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ABOUT COVER

Editorial Board Member of *World Journal of Clinical Cases*, Gagan Mathur, MBBS, MD, Associate Professor, Director, Staff Physician, Department of Pathology, Saint Luke's Health System, Kansas City, MO 64112, United States. gmathur@saint-lukes.org

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Intravascular papillary endothelial hyperplasia as a rare cause of cervicothoracic spinal cord compression: A case report

Hong-Lin Gu, Xiao-Qing Zheng, Shi-Qiang Zhan, Yun-Bing Chang

ORCID number: Hong-Lin Gu 0000-0001-7535-3503; Xiao-Qing Zheng 0000-0003-2882-1356; Shi-Qiang Zhan 0000-0002-8924-2027; Yun-Bing Chang 0000-0003-3581-623X.

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Hong-Lin Gu, Xiao-Qing Zheng, Shi-Qiang Zhan, Yun-Bing Chang, Department of Spine Surgery, Guangdong Provincial People's Hospital, Guangdong Academy of Medical Sciences, Guangzhou 510080, Guangdong Province, China

Corresponding author: Yunbing Chang, MD, Chief Doctor, Department of Spine Surgery, Guangdong Provincial People's Hospital, Guangdong Academy of Medical Sciences, No. 106 Zhongshan Road II, Guangzhou 510080, Guangdong Province, China. sykgspine@163.com

Abstract

BACKGROUND

Intravascular papillary endothelial hyperplasia (IPEH) is a rare benign reactive vascular lesion that grows into an expansile compressing mass. It most commonly involves the skin and subcutaneous tissue. Spinal involvement is rare, with only 11 reported cases in the literature. We report, to our knowledge, the first case of IPEH in the cervicothoracic spinal canal and present a literature review.

CASE SUMMARY

A 27-year-old man presented with acute-onset neck pain, numbness, and weakness in his extremities. Magnetic resonance imaging showed an epidural mass in the cervicothoracic (C6-T1) spinal canal and vertebral hemangioma (VH) involving the C7 vertebral body. C6-T1 Laminectomy and radical excision of the mass were performed. Histopathological examinations revealed papillary proliferation of vascular endothelial cells with thrombus formation, and an IPEH diagnosis was made. By his 6-mo follow-up appointment, his symptoms were relieved without recurrence. The possible pathogenesis, clinical and imaging features, differential diagnosis, and management of IPEH were reviewed.

CONCLUSION

We report, to our knowledge, the first case of IPEH in the cervicothoracic spinal canal, treated *via* complete resection, and showing a favorable outcome. We found a causal relationship between spinal IPEH and VH; this partly explains the mechanism of IPEH.

Key Words: Intravascular papillary endothelial hyperplasia; Cervicothoracic; Spinal cord compression; Thrombosis; Hemangioma; Case report

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Core Tip: Intravascular papillary endothelial hyperplasia (IPEH) is a rare benign reactive vascular lesion that grows into an expansile compressing mass. Spine involvement is rare, with only 11 case reports on its occurrence. We reported the first case of IPEH in the cervicothoracic spinal canal, which was treated *via* complete resection and had a good prognosis. We also found a causal relationship between spinal IPEH and vertebral hemangioma, and this partly explained the mechanism of IPEH.

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INTRODUCTION

Intravascular papillary endothelial hyperplasia (IPEH) was first reported in 1923 by Pierre Masson in a case of an infected hemorrhoidal vein. Initial reports referred to the lesion as a “Masson tumor” or hemangioendothelioma vegetant intravasculaire[1]. In 1975, Clearkin and Enzinger described the lesion as an unusual and exaggerated thrombus reorganization, rather than a true tumor, and the condition was renamed IPEH[2]. IPEHs typically occur in the skin and subcutaneous tissues of the head and neck or limbs[3]; it rarely occurs in the spine. To our knowledge, only 11 cases of IPEH of the spine have been reported. We report the first case of IPEH of the cervicothoracic spinal canal and present a literature review.

CASE PRESENTATION

Chief complaints

A 27-year-old man presented to the Department of Spine Surgery of our hospital with complaints of neck pain, limb numbness, and weakness.

History of present illness

His symptoms started suddenly, 4 d prior to hospital presentation.

History of past illness

The patient had no trauma history. A similar episode of transient limb numbness and weakness occurred 6 years earlier.

Personal and family history

He denied any personal or family history of other diseases.

Physical examination

Physical examination revealed tenderness of the paraspinal muscle of the C6-T1 spinous process, muted sensory responsiveness to touch along the T1 dermatome, and grade IV muscle strength in the four limbs.

Laboratory examinations

The results of all blood analyses — including coagulation markers, inflammatory indicators, and tumor markers — were within normal limits.

Imaging examinations

Spinal radiography and computed tomography demonstrated no obvious bone destruction. Enhanced cervical magnetic resonance imaging (MRI) showed a homogeneously enhanced epidural mass in the C6-T1 spinal canal. The mass compressed the spinal cord and extended into the left C7-T1 foramen. It appeared hypointense on T1-weighted images (T1WIs) and hyperintense on T2WI. Moreover, a 0.5 cm × 0.5 cm × 0.6 cm-sized heterogeneously enhanced hyperintense mass was

found in the C7 vertebral body on T2WI, which was suggestive of a benign vertebral hemangioma (VH) (Figure 1).

DIFFERENTIAL DIAGNOSIS

The mass was possibly an epidural schwannoma; however, we needed to exclude a nonneurogenic tumor diagnosis. The patient required surgical spinal cord decompression for symptom relief. The final diagnosis was confirmed histopathologically.

FINAL DIAGNOSIS

The final diagnosis was cervicothoracic spinal IPEH.

TREATMENT

The patient underwent C6-T1 Laminectomy, left C7-T1 foramen decompression, and radical excision of the epidural mass. A C6-T1 posterior instrumented fusion was performed to stabilize the facetectomy at the spinal level, proximal to the cervicothoracic junction. We observed a dark red, nodular, highly vascularized 3 cm × 1.5 cm × 1 cm mass compressing the spinal cord and left C7 nerve root dorsally. The mass was subsequently excised (Figure 2), and an intraoperative frozen section revealed a diagnosis of benign neoplasm originating from blood vessels. Histopathological examination revealed papillary proliferation of vascular endothelial cells with thrombus formation, consistent with IPEH (Figure 3).

OUTCOME AND FOLLOW-UP

After the surgery, the patient showed gradual neurologic improvement. At his 6-mo follow-up, he was symptom-free, with no spinal cord compression or recurrence on MRI (Figure 4). The clinical timeline of the patient is depicted in Figure 5.

DISCUSSION

IPEH is a rare benign reactive vascular lesion that expands to form a compressing mass. There is no age predilection for IPEH, and its incidence is higher in women than in men[4], with a female-to-male ratio of 4:1 for intracranial lesions[5]. Although spinal presentations are rare, they occur more commonly in men[6]. IPEHs are commonly located in the skin and subcutaneous tissues of the head and neck or limbs[3] but have also been reported in the oral mucosa, lip, thyroid, maxillary sinus, parotid, lung, superior vena cava, adrenal gland, renal vein, forearm, foot, and intracranially[7-11]. There are 11 reported cases of spinal IPEH, including ten cases involving men and one case involving a woman, with patient age ranging from 16 years to 58 years (see Table 1 for details)[6,12-21]. Among these cases, in one case, the mass was located in the vertebral body and in the remaining ten cases, the mass was located in the spinal canal. Only one case of an intradural mass and nine cases of an epidural mass have been reported. The most common site of involvement was the thoracic spinal canal ($n = 7$). Of the three remaining cases, two cases involved the lumbar spine and one case involved the thoracolumbar junction. The mass in one case was multifocal, involving the cervical, thoracic, and lumbar vertebral bodies. The present report is the first report of cervicothoracic spinal IPEH.

The pathogenesis of IPEH remains controversial. Some authors believe that IPEH is an excessive reaction to a normal thrombus reorganization process[2,22-24]. Others proposed that IPEH is a benign proliferation of endothelial cells with secondary thrombosis and fibrin deposition[25]. Few authors believe that there is a causal relationship between VH and spinal IPEH. Mozhdehipanah *et al*[17] reported a case of IPEH in the T4-T6 spinal canal. Two adjacent vertebral bodies (T4 and T5) demonstrated VH. The author speculated that bleeding within the spinal canal

Table 1 Summary of reported cases of spinal intravascular papillary endothelial hyperplasia

No.	Ref.	Year	Age (yr)	Sex	Primary location	Clinical features	RadiologicalFeatures	Treatment	Size (cm)
1	Ali <i>et al</i> [13]	1994	42	M	T8 posterior epidural mass	Paraplegia for 2 wk duration	MRI non-specific T1/T2 signal changes	Radical excision	1.5
2	Porter <i>et al</i> [14]	1995	16	M	T6 posterior epidural mass	Midthoracic radicular back pain with hesitancy for 1-wk duration	CT myelography: extradural thecal compression posteriorly with abnormal lamina	T6 laminectomy with T5-T6 right partial facetectomy	4 × 2 × 1
3	Taricco <i>et al</i> [15]	1999	17	M	T12-L1 posterior epidural mass	Pain, numbness, paresis of left lower limb with bladder dysfunction for 1 mo	Contrast-enhanced CT of spine: hyperdense lesion; MRI: T1-iso, T2-hyperintense with homogeneous contrast enhancement	T12-L1 laminectomy with radical excision of mass	Not mentioned
4	Petry <i>et al</i> [12]	2009	47	M	Multifocal lesions of the spine	Diffuse low back pain	MRI T1-iso, T2-hyperintense with homogeneous contrast enhancement	No surgery	Not mentioned
5	Lanotte <i>et al</i> [16]	2010	33	M	T6-T7 paravertebral mass extending epidural space	Back pain, hesitancy with paraparesis for 2 wk	MRI T1 hypo- T2 hyperintense mass	T6 laminectomy and excision of intracanal mass	4.5 × 2.5 × 2.5
6	Mozhdehpanah <i>et al</i> [17]	2013	58	M	T4-6 posterior epidural mass	Spastic paraparesis and sensory deficit for 1 mo	MRI T2 hyperintense mass	Laminectomy and radical excision of mass	3×1
7	Bhalla <i>et al</i> [21]	2013	51	F	L1 centered on spinous process and involving pedicles	Back pain with paraparesis	MRI L1 centered on spinous process and involving pedicles causing cauda equina compression	Preoperative embolization, incomplete excision and Radiotherapy	4.6×4.3×5.5
8	Singla <i>et al</i> [18]	2016	40	M	T12-L1 dumbbell-shaped mass	Back pain and numbness of the right lower trunk for 2 yr	MRI dumbbell-shaped mass mimicking schwannoma	Radical excision	Not mentioned
9	Behera <i>et al</i> [19]	2017	32	M	T4-5 posterior epidural mass	Paraplegia for 4 mo	MRI T1 hypo- T2 hyperintense mass	Radical excision	5 × 3 × 2
10	Tanaka <i>et al</i> [20]	2018	40	M	L2-3 intradural mass	Low back pain and leg pain beginning approximately 5 yr ago and 1 mo ago	Isointense on T1 and hypointense with partial areas of high signal intensity on T2 without contrast enhancement	L2-3 laminectomy and durotomy with radical excision of mass	2.5 × 1.5 × 1
11	Oktar <i>et al</i> [6]	2019	37	M	T4-5 dumbbell-shaped mass	Dermatomal tingling burning pain with paresis of right lower limb for 1 mo	MRI dumbbell-shaped mass mimicking schwannoma	Radical excision	5 × 2 × 3
12	Present case	2020	27	M	C6-T1 posterior epidural mass	Neck pain and numbness and weakness of the extremities	MRI: T1-hypo-, T2-hyperintense with homogeneous contrast enhancement	C6-T1 laminectomy with C7-T1 left partial facetectomy and radical excision of the mass	3 × 1.5 × 1

MRI: Magnetic resonance imaging; CT: Computed tomography.

subsequently formed an organized thrombus, transformed into Masson's hemangioma and manifested symptoms[17]. According to Petry *et al*[12], IPEH of the vertebral body develops from underlying thrombosis of the basivertebral venous plexus or a preexisting VH. In the present case, spinal cord compression was most severe at the C7 Level. Additionally, a hemangioma was found in the C7 vertebral body, suggesting a possible relationship between spinal IPEH and VH. Therefore, the mechanism behind IPEH could be an excessive reaction to the normal thrombus reorganization process. However, it is difficult to make conclusions based on the few cases. Future, well-powered studies on the relationship between VH and IPEH in the spinal canal are needed.

Preoperative diagnosis of spinal IPEH is challenging because of its non-specific MRI features and the need to differentiate IPEH from arteriovenous malformations, schwannomas, or neurofibromas *via* imaging technology. Reported cases of spinal

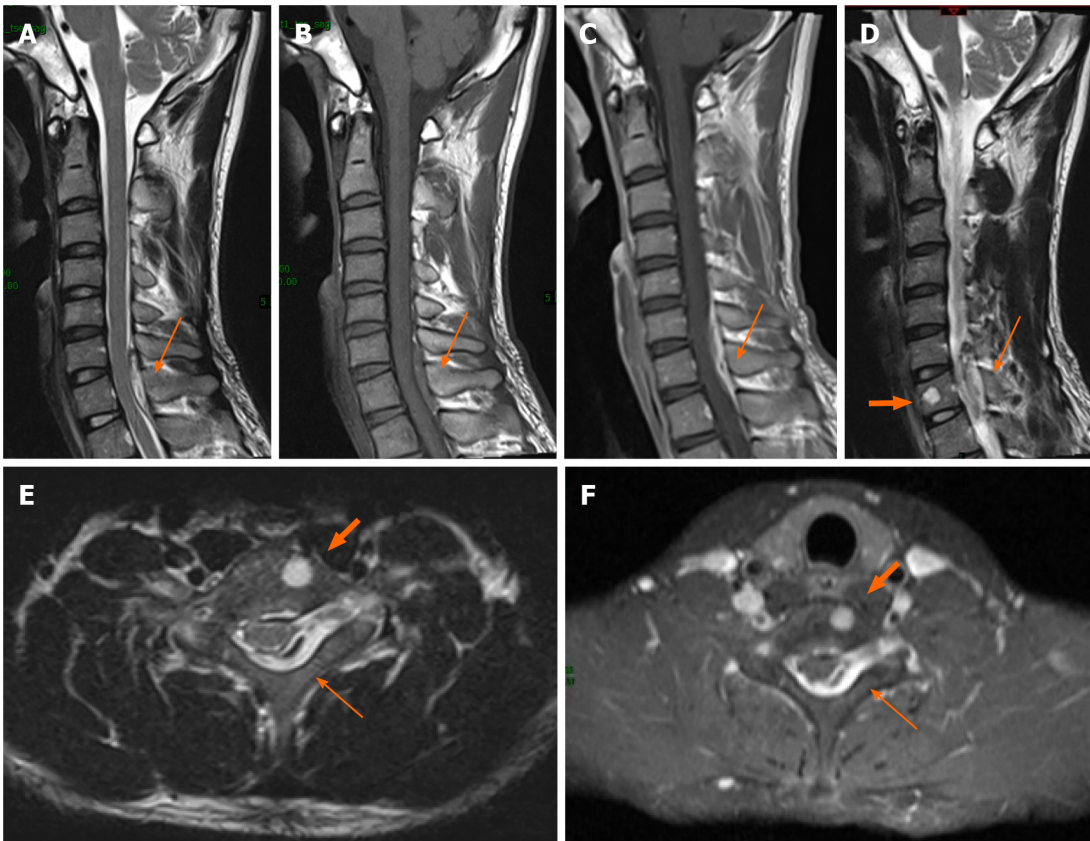


Figure 1 Preoperative magnetic resonance imaging. A and D: Sagittal T2-weighted imaging (T2WI); B: Sagittal T1-weighted imaging (T1WI); C: Sagittal T1WI of the spine with contrast; E: Axial T2WI; F: Axial T1WI with contrast. A posterior spinal epidural mass located from C6 to T1 (thin arrow) appeared high signal intensity on T2WI sagittal and axial images, and low signal intensity on T1WI images. A gadolinium-enhanced scan reveals inhomogeneous enhancement. And a 0.5 cm × 0.5 cm × 0.6 cm-sized round tumor (thick arrow) can be seen on the left side of the C7 vertebral body; high signal intensity is observed on T2WI and homogeneous enhancement is detected on T1WI after contrast agent administration.

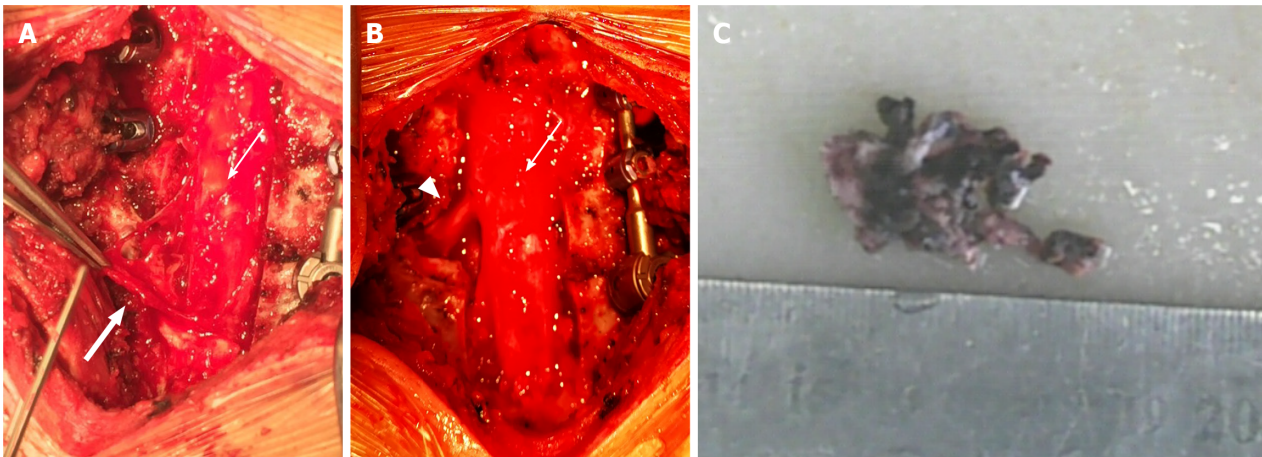


Figure 2 Intraoperative images. A: Operative view of a dark red, nodular, highly vascularized epidural mass (thick arrow) measuring 3 cm × 1.5 cm × 1 cm compressing the left side of the spinal cord (thin arrow) after C6-T1 Laminectomy. B: View of the surgeon after complete resection of the mass and decompression of dura (thin arrow) and left C7 nerve root (triangle). C: Nodular fragment of the lesion.

IPEH mostly exhibited isointensity or low signal intensity on T1WI and high or variable signal intensity on T2WI with contrast enhancement. Three cases presented as dumbbell-shaped masses, mimicking schwannoma[6]. The present patient was preoperatively diagnosed with epidural schwannoma, which typically features papillary proliferation of vascular endothelial cells, localized intravascularly, with normal thrombus formation in the entire papillary tissue[4,23,26]. The pathological findings in our case are consistent with a diagnosis of IPEH. However, IPEH must be

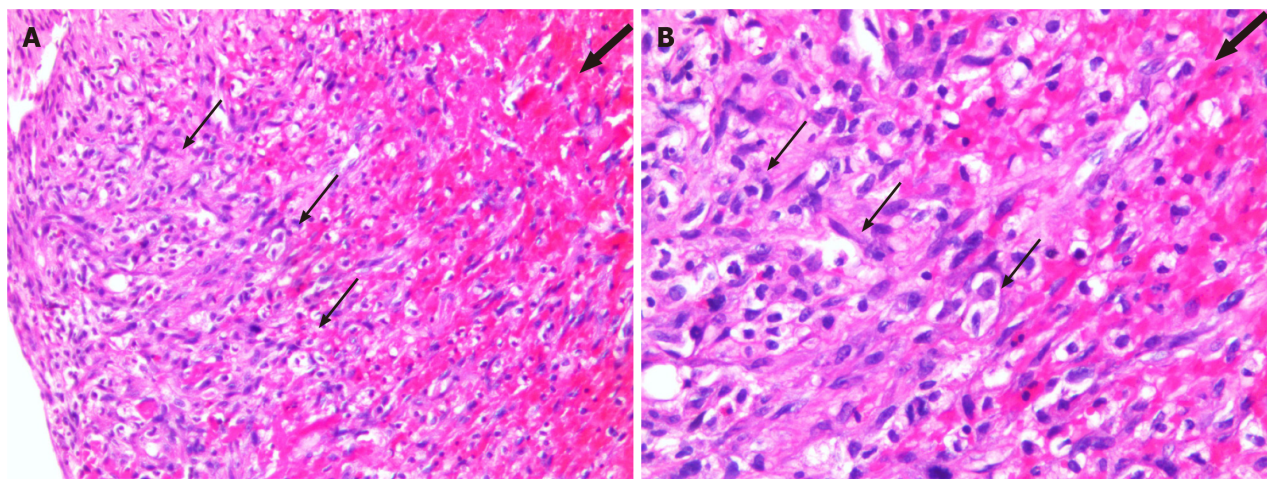


Figure 3 Histological features of the epidural mass. A: Hematoxylin-eosin (HE); $\times 100$; B: HE $\times 200$. Histopathological pictomicrograph shows dilated thin-walled vessels lined by a monolayer of obese endothelial cells (thin arrows). The lumen appears to be filled with organizing thrombi (thick arrow).

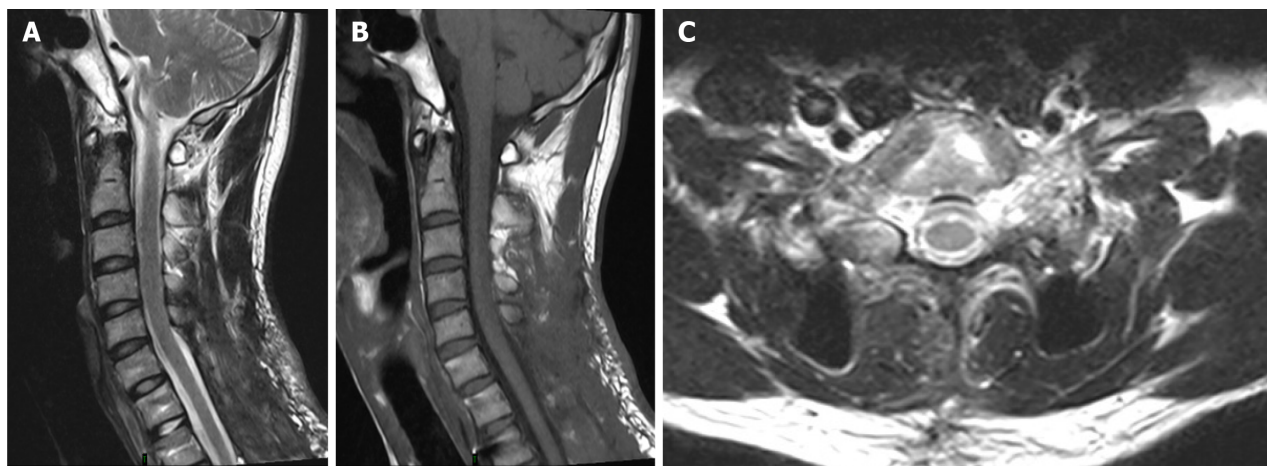


Figure 4 Postoperative magnetic resonance imaging at 6-mo follow-up. A-C: Magnetic resonance imaging showing total relief of the previously noted spinal cord compression and no signs of recurrence.

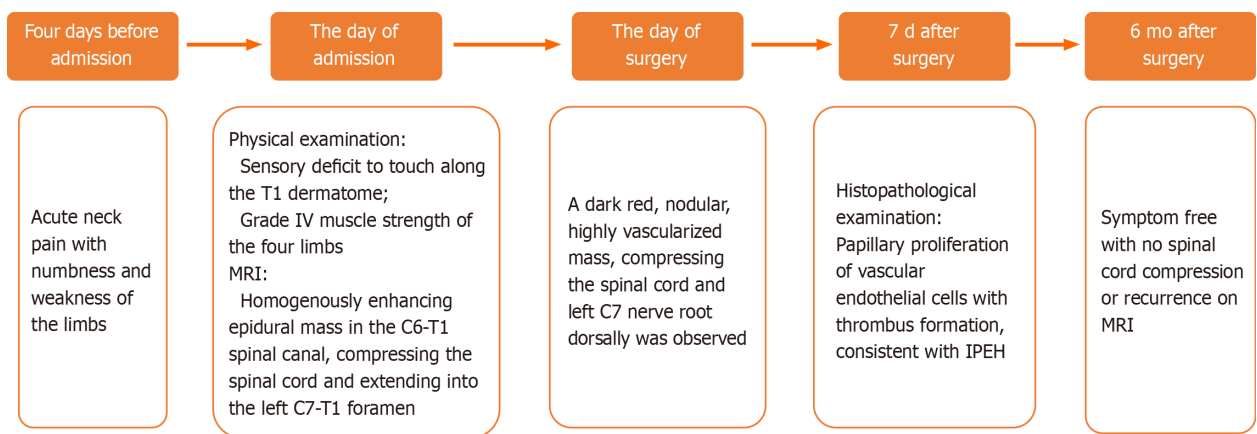


Figure 5 Patient timeline. MRI: Magnetic resonance imaging; IPEH: Intravascular papillary endothelial hyperplasia.

distinguished from other benign and malignant lesions, including cavernous/capillary hemangioma, Kaposi sarcoma, endovascular papillary, and angioendothelioma. Importantly, IPEH should be differentiated from angiosarcoma to avoid unnecessary radiation and surgery[11].

Spinal manifestations of IPEH may be associated with chest or back pain, lower limb numbness, paralysis, and bladder dysfunction caused by spinal cord or cauda equina compression[6]. Treatment is only considered when pain or compression-related symptoms occur, and complete surgical resection is the preferred treatment. Prognosis after complete resection is good, with minimal recurrence. Nine of the 11 patients in the reported cases underwent radical resection, and no recurrence was noted during follow-up. Further, the patient in the present case — who presented with acute-onset numbness and weakness of the limbs owing to spinal cord compression — underwent complete surgical resection. His symptoms were relieved and had not recurred by the 6-mo follow-up visit. Adjuvant radiotherapy can be considered for lesions that cannot be completely removed or that are recurrent. To our knowledge, there is only one reported case of radiotherapy for incompletely resected spinal IPEH. This case showed the potential benefit of radiation following the recurrence of benign IPEH in a patient with epidural disease[21].

CONCLUSION

The main pathological change attributable to IPEH is benign vascular endothelial papillary hyperplasia with thrombosis; however, the mechanism of this relationship remains controversial. Spinal IPEH is rare, occurring more frequently in men, and is localized to the thoracic spine. Importantly, our findings suggest that spinal IPEH is related to VH. There are no prior reports of IPEH in the cervical spinal canal. To our knowledge, this is the first case of IPEH in the cervicothoracic spinal canal which was associated with a favorable outcome after complete resection.

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