

# Cervical Disc Arthroplasty Combined with Interbody Fusion for the Treatment of Cervical Myelopathy with Diffuse Idiopathic Skeletal Hyperostosis: A Case Report

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**Key words:** Cervical Disc Arthroplasty; Cervical Fusion; Cervical Myelopathy; Diffuse Idiopathic Skeletal Hyperostosis

Diffuse idiopathic skeletal hyperostosis (DISH), also known as Forestier's disease, was originally reported by Forestier and Rotes-Querol in 1950. It is a common but underdiagnosed skeletal disease characterized by massive ossification of the anterior longitudinal ligament of several vertebral bodies. The most common symptom of cervical DISH is dysphagia due to mechanical compression of the esophagus. Reports of cervical myelopathy associated with DISH are rare.<sup>[1,2]</sup> Here, we present a case describing a DISH patient with cervical myelopathy who was effectively treated by a carefully designed surgical procedure.

A 50-year-old Chinese man was admitted to our department because of an 18-month history of numbness in both hands and a 1-month history of gait disturbance. The numbness in the patient's hands, especially in his fingertips, was slowly progressive and resulted in disturbed precise motion of both hands. The patient had no discomfort in his neck and shoulders. It is noteworthy that he did not complain of difficulty in swallowing or breathing over the previous 18 months. There was no remission after 1 month of conservative therapy in a local hospital.

On physical examination, the range of motion (ROM) in the neck was almost fully preserved and no apparent mass could be palpated. The sensation was decreased in both hands and also in the right leg. Muscle strength was slightly decreased in the right arm and right leg. The patient's deep reflexes were brisk in the right arm and both legs, but normal in the left arm. Hoffmann's sign was positive on the right side. The patient reported

obvious deterioration in the last 4 weeks. The Japanese Orthopaedic Association (JOA) score was only 8 out of a total 17 points.

The patient did not show any serological or radiological evidence of rheumatoid arthritis or ankylosing spondylitis. The radiographs of the cervical spine revealed giant anterior osteophytes along the ventral aspect of the vertebral bodies from C3 to C6. C4–C5 and C5–C6 segments were fused by a bony bridge. The ROM of the C3–C4 segment was excessive (18.15°), accounting for a large proportion of the total ROM of the cervical spine (C2–C7, 48.65°) [Figure 1a and 1b]. Magnetic resonance imaging showed obvious herniation of the C3–C4 disc compressing the spinal cord with mild herniation at C4–C5 [Figure 1c]. Based on these observations, we made the following diagnoses: (1) cervical myelopathy caused by C3–C4 disc herniation, (2) C3–C4 segment instability, (3) C4–C5 disc herniation, (4) DISH.

Surgery was successfully performed using an anterior approach. After excision of the osteophytes and three discs and thorough neurological decompression, we implanted a Zero-Profile Spacer (Synthes, Oberdorf, Switzerland) into the C3–C4 intervertebral space and two Prestige-LP Artificial

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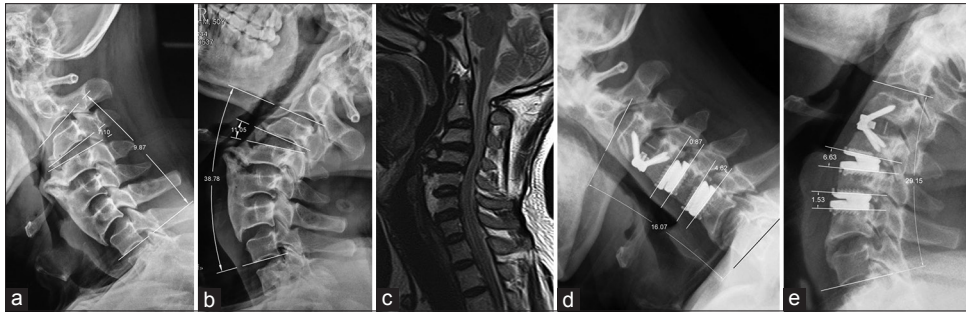
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**Figure 1:** (a and b) Preoperative dynamic radiographs showed the presence of giant osteophytes and the ROMs at C3–C4 and C2–C7; (c) MRI confirmed the disc herniations and spinal cord compression; (d and e) 15 months after surgery, flexion-extension radiographs revealed no recurrence of the osteophytes, good function of internal implants, and preservation of cervical spine ROM. ROM: Range of motion. MRI: Magnetic resource imaging.

Cervical Discs (Medtronic Sofamor Danek, Memphis, TN, USA) into the C4–C5 and C5–C6 intervertebral spaces.

Neurological function improved remarkably after surgery. Numbness in both hands and gait disturbance were significantly reduced and the JOA score improved from 8 to 13. At 15-month follow-up, there was no deterioration of neurological function and no recurrence of the osteophytes. The dynamic radiographs revealed that the ROM of the cervical spine was well preserved ( $45.22^\circ$ ), and C4–C5, C5–C6 segments regained a ROM of  $7.50^\circ$  and  $6.15^\circ$ , respectively [Figure 1d and 1e].

The etiology of DISH remains unclear. Mechanical stress, metabolic conditions, genetic factors, and environmental exposures have been reported as possible underlying causes. The diagnosis of DISH is mainly based on imaging findings. The most widely used diagnostic criteria were proposed by Resnick and Niwayama in the 1970s, including calcification and ossification along the anterolateral aspect of at least four contiguous vertebral bodies, preservation of intervertebral disc height at the involved segments, and absence of apophyseal joint ankylosis and sacroiliac inflammatory changes.<sup>[3]</sup> The patient in this case report was diagnosed to be DISH because anterior osteophytes from C3 to C6 were representative and intervertebral disc height was relatively preserved, and there were no extraspinal manifestations.

Neurological complications are infrequent in typical DISH. Ossification of anterior and posterior longitudinal ligaments sometimes coexists in DISH. Ossification of the posterior longitudinal ligament may result in cervical spinal stenosis, which finally causes cervical myelopathy or radiculopathy. In another rare situation, fusion of the lower cervical spine in DISH is possibly associated with enhanced mechanical stress at the craniocervical region, which could lead to soft tissue mass, odontoid fracture, atlantoaxial subluxation, and pseudotumor and then cause severe spinal cord lesion.<sup>[2]</sup> In the current case, there was no obvious spinal stenosis or craniocervical abnormality. We considered that the myelopathy was mainly due to C3–C4 disc herniation. Biomechanical studies have shown increased intradiscal stress and hypermobility at adjacent

levels after surgical spinal fusion. Many clinical studies have reported accelerated disc degeneration adjacent to the previously fused segments. The apparent hyperostosis in this patient resulted in bony fusion of C4–C5 and C5–C6 segments, thus causing increased stress at the adjacent C3–C4 segment. The excessive stress and motion accelerated degeneration of the C3–C4 disc, leading to disc herniation and spinal cord compression.

Hybrid surgery combining cervical disc arthroplasty and interbody fusion has been widely used for the treatment of multilevel cervical disc herniations in recent years.<sup>[4]</sup> This approach strikes a balance between stability and motion of the cervical spine. In this case, we intended to achieve decompression, resection of osteophytes, and preservation of ROM simultaneously. Because of disc herniation and segment instability, we used a Zero-Profile Spacer for C3–C4 fusion after decompression. On the other hand, the resection of osteophytes damaged the C4–C5 and C5–C6 discs. A procedure was therefore necessary for the reconstruction of C4–C5 and C5–C6 segments. Moreover, we performed C4–C5 and C5–C6 disc arthroplasty to prevent obvious ROM loss of the cervical spine. Although the restoration of motion after fusion has been reported in previous literature,<sup>[5]</sup> a long-term follow-up is required to observe whether C4–C5 and C5–C6 segments could maintain normal ROM.

In conclusion, cervical myelopathy associated with DISH is rarely observed in clinics. The hybrid surgery enabled us to remove the anterior osteophytes to achieve decompression and functional reconstruction of the cervical spine.

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### Conflicts of interest

There are no conflicts of interest.

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