

Minimally invasive surgical treatment for Kimmerle anomaly

ABSTRACT

Introduction: Kimmerle anomaly is the bony ridge between the lateral mass of atlas and its posterior arch or transverse process. This bony tunnel may include the V3 segment of the vertebral artery, vertebral vein, posterior branch of the C1 spinal nerve, and the sympathetic nerves, which results in the clinical symptoms of this disease. Reports on the surgical treatment of Kimmerle anomaly are rare. There are no reports on minimally invasive surgical treatment of this pathology.

Materials and Methods: Six patients with Kimmerle anomaly were treated from 2015 until 2016. Three patients underwent routine surgery through the posterior midline (posterior midline approach [PMA] group). The other three patients underwent decompression with a paravertebral transmuscular approach (PTMA group). The operation time, intraoperative blood loss, clinical symptoms before and after surgery as well as intra- and post-operative complications were compared between the PTMA and PMA groups.

Results: The results of the surgical treatments were assessed at discharge and 1 year later. Blood loss, operation time, and intensity of pain at the postoperative wound area were lower in the PTMA group. There were no postoperative complications. The delayed surgical treatment outcomes did not depend on the method of artery decompression.

Conclusions: Surgical treatment of vertebral artery compression in patients with Kimmerle anomaly is preferable in cases where conservative treatment is inefficient. A minimally invasive PTMA is an alternative to the routine midline posterior approach, providing direct visualization of the compressed V3 segment of the vertebral artery and minimizing postoperative pain.

Keywords: Foramen arcuate, foramen retroarticulare, kimmerle anomaly, paravertebral transmuscular approach, ponticuli posticus

INTRODUCTION

Kimmerle anomaly (foramen arcuate, foramen retroarticulare, ponticuli posticus, canalis Bildungi) is the bony ridge between the superoposterior lateral mass of atlas and its posterior arch or transverse process, which forms because of complete or incomplete ossification of the posterior atlantooccipital membrane. Thus, the V3 segment of the vertebral artery is squeezed in the osseous canal instead of being free in sulcus arteriae vertebralis. This bony canal may also include the vertebral vein, posterior C1 spinal nerve, and posterior branch of the C1 spinal nerve and sympathetic nerves, leading to the clinical signs of this disease.^[1-3]

Kimmerle anomaly occurs frequently according to data from a meta-analysis by Elliott *et al.*^[4] Postmortem examinations

revealed a frequency of 18.8%, computed tomography (CT) scanning showed a frequency of 17.2%, and radiography examination found revealed a frequency of 16.6%. The majority patients with Kimmerle anomaly have no symptoms; the clinical signs of vertebrobasilar insufficiency develop in only 5.5% of patients in the case of cicatricial band strangulation forming in the region of this anomalous osseous ring.^[5]

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There are few reports dedicated to the treatment of compression syndrome in patients with Kimmerle anomaly. Most of the published articles discuss the methods of conservative treatment;^[6,7] meanwhile authors note frequent cases of relapse (50%–60% of patients), requiring repeated treatment courses.

Only a few papers describe the surgical treatment of Kimmerle anomaly: The midline posterior approach for decompression of the V3 segment of the vertebral artery. The regression of the clinical signs of vertebrobasilar conflict was observed in 90% of operated patients.^[5,8-10] There are no articles on minimally invasive surgical treatment of this pathology.

The aim of this study was to optimize a surgical treatment for vertebral artery compression in patients with Kimmerle anomaly.

MATERIALS AND METHODS

We analyzed the surgical treatment outcomes in six patients with Kimmerle anomaly from 2015 to 2016. Three patients underwent decompression of the vertebral artery using the minimally invasive posterior transmuscular approach (PTMA group), and the other three patients underwent the routine posterior midline approach (PMA group).

The severity of the disease was assessed using 3 grades according to the intensity of the clinical symptoms,^[6] as well as considering the modified Rankin scale (mRs) and Rivermead mobility index (Rmi):

- Grade III: Severe; presenting with most clinical symptoms; patients are on bed rest during most of the day; acute attack of clinical disease manifestation while rotating the head; mRs score ≥ 3 ; Rmi ≤ 7
- Grade II: Moderate severity; present with a headache and vertebrobasilar insufficiency several times per year, which increases after head rotation during disease onset; mRs score = 1; Rmi = 8–13
- Grade I: Mild; the appearance of complaints during light physical activity; mRs score = 1; Rmi = 14–15.

The preoperative examination included CT of the craniovertebral region, CT-or magnetic resonance-angiography of the vertebral arteries, and ultrasound examination of the V3 segments of the vertebral arteries with functional tests.

The surgical treatment outcomes were estimated using the modified Glasgow Outcome Scale (GOS), mRs, Rmi, and visual analog scale (VAS). The quality of the decompression of the vertebral arteries was assessed using CT-angiography. The follow-up period was 1 year after the operation.

The operation time, intraoperative blood loss as well as the clinical symptoms before and after surgery were compared between the PTMA and PMA groups.

Surgical technique

The patient was placed in a prone position with the head fixed in a Mayfield frame or Halo vest during surgery. Skin incisions were performed using radiography control in lateral and transoral projections. The soft tissues (skin, subcutaneous fat, and aponeurosis) were cut using a scalpel. The following bundles of trapezius, semispinalis capitis, and rectus capitis posterior major muscles were diverged without cutting using a blunt peanut dissection. All patients had a thin layer of adipose tissue located just below the rectus capitis posterior major muscle, covering the anomalous bony ridge. The tubular wound retractor was placed using the systems of dilators after visualization of ponticulus posticus using an operational microscope ($\times 4-6$) [Figure 1].

Hereafter, the bony ridge was accurately skeletonized from all sides as its incomplete dissection may lead to severe bleeding from the venous collectors, which are located around the vertebral artery. A high-speed drill was used for resection of the external cortical and spongy layers of the ponticulus posticus from the sulcus arteriae vertebralis of the C1 vertebra to the cranial edge of the bony ridge. The internal cortical edge was resected using Kerrison rongeurs, followed by removal of the remaining bony ridge by drilling all the way to the lateral mass of atlas. The main steps of operation are presented in Figure 2.

A band of ossified fibrous tissue was located between the edges of the incomplete bony ridge. Once revealing the type of Kimmerle anomaly, the bony fragments were removed first using a high-speed drill followed by the accurate dissection



Figure 1: Intraoperative image. The tubular retractor is placed through a transmuscular approach directly to the bony ridge

and removal of the fibrous band using Kerrison rongeurs and blunt-pointed microscissors.

RESULTS

The PMA group included 3 female patients aged 30–58 years. The PTMA group included 2 males and 1 female aged 20–58 years.

The clinical presentation of disease was the following in the PTMA group: Grade III in 1, II in 1, I in 1 patient. The mean operation time was 178.3 min with an average blood loss of 300 mL. There were no intra- and post-operative complications. The level of pain at the postoperative wound in the first 3 days after surgery was minimal or absent (VAS = 1). In the 1 year follow-up, examination of the disease severity revealed that a reduction to grade I was achieved in patients previously having grades III and II (modified GOS = 8, mRs = 1, Rmi = 15). The clinical signs of vertebral artery compression were completely regressed after the operation in 1 patient with grade I disease severity (modified GOS = 8, mRs = 0, Rmi = 15).

The clinical presentation of disease was the following in the PMA group: Grade III in 1 patient and II in 2 patients. The mean operation time was 230 min with an average blood loss of 600 mL. There were no intraoperative complications. Marginal necrosis of the upper angle of the operative wound occurred in 1 patient during the postoperative period. The level of pain at the postoperative wound in the first 3 days after surgery was moderately severe (VAS = 5 in two patients and 6 in one patient). Favorable disease course was confirmed 1 year after the operation, revealing a reduction in disease

severity to Grade II in 1 patient (previously Grade III; modified GOS = 7, mRs = 2, Rmi = 13), and grade to I Grade in 2 patients (previously grade II; modified GOS = 8, mRs = 1, Rmi = 15).

Clinical case

We present the case of a 20-year-old male patient, who was an amateur sportsman who had fallen ill 6 months before admission. The patient suffered from a headache and vertebrobasilar insufficiency which arose during light physical activity (Grade I, mRs = 1, Rmi = 15). The frequency and intensity of attacks had constantly progressed despite conservative treatment. A CT of the cervical spine revealed Kimmerle anomaly as an incomplete bony ridge from both sides and compression of the left vertebral artery [Figure 3a]. The ultrasound examination of the V3 segments of the vertebral arteries and functional tests revealed a decrease in the linear velocity of blood flow during contralateral side head rotation.

The patient underwent minimally invasive decompression of the V3 segments of both vertebral arteries [Figure 2]. The band of ossified fibrous tissue compressing the vertebral artery was revealed intraoperatively from both sides. There were no complications during the postoperative period and no development of pain. The control postoperative examination data demonstrate the release of vertebral artery and absence of bone ridge [Figure 3b]. The patient was discharged 7 days after the operation and a control examination 1 year later revealed complete regression of all clinical symptoms (modified GOS = 8, mRs = 0, Rmi = 15).

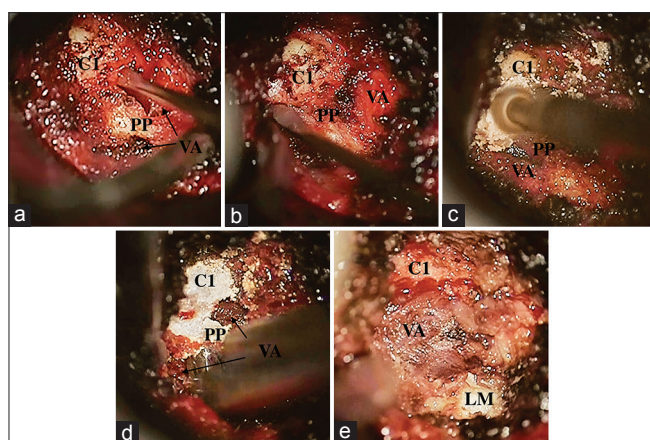


Figure 2: Intraoperative images of the main steps of vertebral artery decompression. (a and b) The bony ridge is accurately skeletonized from both sides; (c) removal of bone tissue to the internal cortical layer using a high-speed drill; (d) resection of the internal cortical layer using Kerrison rongeurs; (e) decompression of vertebral artery is performed. PP, ponticulus posticus; C1, posterior arch of atlas; VA, vertebral artery; LM, superoposterior part of the lateral mass of atlas

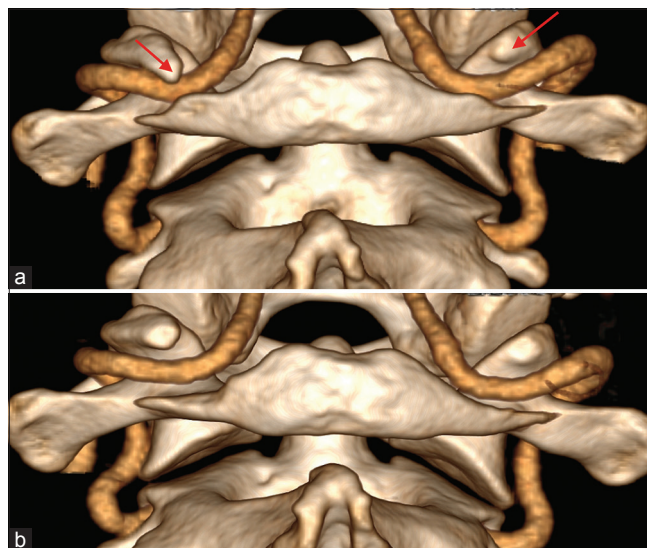


Figure 3: Computed tomography of the craniocervical region of a patient with Kimmerle anomaly before (a) and after (b) surgery. Red arrows indicate the incomplete ponticuli posticus

DISCUSSION

The main pathogenic factors for the development of clinical symptoms in patients with Kimmerle anomaly are as follows: extravasal compression of the vertebral artery, the long-term trauma of the vessel adventitia as well as irritation of the paravascular sympathetic nerves and branches of the occipital nerve.^[7,9] The clinical symptoms of Kimmerle anomaly are variable: headache, dizziness, tinnitus, transient hearing and visual disorders, tiredness, sleep disturbances, unsteady gait, drop attacks, panic attacks, anxiety or asthma, numbness of hands, and seizures.^[7,9,11-14] Such patients often receive ineffective symptomatic therapy for a long time in case timely visualization of the craniovertebral region (using CT or MRI) is not performed. All patients in our study were under the observation of a neurologist. Three patients received conservative treatment for more than 2 years, 2 patients for 1.5 years, and 1 patient for around 6 months before admission to the neurosurgery department. Nonsurgical treatment and physiotherapy were ineffective in all cases; moreover, five patients had an increase in vertebrobasilar symptoms.

The indications for surgical treatment in our study were as follows: Ineffective conservative treatment and changes in the haemodynamics in the vertebral arteries during head rotation in accordance with the results of ultrasound examination with functional tests.

There are very few published papers concerning surgical treatment focusing on the removal of ponticuli posticus.^[5,8-10] The authors used a routine midline approach with an incision from the occipital protuberance to the spinous process of the C3 vertebra with resection of the posterior arch of atlas or the posterior bony ridge of atlas from both sides. The highly efficient rate of symptom regression with surgical treatment is 90%.

We used the above method of surgical treatment in three patients (PMA group) in our study and achieved a favorable outcome with a decrease in the intensity and frequency of disease attacks. However, all patients suffered pain at the region of the postoperative wound of up to a score of 6 (VAS) due to operative trauma. This symptom of pain remained for 3 weeks postsurgery. The length of skin incision was at least 12 cm; moreover, it was necessary to perform a wide opening of the occipital bone (at least 3 cm from the midline) and the arch of the C2 vertebra, with complete dissection of the facet joint for optimal visualization of the anomalous bony ridges. The other disadvantage of the midline posterior approach is that it is not possible to carry out a direct approach to the bony ridge. Visualization of the ponticuli

posticus, despite the type of wound retractor used was always performed at an angle to the sagittal plane, which did not favor full examination of the lateral edge of the bony ridge. Severe blood loss from damaged venous basins around the vertebral artery occurred in 2 cases because of inappropriate visualization of the ponticuli posticus.

We used a paravertebral transmuscular approach with a dilatator system and a tubular wound retractor in the other three patients (PTMA group). The advantages of such an approach are minimal trauma of the cervical muscles, a small area of skeletonisation, and a small skin incision. The low pain score in the area of the operative wound (average VAS = 1) was observed in all patients after minimally invasive decompression of the vertebral artery. The other advantages of the transmuscular approach are a direct visualization of the ponticuli posticus with the possibility of full observation of its lateral and medial edges, which allowed a more accurate dissection and prevented damage to the paravascular venous basins.

In summary, we operated on six patients with vertebral artery compression because of Kimmerle anomaly. Blood loss, operation time, and pain intensity were lower in the PTMA group. The delayed surgical treatment outcomes did not depend on the surgical method of artery decompression. All patients had favorable disease course in terms of neurological deficit regression and a decrease in the frequency of disease attacks.

CONCLUSIONS

The surgical treatment of vertebral artery compression in patients with Kimmerle anomaly is preferable in cases where conservative treatment is inefficient. The minimally invasive PTMA is an alternative to the routine midline posterior approach, which allows direct visualization of the compressed V3 segment of the vertebral artery, decreased blood loss and operation time, and minimizes postoperative pain.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

REFERENCES

1. Senoglu M, Gümüřalan Y, Yüksel KZ, Uzel M, Celik M, Ozbag D, *et al.* The effect of posterior bridging of C-1 on craniovertebral junction surgery. *J Neurosurg Spine* 2006;5:50-2.
2. Kim MS. Anatomical variant of atlas: Arcuate foramen, occipitalization of atlas, and defect of posterior arch of atlas. *J Korean Neurosurg Soc* 2015;58:528-33.
3. Afsharpour S, Hoiriis KT, Fox RB, Demons S. An anatomical study of arcuate foramen and its clinical implications: A case report. *Chiropr Man Therap* 2016;24:4.
4. Elliott RE, Tanweer O. The prevalence of the ponticulus posticus (arcuate foramen) and its importance in the goel-harms procedure: Meta-analysis and review of the literature. *World Neurosurg* 2014;82:e335-43.
5. Lutsik AA, Peganov AI, Kazantsev VV, Ratkin IK. Vertebrobasilar insufficiency due to osseous abnormalities of the craniovertebral junction. *Hir Pozvonoc* 2016;13:49-55.
6. Gulyaev SA, Kulagin VN, Arkhipenko IV, Gulaeva SE. Clinical manifestations and treatment for Kimmerle anomaly. *RMJ* 2013;16:866-8.
7. Buna M, Coghlan W, deGruchy M, Williams D, Zmiywsky O. Ponticles of the atlas: A review and clinical perspective. *J Manipulative Physiol Ther* 1984;7:261-6.
8. Tedeschi G. Surgical trial in the vertebral flow alteration due to Kimmerle anomaly. *J Neurosurg Sci* 1979;23:235-8.
9. Chertkov AK, Klimov ME, Nesterova MV. On surgical treatment of patients with vertebrobasilar insufficiency associated with Kimmerle anomaly. *Hir Pozvonoc* 2005;1:69-73.
10. Limousin CA. Foramen arcuale and syndrome of barre-lieou. Its surgical treatment. *Int Orthop* 1980;4:19-23.
11. Ríos L, Mata-Escolano F, Blanco-Pérez E, Llidó S, Bastir M, Sanchis-Gimeno JA, *et al.* Acute headache attributed to whiplash in arcuate foramen and non-arcuate foramen subjects. *Eur Spine J* 2017;26:1262-5.
12. Taylor WB 3rd, Vandergriff CL, Opatowsky MJ, Layton KF. Bowhunters syndrome diagnosed with provocative digital subtraction cerebral angiography. *Proc (Bayl Univ Med Cent)* 2012;25:26-7.
13. Split W, Sawrasewicz-Rybak M. Clinical symptoms and signs in Kimmerle anomaly. *Wiad Lek* 2002;55:416-22.
14. Mudit G, Srinivas K, Satheesha R. Retrospective analysis of ponticulus posticus in Indian orthodontic patients-a lateral cephalometric study. *Ethiop J Health Sci* 2014;24:285-90.