

Submitted: 12-01-2024 Accepted: 24-03-2024 Posted: 12-04-2024, Vol. (XXII)

Aggressive Thoracic Spine Hemangioma Treated by Total En Bloc Spondylectomy: A Case Report

Kukuh Ali Akbar ¹, Aries Rakhmat Hidayat ¹

¹Orthopedics& Traumatology Department, Faculty of Medicine, Universitas Airlangga/Dr. Soetomo General Hospital, Surabaya, Indonesia

KEYWORDS

Vertebral hemangioma; Spondylectomy; Spinal cord compression

ABSTRACT:

Introduction: Vertebral hemangioma (VH) is the most widely faced tumor in the vertebral column. It is generally asymptomatic and slowly growing. Moreover, the appearance of extraosseous extensions in vertebral hemangiomas is locally aggressive, causing neurological deficits, and should be distinguished from other vertebral hemangiomas based on its prognosis and treatment. Here we present a rare case of vertebral hemangioma caused by thoracic cord compression and healed by total en block spondylectomy

Objectives: We present a case of VH with extraskeletal expansion that caused the progressive neurologic deficit and was medicated by resection of the entire tumor.

Methods: A 33-year-old female came to the Soetomo General Hospital outpatient clinic with the main complaints of back pain and being unable to move her legs since September 2021. In July 2021, the patient felt tingling in her lower extremities. The patient was still able to walk, but she felt the weakness develop. She still can urinate, but she has a defecation problem. There is no history of a palpable mass in some places. The patient is a housewife and she has no previous history of any disease

Results: The patient felt tingling in her lower limbs. She didn't lose her to walk, but she felt the weakness develop. On the physical examination, there was a neurological deficit at the lower extremity. According to radiographic and histopathologic examination, the patient was diagnosed with VH at the T5 level. We then performed total en-block spondylectomy and posterior stabilization. As a result, the motoric part improved but sensory deficits still occurred. Neurological improvement happened during the follow-up in three months. An inevitable challenge awaits in terms of the medication of VH with extraskeletal expansion that causes neurologic deficits. Pre-operative diagnosis including radiologic and pathologic findings is essential for the strategy and decision for combative hemangioma. An unjustified diagnosis may lead to an inappropriate surgical strategy or the emergence of various complications.

Conclusions: Vertebral hemangiomas with extraosseous extension causing spinal cord compression should be considered an aggressive benign tumor, and total excision that includes a tumor margin is indicated.

1. Introduction

Vertebral hemangioma (VH) is the most widely faced tumor in the vertebral column. It is generally asymptomatic and slowly growing (1). In 2–3% of spinal tumors and up to 12 % of general autopsies, a number of cases of VH can be found (2). In spite of that, vertebral hemangiomas that are symptomatic are rare, with a percentage of 0.9–1.2% of all vertebral



Submitted: 12-01-2024 Accepted: 24-03-2024 Posted: 12-04-2024, Vol. (XXII)

hemangiomas. VH is mainly founded in the thoracic spine with the lumbar spine afterward (1,3). The mechanisms of the compression of hemangiomas to the spinal cord include an expansion of the tumor into the extradural cavity, enlargement of the spinal body, compression fracture of an affected spine, and hematoma secondary to the tumor (4). Moreover, the appearance of extraosseous extensions in vertebral hemangiomas is locally aggressive, causing neurological deficits, and should be distinguished from other vertebral hemangiomas based on its prognosis and treatment (5). VH has no symptoms in most of the population and it is founded on imaging Plain examination. radiography shows "corduroy cloth", the palisading view due to the bulking of bony trabeculae. Computed tomography (CT) scan shows a "honeycomb" or a "polka dot" pattern in tumors that have low-density areas with bony tubercles. Of all of these modalities, the most susceptible and greatest modality for diagnosing VH is magnetic resonance imaging (MRI) (6). There are several treatments, whether it is alone or combined in the last 20 years, such as bone cement injection, decompressive surgery with or without tumor resection, radiotherapy, and arterial embolization. However, there is still controversy surrounding what is the greatest option of medication for this disease (7). Therefore, we present a case of VH with extraskeletal expansion that caused the progressive neurologic deficit and medicated by resection of the entire tumor and had improvement result after surgery

2. Objectives

Here we present a rare case of the vertebral hemangioma cause of thoracic cord compression and treated by total en block spondylectomy.

3. Methods

A 33-year-old female came to the Soetomo General Hospital outpatient clinic with the main complaints of back pain and being unable to move her legs since September 2021. In July 2021, the patient felt tingling in her lower extremities. The patient was still able to walk, but she felt the weakness develop. She still can urinate, but she has a defecation problem. There is no history of a palpable mass in some places. The patient is a housewife and she has no previous history of any disease.

4. Results

On the physical examination, there was tenderness at the T5 level. On manual motor testing, the patient had muscle weakness in the lower extremities with 0/0 strength. All muscle groups at the upper extremity remain normal with 5/5 strength. The Pinprick test shows a decline in sensation, whereas normal touch and proprioception are founded at the T5 level. Positive Babinsky pathological reflex happened at both lower extremities.

In radiology examination, no abnormalities showed at plain radiography no abnormality, whereas bone sclerosis of vertebral body T5 level and "polka dot" sign occurred in CT scan, suggesting hemangioma (Figure 1). MRI showed the tumor at T5 with an extraskeletal expansion which cause compression at the spinal cord that appeared hypointense on T1-weighted MRI, hyperintense on T2-weighted and T1-weighted MRI (Figure 2). This imaging suggested vertebral hemangioma. Angiography showed no feeding artery that going through the tumor, while histopathology findings suggest hemangioma (Figure 3).

The neurological deficit and decline of the patient encourage us to do an immediate treatment, therefore we decided to do a



Submitted: 12-01-2024 Accepted: 24-03-2024 Posted: 12-04-2024, Vol. (XXII)

resection of the entire tumor by total en bloc spondylectomy (TES)(Figure 4). For this process, the vertebral body was replaced with a titanium cage filled with an auto bone graft from minced rib not contaminated by the tumor and posterior stabilization composed of a pedicle screw and rod with the placement in the T3 to T7 pedicles (Figure 5). She was sent home 5 days after surgery with thoracolumbar spine orthosis without any perioperative ravelment.

Postoperatively, there was an improvement in the patient's motoric function and the strength in the lower extremity is 2/2. After three month evaluation, the patient's motoric function increased and the strength in the lower extremity is 3/3, however, there was still a sensory deficit

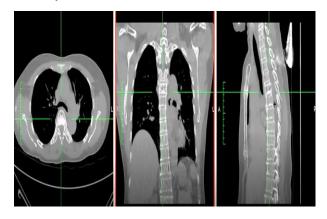


Figure 1. (A) Axial view CT showed bone sclerosis of vertebral body T5. (B) Coronal view showed "polka dot" sign suggests hemangioma. (C) Sagital view showed destruction of vertebral body T5



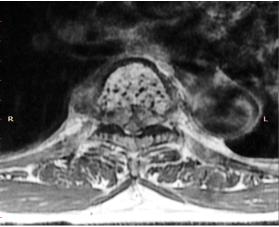


Figure 2. (A) MRI Preoperative showing invasive vertebral tumor with extraskeletal expansion that compresses spinal canal. (B) Axial MRI showed destruction of vertebral body T5 and mass expanded the spinal canal

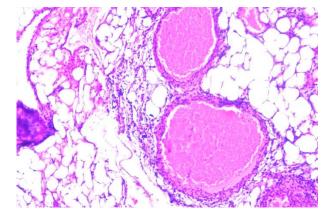
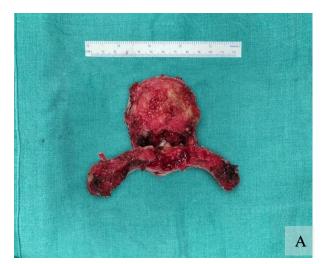


Figure 3. Staining of hematoxylin and eosin Histopathology suggest hemangioma



Submitted: 12-01-2024 Accepted: 24-03-2024 Posted: 12-04-2024, Vol. (XXII)







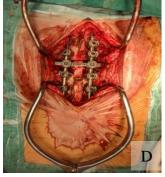


Figure 4. (A)(B) Total Spondylectomy was performed to resect the VH. (C)
Circumferential decompression of the spinal cord showed during en bloc spondylectomy on the intraoperative photograph. (D) posterior stabilization with pedicle screw and rod was performed



Figure 5. (A) AP view of post-operative radiograph showed pedicle screw placement at level T3 to T7. (B) Lateral view showed a titanium cage at T5 filled with an auto bone graft

5. Discussion

This case report shows aggressive vertebral hemangioma because spinal of compression. Its physical examination and diagnostic features are listed. In spite of the scarcity, VH with extraskeletal expansion must be included in the differential diagnosis of cases similar to this one, especially when the characteristic trabecular polka-dot pattern is verifiable, most generally in the thoracal vertebrae. Spinal cord compression at the T5 level based on MRI shows spinal cord compression that leads to a neurological deficit. We planned to perform embolization before the operation to reduce intraoperative bleeding. Nevertheless, the angiography shows no feeding artery that goes through the tumor. During the surgery, the bleeding was tolerable because of the resection with total en bloc spondylectomy (TES). After three months, she had no pain and increased motor function. The patient could do daily activities but there was still sensory deficit.



Submitted: 12-01-2024 Accepted: 24-03-2024 Posted: 12-04-2024, Vol. (XXII)

The main surgical procedure for vertebral hemangioma perennially has always been surgical intrusion resection combined with postoperative radiation since resection of the linked with entire tumor is elevated intraoperative morbidity due to ponderous bleeding. Hence, radical resection has been recommended for hemangiomas with an extraskeletal expansion causing a neurologic deficit (2,8). Laminectomy decompression of the spinal cord is inadequate for thoracic spine tumors. Innumerable bleeding from highly vascular tumors tends to happen in palliative debulking surgery and early recurrence has been declared. TES have several complications during surgery and after surgery like bleeding, dura tear and neurogical deteroration. Murakami et al. stated in a retrospective review of 79 patients with spinal tumors treated with TES that of the patients treated with TES, after the surgery there is no neurologic deterioration. From this study, it could be discovered that TES is the safest procedure without noticing the blood flow of the spinal cord (9).

Refrences

- [1] Kato S, Kawahara N, Murakami H, Demura S, Yoshioka K, Okayama T, et al. Surgical management of aggressive vertebral hemangiomas causing spinal cord compression: Long-term clinical follow-up of fi ve cases. Journal of Orthopaedic Science. 2010;15(3):350–6.
- [2] Fox MW, Onofrio BM. The natural history and management of symptomatic and asymptomatic vertebral hemangiomas. J Neurosurg. 1993;78(1):36–45.
- [3] Hu W, Kan SL, Xu H bin, Cao ZG, Zhang XL, Zhu R sen. Thoracic aggressive vertebral hemangioma with neurologic deficit A retrospective cohort study. Medicine (United States). 2018 Oct 1;97(41).

- [4] Acosta Jr FL, Dowd CF, Chin C, Tihan T, Ames CP, Weinstein PR. Current treatment strategies and outcomes in the management of symptomatic vertebral hemangiomas. Neurosurgery. 2006;58(2):287–95.
- [5] Blecher R, Smorgick Y, Anekstein Y, Peer A, Mirovsky Y. Management of symptomatic vertebral hemangioma: follow-up of 6 patients. Clin Spine Surg. 2011;24(3):196–201.
- [6] Dobran M, Mancini F, Nasi D, Gladi M, Sisti S, Scerrati M. Surgical treatment of aggressive vertebral hemangioma causing progressive paraparesis. Annals of Medicine and Surgery. 2018 Jan 1;25:17–20.
- [7] Ogawa R, Hikata T, Mikami S, Fujita N, Iwanami A, Watanabe K, et al. Total En Bloc Spondylectomy for Locally Aggressive Vertebral Hemangioma Causing Neurological Deficits. Case Rep Orthop. 2015;2015:1–7.
- [8] Rudnick J, Stern M. Symptomatic thoracic vertebral hemangioma: A case report and literature review. Arch Phys Med Rehabil. 2004;85(9):1544–7.
- [9] Shimizu T, Murakami H, Demura S, Kato S, Yoshioka K, Yokogawa N, et al. Total en bloc spondylectomy for primary tumors of the lumbar spine. Medicine (United States). 2018;97(37).