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EVALUATION OF THE EFFICACY AND SAFETY OF SURGICAL TREATMENT OF MULTI-LEVEL VERTEBRAL HEMANGIOMAS CAUSING CORD COMPRESSION

KORD BASISI OLUŞTURAN ÇOK SEVİYELİ VERTEBRA HEMANJİYOMLARINDA CERRAHİ TEDAVİNİN ETKİNLİĞİ VE GÜVENİRLİĞİNİN DEĞERLENDİRİLMESİ

Mehmet Fatih KORKMAZ[1], Kaya SARAÇ[2], Mustafa KARAKAPLAN[1], Gökay GÖRMELİ[1], Mehmet Nuri ERDEM[3], Engin Burak SELÇUK[4]

[1]İnönü University, Medical School, Orthopedics and Traumatology AD, Malatya
[2]İnönü University, Medical School, Radiology AD, Malatya
[3]Kolan International Hospital, Orthopedics and Traumatology AD, Istanbul
[4]İnönü University, Medical School, Family Practice AD, Malatya

SUMMARY

In this study, a 69-year-old woman with more than one vertebral hemangioma causing spinal cord compression at different levels is presented. Posterior surgery was applied due to T12 vertebral compression fractures, and after five asymptomatic years, lower back pain, progressive strength loss in the lower extremities and difficulty in walking occurred for four months, and urinary and fecal incontinence occurred for four weeks. Multiple thoracic and lumbar vertebral hemangiomas were observed in thoracic and lumbar magnetic resonance imaging (MRI). T10 and L4 extraossous extension of the hemangiomas was causing cord compression. Selective embolization was performed preoperatively and in the presence of cord neuromonitorization, and cord decompression was achieved with anterior T10 corpectomy and L4 PVCR (posterior vertebral column resection) with an 18-month interval. Postoperative radiotherapy was performed. The patient's neurological condition improved rapidly after surgery, and she was neurologically intact and able to walk independently. Vertebral hemangiomas can cause serious and progressive cord compression. If the principles of multidisciplinary treatment are applied accurately, this can be treated safely with surgery.

Key words: Hemangiomas, trans-arterial embolization, spinal cord compression, surgical treatment

Level of evidence: Case report, Level IV

ÖZET

Bu çalışmada farklı düzeylerde spinal kord basısı yapan birden fazla vertebra hemanjiomları olan 69 yaşındaki kadın sunuldu. T12 vertebra çökme kırığı nedeniyle posterior cerrahi uygulanmış ve asemptomatik geçen 5 yılın ardından 4 aydır bel ağrısı, alt ekstremitelerde ilerleyici güç kaybı ve yürüme güçlüğü, 4 haftadır idrar ve gaita inkontinansı mevcuttu. Torakal ve lomber Manyetik Rezonans Görüntülemede (MRG) birden fazla torakal ve lomber vertebra hemanjiomları görüldü. T10 ve L4 de hemanjiom ekstraossöz uzantısı kord basısına neden oluyordu. Seçici embolizasyonlar preoperatif yapıldı ve nöromonitörizasyon eşliğinde kordun dekompresyonu anterior T10 korpektomi ile ve L4 PVCR (Posterior Vertebral Kolon Rezeksiyonu) ile 18 ay arayla sağlandı. Postoperatif radyoterapi uygulandı. Hastanın nörolojik durumu ameliyat sonrası hızla düzeldi ve nörolojik olarak sağlam ve bağımsız olarak yürüyebildi. Vertebra hemanjiyomları ciddi ilerleyici kord basısı nedeni olabilirler. Multidisipliner yaklaşımla tedavi prensipleri doğru uygulandığı takdirde cerrahi olarak güvenle tedavi edilebilirler

Anahtar Kelimeler: Hemanjiyom, transarteriyel embolizasyon, spinal kord basısı, cerrahi tedavi

Kanıt Düzeyi: Olgu sunumu, Düzey IV

Address: Mehmet Fatih Korkmaz, İnönü Üniversitesi Tıp Fakültesi, Ortopedi ve Travmatoloji AD, Malatya / Turkey
E-mail: fatih.korkmaz@inonu.edu.tr
Tel: 0422 3410660 / 5103
Fax: 0422 3410036
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INTRODUCTION:

Vertebral hemangiomas represent 2–3% of all spinal tumors that can be detected radiographically[11]. Most of these lesions are clinically silent, and they are detected incidentally during evaluation of other problems. They can be single or multiple, particularly in the lumbar and lower thoracic regions. In symptomatic patients, pain (54%) or variable neurological symptoms (45%) are often seen[4,12]. Here, we aim to present our experience of a unique case with multi-level vertebral hemangiomas causing cord compression at the T10 and L4 levels, and to evaluate the efficacy and safety of surgery in the treatment of multi-level vertebral hemangiomas causing neurological deficit.

CASE PRESENTATION:

The patient was a 69-year-old female who was treated with posterior surgery due to T12 vertebral collapse seven years previously, and who, after five asymptomatic years, had lower back pain, progressive strength loss in the lower extremities and difficulty in walking for four months, and urinary and fecal incontinence for four weeks. On examination, there was sensitivity of the lower thoracic vertebrae with fist percussion, paraparesis signs in the lower extremities, myelopathy, grade 3/5 muscle weakness, and sensory loss from the subjective non-dermatomal bilateral umbilical level. A Romberg test was positive. In the ankles, clonus and bilateral extensor plantar responses were present. Deep tendon reflexes were hyperactive. On rectal examination, it was detected that sensory innervation in the perineum and anus was present, but the patient had had urinary and fecal incontinence for four weeks (Figure-1. a and b).

Magnetic resonance imaging (MRI) was immediately performed in the axial and sagittal planes. In the T7, T10 and L4 vertebral corpuses during MRI screening, areas of involvement were observed that held the posterior elements at the L4 and T10 and facet joint cross, contracted the spinal cord and bilateral neural foramen, and had projections towards the anterior. They were also hypointense in T1W images and hyperintense in T2W images and contrast after IVKM, and had a trabecular view inside (Figure-1. c and d).

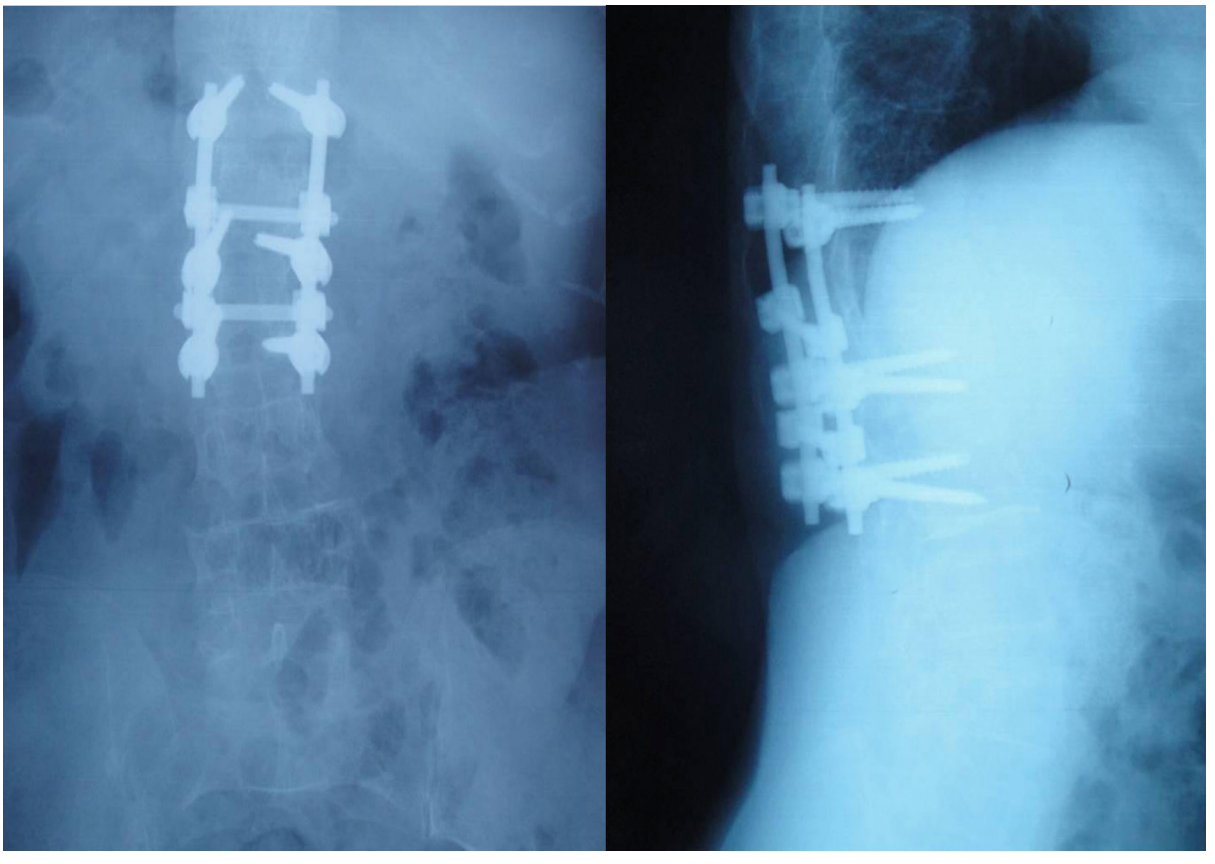

Figure-1.a) Preoperative AP X-ray

Figure-1.b) Preoperative lateral X-ray

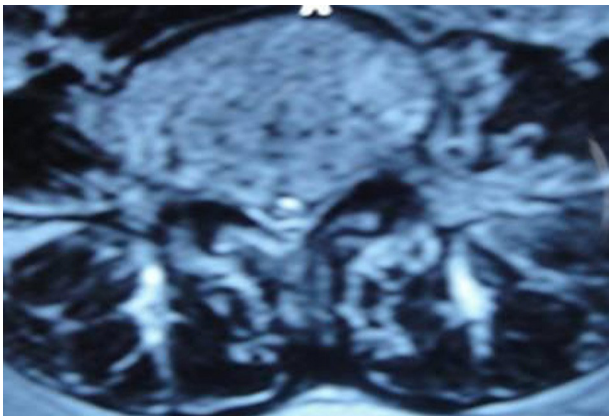

Figure-1. c) Preoperative sagittal MRI

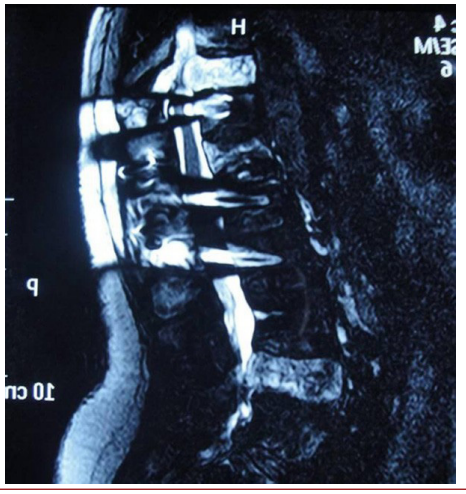

Figure-1. d) Preoperative axial MRI

The laboratory results gave normal values in terms of primary malignancy and metastasis. On screening, a 1.4 cm smooth anechoic cyst in the left lobe of the liver, a 1.5 cm parapelvic cyst in the left kidney, and a 3 cm mesenter herniated under the skin in the umbilical region were observed.

In spinal angiography performed one day before surgery, diffuse massive staining was observed in the T7, T10 and L4 vertebral corpuses. The Adamkiewicz artery went out from the left T9 intercostal artery. Embolization of the hemangioma in the T10 vertebral corpus was performed with polyvinyl alcohol (PVA) particles (300–500 micron) (Bead Block Terumo, UK) and n-butyl cyanoacrylate (Braun, Tuttlingen) from microcatheters placed with microwires from the bilateral T10 intercostal arteries. In the tests performed after embolization, no staining was observed, and embolization was shown to be effective (Figure-2).

Before rib resection, cord decompression was performed with a posterior anterior surgical technique combined with thoracotomy from the left 9–10 intercostal space. The screws at the T11, L1, and L2 vertebrae were removed, and fixation was provided with two rods and screws (bipedicular to T8, T9, T11, L1, and L2). A titanium cage filled with allograft was placed into the corpectomy area (Figure-3).

900 cc of intraoperative bleeding occurred and so blood transfusion of 2 units was performed. Neuromonitorization was carried out during surgery. There were no problems with intraoperative complications or the wound and implant during the follow-up period.

Biopsy samples obtained during corpectomy showing partly degenerated lamellar bone fragments with numerous nested lumens and congested large and small vessels verified the diagnosis of hemangioma (Figure-4).

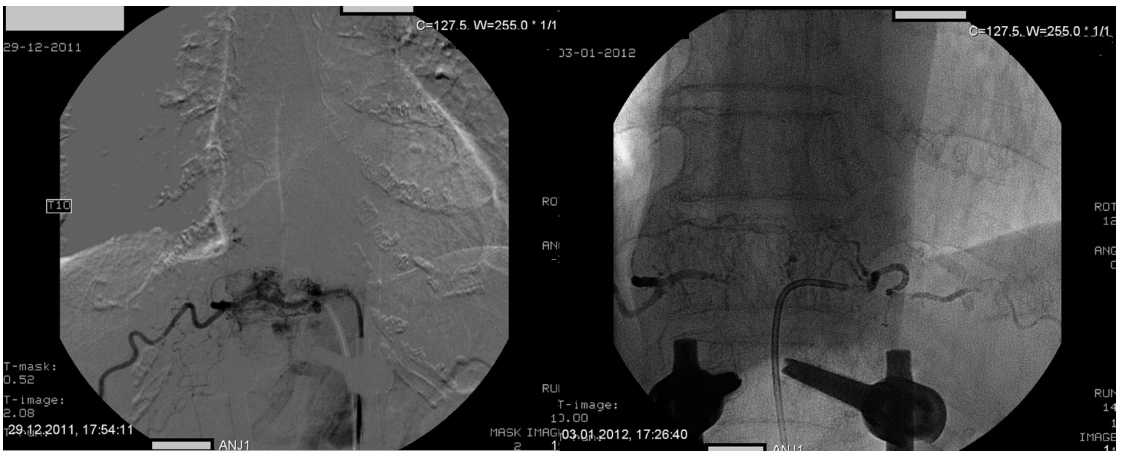

Figure-2. a) On injection performed from the left T10 lumbar vertebral artery, diffuse and massive staining can be observed in the T10 vertebral corpus. **b)** No staining can be observed in control angiography performed after embolization.

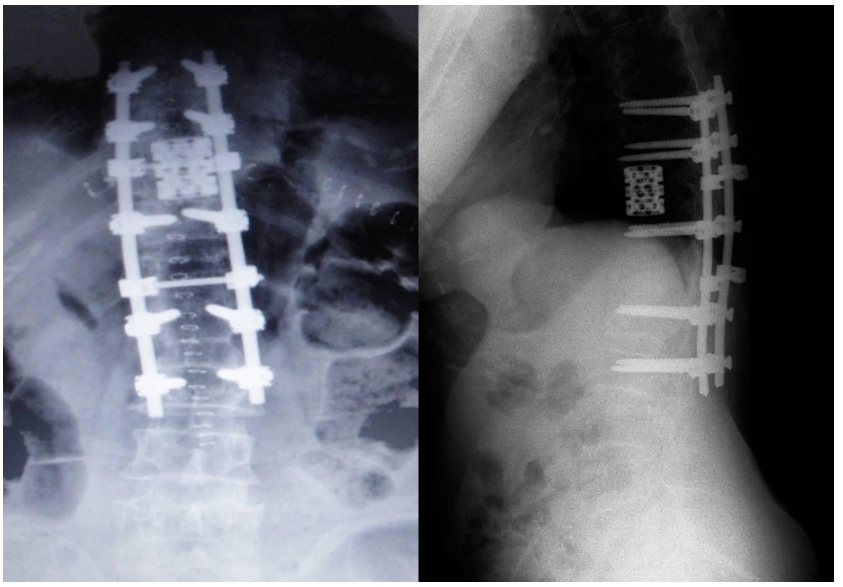

Figure-3.a) AP and **b)** lateral X-rays after T10 corpectomy.

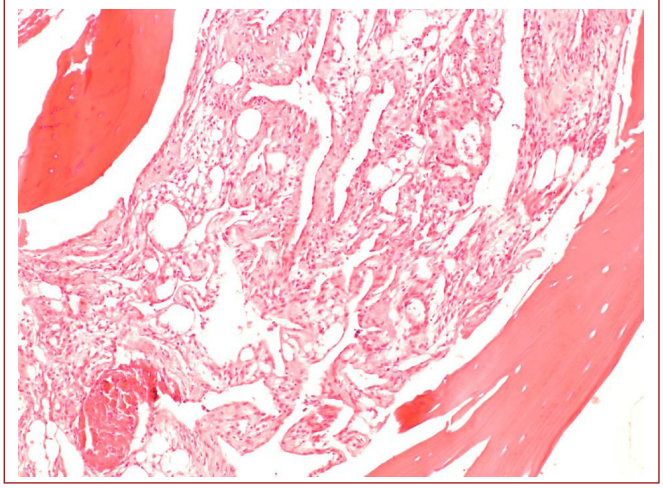

Figure-4. Many vessels (some dilated) between trabecular bone in pathological evaluation of corpectomy material of the patient (H&E 100×).

The patient was mobilized early with thoracolumbosacral orthosis in the postoperative period, and used this device for three months. The neurological signs and findings of the patient recovered rapidly. Four weeks post-operatively, a total of 4000 cGy adjuvant radiotherapy with a 200 cGy fraction dose was applied to the thoracolumbar vertebral region. During clinical follow-up, the patient became neurologically stable and independently mobile. There were no complications or problems with the implant or wound healing during the follow-up.

In a follow-up MRI taken after one year, complete resolution at the T10 level, an intraosseous hemangioma in the L4 vertebral corpus, moderate pressure on the thecal sac due to retropulsion of the posterior cortex, and severe central spinal stenosis were detected, but no severe clinical problems had presented (Figure-5). After 18 months postoperatively, surgical intervention to the L4 vertebral hemangioma was planned due to exacerbation of the pain and a landscape similar to the preoperative period. Surgery was delayed for three months because the patient refused surgery. On examination on the day of surgery, there was sensitivity of the lumbar vertebrae on fist percussion. A Romberg test was positive. The knee and ankle reflexes were bilaterally hypoactive and a straight leg raising test was positive at 45°. There was no walking disorder and the bladder and intestinal functions were not affected, but subjective non-dermatomal sensory loss was recorded on both sides. The laboratory studies gave normal values.

In spinal angiography performed under local surgery one day before surgery, the L4 vertebral corpus was stained diffusely and massively from the bilateral L3–4 lumbar arteries, especially the L4 lumbar artery.

Embolization of the hemangioma was performed with polyvinyl alcohol (PVA) particles (300–500 micron) (Bead Block Terumo, UK) and n-butyl cyanoacrylate (Braun, Tuttlingen) from microcatheters placed with microwire from the bilateral L3–4 lumbar arteries. In tests performed after embolization, no staining was observed and embolization was shown to be effective (Figure-6).

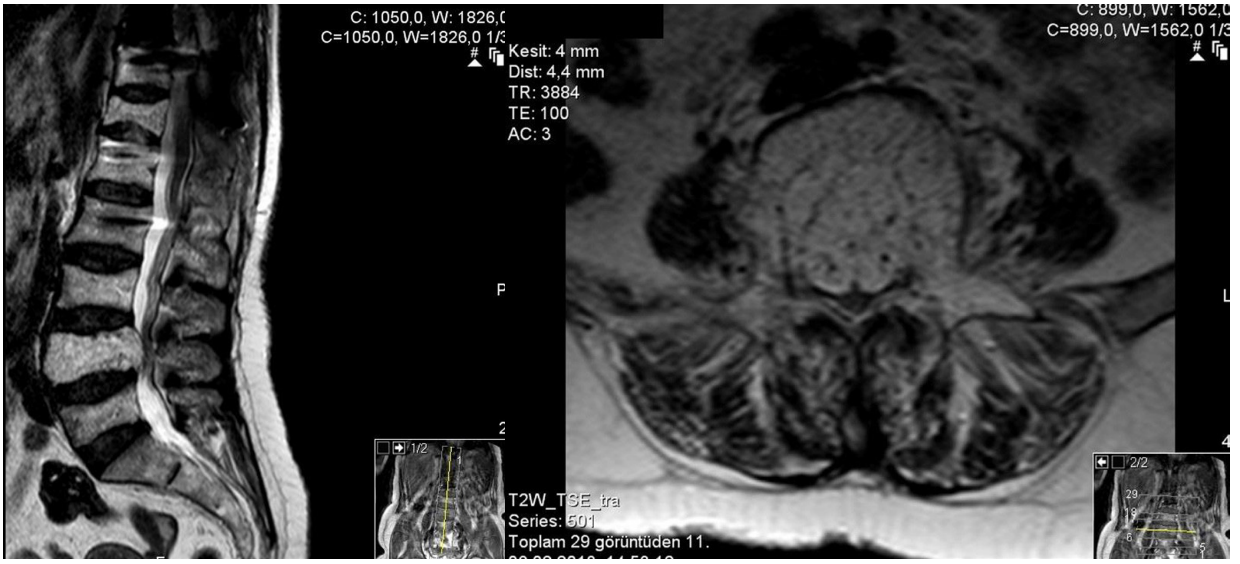

Figure-5.a) sagittal and **b)** axial MRIs one year after T10 corpectomy

Figure-6.a) On injection performed from the left L4 lumbar vertebral artery, diffused and massive staining can be observed in the L4 vertebral corpus. **b)** No staining can be observed in a control angiography performed after embolization.

PVCR was performed at the L4 vertebra and fixation was provided with two rods, two dominos and screws (bipedicular to L3 and L5). A titaniumcage filled with allograft was placed into the corpectomy area (Figure-7).

700 cc of intraoperative bleeding occurred and blood transfusion of 1 unit was performed. Neuromonitorization was carried out during surgery. There were no intraoperative complications or wound and implant problems during the follow-up period. Biopsy samples confirmed the diagnosis of hemangioma. The patient was mobilized early with thoracolumbosacral orthosis in the postoperative period.

The foot numbness and pain symptoms recovered. In the follow-up, the reflexes of the patient returned to normal bilaterally and the related neurological examination was normal. There was no need for adjuvant radiotherapy, as total vertebrectomy was performed. As a result, good clinical and neurological success was obtained.

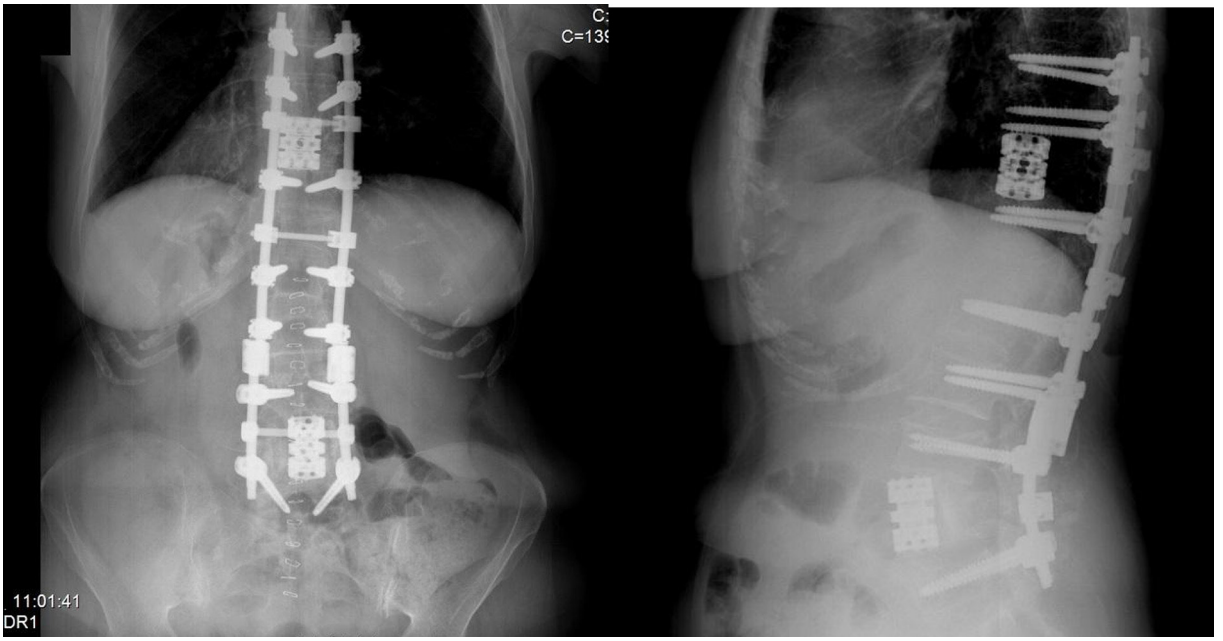

Figure-7.a) AP and **b)** lateral X-rays after PVCR at the L4 vertebra.

DISCUSSION:

Only 0.9–1.2% of all vertebral hemangiomas are symptomatic[4]. Hemangiomas, a small but important subgroup of symptomatic lesions, are characterized by bone expansion, extraosseous tumor extension, local blood flow disorders and rare compression fractures[1].

For every patient admitted due to myelopathic symptoms, early imaging, especially MRI, is important for the clinician to obtain a differential diagnosis of aggressive hemangioma and to determine suitable treatment. Aggressive hemangiomas generally mimic other aggressive spinal lesions, and so extra imaging methods such as CT, angiography and radiography are necessary for a clear diagnosis[20]. Both vertical trabeculation of vertebrae (also known as "prison view") in direct X-rays and spotted images of the vertebral corpus in CT are pathognomonic for these tumors. In addition to routine screening by MRI, determination of the fat content of a hemangioma can be valuable for prognosis. A isointense signal in T1-weighted images and a hyperintense signal in T2-weighted images are related to the hemangioma having a low fat content and large blood build-up, and are important in terms of the progression of the hemangioma[15,18].

There are different treatment approaches to vertebral hemangiomas. In patients with pain and without extraosseous projection or neurological deficit, percutaneous methyl methacrylate injection, radiotherapy, percutaneous intralesional alcohol injection, and transarterial embolization are performed.

For treatment of aggressive hemangiomas with extraosseous projection and neurological deficit, (laminectomy-corpectomy), surgical treatment is necessary. Because hemangiomas are tumors with rich vascularity, preoperative embolization is recommended in order to decrease the mortality and morbidity due to bleeding during surgery[9].

Although the mechanism of action of radiotherapy on vertebral hemangiomas is not exactly known, it is the most common treatment option for painful lesions. Various researchers have suggested a total dose of 30–40 Gy (1.8–2 Gy/fraction) if radiotherapy is used alone. However, delayed effects of radiotherapy cause a risk of radionecrosis in the spinal cord and vertebral corpuses[2]. Extra radiotherapy is suggested after subtotal and decompression surgery[4].

Percutaneous methyl methacrylate injection (percutaneous vertebroplasty) has recently gained popularity in the treatment of vertebral hemangiomas. This technique is ideal for reducing local pain, or stabilizing vertebrae with a risk of collapse. Percutaneous vertebroplasty is not recommended for cases with extraosseous projection or neurological deficit[7].

Transarterial embolization can be used alone for hemangiomas causing spinal cord compression. Endovascular embolization with particulate matter, such as polyvinyl alcohol foam, provides a dramatic but temporary remission[14,17]. One of the most important indications is preoperative embolization, which is performed to decrease severe bleeding, causing intraoperative problems, and postepidural hematoma risk, during the excision of aggressive vertebral hemangiomas developing neurological deficit due to compression[10]. Although transarterial preoperative embolization is a good option, there are studies showing that this has an insufficient effect on decreasing bleeding, due to occlusion of the feeding artery but insufficient embolization of the whole capillary

area[5]. Although ischemic complication has been reported after transarterial embolization in the literature[3], we think that the size of PVA used has an effect. No ischemic complications have been reported when PVA of 300–700 microns in size is used. We used 300–500 micron PVA in this case. Although we performed PVA and glue embolization at different levels and we observed staining in the postembolization controls, 900 cc and 700 cc of perioperative bleeding occurred, and transfusion of 2 units and 1 unit of blood was performed. When corpectomy was performed at two different levels, we think that the perioperative bleeding occurred less than expected and the pre-embolization was effective.

In early reported studies in the literature, surgical intralesional resection and laminectomy with or without fusion are recommended. The aim of surgery is bone decompression and excision of the tumor soft tissue compressing the neural elements by laminectomy or vertebrectomy.

Fox and Onofrio placed the treatment methods of these lesions in order according to the vertebral involvement degree and type. Strut grafted reconstruction and corpectomy is the most suitable intervention according to the degree of frontal vertebral involvement[4].

Patients with multiple noncontiguous vertebral hemangiomas should be closely followed up for other levels of progressive cord compression in future. Karaeminoğulları et al. suggested that preoperative endovascular embolization of these lesions decreases intraoperative bleeding and reduces the risk of postoperative epidural hematoma. They also suggested that surgical decompression and postoperative radiotherapy is an effective treatment for vertebral hemangiomas causing cord compression, which generally provides a full neurological recovery[8].

Stable SSEP/MEP records tell the surgeon that the motor functions are intact and that he can safely continue to surgery. In orthopedic surgery, this provides a safe and sensitive method for observation of spinal cord function[13,19].

In patients with subtotal excision, the addition of postoperative radiotherapy can significantly reduce recurrence. Fox and Onofrio reported that the addition of a postoperative >26 Gy dose of radiotherapy to the treatment can prevent tumor recurrence when the whole tumor cannot be reached[4].

In cases with slow-revealing neurological signs, radiotherapy or non-surgical treatments such as spinal embolization and vertebroplasty can be performed. In cases with cord compression, the neurological result after decompressive surgery is generally excellent, and this is the most commonly used method. We prefer to apply surgical treatment alone for symptomatic situations. We applied endovascular embolization before surgery to reduce the intraoperative and postoperative bleeding and epidural hematoma risk. We followed the patient with neuromonitorization during both surgeries. We preferred to apply postoperative adjuvant radiotherapy.

Vertebral hemangiomas are common benign tumors and are generally asymptomatic. Rarely, they can cause severe progressive cord compression. If treatment principles are properly applied with a multidisciplinary approach, they can be safely treated with surgery.

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