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Case Report

“Dynamic” Rotational Canal Stenosis Caused by Osteoma of the Atlas: A Case Report and Review of Literature

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The upper cervical canal stenosis is relatively rare compared to other cervical regions. We report a rare case of upper cervical canal stenosis caused by osteoma of C1 lamina related to dynamic factor of cervical rotation. A 43-year-old woman had a 2-year history of numbness and pain in the right hand. Because of aggravation of the numbness and loss of the fine movement and strength in the right hand, she visited our outpatient clinic. Computed tomographic (CT) scan revealed an ovoid bony lesion at the right side of the C1 lamina. And magnetic resonance (MR) imagings of the cervical spine showed intramedullary high intensity signals in T_2-weighted imaging at a site slightly distant from the bony lesion. Rotational dynamic myelo-CT scan was performed because aggravation of the radiating pain was observed with neck rotation to the right. Dynamic CT scan of the craniocervical junction with neck rotation to the right revealed that the bony lesion was moved to the dorsal side and posteriorly compressed the spinal cord. The symptoms were relieved following surgical removal of this bony lesion. The histopathological examination was compatible for osteoma. The dynamic rotational factor for cervical canal stenosis should be taken in consideration, especially in dealing with upper cervical lesions.

Keywords: atlas, osteoma, cervical canal stenosis, dynamic myelo-computed tomography, myelopathy

Introduction

The upper cervical spine is an uncommon site for cervical canal stenosis, because the upper cervical spinal canal is wider compared to the middle and lower parts of the cervical region.[1] In this article, we report a rare case with canal stenosis caused by osteoma of the atlas. Cervical canal stenosis related to the atlas has been associated with ossification of its transverse ligament,[2,3] hypoplasia of the atlas,[4–13] atlantoaxial instability,[11,12] and genetic disorders including Down syndrome,[12,13] Klippel–Feil syndrome,[14] and Williams syndrome.[15] On the other hand, canal stenosis caused by neck rotation has been reported in correlation with the hypertrophic atlantal anterior arch[16] and bilateral C-2 nerve root tumors.[17]

However, to our knowledge, there have been no previous reports of the present case in which the canal stenosis was caused by an osteoma located at the lamina of the atlas.

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Case Report

A 43-year-old woman had a 2-year history of numbness and pain in the right hand. She had no congenital abnormalities of the upper cervical spine such as basilar invagination, os odontoideum, and Klippel–Feil syndrome, or history of trauma prior to the onset of such symptoms. Because of aggravation of the numbness, disturbance of fine movement and weakness in the right hand, she visited our outpatient clinic. On her first visit, her manual muscle test score was 4 in the right upper and lower extremities with bilateral patellar hyperreflexia. Sensory disturbance was found in the occipital region, right forearm, and palm. Moreover, rotation of the neck to the right aggravated the radiating pain.

Lateral cervical spine radiograph showed radiodense bony mass at the posterior part of the atlas (Fig. 1). There was no atlanto-axial dislocation and the sagittal diameter of spinal canal at C1 level was 18 mm within the normal range.[18,19] Computed tomography (CT) scan showed a bony lesion on the right side of the C1 lamina which moderately protruded into the spinal canal (Fig. 2A, B). Although the lesion did not cause severe cord compression in the neutral position, intramedullary signal change on T_2-weighted magnetic resonance imaging (MRI) was observed (Fig. 3A, B). The intramedullary signal change occurred at a site distant from the bony lesion.

Fig. 1 Lateral cervical spine radiograph showing radiodense bony mass (asterisk) located at the posterior part of C1.

Rotational dynamic myelo-CT was performed after obtaining informed consent from the patient. Myelo-CT scan revealed that the ovoid bony lesion located at the right dorsal side of the spinal cord in the neutral position (Fig. 4A), which moved to the dorsal side caused compression of the spinal cord from behind when the neck was rotated to the right (Fig. 4B). Compression of the spinal cord decreased when the neck was rotated to the left (Fig. 4C). The site of the cord signal change on T_2-weighted MRI was concordant with the most compressed site of the spinal cord on dynamic myelo-CT scan.

She underwent removal of this bony lesion. The histopathological study revealed the lesion was composed of mature, dense, compact, cortical-like, lamellar bone, and a diagnosis of osteoma was made (Fig. 5). The symptoms relieved postoperatively, and the severe radiating pain caused by rotation of the neck also disappeared. Postoperative CT scan (Fig. 6A, B) and MRI (Fig. 7A, B) documented total removal of the bony lesion.

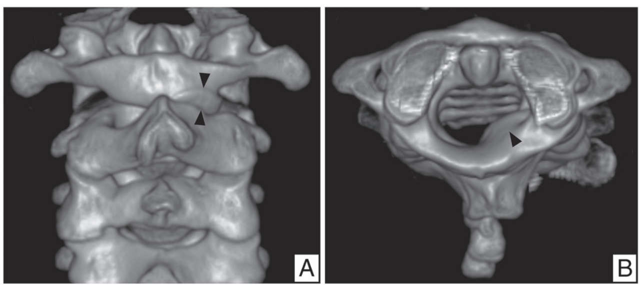

Fig. 2 Preoperative three-dimensional cervical computed tomography scan showing bony lesion (arrowheads) on the right side of the C1 (A) and the bony lesion (arrowhead) protruded into the spinal canal (B).

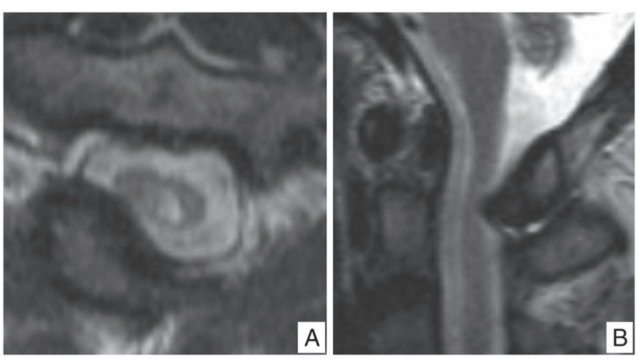

Fig. 3 Preoperative axial (A) and sagittal (B) T_2-weighted magnetic resonance image showing cord compression and intramedullary signal change at the level of the C1.

Discussion

Cervical canal stenosis usually occurs below C3, and rarely occurs above C2 level.[20] The spinal canal at C1 level is relatively wide compared to the middle and lower spinal canal;[18,19] therefore canal stenosis at C1 level is considered to be rare.

It is important to consider not only the static compression factors such as degenerated osteophyte and intervertebral discs, but also dynamic compression factors associated with neck rotation as well as neck flexion and extension especially in dealing with canal stenosis in the upper cervical region, because the mean rotation angle at C1–2 is up to about 80º in the neutral position.[21]

Rotational myelo-CT scan was performed with suspicion of the upper cervical lesion because no obvious cause was apparent in CT scan or MRI in the neutral position. Obviously, rotational MRI can be also useful in elucidating the mechanism, but we abandoned obtaining dynamic MRI because the patient could not endure in a posture of neck rotation to the right for a prolonged time because of the severe radiating pain.

In previous reports, canal stenosis at the level of the atlas was diagnosed by dynamic myelo-CT scan,[16] dynamic MRI,[22] or both of them[17] when no cause for myelopathy was detected with CT scan or MRI in the neutral position. Recent study reported that MR myelography can achieve a reliable diagnosis of lumbar spinal stenosis compared with CT myelogram.[23] However, CT myelography is a valuable investigative examination,[24] and it is considered to be useful to demonstrate the cause of canal stenosis with bony lesion like the presented case.

Besides, osteomas are benign, bone-forming tumors generally arising from the craniofacial skeleton. Osteomas located in the vertebral column are rare and the tumors involved the spine can be complicated by spinal cord or nerve

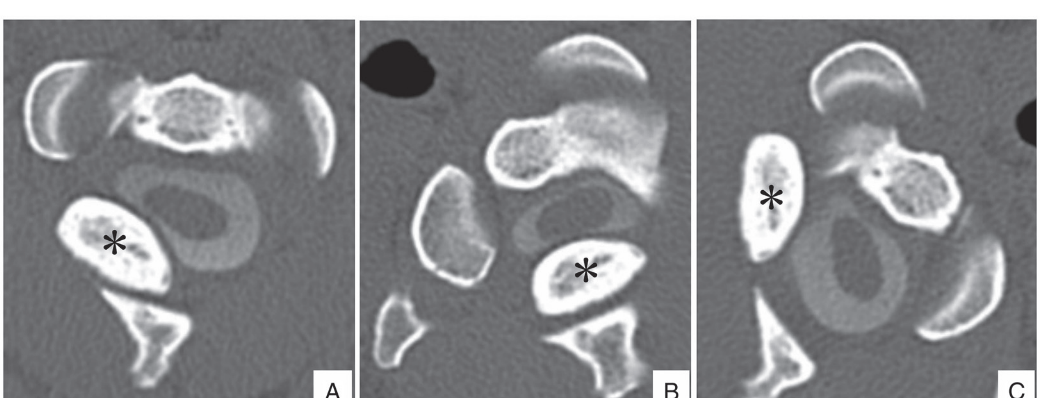

Fig. 4 Axial dynamic myelo-computed tomography scan showing an ovoid, radiodense, sharply-marginated bony lesion (asterisk) located at the right dorsal side with neutral position (A), moving to the dorsal side and compressing the spinal cord with right rotation of the neck (B). No compression of the spinal cord with left rotation of the neck (C).

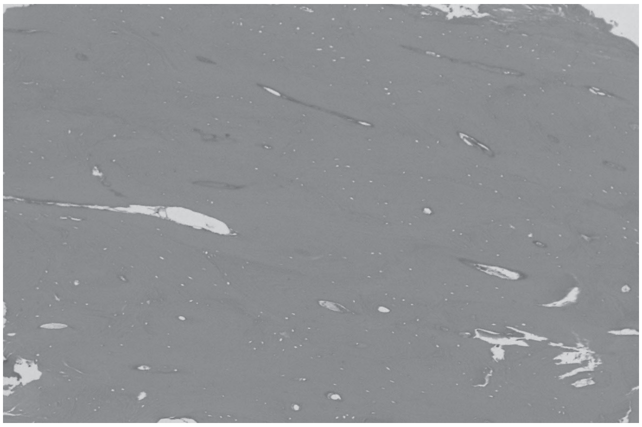

Fig. 5 Photomicrographs of hematoxylin and eosin staining showing surgical specimen composed of mature, uniform dense, compact, cortical-like, lamellar bone. Original magnification ×40.

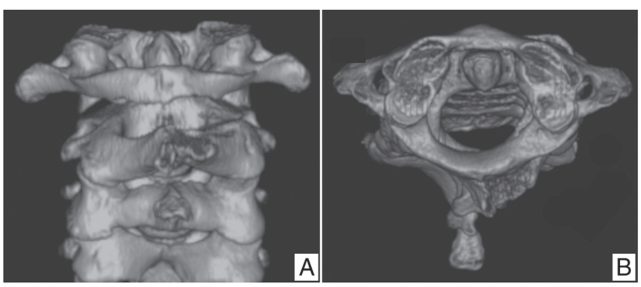

Fig. 6 Postoperative three-dimensional cervical computed tomography scan revealing removal of the bony lesion (A) and no bony lesion protruded into the spinal canal (B).

compression.[25] Among 10 cases of osteomas affecting the spine which have been reported in the English literature (Table 1),[26–30] five cases showed cervical spinal osteomas;[26–28,30] however, there was no report about osteoma located in the altas. Three cases of spinal osteomas, located in the cervical region were discovered by traumatic spinal cord injury.[28,30] Osteomas located in the cervical region can be a cause of traumatic spinal cord injury and symptomatic osteomas are considered for treatment.

In conclusion, we have reported a rare case of upper cervical canal stenosis caused by dynamic factor of cervical rotation. As far as we reviewed, this is the first case of C1 canal stenosis caused by osteoma at C1 lamina. Dynamic cervical canal stenosis is a considerable factor associated with spinal lesions at the atlas.

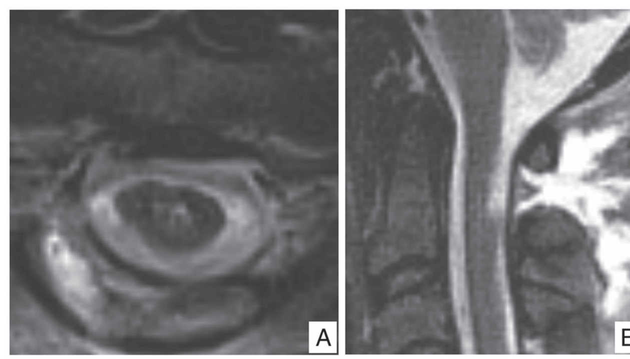

Fig. 7 Postoperative axial (A) and sagittal (B) T_2-weighted magnetic resonance image showing no cord compression.

Table 1 Reported cases of spinal osteomas

First author, year	Age/Sex	Location	Symptoms	Symptoms related to trauma	Treatment	Outcome
Laus et al. (1996)[26]	53/M	C2–3 transverse process	Dysphagia	Unknown	TR	Resolved
Peyser et al. (1996)[27]	44/F	L4 body	Low back pain	Yes	TR	Resolved
	64/F	C4–6 body	Chronic neck pain, weakness, paresthesia	No	TR	Resolved, but pain recurred
	68/F	Ala of sacrum	Chronic low back pain	No	TR	Resolved, but pain recurred
	43/F	S2 body	Low back pain	No	STR	No change
	63/F	L5 body	Chronic low back pain, sciatica	Yes	TR	Resolved
Rengachary et al. (1998)[28]	34/M	C6 pedicle	Weakness, numbness	Yes	TR	Resolved
Kobayashi et al. (2006)[29]	57/M	L5 articular process	Chronic low back pain, sciatica	No	TR	Resolved
Wang et al. (2006)[30]	56/M	C2–3 body	Weakness, hypoesthesia	Yes	STR	Improved
	49/M	C2 lamina	Weakness, hypoesthesia	Yes	TR	Improved
Present case	43/F	C1 lamina	Neck pain, weakness, numbness	No	TR	Improved

F: female, M: male, STR: subtotal resection, TR: total resection.

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Conflicts of Interest Disclosure

All authors have no conflicts of interest in relation to this research and its publication.

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