CASE REPORT

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Piecemeal resection of aggressive vertebral hemangioma using real-time navigation-guided drilling technique

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ABSTRACT

Vertebral hemangiomas are the most common benign vertebral tumors and are usually asymptomatic. Aggressive subtypes of the tumor, called aggressive VHs (AVHs), can become symptomatic with extraosseous extensions and require surgical removal. We present a case of AVH in a 36-year-old man presenting with low back pain and right leg pain that persisted for three months. Imaging studies showed a Th12 vertebral tumor that extended into the spinal canal and was squeezing the spinal cord. Computed tomography (CT)-guided biopsy indicated vertebral hemangimoa. Following preoperative arterial embolization, piecemeal gross total resection was attained under navigation guidance. He was left with no neurological deficit and remained well at the 12-month postoperative folow-up. Since AVHs are benign tumor, piecemeal removal of the tumor can be selected. However, disadvantage of the approach include difficulty of making decision how much to remove the front part of the vertebral body close to thoracic descending aorta. Furthermore, when the tumor tissue is too hard to curett, manipulation in tight spaces near the spinal cord carries the risk of damaging it. Navigation-guided drill is highly helpful for real-time monitoring of ongoing tumor resection. It enables safely resection of the tumor especially in the anterior cortical surface of the vertebral body and easily resection even hard tumors. This method results in reducing residual tumor and maintaining safety resection.

Keywords: aggressive vertebral hemangioma, CT navigation system, preoperative arterial embolization, piecemeal total resection, navigation-guided drill

Abbreviations:
VHs: vertebral hemangiomas
AVHs: aggressive vertebral hemangiomas
TES: total en bloc spondylectomy
3D: three-dimensional
OPLL: ossification of the posterior longitudinal ligament
CT: computed tomography
18F-FDG PET: 18F-fluoro-deoxy-glucose positron-emission-tomography

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INTRODUCTION

Vertebral hemangiomas (VHs) are the most common benign angiomatous tumors in the spinal column.[1] Most VHs are latent and do not require specific treatment; only 1% become active and symptomatic with extension into the spinal canal that leads to compression of the spinal cord.[2] They are called aggressive VHs (AVHs)[2] and require surgical treatment for spinal decompression with stabilization.[3] Treatment for AVHs are technically challenging because they are mainly located in the anterior spinal column. Incomplete resection could lead to local recurrence, and decompression alone without anterior spinal reconstruction could provoke vertebral body collapse. On the other hand, good long-term outcomes are warranted when complete tumor removal with anterior reconstruction is achieved by using either total en bloc spondylectomy (TES) or the piecemeal tumor resection technique.[4] Because of the benign nature of the tumor, we believe piecemeal tumor resection is more feasible and safer if preoperative arterial embolization and intraoperative navigation can be performed. Recently, several reports indicated three-dimensional (3D) navigation system with a real-time navigated drill system (Stealth-Midas™, Medtronic Inc.) enables more accurate screw insertion[5,6] or safety removal of ossification of the posterior longitudinal ligament (OPLL)[7] via anterior approarch. However, there have been no reports describing its application to the removal of vertebral body tumors. We describe a case of AVH that was successfully treated by piecemeal complete tumor resection with real-time navigated drilling technique assisted with preoperative arterial embolization.

CASE PRESENTATION

Patient History and Evaluation

A 36-year-old man with a height of 186 cm, weighing 120 kg with BMI of 34.7, presented with low back pain and right leg pain that persisted for three months. He had a history of surgery for thyroid papillary carcinoma seven years previously, which was periodically monitored without tumor recurrence. He had no muscle weakness in either leg, and was otherwise neurologically intact. Computed tomography (CT) scan showed a "polka dotted" appearance and the tumor protruded into the spinal canal at the Th12 level (Fig. 1 A and B), which was significantly enhanced on CT with contrast material (Fig. 1 C and D). The tumor was depicted as a hypointense lesion with a focal hyperintense area on T1-weighted MRI (T1WI) (Fig. 1 E and F) and a slightly high intensity signal mass on T2-weighted MRI (T2WI) (Fig. 1 G and H). T1WI with gadolinium demonstrated remarkable contrast enhancement with epidural tumor extension and spinal cord compression (Fig. 1 I and J). 18F-fluoro-deoxy-glucose positron-emission-tomography (18F-FDG PET) showed low FDG uptake of the tumor (Fig. 2 A and B). We performed a CT-guided needle biopsy for histological research ahead of tumor removal; the result of which indicated a VH.

Preoperative embolization

We proceeded with surgical tumor removal via the posterior approach with the piecemeal resection technique. To reduce intraoperative bleeding, preoperative arterial embolization preceded the surgery by one day. Selective spinal angiogram showed tumor-feeding vessels arising from the bilateral T11 and T12 segmental arteries (Fig. 2 C, E). Vascular supply to the tumor was markedly reduced after arterial embolization of the bilateral T11 and T12 segmental arteries (Fig. 2 D, F).

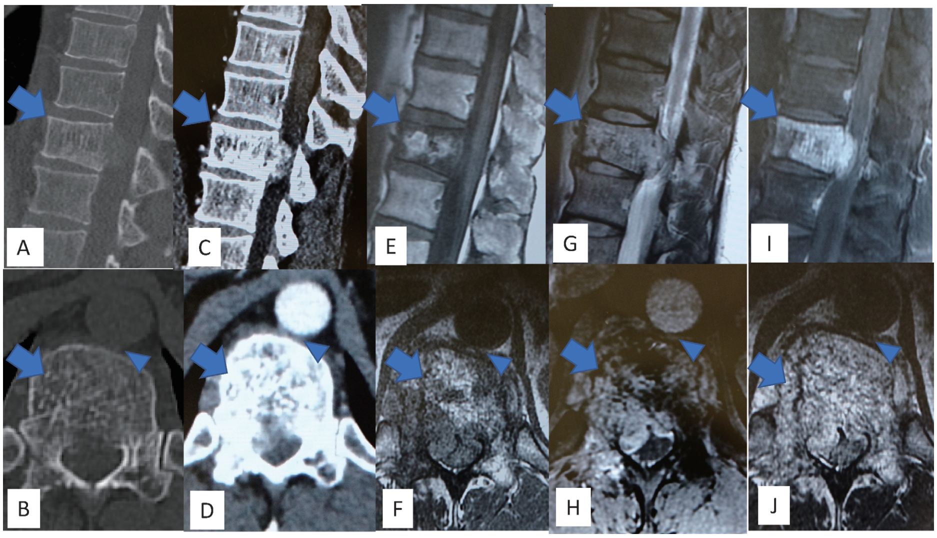

Fig. 1 Preoperative images

Fig. 1A and B (arrow): Computed tomography (CT) scan of thoracolumbar spine shows a "polka dotted" appearance and the tumor protruded into the spinal canal.

Fig. 1C and D (arrow): CT scan with contrast material shows a well-enhanced tumor.

Fig. 1E-J (arrow): Magnetic resonance imaging (MRI) of the thoracolumbar spine showed a mass lesion in the spinal canal and signal change of the vertebral body, high intensity signal on T1-weighted MRI (E and F), and on T2-weighted MRI (G and H). T1-weighted MRI with gadolinium demonstrated remarkable enhancement of the tumor. The surrounding soft tissue of the descending thoracic aorta seemed to be thick and adhered closely to the vessel. (B, D, F, H, J arrowhead)

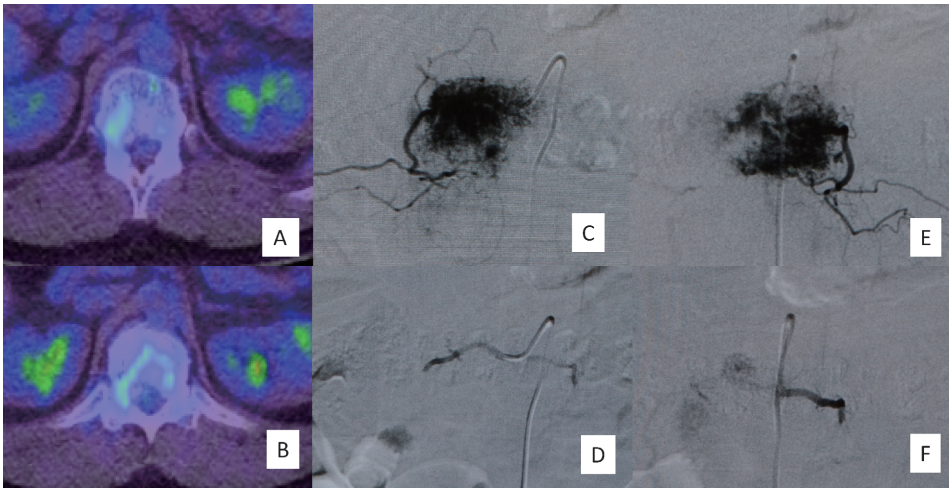

Fig. 2 Pereoperative positron-emission-tomography and angiography

Fig. 2A and B: 18F-fluoro-deoxy-glucose positron-emission-tomography (18F-FDG PET) shows low uptake of FDG.

Fig. 2C-F: Preoperative spinal angiography showing tumor blush via the T12 intercostal artery. (C: right E: left) After embolization, the tumor blush completely disappeared. (D: right, F: left)

Surgical Procedure

The patient was placed in a prone position for posterior approach. The incision performed as a straight posterior midline. Following a subperiosteal dissection, the Th12 vertebra was exposed to the tips of the transverse process. The dissection was then carried out laterally, exposing the Th12 rib. After exposure of posterior elements from Th9 to L3 spine, a reference arm was clamped onto L1 spinous process. Bony surface registration for navigation based on preoperative CT was conducted, we successfully inserted pedicle screws from Th9 to Th11 and L1 to L3 under navigation guidance. These screws were connected on one side with a rod without any attempt at spinal correction. The resection began with a removal of the Th12 posterior elements and we continued to refer to the navigation guidance throughout the procedure for tumor removal. (Fig. 3A) Following a total laminectomy and bilateral total foraminal unroofing to expose the neural elements at T12 level, the transverse process and the corresponding rib on the working side (on the opposite side of the temporary rod) were removed to expose the lateral wall of the pedicle. Pediculectomy was conducted using navigated drill system. Although surrounding periosteum was thickened and adhered to the cortical surface of the tumor, the meticulous subperiosteal dissection was carefully deepened following the lateral wall of the tumor. Cancellous bone of the vertebral body tumor connecting to the pedicles was drilled referring to the real-time navigation. Real-time navigated decancellation drilling and piecemeal tumor resection leaving egg-shell thin layer of cortical wall was performed, which was followed by the remaining egg-shell lateral wall removal by using a small rongeur. (Fig. 3 B-D)

The vertebral body tumor and the intervening discs removal in a piecemeal fashion gradually heads towards the medial side and over to the other half of the vertebral body, keeping a thin shell of bony posterior vertebral wall beneath the dural tube. Following the resection of the posterior wall on the working side, another temporary rod was inserted to the working side and securely locked to the screws. Then the rod on the other side was removed to allow resection on that side. The same procedure was carried out on the opposite side. However, the periosteum was closely adhered to the ventral surface of the vertebral body and descending aorta, therefore the only ventral cortical wall of the tumor adjoining the descending thoracic aorta was intentionally left behind. When an adequate amount of vertebral body tumor was removed, the entire posterior vertebral wall ventral to the dural tube was removed with an Epstein reverse-cutting curette and pituitary forceps. (Fig. 1 and 3B arrowhead). Two expandable cages for lumbar vertebral body replacement were placed side-by-side. The surgery took 585 minutes with a total blood loss of 2650 mL. Histopathological examination showed numerous thin-walled blood vessels without consistent atypia (Fig. 4A,B). The tumor was diagnosed as a spinal hemangioma. He was discharged without any neurological deficits. He was followed up in an outpatient clinic and showed a successful outcome (Fig. 4 C-F).

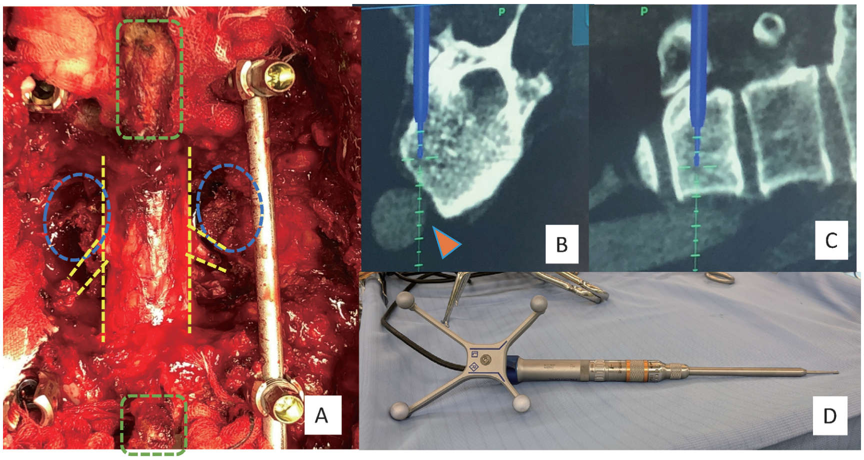

Fig. 3 Description of the procedure

Fig. 3A: Intraoperative photographs (A; green dotted line: spinous process; yellow dotted line: spinal cord; blue dotted line: pedicle). The rod was placed to maintain spinal stability during tumor resection.

Fig. 3B and C: After pedicle screw insertion, tumor removal was performed in a piecemeal fashion using navigation-guided drill. The spinal navigation system enabled safe extraction within a narrow operating window even if the tumor tissue is too hard to curett.

Fig. 3D: Navigated drilling system (Stealth-Midas™, Medtronic Inc.).

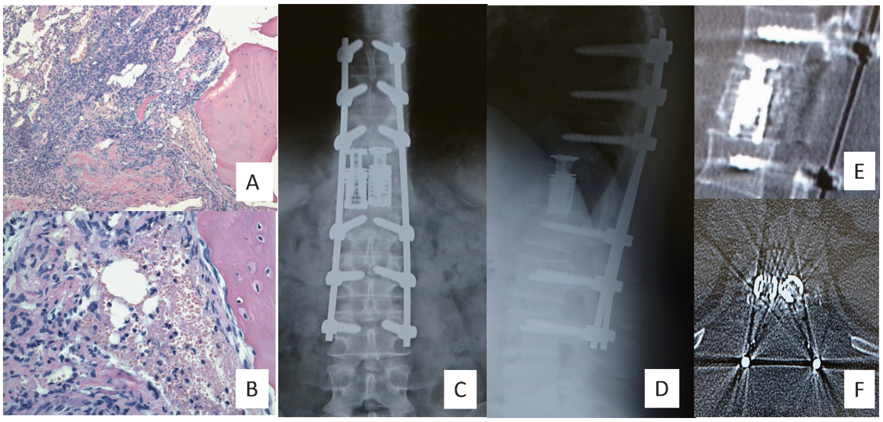

Fig. 4 Pathological results and postoperative radiographic images

Fig. 4A: Histopathological examination showed the scattered epithelioid areas and blood cells (hematoxylin and eosin; original magnification 40).

Fig. 4B: Thin-walled capillary vessels separated without consistent atypia (hematoxylin and eosin; original magnification 200).

Fig. 4C and D: Postoperative radiograph showed excellent positioning of pedicle screws and two mesh cages.

Fig. 4E and F: Postoperative CT scan showed gross total removal of the tumor was confirmed with only the cortical surface adjoining the descending thoracic aorta intentionally left intact.

DISCUSSION

The primary treatment of choice for AVH has been surgical tumor removal, including surgical decompression, piecemeal complete resection, and en bloc spondylectomy. Radiotherapy is usually suggested as an adjuvant therapy after subtotal tumor removal.[8-10] However, local recurrence after subtotal tumor removal combined with radiotherapy is reported and adjuvant radiotherapy can cause radionecrosis, radiation-induced myelitis, and secondary malignancy. Furthermore, radiotherapy could cause further surgeries more difficult to perform.[2,4] From the persepective of selecting a surgical method, en-bloc tumor resection is not necessary because of the low malignant nature of AVHs and technical difficulty.[11] Acosta et al[12,13] reported good outcomes of intralesional spondylectomy with anterior reconstruction and pedicle screw fixation without adjuvant radiotherapy. Kato et al also reported good long-term outcomes of five patients with AVH after complete resection by using either total en bloc spondylectomy (TES) or piecemeal technique.[4] From a technical standpoint, we consider piecemeal tumor resection to be more feasible with the assistance of preoperative tumor embolization and navigation guidance.

Many publications have reported the usefulness of navigation-guided spinal surgery. Firstly, spinal navigation greatly contributes to accurate pedicle screw placement. A single-center evaluation of 4,500 thoracolumbar pedicle screws revealed that the placement accuracy improved from 93.9% to 96.4 % ($P=0.01$) in the lumbar spine and improved from 79.0% to 95.5% ($P<0.01$) in the thoracic spine by using CT-based navigation.[14] In addition, the navigated drill system improves integrity of screw insertion especially in the case of thin pedicles, such as in the cervical spine.[5,6]

Secondly, spinal navigation is also highly helpful for real-time monitoring of ongoing tumor resection,[15-17] which enables a better resection rate of tumors and lower complication rates. It has also been reported that the use of the navigated drilling system improved the resection rate of OPLL via anterior approach.[7] This method can be effectively applied to removing vertebral body tumors. Transpedicular all-posterior vertebral body resection tends to yield only poor visualization of the ventral cortical surface of the vertebral body, which appeared adherent to the descending thoracic aorta in the present case. Intraoperative ultrasound (US) provides a good description of the anterior epidural space of the spinal cord[16-18]; however, the space ventral to the vertebral body is too deep to be visualized. Intraoperative CT-based or O-arm navigation system could provide an excellent description of the surrounding area of the vertebral body, as shown in the present case (Fig. 3B,C). We can easily recognize throughout the procedure where we are and where we are heading by reference to navigation guidance. In addition, the real-time navigated drill system enables safe decancellation of the vertebral tumor without additional force exerted within a narrow operating window beside the spinal cord, This technique is significantly helpful, particularly when we remove rigid vertebral body tumor aiming to leave cortical wall of the tumor adjacent to the thoracic descending aorta to prevent iatrogenic aortic injury.

Hemangiomas are highly vascular tumors, and intraoperative massive bleeding could become a life-threatening complication. Preoperative embolization can contribute to a reduction in intraoperative blood loss with high success rates and a high degree of safety.[19] A recent systematic review, which included 51 cases of surgically-treated AVH, reported the effectiveness of preoperative embolization. Blood loss in the embolized treatment group (980±683 mL) was significantly lower than that in the non-embolized control group (1,629±946 mL).[20] The systemic review includes various types of surgical procedures. They varied from posterior decompression and vertebroplasty to circumferential en-bloc spondylectomy and stabilisation. Despite the fact that the different procedures are not equivalent with regard to invasiveness of the approach, selection was not performed in this context. Therefore, they cannot be simply compared to 2650 ml of bleeding in the present case. In addition, the huge physique with high value of BMI in the present case may have contributed to the increased amount of bleeding despite meticulous attempts to achieve hemostasis throughout the procedure. Segmental arteries at the level of the tumor as well as at least one level above and below the index level should be assessed on diagnostic angiography to identify tumor-feeding vessels and potential intersegmental anastomoses.[21] Experimental studies demonstrated that embolization of the segmental arteries at three successive levels, including the index level and the rostral and caudal adjacent levels, reduced blood flow of the index vertebrae by one-fourth without compromising spinal cord function.[22,23] Thus, segmental arteries of the rostral and caudal adjacent levels as well as the index level should be targeted for embolization with great care to preserve radiculomedullary arteries supplying the spinal cord. Preoperative embolization should be scheduled 24 hours ahead of surgery when it is most effective.[24] Complete embolization is not always possible because AVHs may have blood supply provided by radiculomedullary arteries.[25] In such cases, superselective embolization of the tumor-feeding vessels branching from the radiculomedullary artery is needed to prevent spinal cord ischemia. If embolization is not satisfactory, en bloc spondylectomy might be considered. In the present case, tumor-feeding vessels stemming from segmental arteries at the index level (Th12) as well as one rostral adjacent level (Th11) were successfully embolized. L1 segmental arteries were spared because they provided blood supply to the spinal cord (not shown).

CONCLUSION

Complete resection of AVHs is technically demanding. Preoperative arterial embolization and navigation-guided drilling technique greatly contribute to achieving gross total resection of the tumor safely in a piecemeal fashion.

CONFLICTS OF INTEREST

The authors have nothing to disclose.

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