examination was normal. There were no motor or sensory deficits, the deep-tendon reflexes were normal, and a straight leg raising test was negative on both sides. Bowel and bladder functions and digital rectal examination were all normal. Various laboratory tests showed no abnormalities, but magnetic resonance imaging (MRI) showed a round 2 cm intradural extramedullary mass at the conus medullaris. On T1-weighted images, the signal of the lesion was isointense, and T2-weighted images revealed a hyperintense and homogeneous, strong enhancement on contrast-enhanced T1-weighted images. A spinal angiography with embolization demonstrated a hypervascular mass at the L1 level, just right at the tip of the conus medullaris, arising from an enlarged anterior spinal artery [Figure 1].

After embolization, a T12-L1 laminectomy with total tumor removal was performed with insignificant blood loss. The tumor was found emerging from the anterior spinal artery and attached to the nerve root. After the operation, the patient’s pain decrease, but the postoperative course was complicated by urinary retention. 3 month following the operation the patient recovered his ability to urinate normally. Grossly, the surgical specimen was a lobular, sharp bordered, reddish mass, measuring 2.0 cm in maximum diameter. Microscopically, the capillary hemangioma featured a lobular architecture, with the lobules being composed of tightly packed capillary-sized vessels lined by a single layer of endothelial cells. Diagnosis was confirmed by immunohistochemical staining that positive for CD34 and factor 8 [Figure 2].

Spinal capillary hemangioma is a rare tumor. A few cases have been reported in the literature. This report describes the clinicopathological findings in two cases of spinal capillary hemangiomas, including a capillary hemangioma at the conus medullaris in an adolescent and the another mimicked von Hippel-Lindau disease (VHL) disease which have never been reported before in a middle-aged adult.

**Case Reports**

**Case 1**
A 15-year-old boy presented a 4 month history of coccydynia, associated with left leg pain, both of which had been progressively worsening. The previous medical history and family history were unremarkable, and an neurological examination was normal. There were no motor or sensory deficits, the deep-tendon reflexes were normal, and a straight leg raising test was negative on both sides. Bowel and bladder functions and digital rectal examination were all normal. Various laboratory tests showed no abnormalities, but magnetic resonance imaging (MRI) showed a round 2 cm intradural extramedullary mass at the conus medullaris. On T1-weighted images, the signal of the lesion was isointense, and T2-weighted images revealed a hyperintense and homogeneous, strong enhancement on contrast-enhanced T1-weighted images. A spinal angiography with embolization demonstrated a hypervascular mass at the L1 level, just right at the tip of the conus medullaris, arising from an enlarged anterior spinal artery [Figure 1].

After embolization, a T12-L1 laminectomy with total tumor removal was performed with insignificant blood loss. The tumor was found emerging from the anterior spinal artery and attached to the nerve root. After the operation, the patient’s pain decrease, but the postoperative course was complicated by urinary retention. 3 month following the operation the patient recovered his ability to urinate normally. Grossly, the surgical specimen was a lobular, sharp bordered, reddish mass, measuring 2.0 cm in maximum diameter. Microscopically, the capillary hemangioma featured a lobular architecture, with the lobules being composed of tightly packed capillary-sized vessels lined by a single layer of endothelial cells. Diagnosis was confirmed by immunohistochemical staining that positive for CD34 and factor 8 [Figure 2].

**Key words:** Capillary hemangioma, spinal cord tumor, vascular tumor, von Hippel-Lindau disease
Case 2
A 31-year-old man presented with a 1-month history of left hemiparesis, with aspiration pneumonia associated with dysphagia. He had retinal hemangioma in both eyes and a familial history of his first-degree relatives dying from brain tumors. A neurologic examination revealed left hemiparesis and impairment of cerebellar signs on the left side. A computerized tomography (CT) scan of his brain found a cystic lesion at the fourth ventricle. From the pertinent data, the differential diagnosis in this case was cerebellar hemangioblastoma, syringobulbia and an arachnoid cyst. An MRI brain showed syringobulbia with syringohydromyelia at the upper cervical cord, without Chiari malformation, and a CT scan of the whole abdomen showed multiple pancreatic cysts. An MRI of the whole spine that showed multiple innumerable enhanced nodules along cervicothoracic spinal cord with syringobulbia with syringohydromyelia involving almost the entire spinal cord from the T7 level extending superiorly to involving the brainstem. The intramedullary lesion at C5-6 level evidently the cause of syringobulbia with syringohyromyelia [Figure 3].

Following these investigations, our preoperative diagnosis was VHL. A laminectomy of C4-6 was performed. Intradural exploration revealed a reddish tumor, which was subpial tumor at C4. A posterior midline myelotomy was performed at the C5-6 level which found an intramedullary tumor which was removed completely. After the operation, the patient's dysphagia improved. A postoperative MRI of the brain and whole spine revealed decreased syringobulbia and syringohydromyelia. Histological microscopy showed circumscribed vascular tumor with fibrous connective tissue capsule. The vessels were small, and dilated capillary lined by homogeneous endothelial cell. The vessels separated by fibrous tissue without stromal cell. In this case, a definite diagnosis was considered carefully because the preoperative presentations indicated probable spinal hemangioblastoma with VHL disease. Immunohistochemistry showed positive reactions for the endothelial cell markers. histology and immunohistochemical staining confirmed the diagnosis of capillary hemangioma in this case [Figure 4].

Discussion
Capillary hemangioma is a benign vascular lesion, most often found in the skin and other soft tissues of children. Within the spine, this tumor is commonly found in the vertebral bodies but only a few cases of intradural capillary hemangioma have been reported [Table 1]. The tumor’s characteristics are common in males, located at the thoraco-lumbar level, and clustered in the forth to sixth decades of life [Table 2]. Thus, based on case series of 64 patients, authors propose a classification scheme for the spinal capillary hemangioma classification into five types [Figure 5].

- Pediatric type has the cutaneous hemangioma following to the intradural extramedullary mass via sinus tract and usually form to the intradural extramedullary mass. This type is found in 5%
- Epidural type is extradural mass that usually extend into neural foramen. This type is found 8% and 80% of this type are dumbbell shaped tumors
- Intradural extramedullary type is found in 70% that is the most common type of spinal capillary hemangioma
The classification of spinal capillary hemangiomas (a) Pediatric type, (b) Epidural type, (c) Intradural extramedullary type, (d) Intramedullary type, (e) Hemangiomatosis

• Intramedullary type is found in 14%
• Hemangiomatosis is found in 3%. Roncaroli et al. defined “hemangiomatosis” since 2000. This type is disseminated tumor along entire spinal cord including subpial nodules and intramedullary mass.

The pathogenesis of this disease is still poorly understood. Several hypotheses have been proposed many factors starting hemangioma growth. Some of the key angiogenic factors are highly specific for endothelial cells. Basic fibroblast growth factor and vascular endothelial growth factor (VEGF) are proangiogenic molecules and are often produced by tumor cells. Concurrently, VHL disease is an autosomal dominantly inherited neoplastic syndrome which is associated with various vascular tumors and cysts. The most frequent tumors are retinal and central nervous system hemangioblastomas, clear cell renal cell carcinoma, pheochromocytoma, pancreatic islet tumors, and endolymphatic sac tumors. This syndrome associates with mutation of the VHL gene. The Inactivation of the VHL tumor suppressor protein with loss of function of the VHL protein, and Elongin B, C complex high level of...
hypoxia-inducible factors causes increased transcription of VEGF, platelet-derived growth factor and transforming growth factor-α which is an important step in the development of highly vascular tumors.\textsuperscript{[37]} The hypothesis of VHL disease that associated with capillary hemangioma is a high level of VEGF. This signal protein is an angiogenic factor with critical roles in tumor formation disseminated in multiple organs.

The surgical goal should be total complete tumor removal to decrease proangiogenic factors. Surgical standard procedures consists of a posterior midline approach to the spine, a one- or two-level laminectomy or laminectomy in children, and a midline durotomy. In an intramedullary tumor, a posterior midline myelotomy can avoid a major neurological deficit. This spinal tumor has multiple feeding arteries. The devascularization of the feeding arteries may be required to tumor resection in some situations. Perioperative embolization is an option to avoid massive surgical blood loss, especially in children.\textsuperscript{[38,39]} After total resection, tumor recurrence has rarely been reported.\textsuperscript{[22]} However, Abe et al. reported no recurrence at 13 years after complete resection.\textsuperscript{[19]}

**Conclusion**

We experienced a rare spinal cord tumor with a curious presentation. These lesions pose a challenging diagnostic

| Authors/year | Age (year)/sex | Symptoms (n) | Location | Level of involvement | Foraminal extension |
|--------------|----------------|--------------|----------|---------------------|---------------------|
| Mawk et al., 1987\textsuperscript{[1]} | 7 months/ female | Lower extremity apraxia | Cutaneous hemangioma extended to follow through the fascia, laminar defect at L5 level, the epidural space into the intradural space at conus medullaris | Conus medullaris (L2) | No |
| Hanakita et al., 1991\textsuperscript{[2]} | 58/male | Back and leg pain with sensory deficit at left L5 and bilateral S1 dermatome | Intradural extramedullary | Cauda equina (L1-L2) | No |
| Hida et al., 1993\textsuperscript{[3]} | 50/male | Leg weakness, numbness, bladder and bowel dysfunctions | Intramedullary | C3-T1 | No |
| Gupta et al., 1996\textsuperscript{[4]} | 50/male | Back pain, progressive weakness at left lower extremity, burning pain at right lower extremity, loss of proprioception and vibrator sensation | Epidural | T8-10 | No |
| Mastronardi et al., 1997\textsuperscript{[5]} | 41/male | Low back and leg pain | Intramedullary | L5 | No |
| Zander et al., 1998\textsuperscript{[6]} | 52/female | Back and leg pain | Intramedullary | L4-L5 | No |
| Holtzman et al., 1999\textsuperscript{[7]} | 56/female | Back and leg pain | Intramedullary | Cauda equina (L4) | No |
| Roncaroli et al., 1999\textsuperscript{[8]} | 40-62 Male (6) and female (4) | Leg pain (7) leg weakness (6) bowel and bladder symptoms (1), SLRT (3), paresthesia (2), hyperrelexia (2) | Intramedullary | Cauda equina (B), T5 (1), T1 nerve root (1) | No |
| Nowak et al., 2000\textsuperscript{[9]} | 63/female | Back pain | Intramedullary | T12-L1 | No |
| Roncaroli et al., 2000\textsuperscript{[10]} | 74/male | Weakness of both legs | Intramedullary | Lower thoracic spinal cord and conus medullaris (innumerable) | No |
| Roncaroli et al., 2000\textsuperscript{[11]} | 42/female | Leg weakness | Intramedullary | T11 | No |
| 50/male | Back pain, leg weakness | Intramedullary | T11 | No |
| 53/male | Leg pain | Intramedullary | Conus medullaris | No |
| 64/male | Leg pain, leg weakness | Intramedullary | T10 | No |
| Shin et al., 2000\textsuperscript{[12]} | 66/female | Back pain, leg weakness | Intramedullary | T8-9 | No |
| Choi et al., 2001\textsuperscript{[13]} | 28/male | Back and leg pain | Intramedullary | Cauda equina (L1) | No |
| 52/male | Leg weakness | Intramedullary | T5-T6 | No |
| 51/male | Leg pain, weakness of both legs | Intramedullary | T4-T5 | No |
| Andaluz et al., 2002\textsuperscript{[14]} | 41/male | Back pain and weakness of both legs | Intramedullary | Conus medullaris | No |
| Bozkus et al., 2003\textsuperscript{[15]} | 37/female | Bilateral leg numbness (2) and leg weakness (5) | Intramedullary | Thoracic spinal cord | No |
| 55/male | Leg pain | Intramedullary | Conus medullaris | No |
| Badinand et al., 2003\textsuperscript{[16]} | 40/female | Leg pain, paraparesis, hypoesthesia | Epidural | T2-T4 | Yes |
| 32/female | Paraparesis, progressive lower back pain and paravesthesia | Intramedullary | T10 | No |

Contd...
| Authors/Year | Age (year)/Sex | Symptoms (n) | Location | Level of involvement | Foraminal extension |
|-------------|----------------|--------------|----------|---------------------|-------------------|
| Abe et al., 2004<sup>[19]</sup> | 59/male | Paraparesis, hypesthesia of both legs | Intradural extramedullary | T11 | No |
| | 51/male | Paraparesis, hypesthesia of both legs | Intradural extramedullary | T11 | No |
| | 64/male | Paraparesis, hypesthesia of both legs | Intradural extramedullary | T7 | No |
| | 71/male | Paraparesis, hypesthesia of both legs | Intramedullary | T11 | No |
| | 65/male | Weakness of both legs | Intramedullary | T9 | No |
| | 80/male | Paraparesis, hypesthesia of both legs | Intramedullary with extramedullary component | T7 | No |
| Crispino et al., 2005<sup>[20]</sup> | 65/male | Progressive paraparesis, upper-thoracic back pain | Intradural extramedullary | T1-2 | No |
| | 57/male | Progressive thoracic pain, paraparesis | Intramedullary | T9-10 | No |
| Kim et al., 2006<sup>[21]</sup> | 59/male | Low back pain, leg pain, paresthesia | Intramedullary | Cauda equine (L1-L2) | No |
| Ghazi et al., 2006<sup>[22]</sup> | 42/male | Low back pain with sciatica, increased ICP | Intramedullary | Cauda equine (L3-L4) | No |
| Kang et al., 2006<sup>[23]</sup> | 56/male | Right radiating chest wall pain | Epidural | T2-T4 | Yes |
| Karikari et al., 2007<sup>[24]</sup> | 6 month/female | Lumbar dimple with an overlying raised purple-red hemangioma | Cutaneous hemangioma extended to follow through the fascia, the epidural space into the intradural space at L4 level with a fatty filum terminale | L3-L4 | No |
| | 1-month/female | Lumbar cutaneous hemangioma associated with a dermal sinus tract | Cutaneous hemangioma extended to follow through the fascia, the epidural space into the intradural space at L3-L4 level with a fatty filum terminale | L3-L4 | No |
| Ganapathy et al., 2008<sup>[25]</sup> | 17/male | Low back pain with S1 radiculopathy | Intramedullary | Cauda equina (L2-L3) | No |
| | 20/male | Low back pain radiating to legs, paraparesis, hypesthesia, bladder dysfunction and erectile dysfunction | Intramedullary | Cauda equina (L3) | No |
| Chung et al., 2010<sup>[26]</sup> | 47/male | Back pain on lower thoracic & leg pain, paresthesia at T7 dermatome and hyperreflexia | Intramedullary | T6-T7 | No |
| Hasan et al., 2011<sup>[27]</sup> | 57/male | Back pain, right leg motor weakness, numbness of left leg, band of numbness at the level of the umbilicus on the right | Epidural | T10-12 | Yes |
| Vassal et al., 2012<sup>[28]</sup> | 59/female | Back pain, Rt intercostals neuralgia, motor weakness of lower limbs | Epidural | T5-7 | Yes |
| Sonawane et al., 2012<sup>[29]</sup> | 35/male | Mid back pain, progressive weakness in both lower limbs | Intramedullary | T12 | No |
| Kaneko et al., 2012<sup>[30]</sup> | 48/male | Low back pain, motor weakness of lower limbs, loss of proprioception with positive of Romberg's sign | Intramedullary | T10-11 | No |
| Babu et al., 2013<sup>[31]</sup> | Mean age 53.5 | Pain (3), motor weakness (1), bladder symptoms (1) | Intramedullary (4) | NA | No |
| Wu et al., 2013<sup>[32]</sup> | 49/male | Rt lower limb numbness | Intramedullary | T1-2 | No |
| | 63/female | Low back pain, bilateral lower limbs numbness, weakness, dysuria | Intramedullary | T11 | No |
| | 18/male | Low back pain, bilateral lower limbs weakness, dysuria | Intramedullary | T7-8 | No |
| | 47/male | Rt lower limb numbness and weakness | Intramedullary | C7-T1 | No |
| | 59/male | Back pain, bilateral lower limbs numbness and weakness | Intramedullary with extramedullary component | T3-4 | No |
| Ganazalez et al., 2014<sup>[33]</sup> | 59/male | Progressive numbness of both legs and low back pain | Intramedullary with extramedullary component | T7-T8 | No |
problem, because variable types of this tumor can mimic other spinal tumors. For over 25 years, the tumor has been reported since 1987. It’s important to concern in the differential diagnosis of spinal cord tumor, and surgical resection is the treatment of choice for a definite diagnosis.

Acknowledgment

The authors thank Kittipong Riabroi, MD and Associate Prof. Dr. Kogkun Tungsimmunkong for assistance.

References

1. Pastushyn Al, Slin’ko EI, Mirzoyeva GM. Vertebral hemangiomas: Diagnosis, management, natural history and clinicopathological correlates in 86 patients. Surg Neurol 1998;50:535-47.
2. Mawk JR, Leibrock LG, McComb RD, Trembath EJ. Metameric capillary hemangioma is producing a complete myelographic block in an infant. Case report. J Neurosurg 1987;67:456-9.
3. Hanakita J, Suwa H, Nagayasu S, Suzuki H. Capillary hemangioma in the cauda equina: Neuroradiological findings. Neuroradiology 1991;33:458-61.
4. Hida K, Tada M, Iwasaki Y, Abe H. Intradural disseminated capillary hemangioma with localized spinal cord swelling: Case report. Neurosurgery 1993;33:1099-101.
5. Gupta S, Kumar S, Banerji D, Pandey R, Gujral R. Magnetic resonance imaging features of an epidural spinal haemangioma. Australas Radiol 1996;40:342-4.
6. Mastronardi L, Guiducci A, Frondizi D, Carletti S, Spera C, Maira G. Intraneural capillary hemangioma of the cauda equina. Eur Spine J 1997;6:278-80.
7. Zander DR, Lander P, Just N, Albrecht S, Mohr G. Magnetic resonance imaging features of a nerve root capillary hemangioma of the spinal cord: Case report. Can Assoc Radiol J 1998;49:398-400.
8. Holtzman RN, Brisson PM, Pearl RE, Gruber ML. Lobular capillary hemangioma of the cauda equina. Case report. J Neurosurg 1999;90:239-41.
9. Roncaroli F, Scheithauer BW, Krauss WE. Hemangioma of spinal nerve root. J Neurosurg 1999;91:175-80.
10. Nowak DA, Gunprecht H, Stölze A, Lumenta CB. Intraneural growth of a capillary haemangioma of the cauda equina. Acta Neurochir (Wien) 2000;142:463-7; discussion 467.
11. Roncaroli F, Scheithauer BW, Deen HG Jr. Multiple hemangiomas (hemangiomatosis) of the cauda equina and spinal cord. Case report. J Neurosurg 2000;92:229-32.
12. Roncaroli F, Scheithauer BW, Deen HG Jr. Multiple hemangiomas (hemangiomatosis) of the cauda equina and spinal cord. Case report. J Neurosurg 2000;92:229-32.
13. Shin JH, Lee HK, Jeon SR, Park SH. Spinal intradural capillary hemangioma: MR findings. AJNR Am J Neuroradiol 2001;22:799-802.
14. Andalus N, Balko MG, Stanek J, Morgan C, Schwetschenau PR. Lobular capillary hemangioma of the spinal cord: Case report and review of the literature. J Neurooncol 2002;56:261-4.
15. Bozkus H, Tanriverdi T, Kizilkanli O, Türeci E, Oz B, Hanci M. Capillary haemangiomas of the spinal cord: Report of two cases. Minim Invasive Neurosurg 2003;46:41-6.
16. Badinand B, Morel C, Kopp N, Tran Min VA, Cotten F. Dumbbell-shaped epidural capillary hemangioma. AJNR Am J Neuroradiol 2000;21:954-6.
17. Choi BY, Chang KH, Choe G, Han MH, Park SW, Yu IK, et al. Spinal intradural extramedullary capillary hemangioma: MR imaging findings. AJNR Am J Neuroradiol 2001;22:799-802.
18. Abdulah DC, Rahul Kumar K, Phillips CD, Jane JA Jr, Miller B. Thoracic intradural extramedullary capillary hemangioma. AJNR Am J Neuroradiol 2004;25:1294-6.
19. Abe M, Tabuchi K, Tanaka S, Hodozuka A, Kunishio K, Kubo N, et al. Capillary hemangioma of the central nervous system. J Neurosurg 2004;101:73-81.
20. Crispino M, Vecchioni S, Galli G, Olivetti L. Spinal intradural extramedullary haemangioma: MRI and neurosurgical findings. Acta Neurochir (Wien) 2005;147:1195-8.
21. Kelleher T, Aquilina K, Keohane C, O’Sullivan MG. Intramedullary
capillary haemangioma. Br J Neurosurg 2005;19:345-8.
22. Kim KJ, Lee JY, Lee SH. Spinal intradural capillary haemangioma. Surg Neurol 2006;66:212-4.
23. Ghazi NG, Jane JA, Lopes MB, Newman SA. Capillary haemangioma of the cauda equina presenting with radiculopathy and papilledema. J Neuroophthalmol 2006;26:98-102.
24. Kang JS, Lillehei KO, Kleinschmidt-Demasters BK. Proximal nerve root capillary haemangioma presenting as a lung mass with bandlike chest pain: Case report and review of literature. Surg Neurol 2006;65:584-9.
25. Karikari IO, Selznick LA, Cummings TJ, George TM. Spinal capillary haemangioma in infants: Report of two cases and review of the literature. Pediatr Neurosurg 2007;43:125-9.
26. Ganapathy S, Kleiner LI, Mirkin LD, Hall L. Intradural capillary haemangioma of the cauda equina. Pediatr Radiol 2008;38:1235-8.
27. Mirti SM, Habibi Z, Hashemi M, Meybodi AT, Tabatabai SA. Capillary haemangioma of cauda equina: A case report. Cases J 2009;2:80.
28. Chung SK, Nam TK, Park SW, Wang SN. Capillary haemangioma of the thoracic spinal cord. J Korean Neurosurg Soc 2010;48:272-5.
29. Hasan A, Guiot MC, Torres C, Marcoux J. A case of a spinal epidural capillary haemangioma: Case report. Neurosurgery 2011;68:E850-3.
30. Vassal F, Peoc’h M, Nuti C. Epidural capillary haemangioma of the thoracic spine with proximal nerve root involvement and extraforaminal extension. Acta Neurochir (Wien) 2011;153:2279-81.
31. Sonawane DV, Jagtap SA, Mathews AA. Intradural extramedullary capillary haemangioma of lower thoracic spinal cord. Indian J Orthop 2012;46:475-8.
32. Kaneko Y, Yamabe K, Abe M. Rapid regrowth of a capillary haemangioma of the thoracic spinal cord. Neurol Med Chir (Tokyo) 2012;52:665-9.
33. Babu R, Owens TR, Karikari JO, Moreno J, Cummings TJ, Gottfried ON, et al. Spinal cavernous and capillary haemangiomas in adults. Spine (Phila Pa 1976) 2013;38:E423-30.
34. Wu L, Deng X, Yang C, Xu Y. Intramedullary spinal capillary haemangiomas: Clinical features and surgical outcomes: Clinical article. J Neurosurg Spine 2013;19:477-84.
35. Gonzalez R, Spears J, Bharatha A, Munoz DG. Spinal lobular capillary haemangioma with an intramedullary component. Clin Neuropathol 2014;33:38-41.
36. Hamlat A, Adn M, Pasqualini E, Brassier G, Askar B. Pathophysiology of capillary haemangioma growth after birth. Med Hypotheses 2005;64:1093-6.
37. Shuin T, Yamasaki I, Tamura K, Okuda H, Furihata M, Ashida S. Von Hippel-Lindau disease: Molecular pathological basis, clinical criteria, genetic testing, clinical features of tumors and treatment. Jpn J Clin Oncol 2006;36:337-43.
38. Berkefeld J, Scale D, Kirchner J, Heinrich T, Kollath J. Hypervascular spinal tumors: Influence of the embolization technique on perioperative hemorrhage. AJNR Am J Neuroradiol 1999;20:757-63.
39. Manke C, Bretschneider T, Lenhart M, Stroth C, Neumann C, Gmeinwieser J, et al. Spinal metastases from renal cell carcinoma: Effect of preoperative particle embolization on intraoperative blood loss. AJNR Am J Neuroradiol 2001;22:997-1003.