CASE REPORT

Axillary artery aneurysm combined with brachial plexus palsy due to Takayasu arteritis

Denise Kim,¹ Graham Roche-Nagle²

¹Department of Medicine,
University of Limerick, Limerick,
Ireland

²Peter Munk Cardiac Center,
Toronto General Hospital,
Toronto, Ontario, Canada

Correspondence to
Dr Graham Roche-Nagle,
graham.roche-nagle@uhn.ca

Accepted 20 December 2017

SUMMARY

We report the case of a young female patient who presented with a right axillary artery aneurysm with neurological symptoms due to Takayasuarteritis (TA). Decompression of the aneurysm and arterial repair with an interposition vein bypass resulted in a full recovery. The case emphasises that peripheral aneurysms can occur due to TA and that there may be neurological complications.

BACKGROUND

Takayasu arteritis (TA) is a vasculitis of unknown aetiology that affects the aorta, its main branches and the pulmonary artery.¹ It is a form of panarteritis, beginning with inflammation of the adventitia, followed by inflammation of the media and intima.² It most often results in stenosis and occlusion, although aneurysms can rarely occur.³ We report the case of a female patient who developed an axillary artery aneurysm due to undiagnosed TA and presented with neurological symptoms. We discuss her subsequent management and review the literature surrounding the case.

CASE PRESENTATION

A 25-year-old healthy woman presented with right upper arm ache and subsequently received physical therapy for suspected rotator cuff tendonitis. Her symptoms continued to worsen and she noted progressive numbness and swelling of her right arm and hand. Two months after the onset of arm ache, she incidentally discovered a tender right axillary fossa mass and sought medical advice.

INVESTIGATIONS

On examination, her axillary mass was pulsatile and 2×3 cm in diameter. She had mild swelling of her right arm and reduced light touch in the distribution of the median nerve in the forearm. Ultrasound confirmed the presence of an axillary aneurysm, and CT angiogram demonstrated a fusiform axillary artery aneurysm at the brachial artery junction. The axillary artery was focally narrowed by 50% immediately proximal to the aneurysm (figure 1). There was a 20 mm Hg difference in blood pressure between both arms but pulses were present. There were no other arterial abnormalities noted. The only significant history was blunt trauma from a bicycle accident 1 year prior, which resulted in hyperabduction of her right arm.

DIFFERENTIAL DIAGNOSIS

At the time, the aneurysm was most likely thought to be secondary to trauma.

TREATMENT

Surgical repair of the axillary aneurysm was performed with a longitudinal incision on the right medial upper arm. Exploration revealed thick scar tissue surrounding the aneurysm encasing the median nerve, which was intact and subsequently released. A reversed saphenous vein interposition graft was used to repair the aneurysm (figure 2), and tissue samples were sent to pathology.

OUTCOME AND FOLLOW-UP

The patient made an uneventful recovery and was discharged home on postoperative day 2. At her 6-week follow-up visit, she was asymptomatic, with normal range of shoulder movement and complete recovery of her median nerve neuropraxia.

On histology, there was evidence of patchy mononuclear inflammatory infiltrate and fibrosis with destruction of elastic tissue consistent with a vasculitis (figure 3). Considering her age, the suspected diagnosis was TA, although inflammatory markers were normal. Two years following surgery, she complained of a pulsatile pain in her right mandible and bilateral persistent ache in her sternocleidomastoid muscles. An ultrasound and MR angiography of her neck revealed diffuse thickening of aorta, early aneurysmal change in the common carotid arteries and a diffuse tight right subclavian stenosis. The clinical picture confirmed the suspected diagnosis of TA. She responded to a course of immune therapy (azathioprine) and steroids.

DISCUSSION

TA is a granulomatous inflammatory arteritis and a primary vasculitis of unknown aetiology that affects the aorta and its main branches.⁴ Most patients present in their 20s and the diagnosis is commonly made before 40 years of age. TA has been strongly linked to women and individuals from Asian and Latin American countries.⁴ TA usually presents with stenosis or occlusion of vessels and less commonly with aneurysmatic disease. Early in the disease course, constitutional symptoms such as fever, malaise and weight loss are common. Symptoms later in the disease are more related to the specific vessel affected and ischaemia, and they include upper or lower extremity claudication, gangrene, ulcers, syncope, vertigo and angina pectoris.⁵

CrossMark

To cite: Kim D,
Roche-Nagle G. *BMJ Case Rep* Published Online First:
[please include Day Month
Year]. doi:10.1136/bcr-2017-
221863

Figure 1 CT angiogram reconstructions demonstrating a fusiform axillary artery aneurysm (arrow) at the brachial artery junction. The axillary artery is focally narrowed by 50% immediately proximal to the aneurysm.

Therefore, it is important to complete a thorough history and complete vascular examination from head to toe. For purposes of classification, a patient shall be said to have TA if at least three of these six American College of Rheumatology diagnostic criteria are present: age less than 40 years at diagnosis, claudication of extremities, decreased brachial artery pressure, a difference in upper extremity systolic blood pressures greater than 10 mm Hg, bruit over the aorta or subclavian arteries and arteriogram abnormalities showing narrowing or occlusion of the aorta, its main branches or large arteries of the extremities that is not due to arteriosclerosis or fibromuscular dysplasia.¹ Initial medical management includes corticosteroid and cytotoxic agents,⁵ and surgical intervention for either occlusive or aneurysmal disease involves bypass surgery, angioplasty, stenting⁶ and cardiovascular risk factor management following the acute inflammatory disease phase.

TA most commonly affects the subclavian, carotid, thoracic and abdominal aorta and mesenteric arteries.⁴ The axillary artery is a rare site for this disease, and only one case has been described in the literature of peripheral nervous system involvement because of Takayasu disease.^{7,8} More commonly, reports of neural symptoms due to Takayasu disease have been of the central nervous system secondary to ischaemia involving the carotid or vertebral artery.⁹ This is an unusual case since the axillary artery aneurysm appears to have been the first indication of Takayasu disease 2 years prior to systemic symptoms.¹⁰

The first symptoms of upper extremity aneurysms can involve nerve injury or compression.¹¹ In this case, there is no evidence as to how long the aneurysm had been present before the onset of neurological symptoms. Nonetheless, intraoperative findings demonstrated that the nerve injury in the present case was due to stretching of the median nerve by the enlarging aneurysm.

A clinical diagnosis of an aneurysm based on the patient's history and physical examination can be confirmed with duplex Doppler ultrasonography. Studies suggest that less than 20% of patients with TA require surgery; however, the presence of concomitant neurological symptoms requires surgical exploration and decompression of the aneurysm.⁴ In these cases, an angiography may provide additional information to aid in planning for surgical management. Surgical intervention is normally performed during the quiescent phase when inflammatory markers are not elevated. Depending on the affected arterial bed, a variety of bypass grafting is considered. For example, an aorto-renal bypass is performed for patients who present with renovascular hypertension, while a carotid to subclavian, axillary or brachial artery bypass grafting is necessary for those presenting with arm ischaemia involving the subclavian arteries. Endovascular graft implantation is a new, minimally invasive intervention that can be used for aneurysms and pseudoaneurysms of the peripheral arterial system and for arteriovenous fistulas.¹² However, endovascular repair in a young patient is a secondary choice, and its long-term results are not yet known.

Figure 2 (A) Revascularisation of the injured axillary artery was achieved using an end-to-end interposition saphenous vein graft. (B) The aneurysm opened and feeding vessels ligated.

Figure 3 Infiltration with mononuclear inflammatory cells.

Learning points

- ▶ Takayasu arteritis (TA) is a rare, systemic, inflammatory vasculitis of unknown aetiology that most commonly affects women of childbearing age.
- ▶ TA is an inflammatory disease of large-sized and medium-sized arteries, resulting in stenotic, occlusive or aneurysmal lesions.
- ▶ Treatment focuses on controlling the inflammation using steroids and immunosuppressive therapy. Surgery is considered for symptomatic complications.

Contributors All authors were responsible in drafting the manuscript, revising it critically for important intellectual content and approving of the version to be published.

Competing interests None declared.

Patient consent Obtained.

Provenance and peer review Not commissioned; externally peer reviewed.

© BMJ Publishing Group Ltd (unless otherwise stated in the text of the article) 2018. All rights reserved. No commercial use is permitted unless otherwise expressly granted.

REFERENCES

- 1 Arend WP, Michel BA, Bloch DA, *et al.* The American College of Rheumatology 1990 criteria for the classification of Takayasu arteritis. *Arthritis Rheum* 1990;33:1129–34.
- 2 Perrotta S, Rådberg G, Perrotta A, *et al.* Aneurysmatic disease in patients with Takayasu disease: a case review. *Herz* 2012;37:347–54.
- 3 Matsumura K, Hirano T, Takeda K, *et al.* Incidence of aneurysms in Takayasu's arteritis. *Angiology* 1991;42:308–15.
- 4 Scovell SD. Giant cell and Takayasu arteritis. *Scientific Am Vascular and Endovascular Surgery* 2015:1–15.
- 5 Pereira RMR, Carvalho JF, Levy-Neto M, *et al.* Takayasu arteritis. In: Shoenfeld Y, Cervera R, Gershwin ME, eds. *Diagnostic criteria in autoimmune disease*. Totowa: Humana Press, 2008:81–6.
- 6 Liang P, Tan-On M, Hoffman GS. Takayasu's arteritis: vascular interventions and outcomes. *J Rheumatol* 2004;31:102–6.
- 7 Trullas JC, Márquez F, Andrés O. Takayasu's arteritis with peripheral nervous system involvement: description of a case and a review of the literature. *Neurologia* 2011;26:369–70.
- 8 Ghosn S, Malek J, Shbaklo Z, *et al.* Takayasu disease presenting as malignant pyoderma gangrenosum in a child with relapsing polychondritis. *J Am Acad Dermatol* 2008;59:S84–S87.
- 9 Stepień A, Durka-Kesy M, Warczyńska A. [Compression neuropathy of cranial nerves in the course of Takayasu arteritis]. *Neurol Neurochir Pol* 2007;41:557–61.
- 10 Grayson PC, Maksimowicz-McKinnon K, Clark TM, *et al.* for the vasculitis clinical research consortium. Distribution of arterial lesions in Takayasu's arteritis and giant cell arteritis. *Ann Rheum Dis* 2012;71:1329–34.
- 11 Ergungor MF, Kars HZ, Yalin R. Median neuralgia caused by brachial pseudoaneurysm. *Neurosurgery* 1989;24:924–5.
- 12 Parodi JC, Schönholz C, Ferreira LM, *et al.* Endovascular stent-graft treatment of traumatic arterial lesions. *Ann Vasc Surg* 1999;13:121–9.

Copyright 2017 BMJ Publishing Group. All rights reserved. For permission to reuse any of this content visit <http://group.bmj.com/group/rights-licensing/permissions>.
BMJ Case Report Fellows may re-use this article for personal use and teaching without any further permission.

Become a Fellow of BMJ Case Reports today and you can:

- ▶ Submit as many cases as you like
- ▶ Enjoy fast sympathetic peer review and rapid publication of accepted articles
- ▶ Access all the published articles
- ▶ Re-use any of the published material for personal use and teaching without further permission

For information on Institutional Fellowships contact consortiasales@bmjgroup.com

Visit casereports.bmj.com for more articles like this and to become a Fellow