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Case Report

Spontaneous Ligamentum Flavum Hematoma in the Rigid Thoracic Spine : A Case Report and Review of the Literature

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Ligamentum flavum hematoma is a rare condition. Twenty cases including present case have been reported in English-language literature. Among them, only one case reported in pure thoracic spine. A 72-year-old man presented with thoracic myelopathy without precedent cause. Magnetic resonance images revealed a posterior semicircular mass which was located in T7 and T8 level compressing the spinal cord dorsally. T7-8 total laminectomy and extirpation of the mass was performed. One month later following surgery, the patient fully recovered to normal state. Pathologic result was confirmed as ligamentum flavum hematoma. Ligamentum flavum hematoma of rigid thoracic spine is a very rare disease entity. Most reported cases were confined to mobile cervical and lumbar spine. Surgeons should be aware that there seems to be another different pathogenesis other than previously reported cases of mobile cervical and lumbar spine.

KEY WORDS : Ligamentum flavum · Hematoma · Thoracic spine · Myelopathy.

INTRODUCTION

Myelopathy or nerve root compression induced by ligamentum flavum hematoma (LFH) are extremely rare. Hypertrophy, calcification, ossification and infolding happen more frequently than hemorrhage because ligamentum flavum lack of blood vessels. It serves to bridge the spaces between the laminae of adjacent vertebrae from cervical to the lumbosacral interval[5]. Therefore, LFH may occur in any places from cervical to sacral vertebral column. But, most cases of LFH were reported in the lumbar area. Only one case in pure rigid thoracic spine was reported in the English-language literature[12]. Here the authors describe another case of thoracic LFH.

CASE REPORT

History and Presentation

A 72-year-old man who had been suffering from gait disturbance for 1 month, presented with a 1-day history of abrupt progressive paraparesis and hypoesthesia of the bilateral lower extremities. He had no medical illness such as diabetes mellitus, hypertension and other cardiac disease. He had not been treated with oral anticoagulant or antiplatelet agents. Also, the patient had no recent history of blunt trauma to his abdomen or back. Twenty years prior to presentation, he suffered from mild back pain following fall-down injury. The patient was unable to ambulate without a wheel-chair when his first visit to our outpatient clinic. During 1-month before visit to our clinic, he had been treated with herb medicine and acupuncture by a doctor of oriental medicine under the impression of cerebrovascular accidents. Despite of this alternative medicine, his symptoms were worsened. At last, the day of visit to our hospital, he felt abrupt aggravation of his lower legs weakness.

Examination

On neurologic examination, the motor power of both lower legs was grade II and hypoesthesia below T-10 dermatome was detected. Deep tendon reflexes of both knee and ankle jerks were markedly increased and Babinski signs and ankle clonus were also noted. Intrinsic and extrinsic anal tone were totally relaxed and perianal sensation was lost. But, bulbocavernous reflex was still preserved. Plain X-ray of the thoracolumbar spine revealed no definite abnormal

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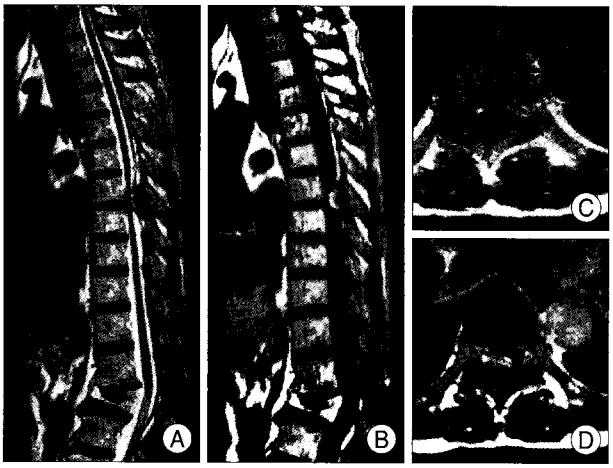

Fig. 1. Magnetic resonance images demonstrating a semicircular extradural mass lesion of the posterior part at T7-8 level with spinal cord compression and central hyperintense signal on the both T1 and T2 weighted images (A : T2 sagittal, B : T1 sagittal, C : T2 axial, D : T1 axial).

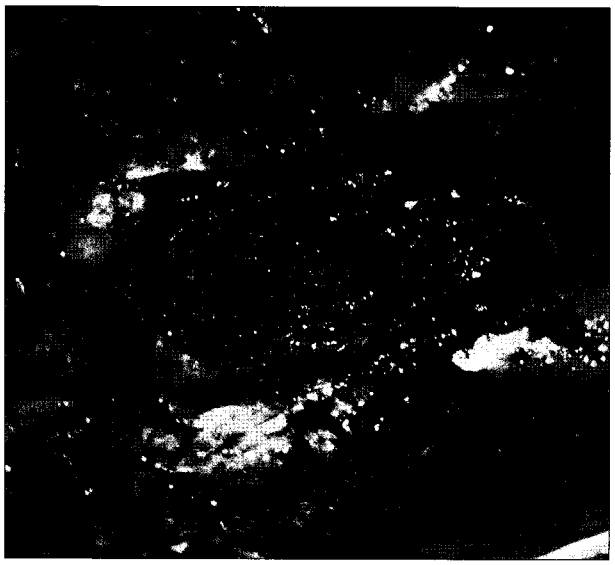

Fig. 2. Appearance of the ligamentum flavum hematoma. Subacute staged-hematoma and thickened dark brownish ligamentum flavum is seen following T7 and T8 total laminectomy.

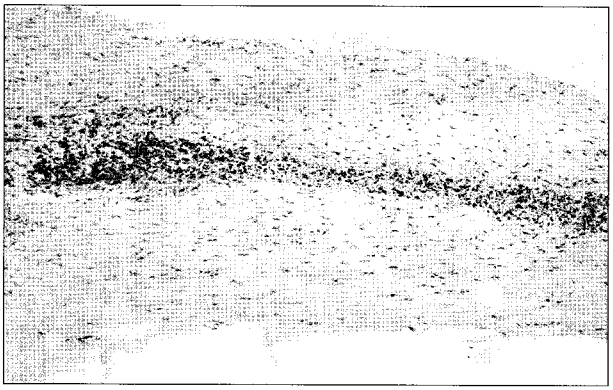

Fig. 3. Photomicrograph showing degenerated ligamentum flavum with loss of elastic fiber including granulation tissue, hemosiderin pigmentation and hemorrhage. Hematoxylin and eosin stain, original magnification, A×100.

findings except old L1 compression fracture which was suspected to be occurred in 20 years ago. The compression fracture was anterior wedge-shaped and compression rate was about 45% and kyphotic angle was about 27 degrees. Magnetic resonance images (MRI) revealed an intraspinal, extradural space-occupying lesion at T7-8 disc space level. This mass lesion was based on the inner surface of T7 laminae and semicircular shape. T2-weighted MRI revealed round low signal intensity rim and high signal intensity in the center. The thick low-signal intensity area was particularly located on the inner surface of T7 laminae and this area was iso-signal to normal ligamentum flavum (Fig. 1A, C). T1-weighted MR images also revealed mixed high and iso-signal intensity and thickened ligamentum flavum (Fig. 1B, D). Contrast-enhanced MR images were not able to be obtained. This mass compressed spinal cord severly and filled entire spinal canal. As shown in the T1-weighted MRI, this lesion was suggestive of hemorrhage.

Operation

The patient underwent emergency operation and was placed prone position. An midline skin incision was done and total laminectomy of T7 was performed with use of high-speed drill to avoid additional cord injury. After removal of entire laminas, thickened dark-brownish ligamentum flavum was encountered in the operative field. It was tough and dark-colored compared to normal ligam-entum flavum. After total removal of ligamentum flavum, subacute hematoma was identified beneath the ligament flavum and chronic organized hematoma was located on the posterior surface of dural sac (Fig. 2). Spinal cord was severely compressed by these hematomas. There were no tumorous or abnormal vascular lesions, ossification and calc-ification. Total hematoma removal was performed and blood tinged-ligamentum flavum was removed untill normal yellow ligament was appeared. Toward the end of the operation, compressed dural sac was restored to original shape.

Pathologic Examination

Histologic findings of the present case were remarkable loss of elastic fibers and intraligamental hemorrhages. We observed granulation tissue with hemosiderin pigmentation. There was no evidence of neoplasm or infection. The histologic findings confirmed diagnosis of a LFH (Fig. 3).

Postoperative Course

Following operation, the patient regained his normal power of bilateral lower legs. The bowel and bladder functions were fully recovered. Four weeks after surgery, the patient had no neurologic abnormalities.

DISCUSSION

Ligamenta flava are the longest of the human ligamentous structures in length, which begins from the second cervical to lumbosacral vertebrae. Its fibers are almost in vertical arrangement and are attached to the ventral surface of the cephalad lamina and to the superior lip of the suprajacent one. Ligamentum flavum consists of elastic fibers (80%) and collagen (20%) and serves as assistance in the maintenance of the erect posture and keeper of the ligament taut during extension[5]. Because ligamentum flavum has abundant in motion, any laxity would permit redundancy and infolding toward the ventrally related structures, as occurs in degenerative lumbar spinal stenosis. Other stenotic lesions which induced by ligamentum flavum are calcifi-cation, ossification and ganglion or synovial cyst[14]. Because ligamentum flavum is poorly vascularized and only a few small vessels pass through it, intraligamentous bleeding is very rare phenomenon[17].

The twenty cases of LFH, including the present case have been reported and are described in Table 1[1-2, 4, 6-13, 15-19]. There are some striking features in these cases. The authors wish to emphasize the following aspects : (1) the patients were relatively older age (average age : 62.9, range : 30-83) and the almost all patients except three cases were more than 60 years old, (2) male patients had more higher incidence rate (male : female=14 : 7) (3) the most frequently involved spinal segment was lumbar area(cervical : 2, rigid thoracic : 2, mobile thoraco-lumbar : 2, lumbar : 14), (4) the onset of symptom was insidious and clinical course was progressively worsened, (5) the most patients experienced minor repeated trivial back injury during daily living lives or sports activities before admission, and (6) all the patients underwent surgery and clinical outcomes and their prognoses were very good following surgery.

From above mentioned characteristics, several features could be extracted. First, LFH occurs in mainly over middle-aged male patients. These epidemiological factors suggest that LFH is a phenomenon associated to age-related ligamental degeneration rather than hypertrophy which generally seen in spinal degenerative diseases. This fact could be identified from the reported pathologic results. The common findings suggested of ligamental degeneration were dispersed ligamental elastic fibers, increased collagen tissues, granulation of fibrous tissues, lymphocyte infiltration and small capillary proliferation[8, 16,18].

Second, more than half of the patients had precipitating factors. These noticable causative factors were repeated trivial abdominal or back trauma which performed during daily living life or sports activities. Coughing and sneezing which could induce increased intraabdominal pressure can also be included in such factors. Increased abdominal pressure transmitted to epidural space resulted in spinal epidural hypertension, which could induce bleeding of

Table 1. Reported Cases of Ligamentum Flavum Hematoma

Case No.	Author	Reported Year	Sex/ Age	Level	Chief Complaint	Pre-admission interval	Precipitating Cause	Treatment	Outcome
1	Sweasey et al[17]	1992	M/43	L4-5	Back & leg pain	2 months	Minor back injury	Operation	Good
2	Sweasey et al[17]	1992	M/60	L2-3	Leg pain & weakness	3 weeks	Stepped up on to box	Operation	Good
3	Cruz-Conde et al[4]	1995	M/67	L4-5	Leg pain	Unknown	Physical exertion	Operation	Good
4	Minamide et al[10]	1999	M/76	L3-4	Leg pain	1 month	Stood upon a train seat	Operation	Good
5	Mahallati et al[9]	1999	M/30	L4-5	Leg pain	5 months	Physical exertion	Operation	Good
6	Yuceer et al[19]	2000	M/67	L2-3	Leg pain	6 weeks	Repaired a car	Operation	Good
7	Hirakawa et al[6]	2000	M/50	L4-5	Low back pain	7 months	Fall-down injury	Operation	Good
8	Maezawa et al[8]	2004	M/66	T11-12	Epiconus syndrome	2 months	Unknown	Operation	Fair
9	Yamaguchi et al[18]	2005	M/62	L3-4	LBP & Gait difficulty	1 month	Repeated minor back injury	Operation	Good
10	Chen et al[2]	2005	M/72	C3-4	Neck & arm pain, weakness	2 months	Unknown	Operation	Good
11	Mizuno et al[13]	2005	F/45	L4-5	Leg pain & weakness	3 months	Lifted a table	Operation	Good
12	Miyakoshi et al[12]	2005	F/66	T9-10	Leg weakness & numbness	2 months	Unknown	Operation	Good
13	Miyakoshi et al[11]	2006	M/67	C3-4	Weakness & numbness	2 weeks	Cervical traction therapy	Operation	Fair
14	Albanese et al[1]	2006	F/70	L1-2	Leg pain & weakness	4 months	Unknown	Operation	Good
15	Keynan et al[7]	2006	F/75	L3-4	Back & leg pain	6 months	Unknown	Operation	Good
16	Shimada et al[15]	2006	F/83	L2-3	Leg pain	2 months	Anticoagulant & antiolatelet agents	Operation	Good
17	Spuck et al[16]	2006	F/64	L2-3	Neurogenic claudication	2 weeks	Unknown	Operation	Good
18	Spuck et al[16]	2006	M/62	L4-5	Leg pain & weakness	6 weeks	Sport exercise	Operation	Good
19	Spuck et al[16]	2006	M/60	T12-L1	Thigh pain	Unknown	Unknown	Operation	Good
20	Present case		M/72	T7-8	Paraparesis	1 month	Unknown	Operation	Good

degenerated ligamentum flavum. This bleeding occurs in proliferating small blood vessels which are frequently seen in ligamental degeneration and these vessels are very small, thin-walled and irregularly dispersed in degenerated ligamentum flavum and are easily broken-down result in LFH[12]. Anatomically, spinal epidural veins lack of valves, thus are easily permitting flow in both directions. Thus, blood from inferior vena cava may be shifted into the vertebral plexus when increased intra-abdominal pressure by physical activity, straining or trauma. Also, the spinal epidural veins receive blood from the intracranial sinuses and veins. Because of these reasons, bleeding from degenerated ligamentum flavum may occur[1].

Third, LFH takes a long clinical course rather than acute spinal epidural hematoma. Preoperative MR images and operative findings of the present case suggested and confirmed that this ligamental hematoma was repeated bleeding and had multi-staged hematoma. Other reported cases also represented similar findings. While acute spinal epidural hematoma have same staged-hematoma (acute or subacute), rapid clinical course and no capsule surrounding hematoma, LFH have opposite features. These are similar to the phenomenon which occur in intracranial chronic subdural hematoma. Minimal and repeated spinal trauma could induce a partial tear in a degenerated ligament and favor intraligamentary hemorrhage. By means of fibrinolytic/hemolytic changes, the hemorrhage would increase significantly in volume inside the ligamentum flavum, causing neural compression. This can explain the insidious onset and progressive clinical worsening of symptoms in the patients of LFH[1]. Also, most patients might have minor trivial back or abdominal injury before symptom onset, but did not remember or forget the time of injury, because there were time interval between trauma and appearance of symptom. Therefore, the patients did not regard that injury as causating and triggering event[3,17,18].

Forth, almost of all cases were seen in mobile lumbar and cervical spine. Only two cases including present case occurred in rigid thoracic spine which is supported by strongly built rib cage[12]. A total of four thoracic cases including present case were reported. Two cases happened in mobile thoracolumbar area and only one case in rigid thoracic spine[2,8,16]. Present case was the second case which occurred in rigid thoracic spine. According to above mentioned mechanisms, cervical and lumbar spine prone to receive pressure transmission to epidural space because they have abundant spinal mobility. But, thoracic spine hardly receives this pressure transmission because it lack of spinal motion. There may also be different mechanisms. In reviewing previous reports, Miyakoshi who was the first reporter of pure rigid thoracic LFH suggested fresh opinion that sagittal imbalance of spine might be involved in this phenomenon[12]. According to his case, thoracic hyperlordosis secondary to type 3 lumbar degenerative kyphosis might be contributed to thoracic LFH. Similarly, our case showed sagittal imbalance as a consequence of the old compression fracture in the first lumbar vertebral body. Kyphosis secondary to compression fracture indicates that greater axial distraction stress or force seems to be loaded to the ligamentum flavum and the facet joint at mid-thoracic area compared with normal spinal alignment[12]. Generally, compression of neural elements by age-related ligamentum flavum hypertrophy or degeneration frequently occurs in mobile lumbar and cervical spine but rarely occurs in pure rigid thoracic spine which is protected by rib cages in keeping the normal spinal alignment and sagittal balance. If sagittal imbalance may happen, thoracic ligamentum flavum could be influenced by axial overstress and degenerative change might be promoted.

Fifth, prognoses following surgery were very good. Authors believe that LFH was formed and compressed in relatively long time, so spinal cord had sufficient time to compensate. Thus, cord restored its original shape immediately after hematoma evacuation backed up this suggestion.

Finally, preoperative MR images are the best choice of diagnostic tools. Common MR findings of reported cases are posterior extradural location, one ligamentum flavum involvement, mainly hyperintensities on T1-weighted images which suggested subacute stage hematoma, continuity between base of hematoma and ligamentum flavum and mixed-stage or multi-stage hematoma signal intensities[9]. If these findings are identified in case of posterior extradural mass coincide with intra-lesional hemorrhage, LFH should be included in differential diagnoses

CONCLUSION

To the best of our knowledge, this is the second case of LFH which occurred in pure rigid thoracic spine. There seems to be different mechanism from mobile lumbar and cervical spine in the development of pure rigid thoracic LFH. The authors believe that spinal sagittal imbalance (thoracic kyphosis secondary to compression fracture) may be the one of the contributors of pathogenesis. Total extirpation of the mass relieved the patient's paraparesis and the diagnosis was histologically confirmed.

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