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Cervical Myelopathy by C1 Posterior tubercle Impingement in a Patient with DISH

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Study Design. A unique case of a patient with diffuse idiopathic skeletal hyperostosis (DISH) associated with C1 posterior tubercle impingement resulting in spinal canal stenosis and cervical myelopathy.

Objective. To describe an uncommon mechanism of spinal cord compression in patient with DISH.

Summary of Background Data. The neurological deficits due to cervical DISH are relatively rare and a few cases of cervical spinal cord compression due to atlantoaxial subluxation, odontoid fracture, pseudotumor, ligamentous hypertrophy and basilar impression have been reported. To the best of our knowledge, there has been no other report of a patient with DISH causing C1 posterior tubercle impingement and cervical myelopathy.

Methods. 75-year-old Japanese man, first diagnosed as hyperostosis of anterior and posterior longitudinal ligament 25 years ago, presented with gradual progression of numbness in both lower extremities, disturbed precise hand motion and urinary function. DISH, OPLL, and C1 posterior tubercle impingement was diagnosed by radiograph, CT, and MRI. Hyperintense signal in the C1 spinal cord on T2 weighted sequence was observed.

Result. Laminectomy from C1 to C3 was performed. Myelocompression and

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myelopathy improved after the surgical intervention. Multilevel fusion of the subaxial cervical spine and increase of the mechanical stress on the craniocervical segment may lead to partial damage of the ligaments and result in C1 posterior arch impingement.

Conclusion. This is the first report of unique C1 posterior tubercle impingement and myelopathy caused by DISH. We should keep it in mind that DISH can cause serious problems in the upper cervical spine even after 25 years of interval.

(254/300 words)

Key words:

Diffuse idiopathic skeletal hyperostosis, myelopathy, ossification of posterior longitudinal ligament, posterior tubercle of atlas

Key Points:

- Neurological deficit due to DISH are relatively rare, and there has been no report of C1 posterior tubercle impingement and myelopathy associated with DISH
- Authors report a case of DISH associated with C1 posterior tubercle impingement resulting in myelopathy. The symptom developed 25 years of interval.
- Laminectomy from C1 to C3 without occipitocervical arthrodesis was performed with

favorable result.

Mini Abstract:

We report a unique case of a patient with diffuse idiopathic skeletal hyperostosis (DISH) associated with C1 posterior tubercle impingement resulting in spinal canal stenosis and cervical myelopathy. (28/50 words)

Diffuse idiopathic skeletal hyperostosis (DISH), also termed Forestier disease, is an ankylosing skeletal disorder characterized by continuous ossification of the anterior longitudinal ligament. The most common symptoms of cervical DISH are compression of the trachea and esophagus¹, and neurological deficits are relatively rare. In this case, the authors describe a unique C1 posterior tubercle impingement causing spinal canal stenosis and cervical myelopathy in patient with DISH and ossification of posterior longitudinal ligament (OPLL). The symptom developed 25 year after the first diagnosis. The radiological characteristics and pathogenesis are also discussed.

Case report

History and Examination

A 75-year-old Japanese man was admitted to our hospital because of 8 months history of gradually progressed numbness in both lower extremities resulting in gait disturbance, disturbed precise motion of the hands, and urinary disturbance. He had been diagnosed as hyperostosis of anterior and posterior longitudinal ligament 25 years ago at another hospital, and has been followed up at our outpatient clinic for recent 6 years. However he had not noticed any symptoms except that he feels difficulty in rotating his neck and had not performed any of the operative maneuver.

Physical examination on admission demonstrated decreased range of motion in the neck.

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Precise motion of the hands were disturbed. Dysesthesia appeared in the right hand and both of lower extremities, and urinary function was moderately disturbed. However, apparent motor weakness, the abnormal reflexes of deep tendons, ankle clonus, and Babinski reflexes were all negative. He did not demonstrate dysphagia nor dyspnea. The patient did not show any serological evidence of rheumatoid arthritis, ankylosing spondylitis, or diabetes mellitus. A lateral cervical radiographs (Figure 1) and cervical computed tomography (Figure 2) showed massive calcification and ossification of the anterior longitudinal ligament (OALL) from C2 to T2 level as well as the continuous OPLL from C2 to C4. Disc height was relatively preserved, and sacroillitis and ankylosis of the sacroiliac joint were absent. The atlantodental interval was 2 mm when the neck was in flexion, and Ranawat's distance was also normal. Marked antero-caudal invagination of the posterior tubercle of atlas resulted in narrowing of the spinal canal was noted (Figure 2). Magnetic resonance imaging (MRI) (Figure 3A) showed narrowing of the spinal canal and the cord compression at the atlanto-axial level, and signal change in the cord on T2-weighted MR image, which was not seen on previous MRI, was revealed. Our diagnosis was cervical myelopathy caused by spinal canal stenosis by C1 posterior tubercle impingement, DISH, and OPLL.

Operation and Postoperative course

Under intra-operative motor evoked potential (MEP) monitoring, prophylactic posterior decompression (laminectomy) between C1 and C3 was performed. No complication has occurred during the perioperative period and the MRI after the surgery showed that the spinal cord had been well decompressed (Fig. 3B). After the surgery, the patient noticed that his finger motion became smooth and the urinary disturbance disappeared, however dysesthesia on both lower extremities was still evident.

Discussion

Diffuse idiopathic skeletal hyperostosis (DISH) was originally described in 1950 by Forestier and Rotes-Querol.² The most widely accepted radiological criteria for DISH was proposed by Resnick and Niwayama in 1976.³ They include flowing calcification along anterolateral aspect of at least four contiguous vertebral bodies, relative preservation of intervertebral disc height in affected areas, and absence of apophyseal joint bone ankylosis and sacroiliac joint erosion, sclerosis, or intraarticular osseous fusion^{3,4}. In our case, the rentgenographic findings and the ossification of acetabular enthesis, along with the clinical spinal manifestations, met the criteria for DISH.

DISH occurs frequently in the middle and elderly man. Its incidence in the general population is estimated to be between 6% and 12% on the basis of autopsy series⁵⁻⁷, and

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DISH is often related to OPLL in up to 50 %.^{8,9}

The majority of people with DISH are asymptomatic,¹⁰ however, in symptomatic cases, with mid- to low-back pain by the lower thoracic lesion is popular. The most common presenting symptoms in cervical DISH are dysphagia and, less frequently, dyspnea, resulting from local mechanical compression of the esophagus and trachea.¹ On the other hand, neurological symptoms such as paresthesia or motor deficits, are found in only 4% of the symptomatic patients.¹¹ These stem from 1) reduced flexibility of the spine, thereby allowing a trivial trauma to result in a fracture through the ossified ligaments. 2) spinal canal stenosis secondary to ossification of anterior and posterior longitudinal ligaments. Patients present with myelopathy as a result. 3) Atlantoaxial subluxation (AAS) and other factors causing myelopathy at the high cervical spine, such as, odontoid fracture¹², pseudotumor^{1,13,14}, ligamentous hypertrophy¹ and basilar impression¹⁵.

However, our patient did not meet any of the aforementioned causes for the upper cervical myelopathy. Instead, our patient presented posterior tubercle impingement of the atlas. In this case, the coexistence of impingement and OPLL, is presumed to have caused progressive myelopathy.

However, the mechanism of atlantoaxial subluxation in the current case is thought to be

same as in the cases of upper cervical myelopathy previously reported^{16,17}. Fusion of the subaxial cervical spine coupled with the mobility of the craniocervical lesion seems to play a pivotal role.¹ The occipito-atlanto-axial joint accounts for 45 degree of flexion and extension and 50% of the rotation in the normal cervical spine, whereas each subaxial level in the cervical spine contributes between 10 to 20 degree of flexion and extension¹⁸. In DISH, all calcification-induced multilevel subaxial fusion concentrates the mechanical stresses at the occipito-atlanto-axial junction. Chronic strain on the supporting ligaments and the surrounding soft tissues results in repeated and continual microtears of the surrounding ligaments.¹

Surgical treatment is capable of eliminating spinal cord compression and stopping neurologic deterioration.¹ From the dynamics radiographs, we estimated the patient has no instability at the craniocervical segment¹⁹, and thus laminectomy from C1 to C3 without occipitoatlantal arthrodesis was performed. The previous reports suggested the necessity of rigid fixation of the cervical spine²⁰, but, we had not recognized any further problems without performing arthrodesis in this case.

Conclusion

A unique case of cervical myelopathy due to C1 posterior tubercle impingement associated with DISH was reported. As the symptoms developed 25 years after the

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first diagnosis, we should keep it in mind that in older patients with pronounced hypersostosis, spinal canal stenosis and myelopathy can be caused even in the chronic stage.

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(1487/1500 words)

Figure 1.

Dynamic plain radiographs of the cervical spine (A: flexion, B: neutral, C: extension) revealed the marked ossification of ALL extending between C2 and T1, and PLL between C2 to C4.

The motion of the subaxial spine was markedly restricted and the most of mobility are contributed to the craniocervical joint.

ALL: anterior longitudinal ligament, PLL: posterior longitudinal ligament

O-A: occipito-atlantal, A-A: atlanto-axial

Figure 2.

3D-CT demonstrates the marked ossification of the ALL (A) and PLL (B) extending from C2 (arrow).

Sagittal reconstructed CT view (C) demonstrates the antero-inferior dislocation of the posterior tubercle of the atlas (arrow) and OPLL causing spinal canal stenosis at the C1-2 level. However the atlantodental interval (ADI) is 2mm and is in normal range.

Figure 3.

Preoperative MRI (A) shows narrowing of the spinal cord canal at the atlanto-axial level

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and cord compression with signal change on T2-weighted MR image was revealed.

Postoperative MRI (B) demonstrates the adequate decompression of the spinal cord.



Fig. 1

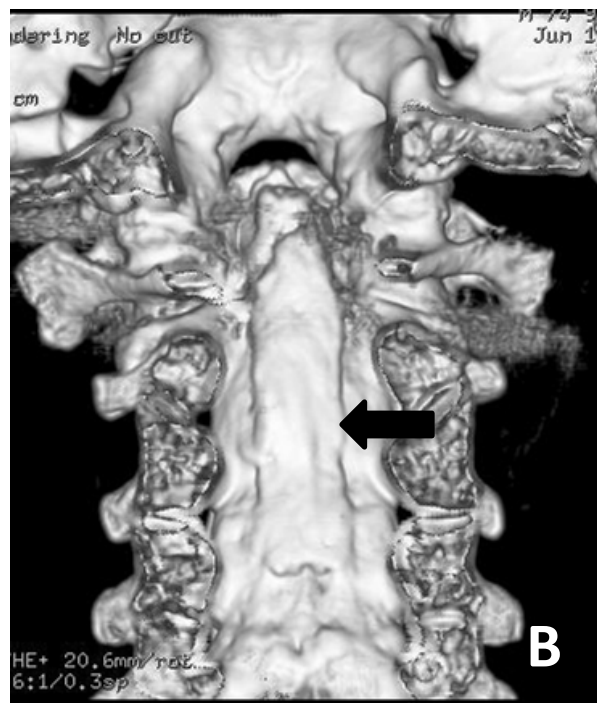


Fig. 2



Fig. 3