Takagi et al. Journal of Medical Case Reports (2017) 11:90
DOI 10.1186/s13256-017-1252-0

Journal of Medical Case Reports

CASE REPORT Open Access

Thoracic spondylolisthesis and spinal cord compression in diffuse idiopathic skeletal hyperostosis: a case report

Yasutaka Takagi[1*], Hiroshi Yamada[1], Hidehumi Ebara[1], Hiroyuki Hayashi[1], Takeshi Iwanaga[1], Kengo Shimozaki[1], Yoshiyuki Kitano[1], Kenji Kagechika[2] and Hiroyuki Tsuchiya[3]

Abstract

Background: Diffuse idiopathic skeletal hyperostosis has long been regarded as a benign asymptomatic clinical entity with an innocuous clinical course. Neurological complications are rare in diffuse idiopathic skeletal hyperostosis. However, if they do occur, the consequences are often significant enough to warrant major neurosurgical intervention. Neurological complications occur when the pathological process of ossification in diffuse idiopathic skeletal hyperostosis extends to other vertebral ligaments, causing ossification of the posterior longitudinal ligaments and/or ossification of the ligamentum flavum. Thoracic spondylolisthesis with spinal cord compression in diffuse idiopathic skeletal hyperostosis has not previously been reported in the literature.

Case presentation: A 78-year-old Japanese man presented with a 6-month history of gait disturbance. A magnetic resonance imaging scan of his cervical and thoracic spine revealed anterior spondylolisthesis and severe cord compression at T3 to T4 and T10 to T11, as well as high signal intensity in a T2-weighted image at T10/11. Computed tomography revealed diffuse idiopathic skeletal hyperostosis at T4 to T10. He underwent partial laminectomy of T10 and posterior fusion of T9 to T12. The postoperative magnetic resonance imaging revealed resolution of the spinal cord compression and an improvement in the high signal intensity on the T2-weighted image.

Conclusions: We report the first case of thoracic spondylolisthesis and spinal cord compression in diffuse idiopathic skeletal hyperostosis. Neurosurgical intervention resulted in a significant improvement of our patient's neurological symptoms.

Keywords: Spondylolisthesis, Spinal cord compression, Diffuse idiopathic skeletal hyperostosis, Thoracic spine

Background

Diffuse idiopathic skeletal hyperostosis (DISH) has long been regarded as a benign asymptomatic clinical entity with an innocuous clinical course [1–3]. DISH rarely causes neurological complications, as evidenced by isolated case reports on the subject; however, if neurological complications do occur, they are often severe enough to warrant major neurosurgical intervention [1–4]. Neurological complications occur in DISH when the pathological process of ossification extends to other vertebral ligaments, causing ossification of the posterior longitudinal ligaments (OPLL) and/or ossification of the ligamentum flavum (OLF) [5]. A retrospective analysis of 74 cases of DISH conducted by Sharma *et al.* found that 11 patients had presented with progressive spinal cord compression or cauda equina syndrome. Of these, OPLL was responsible in nine cases and OLF in two [5]. However, thoracic spondylolisthesis and spinal cord compression in DISH has not previously been reported in the literature. We report the first case of thoracic spondylolisthesis and spinal cord compression in DISH. Neurosurgical intervention relieved the patient's neurological symptoms significantly.

* Correspondence: takagi@p1.coralnet.or.jp
[1]Department of Orthopaedic Surgery, Tonami General Hospital, 1-61 Shintomi-cho, Tonami City, Toyama 939-1395, Japan
Full list of author information is available at the end of the article

Case presentation

A 78-year-old Japanese man presented with a 6-month history of gait disturbance. Magnetic resonance imaging

Fig. 1 MRI revealed anterior spondylolisthesis and severe cord compression at the T3 to T4 and T10 to T11 levels, as well as high signal intensity on a T2-weighted image at the T10/11 level (*white outline arrows*)

(MRI) of his lumbar spine revealed lumbar spinal stenosis (LSS). He could not walk outdoors. A neurological examination of muscle weakness of his iliopsoas and quadriceps femoris suggested possible spinal cord compression. Cervical and thoracic spine MRI revealed anterior spondylolisthesis and severe cord compression at T3 to T4 and T10 to T11, and high signal intensity on a T2-weighted image at T10/11 (Fig. 1). An X-ray revealed intervertebral disc space narrowing and anterior spondylolisthesis at T3/4 and T10/11 (Fig. 2). A myelogram-computed tomography (CT) scan showed anterior spondylolisthesis and severe cord compression at the T10/11 level. OPLL and OLF were not seen at T10/11. DISH was noted above the T10 level (Fig. 3). We determined that the lesion responsible was located at the T10/11 level. He underwent partial laminectomy at T10 and posterior fusion at T9 to T12. He could walk outdoors with one T-cane postoperatively. Postoperative CT detected DISH between T4 and T10 and anatomical repositioning of the anterior spondylolisthesis previously noted at T10. Postoperative MRI revealed resolution of the spinal cord compression and an improvement in the high signal intensity on the T2-weighted image (Fig. 4).

Discussion

DISH is a non-inflammatory skeletal disease characterized by calcification and ossification of soft tissues, primarily ligaments and entheses. DISH is also known as senile ankylosing hyperostosis [6]. DISH involving the spine is identified radiologically by flowing ligamentous ossification and calcification of the anterolateral aspect of the vertebral body with relatively well-preserved disc space [7]. The radiographic criteria, as defined by Utsinger *et al.,* includes: (1) bridging osteophytes extending over four contiguous vertebral bodies; (2) relatively normal intervening disk space height in relation to height in relation to age; and (3) absence of apophyseal joints, bony ankyloses, and absence of erosion, sclerosis, or osseous fusion of the sacroiliac joints [8]. Our patient met all these criteria.

Spinal involvement of DISH is characterized radiologically by flowing ossification of the anterior longitudinal ligament, which is typically separated from the anterior aspect of the vertebral body by a thin radiolucent line [9]. The spinal longitudinal ligaments and entheses slowly ossify and show decreased mobility in the affected region until complete ankylosis results. DISH frequently begins in the lower thoracic spinal

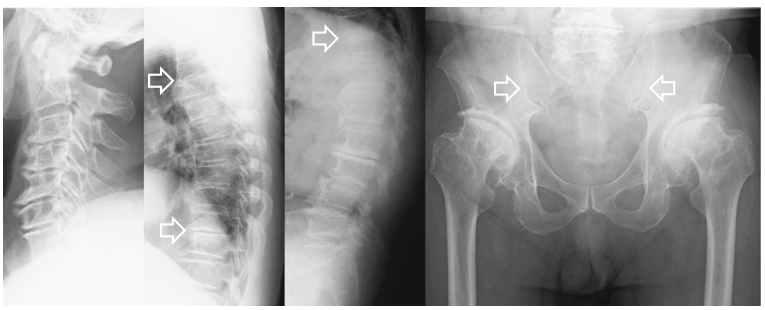

Fig. 2 X-ray revealed T3/4 and T10/11 intervertebral disc space narrowing and anterior spondylolisthesis and no involvement of the sacroiliac joints (*white outline arrows*)

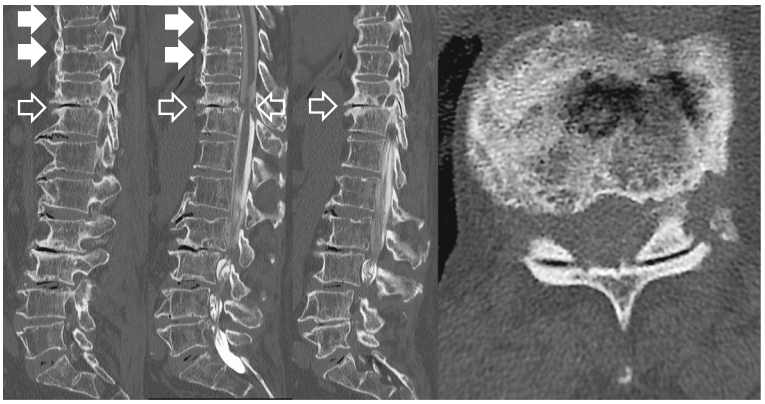

Fig. 3 Myelogram-CT revealed T10 to T11 anterior spondylolisthesis and severe cord compression at T10/11 level (*white outline arrows*). Ossification of the posterior longitudinal ligaments and ossification of the ligamentum flavum were not seen at the T10/11 level. DISH was seen above the T10 level (*white solid arrows*)

segments, before extending into the upper thoracic segments and lumbar spine [10].

DISH results in the fusion of several spinal segments, which amplify the biomechanical load on the unaffected segments. Hypermobility of the spinal segment causes disc degeneration or hypertrophy of the OLF, thus resulting in LSS.

The main point of interest in the present case was the slow progression of myelopathy due to T10 anterior spondylolisthesis in a patient with DISH. In this case, DISH between T4 and T10 caused disc degeneration and anterior spondylolisthesis in T3 to T4 and T10 to T11. Hypermobility of the spinal segment gradually led to spinal cord compression.

To the best of our knowledge, this is the first report of thoracic spondylolisthesis and spinal cord compression in a patient with DISH. Neurosurgical intervention provided significant relief of our patient's symptoms. Patients with significant neurological deficits due to spinal cord compression often require surgical intervention. The type of surgery depends on the site and type of compression.

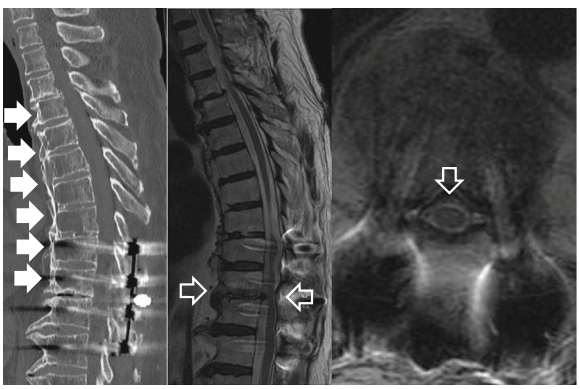

Fig. 4 Postoperative CT revealed diffuse idiopathic skeletal hyperostosis between T4 and T10 (*white solid arrows*). Postoperative MRI revealed that spinal cord compression was well decompressed and high signal intensity in T2-weighted image was improved (*white outline arrows*)

Conclusions

We report the first case of thoracic spondylolisthesis and spinal cord compression in DISH. Neurosurgical intervention resulted in significant improvement of our patient's neurological symptoms.

Abbreviations

CT: Computed tomography; DISH: Diffuse idiopathic skeletal hyperostosis; LSS: Lumbar spinal stenosis; MRI: Magnetic resonance imaging; OLF: Ossification of the ligamentum flavum; OPLL: Ossification of the posterior longitudinal ligaments

Acknowledgements

Not applicable. No assistance was utilized in the writing of the manuscript.

Funding

The authors have no relevant affiliations or financial involvement with any organization or entity with a financial interest in or financial conflict with the subject matter or materials discussed in the manuscript.

Availability of data and materials

Medical imaging data will not be shared because it is not fully anonymous.

Authors' contributions

YT and HH performed the operation. YT, HY, HE, HH, TI, KS, YK, KK, and HT determined the treatment plan. YT and HH conducted the follow-up. YT wrote the draft of the manuscript, which was revised by HT. All authors read and approved the final manuscript.

Competing interests

The authors declare that they have no competing interests.

Consent for publication

Written informed consent was obtained from the patient for publication of this case report and any accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal.

Publisher's Note

Author details
[1]Department of Orthopaedic Surgery, Tonami General Hospital, 1-61 Shintomi-cho, Tonami City, Toyama 939-1395, Japan. [2]Department of Rehabilitation Medicine, Kanazawa Medical University, 1-1 Daigaku, Uchinada-machi, Kahoku-gun, Ishikawa 920-0293, Japan. [3]Department of Orthopaedic Surgery, Graduate School of Medicine, Kanazawa University, 13-1 Takara-machi, Kanazawa City, Ishikawa 920-8641, Japan.

Received: 29 September 2016 Accepted: 1 March 2017
Published online: 01 April 2017

References

1. Reisner A, Stiles RG, Tindal SC. Diffuse idiopathic skeletal hyperostosis causing acute thoracic myelopathy. A case report and discussion. Neurosurgery. 1990;26:507–11.
2. Alegnhat JP, Hallet M, Koha DK. Spinal cord compression in diffuse idiopathic skeletal hyperostosis. Radiology. 1982;142:119–20.
3. Johnson KE, Peterson H, Wollheim FA, Säveland H. Diffuse idiopathic skeletal hyperostosis (DISH) causing spinal stenosis and sudden paraplegia. J Rheumatol. 1983;10:784–9.
4. Stechison MT, Tator CH. Cervical myelopathy in diffuse idiopathic skeletal hyperostosis. Case report. J Neurosurg. 1990;73:279–82.
5. Sharma RR, Mahapatra A, Pawar SJ, Sousa J, Lad SD, Athale SD. Spinal cord and cauda equina compression in 'DISH'. Neurol India. 2001;49:148–52.
6. Forestier J, Rotes-Querol J. Hyperostosis ankylosante vertebrale senile. Rev Rheum. 1950;17:525–34.
7. Resnick D, Shaul SR, Robins JM. Diffuse idiopathic skeletal hyperostosis (DISH): Forestier's disease with extraspinal manifestations. Radiology. 1975; 115:513–24.
8. Utsinger PD, Resnick D, Sapiro R. Diffuse skeletal abnormalities in Forestier's disease. Arch Int Med. 1976;136:163–8.
9. Resnick D, Niwayama G. Radiographic and pathologic features of spinal involvement in diffuse idiopathic skeletal hyperostosis (DISH). Radiology. 1976;119:559–68.
10. Yamada K, Toyoda H, Terai H, Takahashi S, Nakamura H. Spinopelvic alignment of diffuse idiopathic skeletal hyperostosis in lumbar spinal stenosis. Eur Spine J. 2014;23:1302–8.