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CASE REPORT

Spontaneous conus infarction with "snake-eye appearance" on magnetic resonance imaging: A case report and literature review

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Abstract

BACKGROUND

Infarction of the conus medullaris is a rare form of spinal cord infarction. The first symptom is usually acute non-characteristic lumbar pain, followed by lower limb pain, saddle numbness, fecal incontinence, and sexual dysfunction. Spontaneous conus infarction with "snake-eye appearance" on magnetic resonance imaging has rarely been reported.

CASE SUMMARY

We report a 79-year-old male patient with spontaneous conus infarction who had acute lower extremity pain and dysuria as the first symptoms. He did not have any recent history of aortic surgery and trauma. Magnetic resonance imaging revealed a rare "snake-eye appearance." In addition, we reviewed the literature on 23 similar cases and summarized the clinical features and magnetic resonance manifestations of common diseases related to the "snake-eye sign" to explore the etiology, imaging findings, and prognosis of spontaneous conus infarction.

CONCLUSION

We conclude that acute onset of conus medullaris syndrome combined with "snake-eye appearance" should be strongly suspected as conus medullaris infarction caused by anterior spinal artery ischemia. This special imaging manifestation is helpful in the early diagnosis and treatment of conus infarction.

Key Words: Spinal cord disease; Infarction; Paralysis; Dysuria; Magnetic resonance imaging; Case report

Core Tip: Infarction of the conus medullaris is a rare form of spinal cord infarction, and there is no specific examination method in clinic. On the other hand, the "Snake-Eye Appearance" on the diffusion-weighted imaging sequence of magnetic resonance imaging highly suggests spinal cord infarction, although few cases of conus infarction have been reported. This special imaging manifestation is helpful in the early diagnosis and treatment of conus infarction.

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INTRODUCTION

Spinal cord infarctions account for approximately 1.2% of all ischemic strokes[1], and less than 10% of acute non-traumatic myelopathies[2]. The most common infarct sites are the cervical and thoracic segments[3]. The common clinical manifestations of spinal cord infarction are motor and sensory disorders with impaired autonomic nerve function, and the specific manifestations are related to the affected sites[4]. Combined with the observation of the clinical manifestations, magnetic resonance imaging (MRI) and cerebrospinal fluid examination are helpful in distinguishing this condition from optic neuromyelitis pedigree diseases, acute disseminated encephalomyelitis, spinal cord tumors, multiple sclerosis, intervertebral disc herniation, and other spinal cord diseases. However, the lack of specific MRI features in conus medullaris infarction increases the difficulty of early diagnosis and recognition. In this paper, we report a case of spontaneous conus medullaris infarction. Simultaneously providing an analysis of the literature, we focus on the special imaging manifestations and etiological mechanisms of this condition to improve clinicians' understanding of this disease.

CASE PRESENTATION

Chief complaints

A 71-year-old Chinese man with a complaint of sudden onset of pain in both lower extremities and urination disorders for 2 d.

History of present illness

The patient experienced continuous pain on the back of both lower limbs when standing up from the sofa before 2 d, accompanied by muscle spasm, increased pain while sitting, urinary incontinence, and difficulty defecating, but no erectile dysfunction.

History of past illness

The patient had a history of hypertension for 5 years and diabetes for 2 years. In the last year, the patient regular oral administration of Amlodipine besylate tablets was 5 mg qd, and the blood pressure was stable at 17-20/9-11 kPa; Metformin 0.5 g tid and the glycosylated hemoglobin was 6.8%.

Personal and family history

The patient has been smoking for 43 years, about 10 cigarettes a day. The patient denies any family history of disease.

Physical examination

After 48 h of onset, neurological examination showed normal muscle strength and muscle tension in both lower limbs. The bilateral knee reflex was symmetrical, with no hyperactivity and weakness, the bilateral Achilles tendon reflex disappeared, the saddle area sense was normal, anal reflex was weakened, the urinary retention was moderate, and lack of the bilateral Babinski sign.

Laboratory examinations

No obvious abnormalities were found in the test results of perfect nail work, serum immunology, tumor markers, or coagulation images.

Imaging examinations

On the 4th day of onset, lumbar MRI revealed a high signal on the L1 horizontal axial T2-weighted image

with a "snake-eye appearance", and conus infarction was thus considered. On the 5th day of onset, lumbar MRI showed a high signal on L1 horizontal sagittal diffusion-weighted imaging (DWI) (Figure 1).

FURTHER DIAGNOSTIC WORK-UP

Spinal angiography and aortic angiography (CTA) improved, but no definite vascular stenosis or malformation was identified. On the 7th day of onset, the measurement of nerve conduction velocity revealed that the F-wave latency of the left tibial nerve was prolonged.

LITERATURE REVIEW

We used the key terms "conus medullaris infarction," "conus medullaris syndrome," "spinal cord ischemia," "spinal cord infarction," "snake-eye appearance" and "owl-eye appearance" to search the literature on PubMed. We thus identified and summarized 23 cases of conus medullaris infarction from January 1971 to January 2021 (Table 1)[5-21]. The male to female ratio was 1.3:1. The median age at onset was 63 years. Eleven cases (47.8%) were complicated by cardiovascular risk factors, 7 (30.4%) were related to aortic disease, and 3 (13.0%) were secondary to postural changes, similar to the present case. Other causes included dural arteriovenous fistulas, spinal cord venous thrombosis, fibrocartilage embolism, and so on; 9 cases (39.1%) showed a high signal on axial T2-weighted imaging, of which 2 cases showed the typical "snake-eye appearance" high signal on sagittal T2-weighted imaging in the T10 to L1 vertebral segments, 3 cases (13.0%) showed limited DWI, 5 cases (21.7%) involved the vertebral body/muscle/ligament at the same time, and 16 cases (69.6%) showed a partial recovery of neurological deficit, with sequelae of varying degrees, while 5 cases (21.7%) had no improvement, and the overall prognosis was poor.

FINAL DIAGNOSIS

Combined with the patient's medical history, spontaneous spinal cord infarction was confirmed.

TREATMENT

The patient was admitted to the hospital and administered clopidogrel to facilitate antiplatelet aggregation and atorvastatin calcium tablets to reduce blood lipid levels.

OUTCOME AND FOLLOW-UP

After 10 d of treatment, the pain in both lower extremities was relieved, and the symptoms of urinary retention were relieved. After discharge, the patient continued to take clopidogrel and atorvastatin calcium tablets orally. After 3 mo of telephone follow-up, the patient complained of left lower limb pain and prolonged urination time.

DISCUSSION

Conus medullaris infarction is rare, and its incidence is unclear. A study on the clinical and magnetic resonance imaging manifestations and short-term prognosis of patients with spinal cord infarction showed that only 12.5% had isolated conus medullaris infarction[22]. The blood supply to the conus medullaris is very rich and mainly supplied by the anterior spinal artery, posterior spinal artery, and nerve root medullary artery. The anterior spinal artery supplies the first two-thirds of the conus medullaris, and the posterior spinal artery supplies the last one-third. These form a coronary artery ring at the level of the conus medullaris, which then branches from the artery ring into the conus medullaris. In addition, the thick nerve root medullary artery (Adamkiewicz artery) from the intercostal or lumbar artery from T9 to T12 and the desproges gotteron artery originating from the iliolumbar artery are also involved in the blood supply to the spinal conus[6,23-25]. At present, the diagnosis of conus medullaris infarction is mainly based on clinical manifestations and MRI findings. Lumbar puncture cerebrospinal fluid examination is helpful in distinguishing between inflammatory and demyelinating diseases. In

Table 1 Overview of reported cases of conus infarction

Ref.	Age/gender	Risk factors	Pathogeny/mechanism	Prognosis	MRI findings			
					T2WI l high signal (Axial)	T2WI high signal (Sagittal)	DWI high signal	Involve centrum/muscle/ligament
Herrick *et al* [5]	84/M	NA	Aortic dissection aneurysm	Partial improvement, died of rupture of aortic dissection aneurysm on the 18th day of admission	NA	NA	NA	NA
	79/M	Heart failure	Aortic atherosclerosis	No improvement, died of acute myocardial infarction on the 25th day of admission	NA	NA	NA	NA
Anderson *et al* [6]	54/M	Coronary diseaseHeart failure	Aortic balloon pump implantation	Some improved strength in the legs before death 7 wk after the ictus	NA	NA	NA	NA
	75/M	Smoking	Repair operation of abdominal aortic aneurysm	Persistent urinary incontinence with some improvement in bowel function and in motor and sensory signs 16 mo after the ictus	NA	NA	NA	NA
	66/M	Smoking	Aortic atherosclerosis	Some functions recovered 2 mo after the ictus	NA	NA	NA	NA
	51/M	Smoking	NA	Persistent urinary incontinence with some functions recovered 28 mo after the ictus	NA	NA	NA	NA
	47/F	NA	NA	No improvement in 2 yr	NA	NA	NA	NA
Ohbu *et al*[7]	69/F	Hypertension	NA	NA	NA	NA	NA	NA
Andrews *et al* [8]	71/F	NA	NA	Walking independently, mild hypoesthesia, but persistent urinary incontinence 2 mo after the ictus	NA	NA	NA	NA
Mhiri *et al*[9]	28/M	NA	Dural arteriovenous fistula	No improvement	NA	NA	NA	NA
Sinha *et al*[10]	63/M	HypertensionCoronary disease	Coronary artery bypass grafting (CABG)	persistent urinary incontinence 5 yr after the ictus	NA	NA	NA	NA
Greiner-Perth *et al*[11]	66/M	NA	NA	No improvement in 8 mo	NA	T12-L1	NA	NA
Combarros *et al*[12]	69/F	Hypertension	NA	The bladder function returned to normal and can walk with a walker 2 mo after the ictus	NA	NA	NA	NA
Wildgruber *et al*[13]	44/F	NA	Spinal venous thrombosis	Motor function recovered partially and leaving hypoesthesia 6 mo after the ictus	Bilateral anterior horn of gray matter (Snake-eye appearance)	T12-L1	NA	NA
Wong *et al*[14]	79/F	Coronary disease	Aortic atherosclerosis	Partial neurologic recovery	Bilateral gray matter and central white matter	T12-L1	Yes	NA

Konno *et al* [15]	77/F	Hypertension	Spinal venous thrombosis	Symptoms improved rapidly	Diffuse	L1	NA	Yes
Diehn *et al*[16]	24/M	NA	Fibrocartilage embolism	No improvement	Bilateral anterior horn of gray matter	T10-L1	NA	Yes
Alanazy[17]	48/M	NA	Overstretch	Walking resumed on day 105	Diffuse	T11-L1	NA	NA
Hor *et al*[18]	51/F	NA	NA	NA	Bilateral gray matter and central white matter	T12	NA	NA
Kamimura *et al*[19]	70/F	NA	Spinal venous thrombosis	Sensory disturbance improved, leaving numbness in the sellar area and urinary incontinence	Bilateral posterior funiculus, right posterior horn, right lateral funiculus	T12	NA	Yes
Weng *et al*[20]	55/M	Hyperlipidemia	Sofa sedentary	Calf muscle atrophy, perianal hypoesthesia and neurogenic bladder 3 yr after ictus	Bilateral anterior horn of gray matter	T11-12	Yes	Yes
	34/F	NA	Toilet sedentary	Calf muscle atrophy, perianal hypoesthesia and neurogenic bladder 4 yr after ictus	NA	T12	Yes	NA
Breitling *et al* [21]	52/M	NA	NA	Motor function recovered partially, leaving bladder and rectum dysfunction	Bilateral anterior horn of gray matter (Snake-eye appearance)	L1	NA	Yes

DWI: Diffusion-weighted imaging; NA: Not available.

January 2019, Zalewski *et al*[26] proposed the diagnostic criteria for spontaneous spinal cord infarction, emphasizing that the high signal on a MRI intramedullary T2-weighted image is evidence of acute spinal cord infarction, while the DWI/apparent diffusion coefficient diffusion is limited, accompanied by corresponding pyramidal infarction, arterial dissection, or occlusion near the lesion. However, it is important to note that T2-weighted magnetic resonance imaging has a low sensitivity.

In a clinical study[27], only 45% of patients with acute spinal cord infarction showed a high signal on T2-weighted magnetic resonance imaging. The volume of the conus medullaris is smaller, the magnetic sensitivity artifact tendency of DWI is higher, and the detection sensitivity is much lower than those of acute cerebral infarction. Therefore, early neurological function evaluation is very important in identifying patients with negative MRI findings. The MRI results of this patient were consistent with the above standards. It is worth noting that the patient had an isolated "snake-eye appearance" high signal on axial T2-weighted MRI at the level of the spinal cord cone. This "snake-eye appearance" was first described in the results of delayed myelography in 7 patients with compressive cervical spondylotic myelopathy in 1986[28], and is also known as "owl-eye appearance". The main pathological changes which can result in this appearance are cystic necrosis of the central gray matter of the ventrolateral column of the spinal cord and loss of neurons in the anterior horn of the spinal cord[29]. It is usually related to lower motor neuron syndrome, such as Hirayama disease[30,31], spinal muscular atrophy syndrome[32], cervical spondylotic myelopathy[33], amyotrophic lateral sclerosis[34], and anterior spinal artery ischemia[35-37], Detailed identification is shown in Table 2[34,38-41]. There is a watershed area between the sulcus commissural artery and the coronary artery ring sent by the anterior spinal

Table 2 Identification of common diseases related to snake eye sign

Disease	Clinical features	Magnetic resonance performance	Neuroelectrophysiological manifestations
Conus medullaris infarction [38]	The main manifestations are sensory disturbance in the sellar region, bladder and rectal incontinence, bulbar anal reflex weakening or disappearing, erectile dysfunction, root neuralgia and lower limb motor neuron paralysis when combined with cauda equina damage	T12-L1 horizontal magnetic resonance T2WI and DWI high signal, T1W1 low signal	There are few reports about the neurophysiological characteristics of conus medullaris infarction. The reappearance of F wave after infarction may mark the improvement of clinical prognosis
Hirayama disease[39]	The self-limited disease, which is mainly characterized by unilateral muscle atrophy of the distal end of the upper limb, mainly affects the intrinsic muscles of the hand and forearm muscle groups. Typical clinical manifestations also include "cold paralysis", "finger extension tremor" and "muscle bundle tremor"	Asymmetric cervical spinal cord flattening, atrophy and intramedullary T2W1 high signal in cervical flexion position, disappearance of cervical physiological flexion, expansion and increase of epidural venous plexus, and anterior displacement of dural sac after over-extension and over-flexion position	The neurogenic damage of the affected muscle group mainly occurred in the C7-8 sarcomere and T1 sarcomere, while the C5-6 sarcomere (i.e. deltoid, biceps brachii and radial brachii) was not affected
Amyotrophiclateralsclerosis [34]	Malignant degenerative motor neuron disease characterized by multiple or localized progressive muscular atrophy and apraxia is characterized by limb spasms, tendon hyperreflexia, localized or multiple muscle weakness, muscular atrophy and fascicular tremor	T2WI, FLAIR and DWI can find symmetrical high signal in the pyramidal tract of the brain. In a few patients, SWI can see the deposition of hemosiderin along the motor cortex	The muscles innervated by different nerve segments of medulla oblongata, neck, chest and lumbosacral appear progressive denervation and chronic nerve regeneration
Cervical spondylotic myelopathy[40]	Based on cervical degeneration, the main manifestation is atrophy of the proximal or distal muscles of the upper limb, which usually occurs in one side, usually without sensory abnormalities	It is usually manifested as spinal cord thinning, intervertebral disc protrusion or prolapse. Due to long-term compression of the spinal cord, venous hyperemia and infarction can be caused, which can be secondary to cystic necrosis of the anterior horn of the spinal cord, forming T2WI snake-eye sign	Segmental neurogenic damage consistent with the distribution of the injured nerve root
Spinal muscular atrophy syndrome[41]	The most common autosomal recessive disease in childhood is progressive and symmetrical weakness and atrophy of limbs and trunk muscles	Anterior horn of spinal cord α-Degeneration and degeneration of motor neurons led to T2WI snake-eye sign	Typical neuronal damage, fiber fibrillation wave and positive sharp wave can be seen at rest, bundle fibrillation potential can be seen occasionally, and regular spontaneous motor unit activity potential is the characteristic manifestation of its EMG

DWI: Diffusion-weighted imaging; EMG: Electromyography; FLAIR: Fluid-attenuated inversion-recovery; SWI: Susceptibility weighted imaging; T2WI: T2-weighted imaging.

artery. The anterior horn cells of the spinal gray matter in this area are highly sensitive to hypoxia; when local or overall perfusion is insufficient, they are prone to degeneration and necrosis, forming a "snake-eye appearance" high signal limited to the anterior horn of spinal gray matter on the T2-weighted image of the magnetic resonance imaging axis[42,43]. When this "snake-eye appearance" appears during acute onset myelopathy, a vascular origin should be highly suspected. The most commonly used method is aortic CTA. Thoracic and abdominal CTA can help detect aortic atherosclerotic plaques, dissection, aneurysm, and mural thrombosis. If the CTA result is negative, spinal angiography is necessary to exclude dural arteriovenous fistula and spinal intramedullary arteriovenous malformation[4]. Wildgrube *et al*[13] previously reported a case of conus medullaris infarction with "snake-eye appearance", caused by spinal cord venous thrombosis. Thus, it is necessary to evaluate thrombophilic factors and improve spinal cord angiography to distinguish between venous and arterial conus medullaris infarction. The cell number and IgG index of cerebrospinal fluid in patients with spinal conus infarction are usually normal. There is no oligoclonal band, and the protein content in cerebrospinal fluid can be slightly increased in some patients[6-7,20,26]. A previous study[44] proposed two mechanisms of spinal cord infarction: (1) Infarction triggered by mechanical factors (bilateral anterior or posterior spinal cord artery infarction and unilateral infarction); and (2) infarction caused by long-term hypotension or arterial insufficiency (central spinal cord artery infarction and transverse spinal cord infarction). In this case, the patient was associated with cardiovascular risk factors, such as hypertension, diabetes, and smoking, but had no definite history of trauma before disease onset. However, upon presentation, he complained of suffering from bilateral lower extremity pain after completing mechanical action from the sofa, and urination disorder was observed. We speculated that

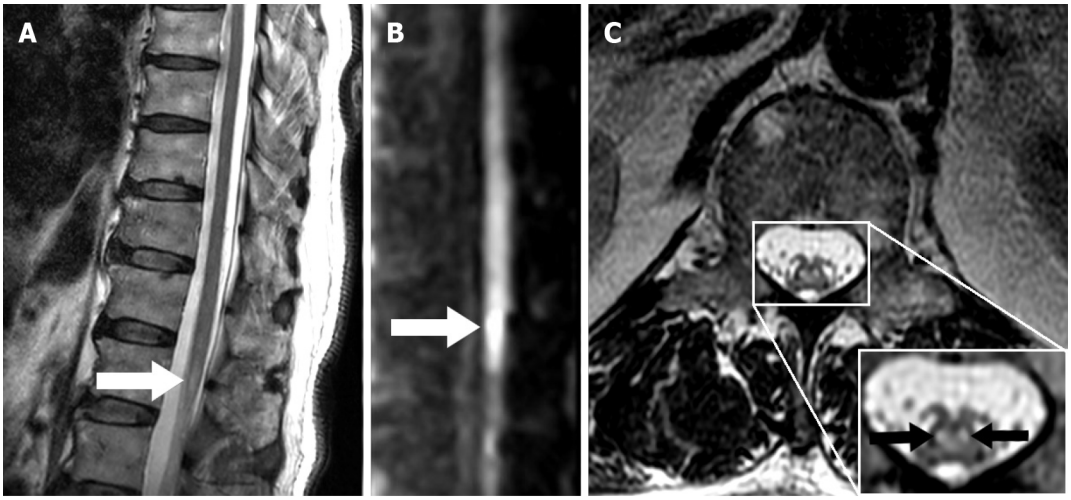

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Figure 1 Conus medullaris magnetic resonance exam and "snake-eye appearance". A: The arrow shows conus infarction on T2 weighted magnetic resonance imaging (MRI) (sagittal position); B: The arrow shows conus infarction on diffusion-weighted imaging image (sagittal position); C: The arrows show "snake-eye appearance" on T2 weighted MRI (axial position).

the possible mechanism was based on atherosclerosis in the anterior spinal cord. Mechanical stress can lead to anterior spinal artery ischemia. At present, the treatment principles for spinal cord infarction mainly refer to the guidelines for acute ischemic stroke. Old age, severe initial neurological deficit, and long segment lesions are considered to be related to poor prognosis[22,27,45,46]. The prognostic value of the "snake-eye appearance" on magnetic resonance imaging in acute myelopathy is unclear, but may be related to the poor prognosis of chronic myelopathy[29,47,48].

Conus medullaris infarction is rare in the clinic and has a high misdiagnosis rate. Detailed medical history and physical examination were the basis of the diagnosis. Although the "snake-eye appearance" is not specific to spinal cord MRI, acute low back pain or bilateral lower limb pain is usually the first symptom, and its clinical manifestation is conus syndrome or cauda equina syndrome. When the axial T2-weighted image of MRI shows "snake-eye appearance", it is necessary to differentiate between spinal conus infarction caused by anterior spinal artery ischemia. Improving aortography, spinal angiography, and cerebrospinal fluid examination will help to clarify the etiology.

The limitations of this report are its short follow-up period and lack of imaging and neurophysiological evaluation results during the follow-up period. Although we have reviewed previously reported cases of conus medullaris infarction, authors may have different descriptions of clinical characteristics and results. More cases need to be analyzed in the future to improve clinicians' understanding of this disease.

CONCLUSION

We conclude that acute onset of conus medullaris syndrome combined with "snake-eye appearance" should be strongly suspected as conus medullaris infarction caused by anterior spinal artery ischemia. This special imaging manifestation is helpful in the early diagnosis and treatment of conus infarction.

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