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EDİTÖRE MEKTUP / LETTER TO THE EDITOR

Pure thoracic spinal epidural cavernous hemangioma (SECH)

Saf torasik spinal epidural kavernöz hemanjiom (SEKH)

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To the Editor,

Hemangiomas are benign vascular malformations. Pure epidural hemangiomas (PEHs) are rare, although they can be seen frequently in the entire spine. PEHs are approximately 4% of epidural lesions. Most spinal hemangiomas are of the cavernous type[1]. Cavernous hemangiomas (CHs) account for 5% to 12% of all spinal vascular malformations. It is usually associated with bone involvement. Although the best diagnosis is made with magnetic resonance imaging (MRI), differential diagnosis can sometimes be difficult. Loss of strength and sensation, radiculopathy, localized pain, urinary and fecal incontinence are the main signs and symptoms[2]. The number of pure spinal epidural cavernous hemangiomas (SECH) reported in the literature until 2010 is only 90[3]. So we wanted to present a very rare case of SECH. Informed consent was obtained from the patient.

A 42-year-old female patient was admitted to our outpatient clinic with complaints of back pain, weakness and numbness in the legs that started 2 months ago. On examination, there was paraparesis causing 2/5 loss of strength and sensory loss below the T8 sensory dermatome. There was no abnormality in the routine laboratory tests of the patient who did not describe urinary and fecal incontinence.

In the radiological imaging of the patient, a solid lesion was observed, measuring approximately 53X28X13 mm in the posterior part of the thoracic spinal canal, starting from the level of the T7 vertebra lower and plateau and continuing to the T9-10 disc distance, displacing the spinal cord anteriorly and compressing the spinal cord anteriorly, while showing moderate contrast enhancement in the postcontrast series. The anterior-posterior diameter of the spinal canal decreased by 3-4 mm, and there was an increased signal that may belong to oedema and myelomalacia in the cord (Figure 1).

The radiology section reported that it could be a meningioma or a nerve sheath tumour. The patient underwent T8/T9 total and partial T7/T10 laminectomy under general anaesthesia. A dark coloured, hemorrhagic encapsulated mass with a soft consistency in the epidural space, adhered to the dura posteriorly, was completely excised by microsurgery (Figure 2). The histopathological diagnosis came as a cavernous hemangioma. Vascular malformation consisting of large, dilated, blood-filled vessels lined with a flattened endothelium was observed (Figure 3A, 3B, 3C, 3D).

The control thoracic MRI was taken on the postoperative 1st day and the postoperative 1st month. No residual mass was detected. In addition, after 1 month, it was observed that the spinal cord regained its normal course and thickness from anterior to posterior (Figure 4). All paraparesis of the patient resolved almost completely in the 1st month postoperatively. The sensory defect was also reduced.

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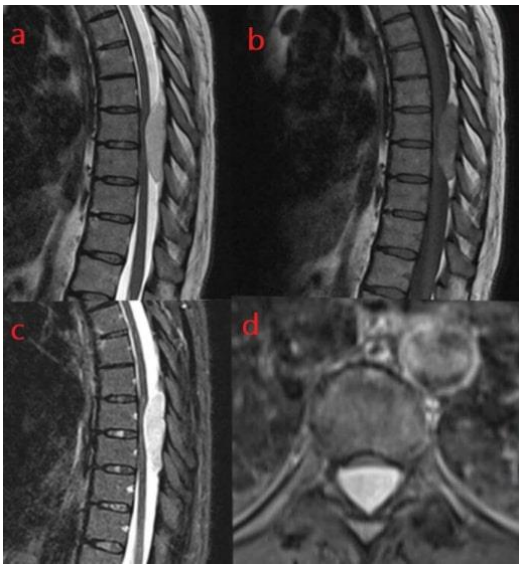

Figure 1. Preoperative thoracic MRI scan demonstrates extradural mass. (a) Sagittal T2-weighted MRI; (b) Sagittal T1-weighted MRI; (c) Sagittal T2 Short-Tau Inversion-Recovery (T2-STIR) MRI; (d) Axial T2-weighted MRI.

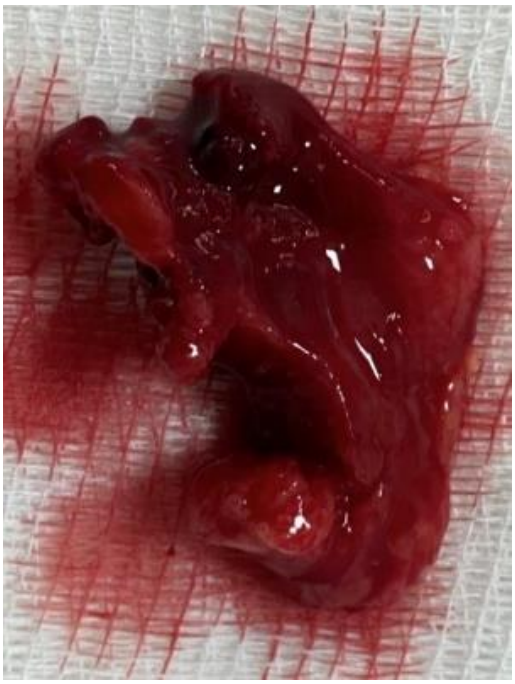

Figure 2. 53X28X13 mm extradural hemorrhagic reddish mass.

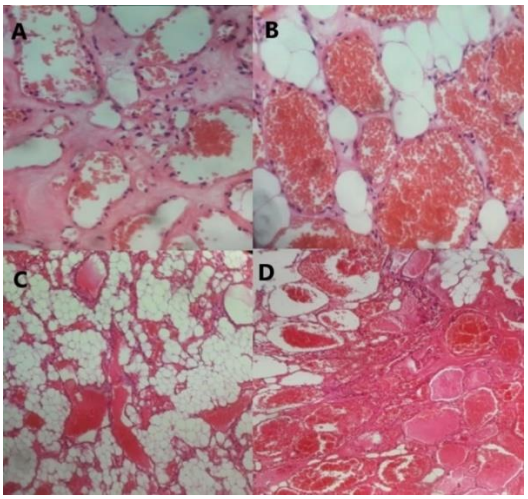

Figure 3. A) Dilated vascular structures in fibroconnective tissue HE, 400x B-C) As the same adipose tissue HE, 400x-100x D) Blood filled vessels lined with flattened endothelium HE, 400x

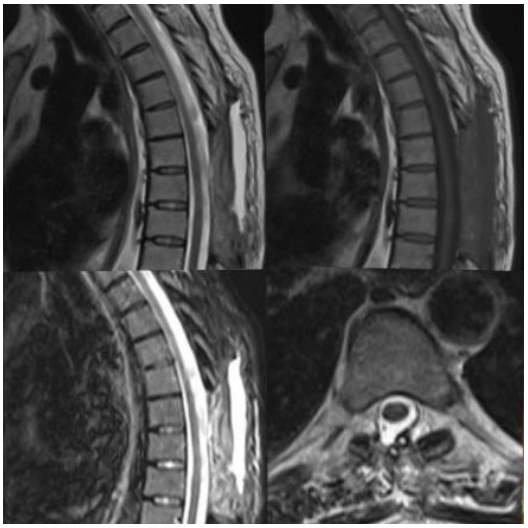

Figure 4. Thoracic MRI scan at 1 month postoperatively shows complete excision of the extradural mass and recovery of the spinal cord.

Cavernous angiomas, which can develop anywhere in the body, most commonly occur in the supratentorial region of the central nervous system (CNS). Cavernous angiomas of the spine are rare and are most common in the vertebral body. Spinal epidural cavernous angiomas were first described by Globus and Doshay in 1929. It can be sporadic or familial. SECHs were detected and published in 2015 in a 50-

year-old woman and a 68-year-old male patient. A total of 128 SECH patients were identified with these two cases published in the literature review conducted until the date of publication of this article. The mean age of cases was 47.7 years and male cases were 1.5 times more common than female cases[4].

Typically, SECHs occur directly through progressive myelopathy, secondary to the phenomenon of compression or vascular stealing. Radiculopathy due to foraminal root compression may also be seen. Bleeding into the lesion may cause sudden clinical findings and complaints. In an article published in 2015, a 27-year-old male patient, who was admitted with bilateral lower extremity numbness, progressive weakness and back pain, was reported[5].

The lesion associated with SECH is usually isointense with the spinal cord on the T1W sequence and hyperintense on the T2W sequence on magnetic resonance imaging (MRI). It shows intense homogeneous contrast after the injection of contrast material. Therefore, neurogenic tumours, metastasis, lymphoma, multiple myeloma, extraosseous Ewing's sarcoma, disc fragment and epidural angiolipoma should be included in the differential diagnosis[6].

Thrombosis and residual bleeding can be observed histopathologically in CHs. The thin walls of the vessels forming the lesions are lined with endothelium. There are also loose ligaments and adipose tissue. A single layer of flattened endothelial cells in collagen tissue, which is elastic and lacks tissue or neurons forms the main structure of the vessel walls. In a very recently published series of nine cases, all patients underwent early surgery and no recurrence was observed at a mean follow-up of 55 months[7]. Also in 2018, SECH in the form of dumbbells and surgical results were published in nine cases. Total resection was performed in six patients and partial resection was performed in three patients. Excellent results were obtained in seven patients[8]. An epidural lesion at the T7-T8 level was detected in a 50-year-old male patient who presented with myelopathy in 2013. The postoperative pathology of the patient, who was thought to be a meningioma, was compatible with CH. The patient improved dramatically after surgery[9]. In 2016, a series of seven cases were published and surgery was performed on all cases. In these patients, the localization is usually thoracic, and no recurrence was observed in long follow-up. Early surgical intervention is recommended in such cases[10]. In 2015, a 55-year-old male patient with SECH who had weakness in the lower extremity was reported. The patient's weakness improved in the early postoperative period[11].

In a large series of 14 cases with primary SECH published in 2015, seven patients were misdiagnosed preoperatively. These diagnoses were schwannoma, meningioma and arachnoid cyst[12]. Our patient had similar prediagnoses. We think that the more experienced, the better the differential diagnosis. In 2014, pure SECH was detected at T3-T4 level in a 64-year-old male patient. Bleeding was detected in the lesion[13]. Bleeding into the lesion can accelerate clinical findings as it rarely increase pressure on the spinal cord. Therefore, it may require emergency surgery.

As you can see, the treatment in the cases reported in the literature is surgery. Most of the patients resulted in healing with surgical intervention in the early period and no recurrence was detected in their follow-up. Pure SECH are rare congenital or developmental vascular malformations in the spinal region. Although spinal MRI is very important in diagnosis, the margin of error is high unless it is considered in the differential diagnosis. Since it is highly vascular, there may be intralesional bleeding before surgery and surgical bleeding during surgery. Early diagnosis and surgical intervention are very satisfactory and there is usually no recurrence in the follow-up. Patients usually recover almost completely. Therefore, pure SECH should be included in the differential diagnosis of tumours located in the spinal epidural region.

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REFERENCES

1. Brasil AVB, Rohrmoser RG, Gago G, Cambruzzi E.

Atypical spinal epidural capillary hemangioma: case report. Surg Neurol Int. 2018;9:198.
2. Ozkal B, Yaldiz C, Yaman O, Ozdemır N, Dalbayrak S. Extraosseous, epidural cavernous hemangioma with back pain. Pol J Radiol. 2015;80:206-9.
3. Koktekir E. Pure cervicothoracic epidural cavernous hemangioma presenting with neurologic deficit. J Kartal TR. 2010;21:103-6.
4. Bayri Y, Ekşi MŞ, Yalçınkaya Koç D, Konya D. Spinal epidural cavernous angioma: two case reports and review of the literature. Acta Orthop Traumatol Turc. 2015;49:459-64.
5. Meng Y, Shamji MF. Solitary spinal epidural cavernous haemangiomas as a rare cause of myelopathy. BMJ Case Rep. 2015;2015:bcr2015211644.
6. Khalatbari MR, Abbassioun K, Amirjmshidi A. Solitary spinal epidural cavernous angioma: report of nine surgically treated cases and review of the literature. Eur Spine J. 2013;22:542-7.
7. Zhao L, Jiang Y, Wang Y, Bai Y, Sun Y, Li Y. Spinal epidural cavernous hemangiomas: a clinical series of 9 cases and literature review. Front Oncol. 2021;11:572313.
8. Zhang L, Zhang Z, Yang W, Shang J, Jia W, Yang J et al. Spinal dumbbell-shaped epidural cavernous hemangioma (CM): report of nine surgical cases and literature review. Chin Neurosurg J. 2018;4:3.
9. A L H, T R, Chamarthy NP, Puri K. A pure epidural spinal cavernous hemangioma - with an innocuous face but a perilous behaviour!! J Clin Diagn Res. 2013;7:1434-5.
10. Esene IN, Ashour AM, Marvin E, Nosseir M, Fayed ZY, Seoud K et al. Pure spinal epidural cavernous hemangioma: a case series of seven cases. J Craniovertebr Junction Spine. 2016;7:176-83.
11. Yaldiz C, Asil K, Ceylan D, Erdem S. Thoracic extraosseous epidural cavernous hemangioma. J Korean Neurosurg Soc. 2015;57:65-7.
12. Li TY, Xu YL, Yang J, Wang J, Wang GH. Primary spinal epidural cavernous hemangioma: clinical features and surgical outcome in 14 cases. J Neurosurg Spine. 2015;22:39-46.
13. Jang D, Kim C, Lee SJ, Ryu YJ, Kim J. Pure spinal epidural cavernous hemangioma with intralesional hemorrhage: a rare cause of thoracic myelopathy. Korean J Spine. 2014;11:85-8.