CASE REPORT

Aberrant cervical vasculature anastomosis as cause of neck pain and successful treatment with embolization technique

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Accepted 6 January 2016

SUMMARY

We report a patient with non-dermatomal radiating neck pain without focal neurologic deficit. Traditional workup could not identify an anatomic or biomechanical cause. Imaging showed a deep cervical vessel centered in the region of pain. Angiography later identified an aberrant anastomosis of this vessel with the occipital artery. Subsequent endovascular embolization of this arterial trunk resulted in complete pain relief.

BACKGROUND

Cervicalgia without focal neurologic findings has a large differential diagnosis. Sources include trauma, including chiropractic manipulations; mechanical instability; referred musculoskeletal pain; malignancy; and rheumatologic disease.¹⁻⁴ However, often no substantive diagnosis can be made nor operative lesion identified, and patients are often sent to pain specialists for long term pain management. We present a patient who presented with non-dermatomal radiating neck pain caused by a hypertrophied vessel that improved after embolization.

CASE PRESENTATION

The patient (aged in their 70s) had a prior C2 dens injury and odontoid screw fixation performed a decade prior after a traumatic accident. The patient presented with a 1 year history of right lower lateral neck pain. The pain was described as sharp, with radiation towards the right neck anteriorly and into the right clavicular region. The patient was seen previously by a neurologist who did not feel that the pain was dermatomal or radicular in nature and recommended surgical consultation. The patient was previously evaluated by two neurosurgeons for neck pain and an orthopedist for the right shoulder, which was normal per imaging and examination.

The pain was significantly impairing quality of life, requiring oxycodone/acetaminophen several times per day without relief, and impeding on daily activities, including work. On examination, there was full strength in the upper extremities bilaterally; deep tendon reflexes were normal. Initial cervical spine flexion/extension X-rays showed only mild degenerative changes, the prior odontoid screw, without abnormal motion. Cervical spine MRI demonstrated spondylotic changes without significant stenosis or neuroforaminal encroachment. However, an enlarged deep right-sided vessel

in the deep posterior cervical musculature was seen (figure 1). This vessel originated around C7, ascending to the skull base, and CT angiography confirmed the presence and course of this vessel. It was unclear if this vessel represented an aberrant venous drainage or arterial anomaly. While the patient's pain did not fit a dermatomal distribution, the distribution is similar to the cutaneous area supplied by supraclavicular nerves that travel in the posterior triangle of the neck.

INVESTIGATIONS

The patient was referred to a neurovascular specialist. Diagnostic angiography of the head and neck was obtained. Imaging confirmed a normal appearing right innominate and right subclavian artery (SA) with a very robust right thyrocervical trunk taking off just distal to the right vertebral artery. This enlarged right thyrocervical vessel was nearly the same caliber in diameter as its vertebral artery counterpart and directly anastomosed with the right occipital artery. No other abnormalities were found in the subclavian or thyrocervical takeoff (figure 2). Selective catheterization of the right thyrocervical trunk confirmed this aberrant connection to the occipital artery and also showed reflux into the proximal external carotid artery. No sign of arteriovenous malformation, fistula, or shunting was identified. Selective catheterizations of the right internal carotid and vertebral artery did not show any anastomosis with this enlarged thyrocervical trunk. Remaining projections of the left intracranial, cervical, and subclavian vessels showed normal vessels.

TREATMENT

The patient was counseled about these findings and accepted the offer for attempted embolization of this hypertrophied right thyrocervical vessel. The patient was brought to the angiography suite and the procedure performed under monitored anesthesia care, which was the preference of the neurointerventionalist. Groin access was obtained in the usual fashion. A NeuronMax (Penumbra) was advanced over a 125 cm Vitek catheter (Cook) to the aortic arch, and selective catheterization of the right SA was performed. A V-18 buddy wire (Boston Scientific) was brought up to provide stability to the NeuronMax catheter, and a Echelon 10 microcatheter (ev3) was advanced over a Synchro standard microwire (Stryker) to selectively catheterize the right thyrocervical vessel. The

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To cite: He L, Ladner TR, Cobb M, et al. *BMJ Case Rep* Published online: [please include Day Month Year] doi:10.1136/bcr-2015-012105

Figure 1 MRI of the cervical spine without contrast. (A) Coronal T2 view with the white arrow indicating the right-sided aberrant posterior cervical vessel. (B) Sagittal T2 view localizing the level to C3; note the metal artifact in the anterior cervical space at C2 from the patient's prior odontoid screw. The axial sequences of the MRI encompassed the entire neck from the skull base downwards; no additional soft tissue MRI of the neck was obtained.

microcatheter was positioned approximately 5 cm distal to the vessel origin and an Axiom 2 mm×6 cm coil, followed by an Axiom 3 mm×8 cm coil (ev3) were deployed. Next, embolization of the proximal 4 cm of the thyrocervical artery was undertaken with Onyx 34 (ev3) (figure 3). Given the relatively straight segment of vessel and no intervening 'nidus' to allow for control of liquid embolic forward migration, these aneurysm coils were placed first to prevent distal extravasation of the liquid embolic material while allowing for complete filling and occlusion of the proximal right thyrocervical trunk. More proximal occlusion of the right thyrocervical trunk was thought safer given the collaterals to the vertebral artery in the distal cervical region. Care was taken to avoid reflux of Onyx into the SA.

Follow-up selective angiography of the right thyrocervical trunk did not demonstrate further anterograde flow from the origin distally or filling of the occipital artery. Next, selective catheterization of the right common carotid artery was undertaken and there was again retrograde filling from the occipital branch distally towards the thyrocervical trunk that arrested at the coil mass (C2–3) without further filling after this point.

Figure 2 (A) Right lateral subclavian projection showing the very robust thyrocervical trunk with superior flow and anastomosis to the right occipital artery. (B) Right anteroposterior selective projection of the thyrocervical trunk showing tortuosity, very robust filling, and again drainage into the occipital artery superiorly; early reflux into the external carotid artery can also be seen.

Figure 3 (A) Pre-embolization selective projection showing the abnormal thyrocervical trunk (white arrow) and vertebral artery (black arrow). (B) Embolized thyrocervical trunk without further filling of the aberrant vessel; note that the right vertebral artery continues to fill.

OUTCOME AND FOLLOW-UP

Postoperatively the patient was monitored in the neurointensive care unit overnight, per our institution's standard protocol after embolization procedures, and was discharged the following day. Immediately postoperatively the patient noted dramatic improvement of the right-sided neck pain. This pain relief was enough to allow the patient to resume daily routines, including return to work 1 week after the embolization procedure. Additional longer term follow-up was attempted with the patient, however this was unsuccessful.

DISCUSSION

Approximately 10% of adults will develop neck pain, with a low likelihood (<1%) of neurologic deficit.³ Vascular causes for neck pain are a minority, and nearly always include dissection secondary to trauma and concurrent headaches or other deficits.^{4–7}

Our patient presented without recent trauma, without deficit or imaging evidence of dissection or vasculitis. We suspect the abnormal right thyrocervical trunk was not a congenital development, given the onset of his symptoms 1 year prior to the intervention. Further, there was moderate stenosis of the right external carotid artery near the carotid bifurcation. Over time, this stenosis may have precipitated significant augmentation of the right thyrocervical trunk to provide collaterals through the occipital artery into the external carotid artery. This augmented vessel may have caused compression on surrounding sensory nerve fibers, specifically to the cutaneous supraclavicular nerve branches in the posterior triangle of the neck, leading to the patient's pain syndrome, as it appeared that the pain followed this abnormal vessel.

The patient's pain syndrome may be similar in mechanism to other vascular compression syndromes, as seen in trigeminal neuralgia.⁸ Two prior articles describe pain secondary to neurovascular compression, both involving sciatic nerve pain.^{9 10} In these cases, decompression of the offending vessel off the nerve alleviated the pain. In our patient, we believe that shutting down flow and pulsatility from the hypertrophied thyrocervical trunk and aberrant connection of the occipital artery relieved aggravation of nociceptors and alleviated the pain—an effect similar to a direct surgical decompression. Additionally, a trial of balloon test occlusion may be helpful to predict if arrest of forward pulsatile arterial flow alleviates

pain prior to committing patients to embolization, especially if these anastomoses are located at more concerning areas, such as in regions with connections to the intracranial vasculature.

It should be noted that other vascular pathologies, including dural arteriovenous fistulas and facial vascular malformations, show evidence of cervical arterial enlargement without pain. In our patient, we believe that the right thyrocervical trunk enlargement and associated anastomosis with the occipital artery occurred in a more acute fashion (onset of pain occurred over the past year) than is typically associated with fistula or vascular malformation formation and may explain why pain was present.

Learning points

- ▶ Clinicians should consider aberrant vascular anastomosis or anatomy as a potential cause of pain when other causes are ruled out.
- ▶ To our knowledge, this is the first reported case of vascular embolization as treatment for atypical neck pain.
- ▶ Severe neck pain may be caused by a vascular arterial source, and endovascular embolization is a safe and robust treatment option.

Contributors LH conceived the project, gathered the data, prepared the figures, wrote and edited the manuscript, and is the guarantor. TRL critically revised the manuscript and approved the final manuscript. MC conceived the project, edited the manuscript, and approved the final manuscript. JM conceived the project, edited the manuscript, and approved the final manuscript.

Competing interests None declared.

Patient consent Not obtained.

Provenance and peer review Not commissioned; externally peer reviewed.

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