



# Impending Rupture Traumatic Large Pseudoaneurysm of (V2) of Vertebral Artery: A Successful Hybrid Approach and Outcome. A Vascular Surgeon View

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## Abstract

**Objectives:** Vertebral artery aneurysms are rare clinical findings, representing 1% of supra-aortic aneurysms, and the most common cause of Extracranial Vertebral Artery Aneurysms (EVAA) is penetrating neck trauma, but they can also occur secondary to dissection, atherosclerosis, infection, collagen vascular diseases, and inherited connective tissue disorders. Extracranial VA aneurysms are very uncommon accounting for 0.5% of all arterial aneurysms; they generally affect the most mobile segment, the V3 segment, followed by the V1 segment. The rupture of an EVAA can lead to catastrophic bleeding and pose a diagnostic and therapeutic challenge. VA injuries constitute less than 1% of all the vascular injuries and less than 1% to 6% of all the vascular injuries in the cervical region penetrating vertebral artery injuries leading to serious external bleeding are rare, and their injuries were previously missed prior to the routine use of angiography in diagnosing penetrating neck injuries. This frank hemorrhage was the impetus for the decision for operative exploration. Here, in our article we report the largest pseudoaneurysm reported in the literature. It was impending to rupture and arising from the second part of the right vertebral artery (V2) in a 23-years-old male. Duplex study and Urgent CT angiography confirmed the presence of an extracranial pseudoaneurysm involving the 2<sup>nd</sup> part of right VA, measuring 10 cm × 7.5 cm × 4.8 cm.

**Aim:** The aim of this article is focusing and highlighting on a very rare vascular injury that any vascular or general surgeon could encounter throughout his daily work and highlighting the treatment options in emergency situations, putting into consideration the availability of different tools.

**Methods:** We went through a hybrid approach where endovascular proximal parent vertebral artery was controlled with a 3 mm balloon diameter 30 mm length, through a rapid right trans brachial access followed by surgical exposure and ligation of V1 part that ameliorated the risk of rupture for a great extent, but postoperative duplex 14 days later showed retrograde filling of the aneurysmal sac that necessitated distal V3 control.

**Results:** The first and second postoperative periods were free of any cerebrovascular or vertebrobasilar ischemic or neurological signs.

**Conclusion:** The consequences of VA injury are fetal and even result in death because of the difficulty in controlling the pulsating hemorrhage which cause severe hypotension resulting in cardiac arrest and finally death. Therefore, timely diagnosing and intervention is crucial in determining the interventional outcome. Whether the treatment of VA injury is surgery or endovascular approach depends on the location and radius of the lesion. VA is divided into four segments along its course and the decision of intervention, the postoperative outcome is determined according to the VA segment affected. Finally If the patient has a patent contralateral VA or well-developed collateral circulation from posterior circulation. If the contralateral VA is underdeveloped or slender, vessel repair or endovascular restoration of flow is a must.

**Keywords:** Aneurysm; Cerebral revascularization; Pseudo aneurysm; Vertebral artery

## Background

Vertebral Artery (VA) aneurysms are rare clinical finding accounting for 0.5% of all arterial aneurysms, and the most common cause of Extracranial Vertebral Artery Aneurysms (EVAA) is penetrating neck trauma, they can also occur secondary to dissection, atherosclerosis, infection,

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collagen vascular diseases, inherited connective tissue disorders and *NF1 (Neurofibromatosis Type 1)*, where only eight cases were reported in the revised literature [1,2].

They affect the most mobile segments, (V3), followed by (V1) segment. The rupture of an EVAA can lead to catastrophic bleeding and pose a diagnostic and therapeutic challenge. However, extracranial vertebral artery aneurysms are also recognized as a cause of ischemic strokes, especially in young patients less than 45-year-old and accounts for approximately 25 percent of such cases [3], and it also carries 13% risk of intracranial embolic events and death [4]. VA injuries constitute less than 1% of all the vascular injuries and less than 1% to 6% of all the vascular injuries in the cervical region [5]. Penetrating vertebral artery injuries leading to serious external bleeding are rare, and their injuries were previously missed prior to the routine use of angiography in diagnosing penetrating neck injuries and this frank hemorrhage is the impetus for the decision for operative exploration [6]. V2 of vertebral artery could be injured during anterior cervical spine surgery which is exceptionally rare, occurring in 0.5% of such procedures according to a series analysis [7]. Pseudoaneurysm develops when there is a tear between the media and adventitia layers of any vessel; hematoma and circumferential fibrotic tissue from the wall of the aneurysm. Despite the presence of adventitia layer in true aneurysms, there is no adventitia layer in PAs [8]. Thrombosis with subsequent resolution, arteriovenous fistulas, and intrathoracic hemorrhage following rupture of extracranial vertebral artery aneurysm are the major complications of VA aneurysms and its fate [9-11]. Also, anatomical variations in vertebral artery origin and course have been also reported in Klippel-Feil syndrome which is characterized by the fusion of one or more cervical vertebrae and anomalous vertebral artery course and origin where the artery can be injured during surgical screw placement [12]. VA injury and subsequent pseudoaneurysm formation has been reported during internal jugular vein catheterization, pathological and traumatic fracture dislocation or even during transarticular screw fixation, chiropractic manipulation, falls, sport activity, prior head injury without direct neck trauma also have been implicated in vertebral artery tortuosity or aneurysm formation and recently VA hypoplasia and aplasia are considered potential risk factors for vertebral-posterior inferior cerebella artery aneurysms [13-16]. In our article, we have managed the largest traumatic vertebral artery pseudoaneurysm reported in the literature, and the location in the foraminal part of vertebral artery (v2) and its presentation with rapid enlargement in size (impending rupture) added more challenge for urgent decision and management. The Aim of this article is focusing on a very rare vascular injury that a vascular surgeon could encounter throughout his daily work and highlighting the treatment options in emergency situation, putting into consideration the availability of different tools.

## Case Description

A23-years-old male, with No history of hypertension, non-diabetic, non-smoker was presented at our emergency department with 36 h history of a stab injury of his neck on the right side. On examination; a large pulsating swelling with bruit on auscultation, no palpable thrill (Figure 1), the mass reached 10 cm × 7.8 cm by Color Duplex that showed swirling flow and decreased linear flow velocity. CT angiography confirmed the presence of an extracranial pseudoaneurysm involving the 2<sup>nd</sup> part of right VA, measuring 10 cm × 7.5 cm × 4.8 cm. CT Angiography indicated an intact Circle of Willis



Figure 1: Large pulsating pseudoaneurysm on right posterior aspect of neck.



Figure 2: Giant pseudoaneurysm arising from v2 (intraforaminal) part of right vertebral artery with normal diameter left vertebral artery.

without any stenosis of cervical arteries. The left VA was of normal caliber (>3.5 mm) and both vertebral arteries were dominant in the vertebrobasilar system (Figure 2). The patient was resuscitated with full laboratory investigations, biochemistry tests that revealed marked hematocrit level reduction. Chest radiography, electrocardiography were all normal and neurological evaluation was free, there was no manifestations of cerebral or vertebrobasilar insufficiency. Six hours later the swelling was obviously reached its maximum in all dimensions and the patient suffered severe pain and headache with the overlying skin was severely ecchymotic, bluish in color with thinning of overlying skin. Urgent interference was decided. In this case, and in such situation, the following two questions should be answered: Will we go on for restorative surgery for the vertebral artery flow or for control of the parent vertebral artery (V1) either surgical ligation or endovascular coiling. Second question: open or totally endovascular intervention? So, restoration of flow excluding the aneurysm with endovascular intervention includes endovascular stent graft [17], Stent-supported Coil Embolization of the aneurysm sac [18]. The open surgery include excision with autogenous interposition graft, ligation of v1 (parent artery) and a bypass from the occipital artery to the atlas loop of the vertebral artery (v3) in the sub occipital triangle [19]. We excluded surgical excision and interposition graft due to



**Figure 3:** Temporary proximal v1 balloon control.



**Figure 4:** Supraclavicular exposure and ligation of first part of right vertebral artery (the blue vessel loop).

very difficult exposure of v2 (intraforaminal) in such situation. Most patients have a dominant VA, the left VA accounts for 50% to 60% of the population whereas the right VA accounts for in approximately only 25% and the remaining 25% of the population have equivalent vertebral arteries [20] as in our patient, and putting into consideration the risk of rupture that arise from the foraminal part (v2) which is the most difficult to approach by open exposure so we excluded surgical excision and interposition graft in such situation. Finally we decided to go for the following approach: We choose the hybrid operating room, Under local anesthesia rapid right brachial artery access, a (5F) sheath was inserted, a 0.14 terumo wire was used to cannulate right vertebral artery guided by a vertebral catheter (4F), we tried to pass the wire more distally where a covered stent graft 4 mm that used for coronary perforation (biotronic) was the only available in such time, unfortunately our trials failed to cross the site of tear, so a temporary occlusion balloon (3F), 30 mm length was used for v1 control and the balloon was inflated where a successful proximal flow arrest was achieved (Figure 3), and the pulsation of the aneurysm was markedly diminished and the local anesthesia allowed us to talk with patient and test for vertebrobasilar insufficiency symptoms, and fortunately the affected vertebral artery was not dominant. And due to the high cost of coils, and under local anesthesia with sedation, exposure of the V1 segment was obtained through a small transverse (4 cm) supraclavicular approach, dissection downward guided by pulsation of the subclavian artery, and it was encircled by vessel loop, more medial dissection the vertebral artery was identified, and feeling of the inflated balloon guided us to reach the v1 part in very short time, and it was ligated (Figure 4). Closure of the wound was done after ensuring absence of any pulsation signals inside the aneurysmal sac leaving it to thrombose. Our patient postoperative period was smooth, free of any signs or symptoms of focal neurological ischemic insult or vertebrobasilar insufficiency. He was discharged 72 h postoperatively, and strict follow-up revealed considerable decrease in

size of the aneurysm and free neurological condition. Unfortunately postoperative duplex 14 days later showed retrograde filling of the partially thrombosed aneurysmal sac from the contralateral vertebral artery through distal V3 segment that necessitated exposure and ligation. Exposure of the VA V3 segment was done through an anterolateral approach [21] by passing medial to sternomastoid muscle, the spinal accessory nerve is identified, it was retracted anteriorly, the lavatory scapulae and underlying splenius cervicis muscles are readily identified in the posterior wound, vertebral artery was accessed this inter space, the C1 transverse process is identified below and in front of the mastoid tip. The small muscles that insert on it are cut to expose the C1-C2 portion. The inferior aspect of the horizontal portion is safely separated from the atlas groove by elevating the subperiosteal plane and the superior aspect is freed by a cut a few millimeters above the VA on the occipital condyle. Complete De roofing of the C1 transverse foramen is achieved by resecting the bone and V3 was identified and, ligated.

## Discussion

Extracranial VA pseudo aneurysms are generally caused by traumatic factors, which include penetrating or blunt injury, and apart from local compression effect or ischemic posterior circulatory strokes, they may firstly present with rupture leading to cervical hematoma, hypotension, dyspnea, respiratory failure and even hemothorax which indicate a very poor prognosis. While the old Denver classification system was often used to characterize blunt traumatic cerebrovascular injuries and in the setting of penetrating trauma, transection (grade V) and pseudoaneurysm (grade III) are particularly common injury patterns and may result in active hemorrhage or vertebral arteriovenous fistula [22]. A more recent study made by William W. Scott, 2015, it was found that Grade 3 and 4 BCAIs carry the highest stroke risk of the blunt cerebrovascular injuries; the authors define Grade 3 injuries as stenosis of the vessel greater than 50%, or the development of a pseudo aneurysm, and Grade 4 injuries as complete vessel occlusion [23]. According for those classifications, our patient was graded as grade 3 that has the worst prognosis, and that may explain failure to pass a wire across the tear, may be due to the large compressing hematoma or nearly total vertebral artery transection that not allowed us to pass the wire, so we could not complete the procedure totally endovascular. Hanifi Bayarogullari et al. [24], 2016 reported two Syrian 12- and 17-year-old male patients with giant pseudo aneurysms of vertebral arteries the size of the aneurysm was 70 mm × 42 mm × 38 mm at its largest radius. Originating from proximal part of second vertebral part between cervical 7 and 6, its etiology was a shrapnel injury during Syria civil war the size of the other aneurysm was 81 mm × 69 mm × 51 mm at the level of the corpus of the 7<sup>th</sup> vertebra. After thorough review of literature, we think that this case may be the largest reported traumatic pseudoaneurysm of vertebral artery in addition to its anatomical location up till now. Whether the treatment of VA injury is surgery or interventional approach depends on the location and radius of the lesion. VA is divided into four segments along its course. The type of treatment is determined according to the location of PA in these segments [25]. Furthermore, the consequences of VA injure can be fetal and even result in death because of the difficulty in controlling the pulsating hemorrhage which can cause severe hypotension resulting in cardiac arrest and finally death. Therefore, timely diagnosis and intervention of these spontaneous occurrences is crucial in determining the interventional outcome, if the patient has a patent contralateral VA or well-developed collateral circulation

from posterior circulation, an injured vertebral artery could be occluded by the neurosurgeon or endovascular team directly. If the contralateral VA is underdeveloped or slender, vessel repair is the preferred treatment modality: However, ligation of the VA distal to the lesion is necessary on account of successive perfusion from distal collaterals. According to literature and to this paradigmatic case, in order to definitely exclude potential embolic sources and avoid long-term complications of aneurysm revascularization, both proximal and distal occlusions of the aneurysm sac have to be considered for extracranial VA aneurysms. Whenever complete definitive exclusion of the aneurysm cannot be achieved in a single endovascular intervention, a combined approach with surgical distal occlusion of the parent artery closely following the endovascular procedure is recommended.

## Conclusion

We could manage this huge, about to rupture, largest reported traumatic pseudoaneurysm in urgent situation as regard vertebral artery injuries or rupture aneurysm. A hybrid approach, endovascular proximal flow arrest by balloon occlusion of V1, under local anesthesia was an excellent first step procedure as it gave us a diagnostic, and therapeutic value in such urgent, difficult anatomical vertebral artery traumatic injury, and extracranial VA scarifying surgery could be well tolerated in this young age patient, with attention to the other vertebral artery in presence of complete circle of Willis, and the cost and availability of many so expensive tools.

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