

Young Woman With Sudden-onset Neck Pain and Quadriplegia: A Case Report

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Case report

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Abstract

Introduction: Sudden-onset quadriplegia is a neurologic emergency that requires immediate management. Anterior spinal artery syndrome (ASAS) is rare, but can cause sudden-onset quadriplegia. Magnetic resonance imaging (MRI) is an essential imaging modality for diagnosing ASAS.

Case presentation: A 31-year-old woman without previous medical history was transferred to our facility for further workup of sudden-onset neck pain with quadriplegia. Diffusion-weighted imaging (DWI) revealed hyperintense signals predominantly in grey matter with a decreased apparent diffusion coefficient (ADC). These findings strongly suggested ASAS with spinal cord infarction. Edoxaban, heparin, and aspirin with steroid pulse therapy was immediately initiated. Twelve days in an intensive care unit was required for dyspnea. Symptoms gradually resolved with rehabilitation, but the patient was wheelchair-bound without bladder control and was transferred to a rehabilitation facility on hospital day 48.

Conclusion: A combination of DWI with ADC mapping of the spine should proceed as soon as possible to ensure appropriate management. Early treatment with edoxaban, aspirin and steroid pulse therapy may prove beneficial for ASAS.

Introduction

Abrupt onset of quadriplegia is a neurologic emergency that requires immediate management.^{1,2} Several etiologies can cause the sudden onset of quadriplegia or quadriparesis.² Anterior spinal artery syndrome (ASAS) is one of these differential diagnoses, and is a lesion affecting the anterior two-thirds of the spinal cord including the anterior horns of the gray matter, the spinothalamic tract, and the corticospinal tract, with weakness and sensory loss but spared proprioception (body position in space and vibratory sense).³ Early treatment with edoxaban, aspirin and steroid pulse therapy may be beneficial for ASAS. A combination of diffusion-weighted imaging (DWI) with apparent diffusion coefficient (ADC) mapping of the spine is essential to detect the cause of ASAS, such as spinal cord infarction, spinal cord angioma, metastatic spinal carcinoma or acute epidural hematoma.^{4,5} However reading MRI of the spine is difficult, especially for primary-care physicians treating early-stage disease.⁶ Here, we describe the case of a previously healthy young woman in whom ASAS with spinal cord infarction was detected on magnetic resonance imaging (MRI) soon after arrival in our emergency room (ER). We obtained verbal informed consent from the patient and written informed consent from her husband and father to report this case.

Case Report

A previously healthy 31-year-old woman experienced sudden onset of neck pain with left arm discomfort 2 days before admission, but did not seek medical attention. She had no history of trauma, and her familial medical histories included breast cancer in her older sister and maternal grandmother, and rheumatoid arthritis and diabetes mellitus in her maternal grandfather. On the morning of her admission,

she developed very severe neck pain in the morning with acute flaccid paralysis of the legs. She was brought to the nearest hospital in an ambulance, was diagnosed with sprained neck after cervical X-ray, and was discharged to home. However, paralysis of the legs deteriorated with neck pain and she was brought to another hospital and underwent computed tomography (CT) of the brain and neck, showing no abnormalities. The patient was then transferred to our facility for further workup of neck pain with quadriplegia. Vital signs on arrival at the ER were: body temperature, 36.4 °C; heart rate, 98 beats/min; respiratory rate, 18 breaths/min; blood pressure, 107/67 mmHg; and peripheral oxygen saturation (SpO₂), 98% on ambient air. Speech, cognition, and cranial nerve function remained normal. Manual muscle testing (MMT) showed left-right symmetry of findings (Table 1). Passive range of motion in all extremities was complete, with no pain or spasticity. Reflexes were bilaterally absent for the biceps, triceps, and brachioradialis, and of the patellar and Achilles tendon reflexes, and Babinski sign was also absent bilaterally. Proprioception (body position in space and vibratory sense) was preserved, but pain and temperature sensation were lost below the T4 level bilaterally. Urinary incontinence also developed and a urethral catheter was inserted. Whole-body CT findings including the cervical, thoracic, and lumbar spine were normal. Initial MRI examination 30 min after arrival revealed abnormal T2 signals in the cervical spine from C3 to T1 without cord compression (Fig. 1A, B). A neurologist was consulted under a potential diagnosis of ASAS. Repeat MRI with DWI and ADC mapping 3 h later revealed hyperintense signals predominantly in the grey matter, and a contrast T2 signal abnormality with decreased ADC (Fig. 1C, D). Cerebrospinal fluid (CSF) obtained via lumbar puncture on the day of admission showed: white blood cells, 0 cells/mm³; red blood cells, 1 cell/mm³; total protein, 53.9 mg/dL (normal, 15–45 mg/dL); and glucose, 57 mg/dL (normal, 70–110 mg/dL). Hematological and biochemical findings including autoimmune-associated vasculitis, infections and hypercoagulopathies were mostly within normal ranges, except for mildly decreased potassium (3.4 mmol/L; normal range 3.6–4.8). ASAS was finally diagnosed after excluding other diagnoses. We started edaravone, heparin and aspirin with steroid pulse therapy (methylprednisolone, 1000 mg/day) to prevent swelling of the spine, but this did not completely improve the neurological deficits. The next day, she complained of dyspnea and aspiration occurred after drinking water, and was transferred to the intensive care unit (ICU) to secure the airway. Intubation was planned initially, but was not performed, because she could breathe by herself. She was therefore moved to a general ward after 12 days in the ICU. DWI on hospital day 13 revealed enlargement of hyperintense signals predominantly in the grey matter from C3 to T3, and contrast T2 signal abnormality with a decreased ADC (Fig. 2). MRI findings on hospital days 18 and 28 were better and infarctions showed decreased size compared with the findings from hospital day 13 (Fig. 3). The patient started to gradually recover and could eat by herself using her left hand, although she needed a wheelchair and urethral catheter. She was transferred to a rehabilitation facility on hospital day 48.

Table 1
Findings from manual muscle testing.

	On arrival		48 days later	
	Right	Left	Right	Left
Shoulder abduction	4/5	3/5	5/5	4/5
Elbow flexion	0/5	0/5	4/5	3/5
Elbow extension	0/5	0/5	3/5	3/5
Wrist extension	0/5	0/5	3/5	3/5
Wrist flexion	0/5	0/5	3/5	3/5
Grip strength	0/5	0/5	3/5	3/5
Thumb adduction	0/5	0/5	3/5	3/5
Thumb abduction	0/5	0/5	3/5	3/5
Interossei muscles	0/5	0/5	3/5	3/5
Hip flexion	0/5	0/5	3/5	3/5
Hip extension	0/5	0/5	3/5	3/5
Hip adduction	0/5	0/5	3/5	3/5
Hip abduction	0/5	0/5	3/5	3/5
Knee flexion	0/5	0/5	3/5	3/5
Knee extension	0/5	0/5	3/5	3/5
Ankle dorsiflexion	0/5	0/5	3/5	3/5
Ankle plantarflexion	0/5	0/5	3/5	3/5

Discussion

ASAS due to spinal cord infarction is very rare and the diagnosis is challenging when encountered in a previously healthy young patient. Our patient was previously healthy, and extensive investigations including laboratory screening for autoimmune-associated vasculitis, infections, hypercoagulopathies, and CSF analysis identified no risk factors. Severe neck pain 2 days before admission was an alarming sign of ASAS, but it might be impossible to consider ASAS in a patient without any risk factors and MRI did not seem to reveal any abnormalities on admission. Young patients have been described in several previous studies, but those cases mostly involved children, adolescents, or young patients with previous

trauma or angioma.⁷⁻⁹ Emergency physicians must therefore consider ASAS when encountering a patient with sudden onset of quadriplegia, even if the patient is previously healthy and young.

MRI findings in the early stage are usually ambiguous, and correct interpretation is difficult for emergency physicians, but ASAS must be considered once spinal epidural hematoma has been ruled out by CT and MRI. We have previously reported that MRI can detect ASAS using a combination of DWI with ADC mapping in the early stage.⁶

No standardized guidelines have been set for the treatment of ASAS. A previous retrospective study did not show any difference in the clinical course of patients who received antiplatelet therapy, even if they received treatment with corticosteroids or anticoagulation (heparin).¹⁰ We used edaravone for 14 days, heparin for 21 days, aspirin and steroid pulse therapy (methylprednisolone 1000 mg/day for 5 days first and 3 days later) in the early stage. She was able to move her left arm with biceps MMT 4/5, and right arm with biceps MMT 3/5 (Table 1) on hospital day 48, although sensory deficit remained below the T4 level, which was much better than we had predicted. These effects might have been due to early combination therapy with edaravone, heparin, aspirin, and steroid usages. In contrast, no previous ASAS cases used such a combination therapy and neurological outcomes were unfavorable. Long cord lesions, severe initial impairment with weakness and female sex have been reported as predictors of unfavorable outcomes.^{11,12} In our case, the patient had all these unfavorable predictors, but could move her left hand and eat unassisted. However, she could not stand unaided, was wheelchair-bound and was still experiencing bladder dysfunction when transferred from our facility. Symptoms were better than we predicted based on initial MRI showing long cord lesions from C3 to T1 and complete flaccid quadriplegia without pain or temperature sensation.

In conclusion, ASAS can cause sudden onset of neck pain with quadriplegia in a young healthy woman, and DWI of the spine should be considered for early diagnosis of ASAS. Early combination treatment with edaravone, antiplatelet agents, corticosteroids and anticoagulants heparin, and aspirin with steroid pulse might be beneficial for patients with ASAS.

Abbreviations

ADC, apparent diffusion coefficient; ASAS, anterior spinal artery syndrome; CSF, cerebrospinal fluid; CT, computed tomography; DWI, diffusion-weighted imaging; ER, emergency room; ICU, intensive care unit; MMT, manual muscle testing; MRI, magnetic resonance imaging

Declarations

Ethics approval and consent to participate: N/A

Consent for publication: We obtained verbal informed consent from the patient and written informed consent from her husband and father to report this case.

Availability of data and material: The datasets used during the current study are available from the corresponding author on reasonable request.

Competing interests: The authors declare that they have no competing interests.

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Authors' contributions: KoO, SO, YO, MO, NO, YS and HY treated the patient, made the clinical diagnosis, KoO, KaO and AT wrote and revised the manuscript. All authors have read and approved the manuscript.

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Tables

Table 1. Findings from manual muscle testing.

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Wrist extension	0/5	0/5	3/5	3/5
Wrist flexion	0/5	0/5	3/5	3/5
Grip strength	0/5	0/5	3/5	3/5
Thumb adduction	0/5	0/5	3/5	3/5
Thumb abduction	0/5	0/5	3/5	3/5
Interossei muscles	0/5	0/5	3/5	3/5
Hip flexion	0/5	0/5	3/5	3/5
Hip extension	0/5	0/5	3/5	3/5
Hip adduction	0/5	0/5	3/5	3/5
Hip abduction	0/5	0/5	3/5	3/5
Knee flexion	0/5	0/5	3/5	3/5
Knee extension	0/5	0/5	3/5	3/5
Ankle dorsiflexion	0/5	0/5	3/5	3/5
Ankle plantarflexion	0/5	0/5	3/5	3/5

Figures

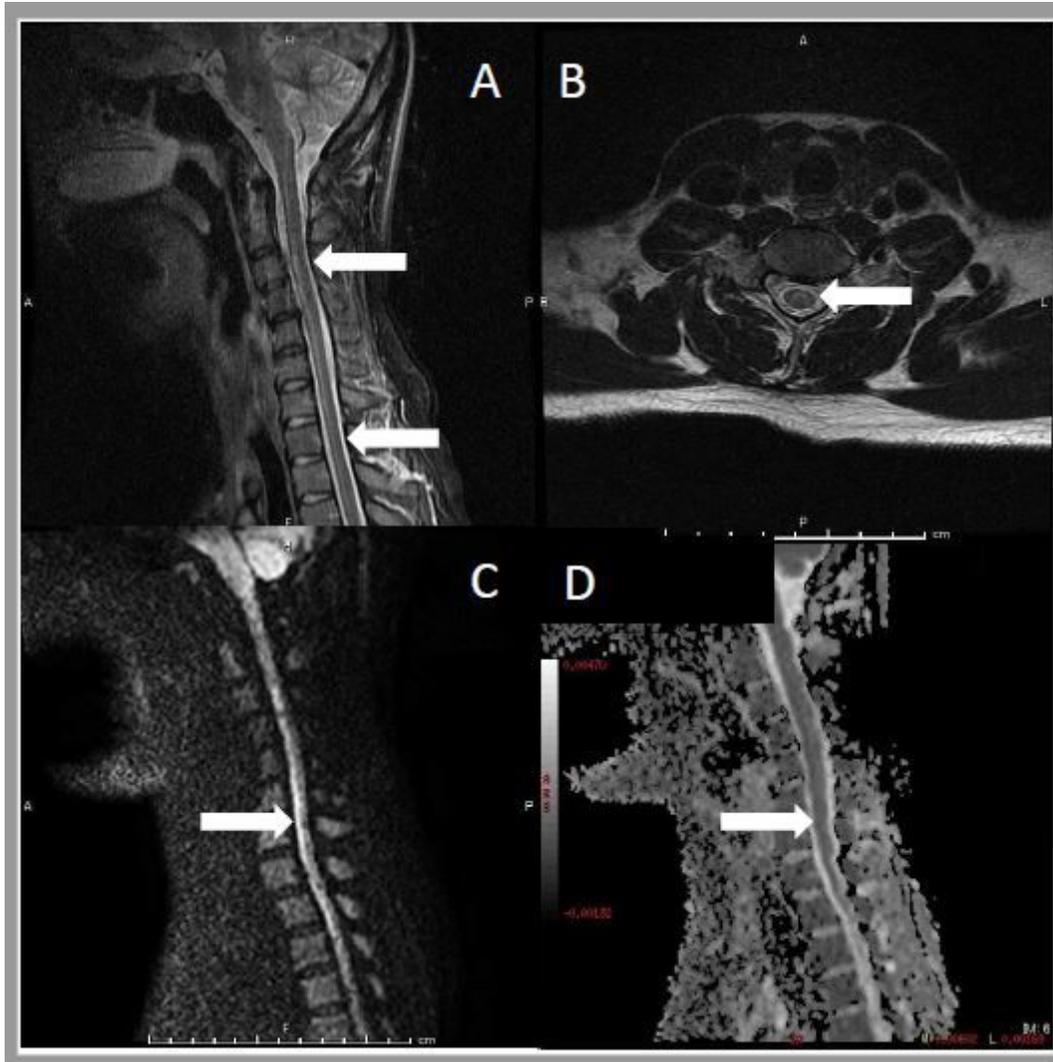


Figure 1

MRI findings on admission and after 3 h. (A) Sagittal T2-weighted imaging (Repetition Time (TR) 3000 ms, Echo Time (TE) 106.20 ms) shows signal hyperintensity from C3 to T1 (arrows). (B) Axial T2-weighted imaging (TR 3116.67 ms, TE 93.16 ms) shows signal hyperintensity with involvement of grey matter and adjacent central white matter (arrow). (C) Sagittal DWI imaging (TR 7000 ms, TE 72.70 ms) shows signal hyperintensity from C3 to T1 (arrow), as in A. (D) Axial ADC mapping (TR 7000 ms, TE 72.70 ms) shows a slightly dark area from C3 to T1 (arrow), as in A and C.

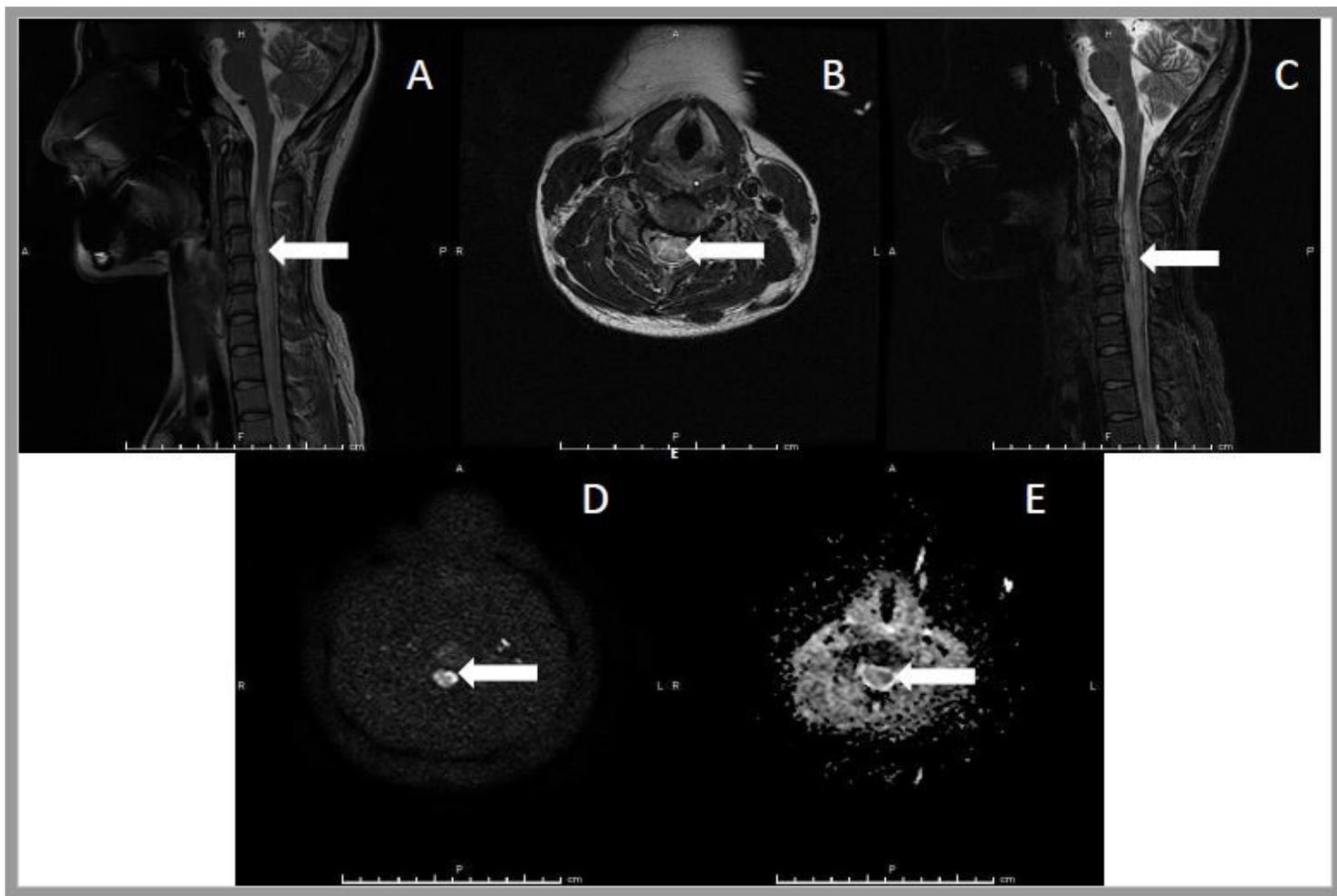


Figure 2

MRI findings on hospital day 13. Signal hyperintensity with spinal swelling from C3 to T1 (arrow) on (A) sagittal T2-weighted imaging (TR 3000 ms, TE 104 ms) image, (B) axial T2-weighted (TR 3716.67 ms, TE 102.58 ms) image (arrow), and (C) sagittal short TI inversion recovery imaging (TR 3500 ms, TE 81.58 ms) (arrow). (D) Axial DWI (TR 6575 ms; TE 79.70 ms) shows signal hyperintensity (arrow) and (E) ADC mapping (TR 6575 ms, TE 79.70 ms) shows a dark area (arrow).

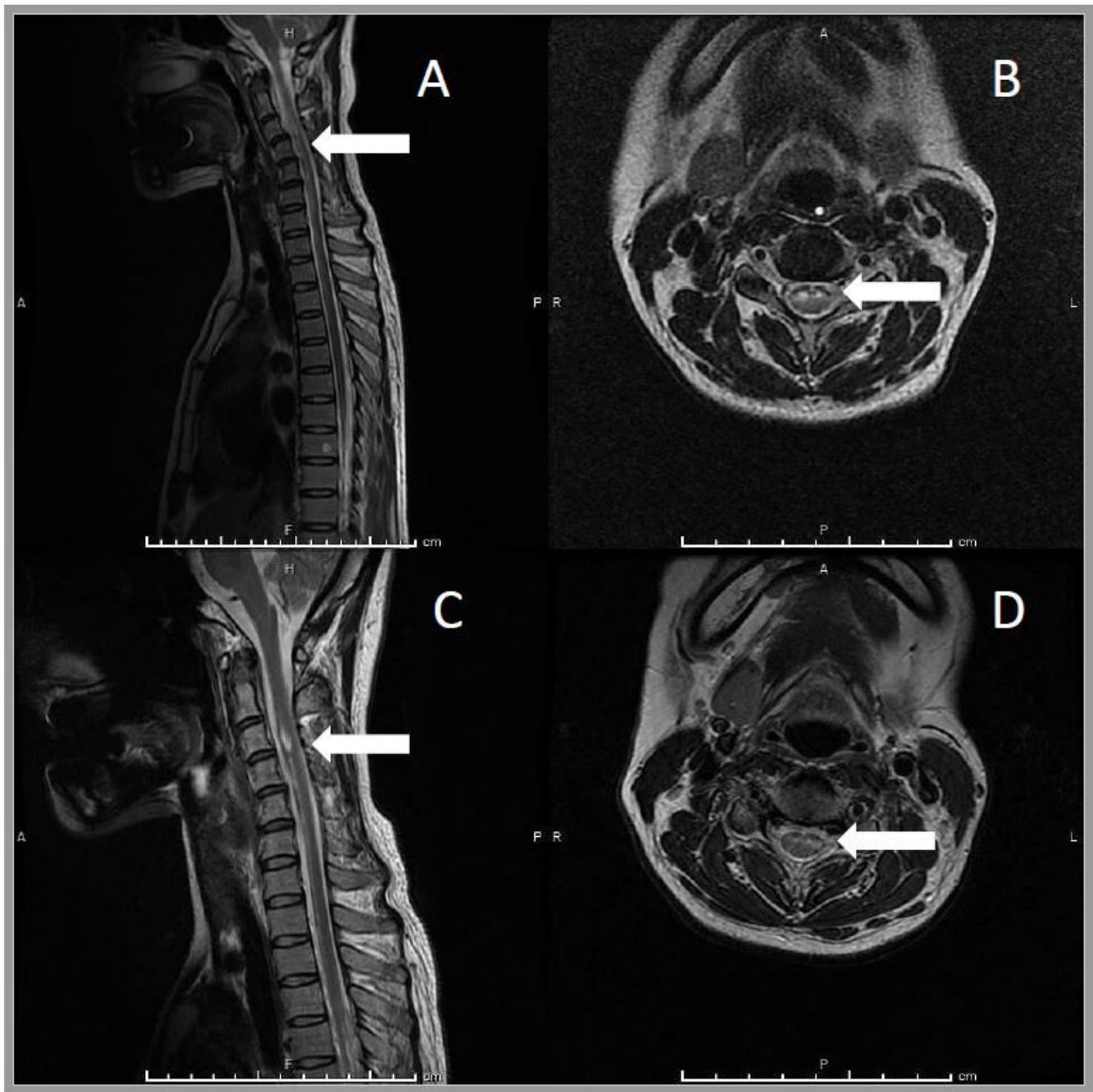


Figure 3

MRI findings on hospital days 18 and 28. (A) Hyperintense signal persists on sagittal T2-weighted (TR 3000 ms, TE 101.64 ms) and (B) axial T2-weighted imaging (TR 4600 ms, TE 104.16 ms) (arrow) on hospital day 18. (C) Signal hyperintensity is reduced on sagittal T2-weighted (TR 3350 ms, TE 84.48 ms) and (D) axial T2-weighted imaging (TR 2700 ms, TE 102.49 ms) (arrow) on hospital day 28.