostosis (DISH) associated with ossification of posterior longitudinal ligament (OPLL) and dens hypertrophy resulting in spinal canal stenosis and progressive hemiplegia

Objective: To demonstrate the excellent functional outcome following posterior spinal decompression and fusion.

Methods: In December 2012, a 72-year-old male patient, who was diagnosed with hyperostosis of anterior and posterior longitudinal ligament 6 months ago, presented with rapidly increasing motor deficit of the left arm and unsecure gait pattern. X-rays, CT, and MRI diagnosed DISH and OPLL. Hyperintense signal in the C1 spinal cord on T2 weighted sequence was also observed. Patient underwent decompressive foraminotomy of foramen magnum and laminectomy of C1-C4 with Occiput – C5 fusion.

Results: Neurological status and myelopathy improved to a great extent after surgical intervention.

Conclusion: The current case demonstrates the scenario of DISH associated hemiplegia and the need for a posterior spinal decompression and fusion.

Keywords: Cervical myelopathy; Occipito-cervical fusion; Diffuse idiopathic skeletal hyperostosis (DISH); OPLL; Hemiplegia

Introduction

DISH (Diffuse idiopathic skeletal hyperostosis) is a non-inflammatory enthesopathy ossifying the anterolateral spine and sparing the disc and joint space in elderly men, mostly at thoracic levels. In rare cases of extensive ossification of the cervical spine, compression of the oesophagus and less often the trachea by anterior longitudinal ligament can lead to dysphagia, hoarseness, stridor and dyspnoea. Symptoms occur as a result of direct bony compression of the oesophagus. However, they may also be caused by compression neuropathy of laryngeal nerves [1].

Flexibility of the spinal column in DISH is markedly reduced as a result of ossification of anterior longitudinal ligament. Ankylosis renders the spine susceptible to skeletal injury even after trivial trauma, cervical spine being most commonly affected [2,3]. Spinal fractures are easily missed, because initial presentation may be limited to moderate pain without a neurological deficit [3]. Because of fracture instability, neurological deterioration usually occurs after a certain period of time. This is one of the main reasons why mortality rate in DISH patients with spinal fracture tends to be as high as 20% [3].

Occipitocervical fixation refers to instrumentation and fusion of the occiput to any area of the cervical spine. The main surgical indications are pain, instability, and neurologic compromise. The treatment goals are pain relief, correction or prevention of instability, and decompression of the neural canal. Occipito-cervical fixation confers the advantages of rigid fixation without the use of a halo vest, higher fusion rates, and becoming a salvage procedure for failed non-instrumented occipito-cervical fusion.

We report a case of DISH associated with OPLL and dens hypertrophy leading on to rapid progressive hemiplegia. Posterior decompression and fusion was indicated.

Case Report

Clinical presentation and history

A 72-year-old male patient presented to our hospital in June 2012 with complaints of mild cervical pain, unsecured gait pattern and hypo-sensibility of both hands and left distal forearm. History of anterior cervical decompression and fusion C4-C7 (ACDF) done for similar complaints at a different institution in 1982. There was no associated history of dysphagia.

Motor evoked potential studies were performed and confirmed the diagnosis of cervical myelopathy. Patient was advised to undergo surgery for his clinical symptoms, but he refused. In early December 2012, patient returned to our hospital with complaints of rapidly increasing motor deficit of the left arm with progressive unsecure gait pattern.

Evaluation and management

The patient was admitted in the emergency department and evaluated both clinically and radiologically. Clinical examination revealed defect in fine motor skills in both hands. Motor examination of both lower limbs was normal and there were no signs of paresis.

Additional expertise from neurologist elicited a decrease in temperature sensation on the left side of the body and difference in muscle reflex in the arm (right > left).

Motor examination findings elicited at the emergency department are as follows:

***Corresponding author:** Imtiaz Ghani, Consultant Spine surgeon, Apollo Hospitals, India, Tel: 919962078643; E-mail: imtiazghani@gmail.com

Received July 11, 2014; **Accepted** November 06, 2014; **Published** November 08, 2014

Citation: Ghani I. (2014) Functional Outcome in Dish Asssociated with Opll and Dens Hypertrophy Resulting in Rapidly Progressive Hemiplegia – A Case Report. J Spine 3: 191. doi:10.4172/2165-7939.1000191

	RIGHT	LEFT
Deltoid muscle	3-4/5	0/5
Biceps	4+/5	0/5
Triceps	4+/5	0/5
Finger abduction	4/5	0/5
Hand clasp	4/5	0/5
	RIGHT	**LEFT**
Hip Flexion	5/5	3/5
Knee extension	5/5	4/5
Knee flexion	5/5	3-4/5
Foot dorsi-flexion	5/5	0/5
Foot plantar flexion	5/5	0/5
Toe flexion	5/5	0/5

Hyposensibility of left arm and both hands were noted. Also decrease in temperature sensation was noted on the left half of the body.

Radiological findings

The Radiological findings are clearly explained in Figures 1 and 2.

Surgery

Patient underwent decompressive foraminotomy of foramen magnum and laminectomy of C1-C4 with Occiput – C5 fusion. Intraoperative neuro-physiological monitoring was used Figure 3.

Clinical outcome

Patient was clinically evaluated 1 month, 3 months and 6 months post-operatively to assess the functional outcome.

At the end of 1 month post-surgery, temperature sensation on the left side of the body improved. Sensation of left arm and right hand improved as well.

Following a post-operative period of 3 months, sensation improved in left hand. Also patient demonstrated good response to tendon reflexes in left arm.

The 6 months post-operative neurological findings are as follows,

	RIGHT	LEFT
Deltoid muscle	4/5	4/5
Biceps	4+/5	3/5
Triceps	4+/5	4+/5
Finger abduction	4/5	3/5
Hand clasp	4/5	3/5
	RIGHT	**LEFT**
Hip Flexion	5/5	4/5
Knee extension	5/5	4/5
Knee flexion	5/5	4/5
Foot dorsi-flexion	5/5	3/5
Foot plantar flexion	5/5	3/5
Toe flexion	5/5	2/5

Discussion

Pathologies affecting the craniocervical junction include: traumatic and pathological fractures from metastases, inflammatory conditions (such as rheumatoid arthritis), infections, and congenital and developmental anomalies.

Only fusion techniques did not include any instrumentation and required rigid external fixation with a prolonged use of a halo vest. Halo vests were tolerated especially badly in the elderly population [4]. Internal fixation techniques offer several advantages, namely easier nursing care, earlier mobilization and ambulation, and a higher union rate [5].

In a cadaveric biomechanical testing, C1-C2 trans articular screws or C2 pedicle screws conferred superior results on axial rotation, flexion/extension, lateral bending, and anterior-posterior translation [5]. The fusion rates of these methods were 100% [6-8]. The first preliminary report on successful use of a screwrod stabilizing system posteriorly applied to the occipitocervical junction was in 1996 by Jeanneret et al. [9].

The majority of people with DISH are asymptomatic. Of those with

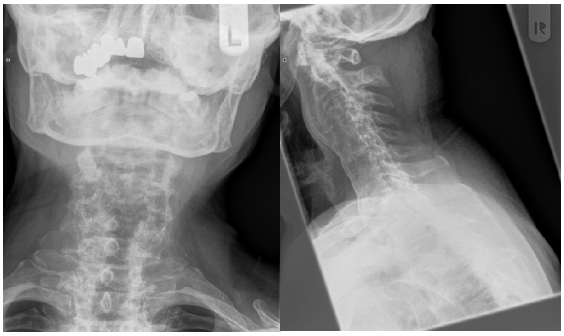

Figure 1: Pre – operative AP and Lateral view of the cervical spine showing exuberant ossification with irregular margins along anterior margins of C2, C3, and C4 vertebral bodies suggestive of Diffuse idiopathic skeletal hyperostosis (DISH)

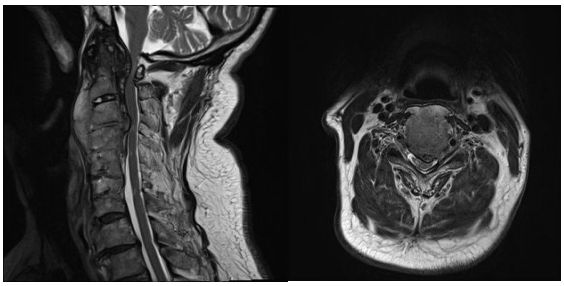

Figure 2: Pre – operative sagittal – enlargement of dens and atlantoaxial joint with calcified thickening of the posterior common ligament resulting in anterior spinal cord compression and coronal MRI reconstruction of the cervical spine showing complete stenosis at C1 level (more on the left side), caused by anterior ossification. Also relative stenosis of C3/4 noted, pronounced on left side

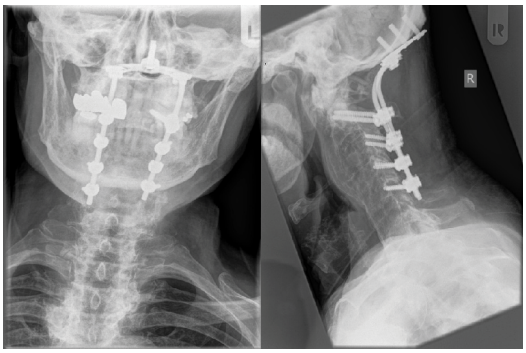

Figure 3: Post – operative AP and Lateral view of the cervical spine demonstrating posterior instrumentation and fusion between Occiput-C5

clinical manifestations, neurological symptoms are the most common. However, subjects do present with other symptoms, including mechanical dysphagia, dyspnoea, stridor, thoracic outlet syndrome and heterotopic ossification [10,11]. Neurological manifestations secondary to DISH are commonly due to involvement of the cervical spine, which is affected in 75% of patients with DISH [12].

Conclusion

The neurosurgical community should become better aware of Forestier's disease. Prior knowledge of the existence of cervical DISH should alert the clinicians for possible complications, at times severe, during invasive procedures in the neck region. The current case demonstrates the rare association of DISH associated hemiplegia and the need for posterior spinal decompression and fusion.

References

1. McCafferty RR, Harrison MJ, Tamas LB, Larkins MV (1995) Ossification of the anterior longitudinal ligament and Forestier's disease: an analysis of seven cases. J Neurosurg 83: 13-17.
2. Sreedharan S, Li YH (2005) Diffuse idiopathic skeletal hyperostosis with cervical spinal cord injury -a report of 3 cases and a literature review. Ann Acad Med Singapore 34: 257-261.
3. Westerveld LA, Verlaan JJ, Oner FC (2009) Spinal fractures in patients with ankylosing spinal disorders: a systematic review of the literature on treatment, neurological status and complications. Eur Spine J 18: 145-156.
4. Lennarson PJ, Mostafavi H, Traynelis VC, Walters BC (2000) Management of type II dens fractures: a case-control study. Spine (Phila Pa 1976) 25: 1234-1237.
5. Oda I, Abumi K, Sell LC, Haggerty CJ, Cunningham BW, et al. (1999) Biomechanical evaluation of five different occipito-atlanto-axial fixation techniques. Spine (Phila Pa 1976) 24: 2377-2382.
6. Deutsch H, Haid RW Jr, Rodts GE Jr, Mummaneni PV (2005) Occipitocervical fixation: long-term results. Spine (Phila Pa 1976) 30: 530-535.
7. Gluf WM, Brockmeyer DL (2005) Atlantoaxial transarticular screw fixation: a review of surgical indications, fusion rate, complications, and lessons learned in 67 pediatric patients. J Neurosurg Spine 2: 164-169.
8. Gluf WM, Schmidt MH, Apfelbaum RI (2005) Atlantoaxial transarticular screw fixation: a review of surgical indications, fusion rate, complications, and lessons learned in 191 adult patients. J Neurosurg Spine 2: 155-163.
9. Jeanneret B (1996) Posterior rod system of the cervical spine: a new implant allowing optimal screw insertion. Eur Spine J 5: 350-356.
10. Wertheim SB, Bohlman HH (1987) Occipitocervical fusion. Indications, technique, and long-term results in thirteen patients. J Bone Joint Surg Am 69: 833-836.
11. Matgé G (2005) [Surgical management of cervical radiculopathy in Forestier's disease. Case report and review]. Neurochirurgie 51: 15-18.
12. Martínez-Berenguer L, Gómez-Calcerrada D, Martínez-López JJ (1995) Cervical cord compression in diffuse idiopathic skeletal hyperostosis. J Am Board Fam Pract 8: 331-334.