Abstract

Background Cervical radiculopathies are rarely caused by vertebral artery loop formation, which is estimated to be present in less than 3% of patients. It is uncertain what causes the loop formation: some propose an association with spondylotic changes or trauma, whilst others suggest hypertension and atherosclerosis may be responsible.

Case Report 1 A 35-year old male patient presented with signs and symptoms of cervical radiculopathy that was not improved with anterior cervical discectomy and fusion surgery performed two years beforehand. Vertebral artery loop was discovered at the level C5/6 on the MRI. Vertebral artery transposition surgery via a lateral approach was performed at the level of the left C5/6 for symptoms of left C6 radiculopathy. Deroofing of the transverse process was performed with post-surgical complete improvement in weakness and pain.

Case Report 2 A 48-year old female patient presented with a ten-year history of left shoulder pain with occasional radiation into her middle three fingers accompanied by intermittent paraesthesia and weakness. Numerous shoulder surgeries, botox injections and suprascapular nerve blocks had not provided any significant benefit. A vertebral artery loop was identified at the level of C3/4 and C4/5 on the left with cervical MRI. Transposition surgery of these two levels provided some post-surgical improvement in pain.

Conclusion Vertebral artery loop formations are a rare but potential cause for cervical radiculopathy. In two cases, the loop formations were not radiographically reported on MRI, thus clinicians should be aware of this as a differential diagnosis in the management of cervical radiculopathy. The presented surgical approach may be useful in managing future cases of vertebral artery loop formation causing cervical radiculopathy resistant to conservative measures.

Key Words: vertebral artery loop; cervical radiculopathy, vertebral artery transposition

Cases Presentation

Case 1

A 35-year old male presented initially in June 2012 with symptoms in keeping with a left C7 radiculopathy. A targeted nerve root block to this level provided 48 hours of relief, but a C6/7 anterior cervical discectomy and fusion (ACDF) did not change his symptoms significantly. He re-presented in 2014 with similar symptoms and a vertebral artery loop at the level C5/6 was confirmed. On examination there was no loss of cervical range of movement. Neurological examination demonstrated decreased sensation in C6, 7 and C8 on the left to light touch, with no loss of strength or abnormal reflexes. Nerve conduction studies were normal for the upper limbs and a recent cervical MRI confirmed a satisfactory decompression of the C6/7 foramina. The patient underwent a transposition of the left vertebral artery loop compressing the C6 nerve root. Postoperatively gradual and significant improvement in pain and sensation was observed. The arm pain improved from 8/10 to 0/10 on the Visual Analog Score (VAS) whilst neck pain reduced from 8/10 to 3/10 postoperatively. Similarly, the Neck Disability Index (NDI) dropped from 38% to 16% postoperatively.

Case 2

A 48-year old female presented with a ten-year history of left shoulder pain radiating into her middle three fingers with occasional paraesthesia and weakness. Original assessment by a shoulder surgeon suggested a rotator cuff tear, however, two arthroscopic shoulder decompressions, numerous Botox injections and suprascapular nerve blocks and intensive physiotherapy failed to provide any long-term benefit. On examination she had significant elevation of her left shoulder with maintained cervical range. Shoulder abduction was reduced to 45 degrees, and flexion to 90 degrees limited by pain. Neurological examination revealed hyper-reflexic left upper limb reflexes, with intact sensation and no significant weakness apart from marked medial border winging of her left scapula. Spurling’s sign was positive.

Investigations revealed a normal shoulder MRI with left C3/4 and 4/5 vertebral artery loop causing radicular compression. Nerve conduction studies were normal. A targeted C5 nerve root block provided good but transient pain relief. The patient underwent a C3-5 left vertebral artery transposition with no complications. Long-term, the patient continued to report persistent shoulder pain, but this may partly be due to the unnecessary shoulder surgeries she had undergone. The pain radiating to her fingers, paraesthesia and weakness did not recur. The arm pain improved from 9/10 to 1/10 on the Visual Analog Score (VAS) whilst neck pain reduced from 9/10 to 6/10 postoperatively. Similarly, the Neck Disability Index (NDI) dropped from 60% to 48% postoperatively.

Diagnostic imaging

In both cases the diagnosis was made based on the magnetic resonance imaging (MRI) showing a tortuous vertebral artery on the symptomatic side with the characteristic flow-void phenomenon proving its patency (Figure 1). The studies were discussed with the neuroradiologists who confirmed the findings and possible compression to the relevant nerve roots by the vessels. The need for the further neurovascular imaging was contemplated. In Computerised tomography angiogram (CTA) was performed in both cases allowing to assess clear the morphology of the VA loop and topographical relations between the loop and surrounding bony structures. Magnetic resonance angiogram (MRA) was initially performed in Case 1 as a less invasive modality and was followed by CTA prior to the operation (Figure 2).

Historical review of the condition, epidemiology, diagnosis, pathology and differential diagnosis

The vertebral artery, originating from the subclavian, passes through the transverse foramina of the C2-C6 vertebrae in the cervical spine, and is referred to as the V2 segment in this region [1]. The limited space for the artery to traverse may result in anomalies or vascular abnormalities causing either compression of the artery (with symptoms of vertebrobasilar insufficiency) or compression of the cervical nerve roots (resulting in pain or radicular symptoms) [2]. An anomalous V2 segment may present as a tortuous vertebral artery; a medial location of the vertebral artery or an abnormal vertebral artery entrance [1]. Hadley was the first to report a tortuous vertebral artery loop with bony erosions in the cervical vertebrae of four out of 21 cadavers [3].

Epidemiology

The incidence of vertebral artery loop formation varies across the literature depending on the criteria used: Paksoy et al. reported an incidence in symptomatic patients of 7.5% [4]; Park et al. excluded bony erosion or widened intervertebral foraminae (typical identifiers) but required a third of the axial aspect of the vertebral artery to be located in the intervertebral foramen and reported an incidence of 0.6% [1]. In contrast, Curylo et al. in a cadaveric study reported an incidence of 2.7% in 222 specimens [5].

Vertebral artery loop formations appear to affect males and females equally, with unilateral lesions most commonly found, followed by multiple levels and rarely bilateral lesions [6]. The most commonly reported affected level is C4/5 followed by C3/4 and C5/6 [6, 7].

Pathogenesis

The pathogenesis of the VA loop formation remains unclear. In his review paper Benny et al. quotes several authors claiming that it is mostly related to the relative elongation of the vertebral artery caused by narrowing of the degenerating disc space [8-10]. Very uncommonly, a VA loop causes direct nerve root compression into the nerve root foramen.

Repeated micro-injuries caused by pulsation of the vessel would lead to a progressive demyelination and subsequent increased irritability resulting in pain. It is a relatively well documented phenomenon observed in cases of trigeminal neuralgia caused by arterial compression of the fifth cranial nerve at the dorsal root entry zone [11]. Therefore, the concept of a direct microvascular decompression appears to be a sensible option while dealing with the cervical radiculopathy related to the VA loop [7, 8, 12-14].

Clinical Diagnosis

Patients with vertebral artery tortuosities may be asymptomatic, wherein they are often incidentally identified, or they may present with symptoms ranging from cervical radiculopathy or neck pain to vertebrobasilar insufficiency or stroke syndromes [2, 7, 13, 15]. MRI and magnetic resonance angiography (MRA) are useful diagnostic tools in the diagnosis of vertebral artery loop formation and are recommended as the first choice. Epstein et al reported vertebral artery loop formations and other vascular malformations as signal void tubular structures on MRI, requiring MRA to differentiate the nature of the lesion [6]

Differential Diagnosis

Vertebral artery loop formation is a rare cause of cervical radiculopathy which can also be attributed to congenital, cystic, vascular and neoplastic conditions [16]. However, differential diagnoses of vertebral artery loop formation due to radiographically widened neural exit foramen may include benign peripheral nerve sheath tumour, congenital absence or hypoplasia of the pedicle, dural ectasia (such as in Marfan syndrome, Ehlers Danlos or neurofibromatosis), intra-spinal neoplasm, meningocele or arachnoid cyst, or metastatic destruction of the pedicle [6]. Neurofibromas most commonly produce a similar presentation of cervical radiculopathy with widened intervertebral foramen however other neoplastic conditions may include meningioma, multiple myeloma or metastatic disease. Arteriovenous malformations and aneurysms should also be considered in cervical cases associated with myelopathy, radiculopathy and or widening of the intervertebral foramen [7].

Management

Several reported cases of vertebral artery loop formation were successfully managed conservatively [1]. This may include non-steroidal anti-inflammatory treatment, physiotherapy and or injection therapy as required [6, 13]. However, in cases where this has failed, surgical treatment appears to be successful and has been reported via a posterior cervical approach [7, 17], anterolateral foraminotomy with sectioning of the compressed rootlet [2, 13, 14] and vascular reconstruction [10].

This paper reports on 2 cases with a lateral approach and deroofing of the transverse process in order to free the affected nerve root.

Procedure

In both cases direct lateral approach to the spine was used (modified Verbiest's approach) [18, 19]. The patients were supine on the table, the head was turned in the direction opposite to the surgical site and immobilized with the Mayfield's clamp (Figure 3A). Once level of the operation was confirmed with the XR linear skin incision was performed along the lateral margin of sternocleidomastoid (SCM) muscle on the relevant side. The lateral aspect of transverse processes (TP) was visualised by gentle retraction of the SCM, strap muscles and neurovascular bundle of neck medially. Self-retain retractor system with short (60 mm) blades was used (SynFrame Ring Clamp, DePuy Synthes, Zuchwil, Switzerland). Fine attachments of the longus coli, longus capitis and anterior scalene muscles were dissected out of the anterior tuberculae of TPs at the level of interest, one level above and one level below. This allowed visualisation of the vertebral artery (VA) confined by venous plexus (VP). The TPs of the vertebrae above and below the loop were de-roofed by removing the anterior tubercula and costotransverse lamellae (bony bridge connecting anterior and posterior tubercula of the TP) with the use of Kerrison rongeur No. 1 (Figure 3B). The VP of the VA was cauterised with bipolar coagulation in order to minimize bleeding during further manipulations. In both cases the tortuous VA loop was visualised and mobilised out of the deroofed transverse foramen and away from the relevant spinal nerve (Figure 3C). A small ($1cm^2$) patch of the Dacron® vascular graft was placed between the nerve and VA in order to isolate the neural structure from the ongoing pulsation of the vessel (Figure 3D) [20, 21]. Classically implanted Teflon sponge was not used due to the well-documented risk of inflammatory giant-cell foreign body reaction [22-24]. No major technical difficulties nor severe bleeding were encountered during either procedure. The wound was washed out with normal saline and closed in layers with a small deep suction drain being left for 24 hours. Patients were discharged from the hospital 48 hours postoperatively once appropriate pain control and level of mobility were obtained.

Outcome and follow up

Case one improved post-operatively with complete resolution of pain and sensation within three months. Nine months later he had maintained his improvement.

Case two had some reduction in pain six months postoperatively, but continued to describe persistent shoulder pain which is thought to be attributed to her numerous shoulder surgeries. Weakness had also not resolved but this was thought to be due to the duration of symptom presentation.

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Figure legends

Figure 1. Diagnostic T2-weighted MRI scans. Top row: Case 1 demonstrating left sided vertebral artery loop (white arrow). Left – sagittal section. Right - axial section at the level C5/6. Bottom row: Case 2 demonstrating the vertebral artery loop (white arrow) on the left with erosion of the vertebral body and intervertebral foramen (white asterisk). Left – coronal section. Right - axial section at the level C4/5.

Figure 2. Neurovascular studies performed in both cases. Left: Lateral reconstruction of the Magnetic Resonance Angiogram (MRA) of Case 1 showing left carotid artery (large vessel on the left) and left vertebral artery (VA). Note remarkably tortuous course of the VA and presence of the loop (white arrow). Right: A Computerised Tomography Angiogram (CTA) of Case 2 demonstrating left sided vertebral artery loops (black arrows) scalloping the vertebral body of C4 and (to a lesser degree C6).

Figure 3. Intraoperative photographs of the Case 1. A. Head and neck position. Note medial and lateral border of the sternocleidomastoid (SCM) muscle (dotted marks on the skin) as well as the level C6/7 (white arrow) which has been marked with the XR. Planned skin incision marked with the while line on the lateral border of the SCM muscle. B. Intraoperative view. Cranial – right. Note: vertebral artery [VA] (black asterisk), longus coli muscle (white asterisk), rim of the C7 transverse foramen [TF] (white arrow), lateral margin of the SCM and strap muscles (black arrows), blades of the SynFrame Ring Clamp system and suction tip are also visible. C. Intraoperative view on the mobilised VA (black asterisk) and C7 nerve root (white arrow). D. Intraoperative view on the VA (black asterisk) isolated from the nerve root by the patch of vascular graft (white).