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Ossification of posterior atlantoaxial membrane causing spinal stenosis – A case report

Yen-Ting Liu [a], Chon-Haw Tsai [a,b], Po-Hao Huang [c], Po-Chang Wu [a,c,*]

[a] School of Medicine, College of Medicine, China Medical University, Taichung, Taiwan
[b] Department of Neurology, China Medical University Hospital, Taichung, Taiwan
[c] Rheumatology and Immunology Center, China Medical University Hospital, Taichung, Taiwan

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ABSTRACT

Background: Ossification of the posterior atlantoaxial membrane (PAAM) is a rare cause of spinal cord compression.
Case presentation: A 46-year-old woman with rheumatoid arthritis (RA) and a 2-year history of slowly progressive gait disturbance underwent surgery for right knee stiffness and right lower limb mild weakness. A neurologic examination revealed brisk deep tendon reflexes (DTR) and spasticity in her four limbs. A computed tomography (CT) scan revealed spinal stenosis caused by ossification of the PAAM, a rare cause of spinal cord compression. The patient's lower limbs weakness and walking capability were ameliorated post-surgery.
Conclusions: Although the exact mechanism of ossification of PAAM remains unclear, chronic mechanical stress as well as persistent atlantoaxial instability may promote the development of the ossification.

1. Background

In patients with rheumatoid arthritis (RA) presented with the symptoms of cervical spinal cord compression, the cause is often attributed to atlantoaxial subluxation. However, ossification of the posterior atlantoaxial membrane (PAAM) causing spinal stenosis should also be considered. Herein, we reported a case of this rare cause of spinal stenosis. This case report was described according to the CARE guidelines [1].

2. Case presentation

A 46-year-old woman with RA for 7 years presented to our outpatient department with a 2-year history of slowly progressive gait disturbance associated with right knee stiffness and right lower limb mild weakness. She did not receive any intervention for her symptom and denied family history or psycho-social history. Two months prior to the visit, mild left knee stiffness and left limb weakness also developed. Moreover, her right lower limb weakness worsened. A neurologic examination revealed brisk deep tendon reflex (DTR) and spasticity in her four limbs. Her proprioception was decreased in her left lower limb. The plantar responses were extensor, while there was no enhancement of jaw jerk. With the clinical findings and the history of RA gathered, magnetic resonance imaging (MRI) was arranged to exclude C1–C2 subluxation. However, the images disclosed severe canal stenosis at the C1 level due to

* Corresponding author. China Medical University Hospital, No. 2, Yude Road, North District, Taichung City, Taiwan.
E-mail address: wpc625531@gmail.com (P.-C. Wu).

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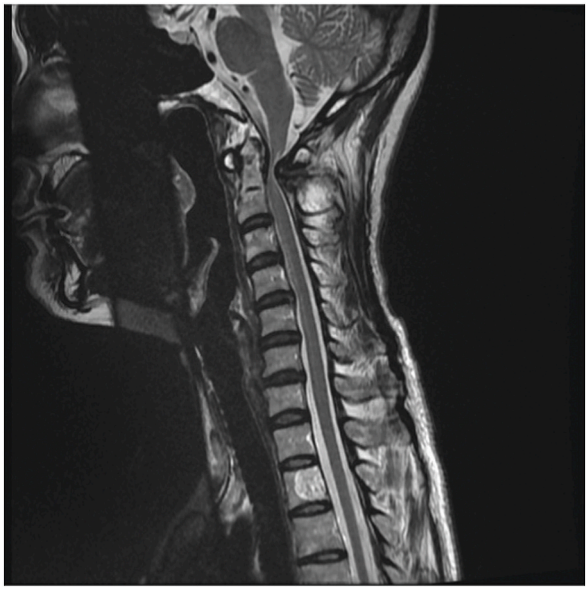

Fig. 1. C1 level due to posterior compression.

posterior compression (Figs. 1 and 2). A computed tomography (CT) scan was arranged to investigate bony lesions, and ossification of the PAAM causing spinal stenosis was revealed (Figs. 3 and 4). Surgical intervention was performed for spinal decompression, and the pathology proved the diagnosis of PAAM ossification. There were no complications or adverse events noted during her follow-up in the outpatient clinic. Neurologic examination disclosed reduced spasticity of four limbs, normal deep tendon reflex, and flexor plantar response. After the surgery, her lower limbs weakness and walking capability were ameliorated. The presentation, diagnosis, and treatment of this case were summarized in Table 1.

3. Discussion and conclusions

Instability between C1 and C2 vertebrae occurs in more than 40% of patients with RA at an average 6-year follow-up [2]. In severe cases, this instability can lead to atlantoaxial subluxation. Ossification of the PAAM is a rare cause of spinal cord compression. To date, only eight patients have been reported [3–10]. Most patients presented with progressive myelopathy. Although the exact mechanism

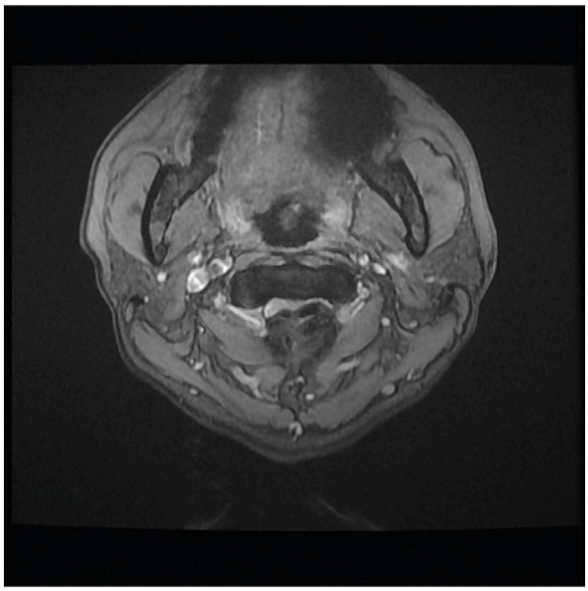

Fig. 2. C1 level due to posterior compression.

Fig. 3. Spinal stenosis caused by PAAM ossification.

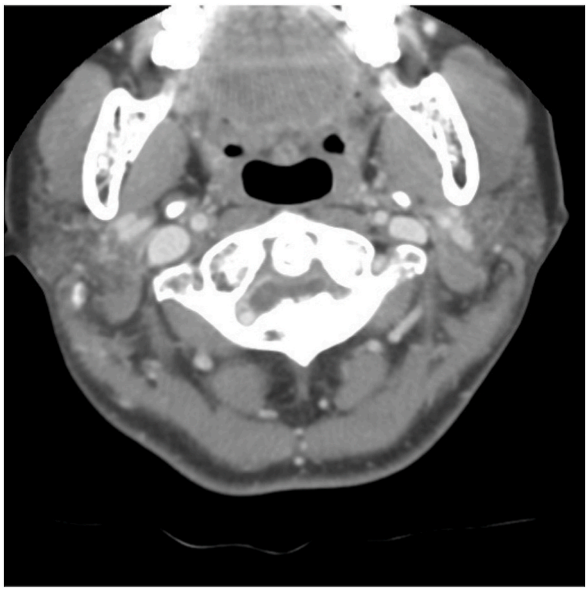

Fig. 4. Spinal stenosis caused by PAAM ossification.

Table 1
Summarized timeline of the case.

Past medical history	• RA for 7 years under medication
Symptom	• Slowly progressive gait disturbance with right lower limb weakness since two years ago • Left limb weakness and right lower limb weakness worsened since two months ago
Diagnosis	• Neurologic examination: Brisk DTR and spast icity in her four limbs, decreased proprioception in her left lower limb, extensor plantar response • CT scan: Ossification of PAAM • MRI: Severe canal stenosis at Cl level due to posterior compression
Treatment	• Surgical intervention for spinal decompression
Follow-up	• Her symptom ameliorated after operation

of ossification of the PAAM remains unclear, one postulates that chronic mechanical stress as well as persistent atlantoaxial instability may promote the development of the ossification [8,11,12].

This case report has several limitations. First, all publications were issued from various institutions, with a significant proportion dating back to before 2010. Variations in the clinical approach could potentially give rise to concerns regarding the heterogeneity of the cases included. Besides, the total published case number was small, which limited the clinical applicability of the diagnostic approach and treatment. To the best of our knowledge, this is the first case that presented with RA history and the diagnosis of ossification of PAAM. The rare cause of spinal stenosis should be considered in the patient presented with the related neurologic symptom. In our patient, compression of the spinal cord was caused by ossification of the PAAM which may be secondary to RA-associated chronic atlantoaxial instability. The mainstay of treatment is a surgical intervention for spinal decompression, and the outcome is satisfactory.

Ethics approval and consent to participate

The institutional review board of China Medical University Hospital approved this study.

Consent for publication

Written informed consent and consent for publication were obtained from the patient.

Competing interests

The authors declare that they have no competing interests.

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Author contribution statement

All authors listed have significantly contributed to the investigation, development and writing of this article.

Data availability statement

Data included in article/supp. material/referenced in article.

Declaration of competing interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper

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Not applicable.

List of abbreviations

CT	computed tomography
DTR	Deep tendon reflex
MRI	Magnetic resonance imaging
PAAM	posterior atlantoaxial membrane
RA	Rheumatoid arthritis

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