

CASE REPORT

Multiple Fractures of Cervical Vertebrae Combined with Arcuate Foramen and Vertebral Artery Occlusion: A Case Report and Literature Review

Wei-hao Wang, BM[†], Zhao-yong Liu, MS[†], Huan-cheng Guo, BM[†], Hu Wang, PhD, MD 

Department of Orthopaedics, First Affiliated Hospital of Shantou University Medical College, Shantou, China

Background: The arcuate foramen is a complete or partial bony bridge over the vertebral artery groove of atlas. The mechanism of the arcuate foramen is not clearly understood. Omission of the arcuate foramen sometimes causes lethal iatrogenic injury during spinal surgery.

Case Presentation: We describe a patient who was diagnosed with multiple fractures of the cervical vertebrae, arcuate foramen, and right vertebral artery occlusion based on clinical and radiological exams. After conservative treatment, he resumed a normal and productive life.

Conclusions: Arcuate foramen is a common variation that causes symptoms such as dizziness, headache, and migraine. If the patient does not develop severe symptoms, conservative treatment can achieve very good results without the necessity to remove the bone bridge. When serious symptoms occur, surgical treatment to resect the bony ridges can relieve the symptoms dramatically.

Key words: Arcuate foramen; Multiple fracture; Spine; Vertebral artery occlusion

Introduction

The arcuate foramen (AF) is a complete or partial bony bridge over the vertebral artery groove of atlas. The mechanism of variation in the pathology is not clearly understood. The overall pooled prevalence of a complete AF was 9.1% and that of an incomplete AF was 13.6%¹. Some patients show symptoms, including dizziness, headache, migraine, neck pain, shoulder–arm pain, vertebrobasilar insufficiency, neurosensory-type hearing loss, and bow hunter's syndrome^{2–4}. When serious symptoms occur, surgical treatment to resect the bony ridges has been deemed an acceptable solution to the problem.

We describe a 52-year-old man who fell from a high place and then felt dizziness and neck pain. This patient was diagnosed with multiple fractures of the cervical vertebrae, arcuate foramen, and right vertebral artery occlusion based on clinical and radiological exams. There are many excellent

reviews in the literature reporting research on the arcuate foramen. Unfortunately, no studies have reported the incidence of a atlas posterior arch fracture combined with bilateral arcuate foramen and right vertebral artery occlusion. The aim of this article is to report on a patient with arcuate foramen, multiple fractures of the cervical vertebrae, and right vertebral artery occlusion. Besides, we made a comprehensive and detailed analysis of the literature of arcuate foramen.

Case Presentation

A 52-year-old man fell from a 3-metre-high ladder, his head hit the floor, and he then felt dizziness and neck pain, without slipping into a coma. The physical examination showed that manual muscle testing (MMT) of the arms and legs was grade V, and muscle tension was normal, without any sensory disorders or pathological reflexes. X-ray and three-

Address for correspondence Hu Wang, PhD, MD, Department of Orthopaedics, the First Affiliated Hospital, Shantou University Medical College, No.57 Changping Road, Shantou, Guangdong, China 515041 Tel: +86-13715965576; +86-0754-88905197; Email: wanghu0754@163.com

[†]These authors contributed equally to this study.

Grant Source: The study was funded by the Science and Technology Planning Project of Shantou City (no. st_20160306) by Hu Wang.

Received 10 June 2020; accepted 18 October 2020



dimensional (3D) computed tomography (CT) showed multiple complex fractures of atlantoaxial vertebrae comprised of left posterior arch fracture of the atlas, comminuted fracture of the epistropheus, left laminae fracture of the epistropheus, dens fracture, and bilateral arcuate foramen (Figs 1 and 2). 3D computed tomography angiography (CTA) revealed the running position of the vertebral artery, which passed under the bony bridges on the posterior arch of the atlas. The severe problem affecting our treatment plan was right vertebral artery occlusion (RVAO), and the isolated left vertebral artery was dominant (Fig. 3A). Magnetic resonance imaging (MRI) indicated no obvious compression or hydroptic degeneration of the cervical spinal cord (Fig. 3B). Fortunately, the patient did not present with significant symptoms

of cervical spinal cord compression or cerebral ischaemia, and the bony fragments had not moved into the canalis vertebralis. Surgery or interventional operation may cause vasospasm or injury of the left vertebral artery, which was the only blood supply to the brain. For fear of this fatal condition, our medical team chose the expectant treatment, including jaw-occipital belt traction with a 4-kg weight, anti-coagulation, and clinical observation of sensory-motor function and respiratory conditions. During the 6-week expectant treatment period, the patient did not experience any cervical spinal cord compression or cerebral ischaemia. The results of the physical examinations were normal, and CTA showed that the fracture line was blurred due to bone formation, but RVAO existed (Fig. 4). He was informed of the necessity for

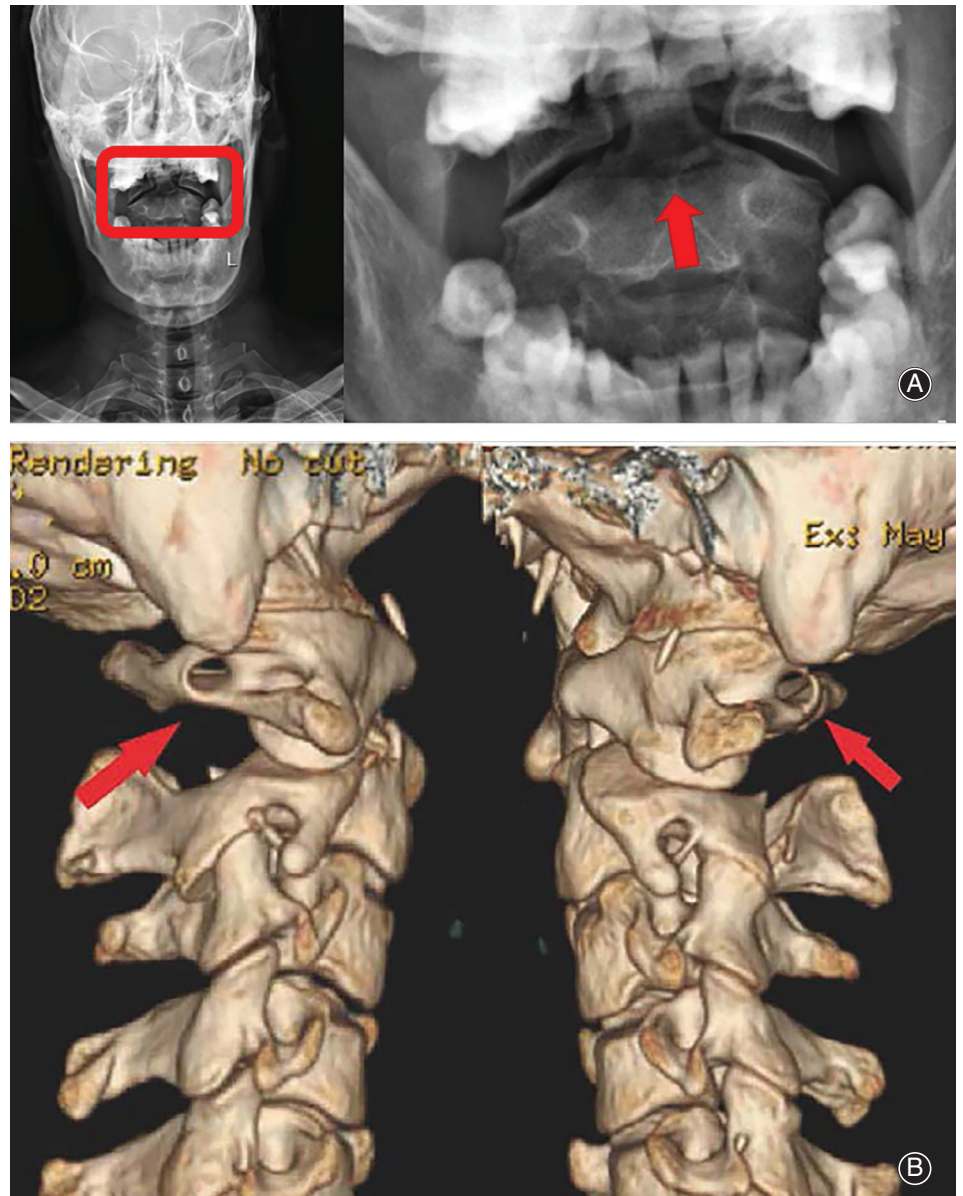


Fig. 1 (A) X-ray showed fractures of epistropheus (arrows). (B) 3D-CT also showed a dormant variation of bilateral AF (arrows).

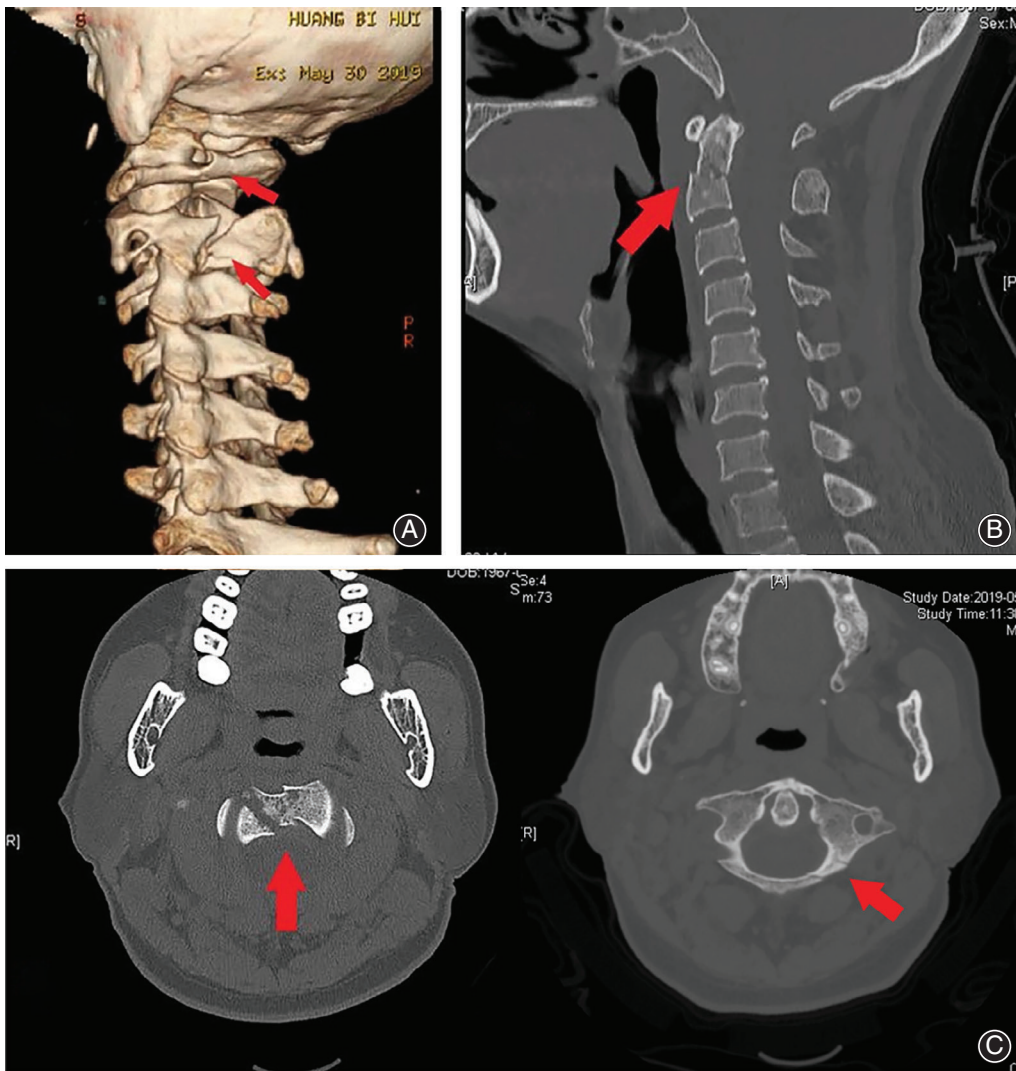


Fig. 2 CT and 3D-CT showed multiple complex fractures of atlas and epistropheus (arrows).

strict restriction of neck movement, received 6 weeks of cervicothoracic orthosis, and periodically returned to the hospital for medical examinations. Telephone follow-ups at 3 and 6 months showed that the patient resumed a normal and productive life with his family and in his workplace.

Discussion

Although the arcuate foramen has been intensively investigated, the mechanism of variation in the pathology is not clearly understood^{2, 5-7}. Previous studies have examined the arcuate foramen as a rare variation, but the prevalence has increased in recent years as clinicians and radiologists have placed a large focus on this anomaly. Due to the different reference populations, the prevalence of the arcuate foramen is highly variable (Table 1). A study from Japan analyzed 153 CTA images and showed a prevalence of 12.0%⁸. Four studies that focused on Korean individuals demonstrated a prevalence of 7.1%–26.0% in the CT scan group and 6.96%–14.0% in the X-ray group, and the arcuate

foramen was more prominent on the left side and in male patients. Moreover, Hong *et al.* inferred that age might be related to the formation of the bony bridge^{3, 9-11}. Two Indian studies of different populations showed a wide variation between 15.8% and 60.0%^{12, 13}. Three studies in Turkey on different populations also showed wide variations, which were 9.4%, 14.3%, and 43%. However, one of them was a cadaver study, and another focused on orthodontic patients¹⁴⁻¹⁶. Another study that investigated 180 3D-CT images showed incidences of 5.8%, 14.2%, and 5.0% in Chinese, Indian, and Malaysian populations, respectively¹⁷. A study of 221 X-ray images from Northern Italian orthodontic patients showed a prevalence of 9.0% for a partial AF and 7.7% for a complete AF¹⁸. Ahmed *et al.* analyzed 2917 patients from the north-eastern United States and found a prevalence of 22.5%, and the most common classification was a complete bony bridge on both sides¹⁹. A meta-analysis estimated that the overall pooled prevalence of a complete AF was 9.1% (95% CI, 8.2%–10.1%) and that of an

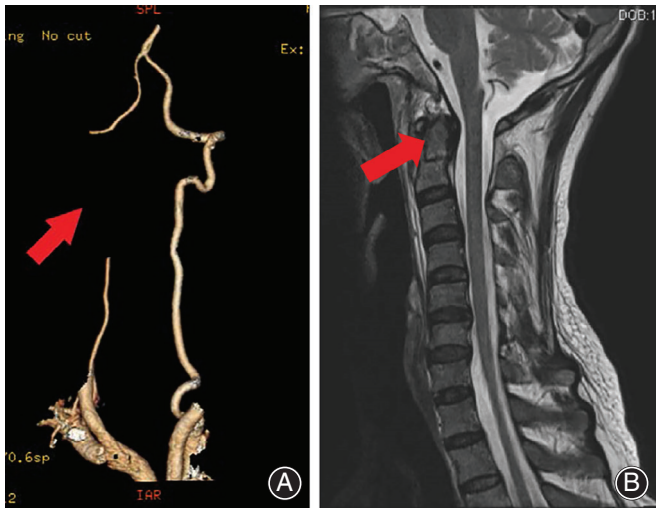


Fig. 3 (A) 3D-CTA showed right vertebral artery occlusion (arrows), and the isolated left vertebral artery was dominant. (B) MRI indicated no obvious compression or hydroptic degeneration of the cervical spinal cord (arrows).

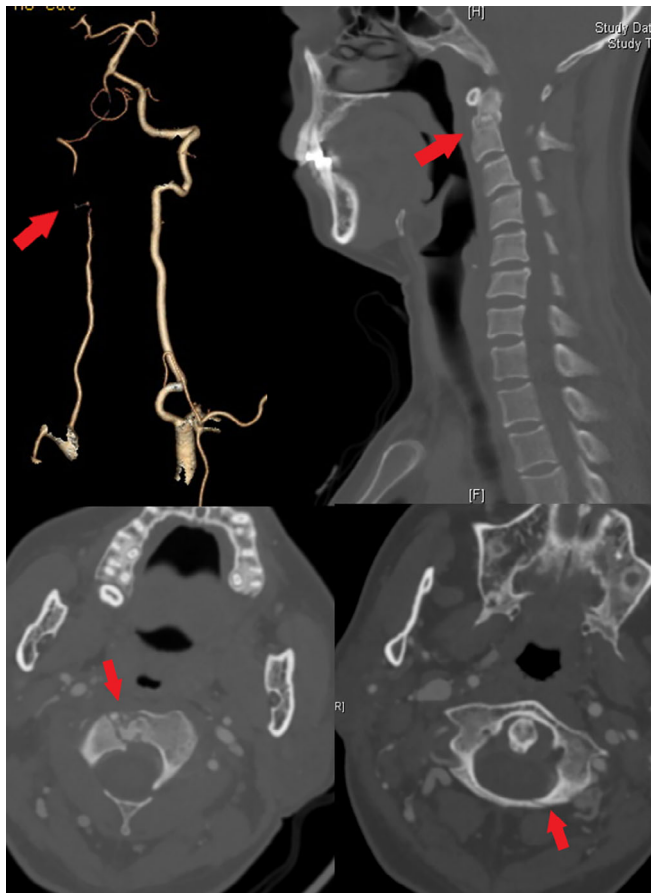


Fig. 4 CTA of 6 weeks after injury showed that the fracture line was blurred due to bone formation, but RVAO existed (arrows).

incomplete AF was 13.6% (95% CI, 11.2%–16.2%)¹. After reviewing these articles, we found a wide variation in the prevalence of the AF, and it is necessary to conduct further large-scale survey-based epidemiological studies.

The mechanism of the variation is not clearly understood. Based on the literature, ossification of the connective tissue surrounding the vertebral artery or the lower edge of the atlanto-occipital membrane might be one of the possible reasons for the variation^{2, 5}. Some researchers believe that the arcuate foramen seems to be produced by the persistence of a vestigial structure lost during hominoid evolution. The disappearance of the bony bridge on the posterior arch of the atlas was the result of natural selection⁷. Consanguinity might play an important role in the presence of the AF⁵.

At present, there are still few reports on the treatment of AF, and clinical features and treatment weight needs to be studied further. We had reviewed the reports on the clinical features and treatment methods of AF (Table 2). Some patients show symptoms including dizziness, headache, migraine, cervico-genic headache, neck pain, shoulder–arm pain, vertebrobasilar insufficiency, neurosensory-type hearing loss, bow hunter's syndrome, Barre–Lieou syndrome, eye pain, and photophobia^{2, 4, 12, 20, 21}. When serious symptoms occur, surgical treatment to resect the bony ridges has been deemed a solution to the problem^{2, 20, 21}. Victor *et al.* reported a patient complained of continuous dizziness and losing consciousness while rotating her head to the left. CTA revealed the formation of ponticulus posticus and ponticulus lateralis with acute-angled C-shaped kinking of the vertebral artery. After minimally invasive surgery to resect these bony ridges, the patient's symptoms resolved².

After reviewing the previous literature, we found that no study reported the presence of a left atlas posterior arch fracture combined with bilateral arcuate foramen and right vertebral artery occlusion. Juan *et al.* declared that the AF resulted in thickened cortical bone and the avoidance of fracture⁴. This article is the first to report this rare and complex clinical case, and the successful treatment experience may provide a reference for such cases that may occur in the future. We suspect that movement of the vertebral artery is restricted by the AF when cervical vertebrae are injured, which may cause fatal vertebral artery injury, and patients may die before reaching the hospital for further medical examinations.

Through this case and literature review, we summarize several suggestions. When patients have unknown symptoms, such as headache or the other symptoms mentioned above, doctors should not omit the existence of the AF. When patients are suspected to have an AF and when the X-ray is negative, 3D-CT should be performed actively. When AF is discovered, CTA is mandatory, as it can show the vertebral artery course and vertebral artery occlusion. Although patients had no symptoms, primary physicians must inform patients of the existence of the AF and the resulting symptoms so that patients can tell specialists when seeking medical treatment. When patients have atlas fracture

TABLE 1 The prevalence of the arcuate foramen is highly variable

Study	Year	Country	Object	Inspection techniques	Prevalence (%)
Kim <i>et al.</i> ¹⁰	2007	Korea	312 patients(X-ray), 2225 patients(3D-CT)	X-ray, 3D-CT	14.0(X-ray), 26.0(3D-CT)
Simsek <i>et al.</i> ¹⁶	2007	Turkey	158 cadavers	Anatomical Observations	9.5
Hong <i>et al.</i> ⁹	2008	Korea	1013 patients	CTA	15.6
Cho ³	2009	Korea	155 patients(X-ray), 200 patients(CT)	X-ray, CT	7.0(X-ray), 15.5(CT)
Chitroda <i>et al.</i> ¹²	2013	India	500 patients	X-ray	60.0
Gibelli <i>et al.</i> ¹⁸	2015	Italy	221 patients	X-ray	16.7
Buyuk <i>et al.</i> ¹⁵	2017	Turkey	374 patients	3D-CT	43.0
Lee <i>et al.</i> ¹⁷	2017	China, India, Malaysia	60 patients(China), 60 patients(India), 60 patients(Malaysia)	3D-CT	5.8(China), 14.2(India), 5.0 (Malaysia)
Song <i>et al.</i> ¹¹	2017	Korea	2628 patients	3D-CTA	7.7
Tambawala <i>et al.</i> ¹³	2017	India	500 patients	X-ray	15.9
Arslan <i>et al.</i> ¹⁴	2018	Turkey	200 patients	3D-CTA	14.3
Isaji <i>et al.</i> ⁸	2018	Japan	153 patients	CT, CTA	12.0
Saleh <i>et al.</i> ¹⁹	2018	USA	2917 patients	CT	22.5

CT, computed tomography; 3D-CT, three-dimensional CT; CTA, computed tomography angiography; 3D-CTA, three-dimensional CTA.

TABLE 2 The review of the clinical features and treatment of arcuate foramen

Study	Year	Patients	Average age (years)	Symptom	Treatment	Prognosis
Tubbs <i>et al.</i> ²¹	2007	NA	NA	Headache, phonation, Hypothesized	Dissection of bony bridge and periarterial sympathectomy	Symptomatic relief
Koutsouraki <i>et al.</i> ²⁰	2010	1	18	Chronic headache Unilateral (left) hearing loss	NA	NA
Chitroda <i>et al.</i> ¹²	2013	432	27.76 ± 10.74	Migraine or tension type headache	NA	NA
Lukianchikov <i>et al.</i> ²	2018	1	34	Neck pain Bow hunters syndrome	Surgical treatment	Symptomatic relief

NA, non mentioned.

and the appearance of neurological symptoms, physicians should distinguish between the occurrence of spinal cord compression and vertebral artery occlusion.

Disclosure

The author declares that he has no conflict of interest.

References

- Pekala PA, Henry BM, Pekala JR, *et al.* Prevalence of foramen arcuale and its clinical significance: a meta-analysis of 55,985 subjects. *J Neurosurg Spine*, 2017, 27: 276–290.
- Lukianchikov V, Lvov I, Grin A, Kordonskiy A, Polunina N, Krylov V. Minimally invasive surgical treatment for vertebral artery compression in a patient with one-sided ponticulus posticus and ponticulus lateralis. *World Neurosurg*, 2018, 117: 97–102.
- Cho YJ. Radiological analysis of ponticulus posticus in Koreans. *Yonsei Med J*, 2009, 50: 45–49.
- Sanchis-Gimeno JA, Llido S, Guede D, *et al.* Atlases with arcuate foramen present cortical bone thickening that may contribute to lower fracture risk. *World Neurosurg*, 2018, 117: e162–e166.
- Sanchis-Gimeno JA, Llido S, Miquel-Feutch M, *et al.* The decreasing prevalence of the arcuate foramen. *World Neurosurg*, 2018, 110: 521–525.
- Tubbs RS, Johnson PC, Shoja MM, Loukas M, Oakes WJ. Foramen arcuale: anatomical study and review of the literature. *J Neurosurg Spine*, 2007, 6: 31–34.
- Travan L, Saccheri P, Gregoraci G, Mardegan C, Crivellato E. Normal anatomy and anatomic variants of vascular foramina in the cervical vertebrae: a paleo-osteological study and review of the literature. *Anat Sci Int*, 2015, 90: 308–323.
- Isaji T, Yasuda M, Kawaguchi R, *et al.* Posterior inferior cerebellar artery with an extradural origin from the V₃ segment: higher incidence on the nondominant vertebral artery. *J Neurosurg Spine*, 2018, 28: 154–159.
- Hong JT, Lee SW, Son BC, *et al.* Analysis of anatomical variations of bone and vascular structures around the posterior atlantal arch using three-dimensional computed tomography angiography. *J Neurosurg Spine*, 2008, 8: 230–236.
- Kim KH, Park KW, Manh TH, Yeom JS, Chang BS, Lee CK. Prevalence and morphologic features of ponticulus posticus in Koreans: analysis of 312 radiographs and 225 three-dimensional CT scans. *Asian Spine J*, 2007, 1: 27–31.
- Song MS, Lee HJ, Kim JT, Kim JH, Hong JT. Ponticulus posticus: morphometric analysis and its anatomical implications for occipito-cervical fusion. *Clin Neurol Neurosurg*, 2017, 157: 76–81.

- 12.** Chitroda PK, Katti G, Baba IA, *et al.* Ponticulus posticus on the posterior arch of atlas, prevalence analysis in symptomatic and asymptomatic patients of Gulbarga population. *J Clin Diagn Res*, 2013, 7: 3044–3047.
- 13.** Tambawala SS, Karjodkar FR, Sansare K, *et al.* Prevalence of ponticulus posticus on lateral cephalometric radiographs, its association with cervicogenic headache and a review of literature. *World Neurosurg*, 2017, 103: 566–575.
- 14.** Arslan D, Ozer MA, Govsa F, Kitis O. The ponticulus posticus as risk factor for screw insertion into the first cervical lateral mass. *World Neurosurg*, 2018, 113: e579–e585.
- 15.** Buyuk SK, Sekerci AE, Benkli YA, Ekizer A. A survey of ponticulus posticus: radiological analysis of atlas in an orthodontic population based on cone-beam computed tomography. *Niger J Clin Pract*, 2017, 20: 106–110.
- 16.** Simsek S, Yigitkanli K, Comert A, *et al.* Posterior osseous bridging of C1. *J Clin Neurosci*, 2008, 15: 686–688.
- 17.** Lee CK, Tan TS, Chan C, Kwan MK. Is C1 lateral mass screw placement safe for the Chinese, Indians, and Malays? An analysis of 180 computed tomography scans. *J Orthop Surg (Hong Kong)*, 2017, 25: 2309499017692683–2230949901769268.
- 18.** Gibelli D, Cappella A, Cerutti E, Spagnoli L, Dolci C, Sforza C. Prevalence of ponticulus posticus in a northern Italian orthodontic population: a lateral cephalometric study. *Surg Radiol Anat*, 2016, 38: 309–312.
- 19.** Saleh A, Gruber J, Bakhsh W, Rubery PT, Mesfin A. How common is the ponticulus posticus?: a computed tomography based analysis of 2917 patients. *Spine (Phila pa 1976)*, 2018, 43: E436–E441.
- 20.** Koutsouraki E, Avdelidi E, Michmizos D, Kapsali SE, Costa V, Baloyannis S. Kimmerle's anomaly as a possible causative factor of chronic tension-type headaches and neurosensory hearing loss: case report and literature review. *Int J Neurosci*, 2010, 120: 236–239.
- 21.** Tubbs RS, Shoja MM, Shokouhi G, Farahani RM, Loukas M, Oakes WJ. Simultaneous lateral and posterior ponticles resulting in the formation of a vertebral artery tunnel of the atlas: case report and review of the literature. *Folia Neuropathol*, 2007, 45: 43–46.