

Delayed epidural pseudoaneurysm following cervical laminectomy and instrumentation in a patient with canal stenosis secondary to skeletal fluorosis

A case report

Yinze Diao, MD, Yu Sun, MD*, Shaobo Wang, MD, Fengshan Zhang, MD, Shengfa Pan, MD, Zhongjun Liu, MD

Abstract

Rationale: The typical intraoperative presentation of vertebral artery injury (VAI) usually involves profuse bleeding and requires immediate treatment. However, an occult VAI may occur intraoperatively and result in delayed life-threatening epidural pseudoaneurysm several days postoperatively.

Patient concerns: A 21-year-old man with compressive cervical myelopathy resulting from canal stenosis of skeletal fluorosis underwent decompression of C1 to C7 and instrumentation from C2 to C7. No impressive bleeding event occurred during the operation. On postoperative day 40, progressive quadriplegia developed.

Diagnoses: Pseudoaneurysm of the VA was established by angiography.

Interventions: After occlusion of the right VA, the patient underwent hematoma clearing.

Outcomes: Fortunately, the patient experienced significant recovery of neurologic function after the second surgery.

Lessons: From this case, we realize even in the absence of obvious signs of VAI during a cervical operation, postoperative evaluation should be mandatory for suspected bleeding events occurring at VAI-prone sites during surgery. Moreover, the bone morphological abnormality of skeletal fluorosis was determined to be the most important risk contributing to VAI in this case. The safety limits of bone removal should be determined preoperatively to avoid the effects of bone morphological abnormalities.

Abbreviations: mJOA= modified Japanese Orthopaedic Association, VA = vertebral artery, VAI = vertebral artery injury.

Keywords: cervical spine surgery, late-onset bleeding, pseudoaneurysm, skeletal fluorosis, vertebral artery injury

1. Introduction

Iatrogenic vertebral artery injury (VAI) is a rare but catastrophic complication of cervical or craniocervical surgery, with an overall incidence between 0.14%^[1] and 0.2%^[2] in large series. Usually, the typical intraoperative presentation of VAI is sudden and profuse bleeding that is difficult to control. However, in the current case, VAI during operation presented as a mild hemorrhage and was easily controlled by compression with pieces of gelatin sponge, similar to venous plexus rupture in the upper cervical region. This led to the neglect of possible VAI by

the surgical team and resulted in the development of a life-threatening late-onset pseudoaneurysm. Hypertrophy of bone and cervical canal stenosis secondary to skeletal fluorosis contributed to the injury, and this possibility has not been thoroughly discussed in the literature.

2. Case report

The study was approved by the Institutional Review Board and Ethics Committee of Peking University Third Hospital. Informed consent was obtained from the individual participant included in the study.

A 21-year-old male from a known high fluorine area presented progressing neurologic dysfunction. Imaging examinations demonstrated bone hypertrophy and canal stenosis (Fig. 1). The patient underwent posterior decompression with resection of the posterior arch of atlas as well as laminectomy from C2 to C7. Pars screws were applied on C2. Lateral mass instrumentation was performed from C3 to C7. No severe bleeding occurred during the operation, and the estimated blood loss was 500 mL. The mJOA score recovered from 4.5 points preoperatively to 13 at 4 weeks postoperatively.

On postoperative day 40, however, the patient developed progressive quadriparesis within 4 hours. A cervical MR examination demonstrated a large epidural hematoma and spinal cord compression (Fig. 2A). A bedside ultrasonography showing swirl-like arterial blood flow signals in the hematoma

Editor: Bernhard Schaller.

The authors have no conflicts of interest to disclose.

Department of Orthopaedics, Peking University Third Hospital, Beijing, China.
Beijing Key Laboratory of Spinal Disease, Beijing, China.

* Correspondence: Yu Sun, Department of Orthopaedics, Peking University Third Hospital, Beijing Key Laboratory of Spinal Disease, 49 North Garden Rd, Haidian District, Beijing 100191, China (e-mail: puthresearch@sina.com).

Copyright © 2018 the Author(s). Published by Wolters Kluwer Health, Inc. This is an open access article distributed under the Creative Commons Attribution-ShareAlike License 4.0, which allows others to remix, tweak, and build upon the work, even for commercial purposes, as long as the author is credited and the new creations are licensed under the identical terms.

Medicine (2018) 97:8(e9883)

Received: 16 July 2017 / Received in final form: 11 December 2017 / Accepted: 24 January 2018

<http://dx.doi.org/10.1097/MD.0000000000009883>

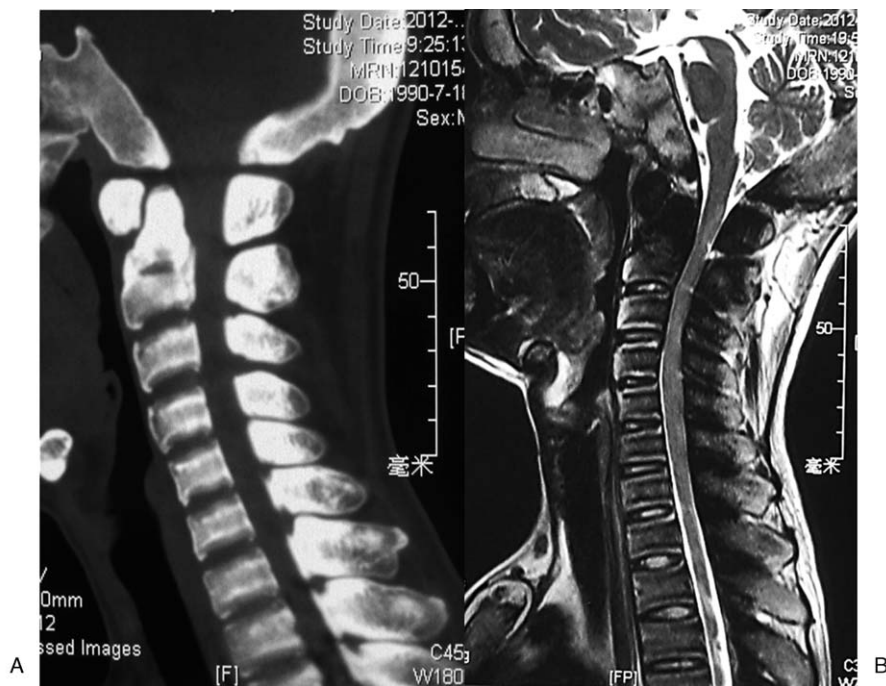


Figure 1. Preoperative CT and MRI of the cervical spine. (A) Preoperative CT demonstrated significant hypertrophy of bone and canal stenosis. (B) Preoperative MRI demonstrated cervical cord compression.

indicated a pseudoaneurysm from the right VA. Angiography showed bleeding of the right V3 segment in the vertebral artery groove of the atlas (Fig. 2B). The right VA was occluded with embolization microcoils. The patient then underwent hematoma clearing and experienced significant recovery of neurologic function after the second surgery, with the mJOA score reaching 13 points after 6 weeks.

3. Discussion

Intraoperative VAI occurs in cases of incorrect procedures entering the area occupied by a normal VA course or of correct procedures applied to a VA following an aberrant course.^[3–5] Patients are at the greatest risk when undergoing posterior instrumentation in upper cervical spine surgery.^[2] During cervical procedures adjacent to the VA course, sudden, rapid,



Figure 2. Cervical MRI and angiography on postoperative day 40. (A) MRI examination demonstrated a large epidural hematoma and extensive spinal cord compression. (B) The bleeding point was located at the right V3 segment in the VA groove of the atlas.

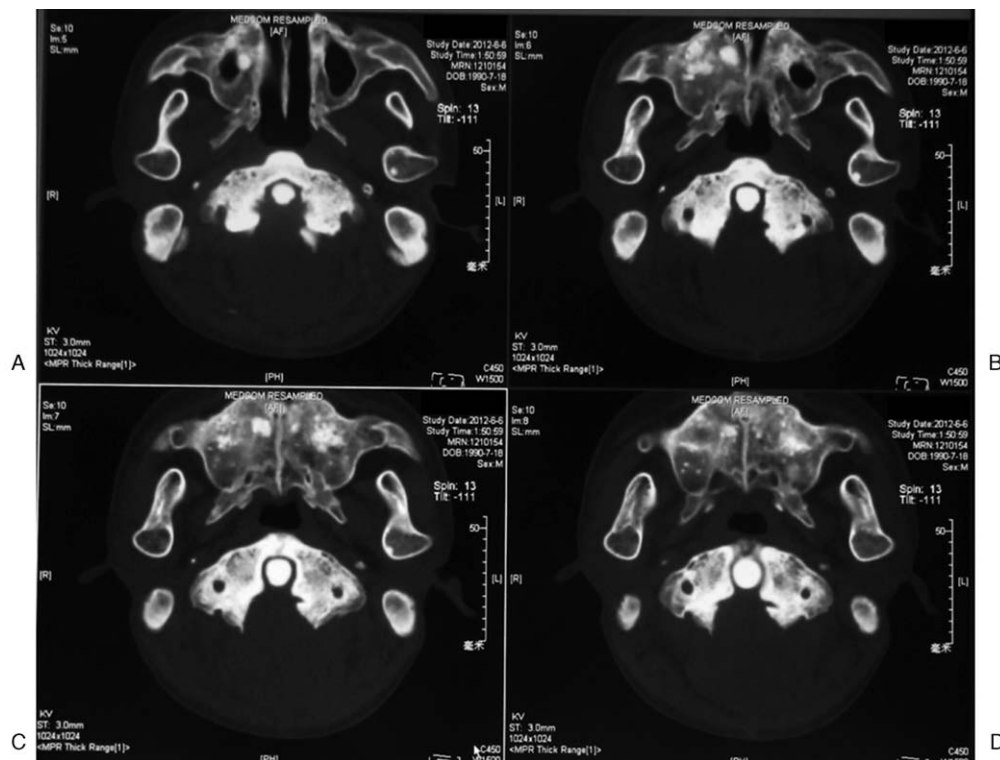


Figure 3. Continuous CT scans at the level of the atlas posterior arch. (A) The medial bone of the right VA groove was ground, whereas the left was preserved. (B–D) The bone groove ground during resection of the posterior ring was V-shaped, and the shallow grinding width was much larger than that deeper in the bone. VA = vertebral artery.

and sometimes uncontrollable massive bleeding is the typical presentation of VAI. This typical presentation is impressive enough to establish a diagnosis of iatrogenic VAI, which is usually associated with a large amount of blood loss. A survey of large series demonstrated that blood loss ranges from 800 mL to >8500 mL (>3850 mL on average) when VAI occurs in an open field.^[1]

In the present case, such noticeable bleeding did not occur. VAI probably occurred during the drilling of the posterior ring of the atlas according to photographs taken during surgery. Hemostasis was achieved by compression with only a few pieces of gelatin sponge, so the surgical team neglected the likelihood of a tiny rupture of the VA. To find the occult VAI in a timely manner, operators should be sensitive and pay sufficient attention to suspected bleeding at particular VAI-prone sites. Further examination might help to determine the potential risk.

To our knowledge, this is the first case of an occult VAI presenting during an open cervical procedure in a patient with skeletal fluorosis. The morphological abnormality of bone may have contributed to the occurrence of iatrogenic VAI. In skeletal fluorosis with hypertrophy of the posterior ring and lamina, the operator tends to assume the lateral boundary of the spinal canal is wide, and thus, the grinding width is often larger than necessary. In the present case, as confirmed by CT, the groove was V-shaped (Fig. 3), and the shallow grinding width was much larger than that deeper in the bone to obtain a better vision and operation field during the operation. For C2 to C7, the widened groove in the shallow bone would not lead to problems as long as enough bone was reserved for instrumentation and fusion. However, when this groove was extended to C1, the expanded grinding procedure was more likely to injure the VA. Careful preoperative evaluation of the

VA and identification of safe operation boundaries seem to be the key to preventing iatrogenic VAI, particularly in cases of morphological abnormality of bone or anomalous VA course.

4. Conclusion

VAI may occur during surgery without typical massive bleeding and lead to life-threatening late-onset pseudoaneurysm several weeks postoperatively. Surgeons should be sensitive and pay sufficient attention to suspected atypical bleeding at VAI-prone sites. The possibility of VAI should be considered when incision hematoma occurs after cervical surgery. In skeletal fluorosis, significant bone hypertrophy increases the risk of VAI in the upper cervical region. Compliance with the predetermined boundaries during the surgery will help to avoid VAI related to morphological abnormalities of the skeletal structure.

References

- [1] Neo M, Fujibayashi S, Miyata M, et al. Vertebral artery injury during cervical spine surgery: a survey of more than 5600 operations. *Spine (Phila Pa 1976)* 2008;33:779–85.
- [2] Maughan PH, Ducruet AF, Elhadi AM, et al. Multimodality management of vertebral artery injury sustained during cervical or craniocervical surgery. *Neurosurgery* 2013;73: ons271–81; discussion ons281–2.
- [3] Curylo LJ, Mason HC, Bohlman HH, et al. Tortuous course of the vertebral artery and anterior cervical decompression: a cadaveric and clinical case study. *Spine (Phila Pa 1976)* 2000;25:2860–4.
- [4] Gantwerker BR, Baaj AA, Maughan PH, et al. Vertebral artery injury during cervical discectomy and fusion in a patient with bilateral anomalous arteries in the disc space: case report. *Neurosurgery* 2010;67:E874–5. discussion E875.
- [5] Paramore CG, Dickman CA, Sonntag VK. The anatomical suitability of the C1-2 complex for transarticular screw fixation. *J Neurosurg* 1996;85:221–4.