Learning from errors

Death from axillary haemorrhage during haemodialysis in a patient with a history of microscopic polyangiitis

Kana Unuma,¹ Hiroshi Uozaki,² Ryouhei Kuroda,³ Koichi Uemura,¹ Ken-ichi Yoshida³

¹Section of Forensic Medicine, Graduate School of Medical and Dental Sciences, Tokyo Medical and Dental University, Tokyo, Japan;

²Department of Pathology, Graduate School of Medicine, University of Tokyo, Tokyo, Japan;

³Department of Forensic Medicine, Graduate School of Medicine, University of Tokyo, Tokyo, Japan

Correspondence to Dr Kana Unuma, unumlegm@tmd.ac.jp

Summary

An older female with a history of microscopic polyangiitis underwent haemodialysis through an end-to-side anastomosis between the left basilica vein and brachial artery. During the last haemodialysis session, repeated punctures induced haemorrhage that required brachial compression. Twenty min posthaemodialysis, haemorrhage had expanded from the axilla to the left lateral thorax. Autopsy disclosed axillary haematoma. The haemorrhage was not derived from punctured vessels or the left axillary artery. Although neither an alveolar nor a glomerular microscopic polyangiitis lesion was detected, fragility of the axillary small vessels due to microscopic polyangiitis, ageing, atherosclerosis and steroid therapy were underlying factors in the haematoma. Aspirin and heparin may have promoted haemorrhage, while shunt vessel stenosis with disturbed flow may have increased the axillary vessel pressure when the shunt vessels were compressed for haemostasis. This is the first report of a death due to haemorrhage from ruptured axillary vessels related to haemodialysis or microscopic polyangiitis.

BACKGROUND

In 1982, Davies *et al* first described systemic polyangiitis in the small arteries, arterioles, venules and capillaries based on positive staining with antineutrophil cytoplasmic antibody (ANCA).¹ ANCA is closely associated with microscopic polyangiitis (MPA), Wegener's granulomatosis and Churg–Strauss syndrome.² The glomerular and pulmonary alveolar capillaries are frequently affected by fibrinoid necrosis and leucocyte infiltration, which result in pulmonary haemorrhage, interstitial pneumonia and crescentic glomerulonephritis.³

Here, we describe the first reported case of an older person who died from axillary vessel haemorrhage related to MPA or haemodialysis, with potential contributions of steroids, aspirin and heparin. Procedures related to problems during haemodialysis might have triggered the haemorrhage. This paper should constitute a warning regarding performance of haemodialysis for patients with this condition and prevent similar accidents.

CASE PRESENTATION

A female in her late 70s was diagnosed with ANCA-associated glomerulonephritis based on high serum levels of MPO-ANCA (32 IU/l) and creatinine (4 mg/dl), but without histological confirmation. She was treated with prednisolone and her condition was stable. Three years later, severe renal dysfunction with pleural and abdominal effusion prompted initiation of haemodialysis. After a further year, vascular access for haemodialysis had become difficult and an end-to-side anastomosis was made between the left basilica vein and brachial artery. An additional shunt was produced by pulling a patent deep vein of the pre-existing shunt to the surface. The patient continued to undergo dialysis three times weekly at a clinic.

The accident described in this report occurred 2 years after the introduction of haemodialysis. After about 50 min of the last haemodialysis session, swelling and pain developed in the left brachium. The physician conducted brachial compression, cooling and pressor infusion until the end of dialysis. The patient complained of pain 20 min posthaemodialysis and the haemorrhage had expanded from the left axillary to lateral thorax. Because of progressive haemorrhagic shock, she was referred to an emergency hospital, at which she was temporarily resuscitated before death. A subsequent review showed that she had experienced a similar haemorrhagic accident during haemodialysis about 1 year earlier.

INVESTIGATIONS

Autopsy findings

A small (146 cm tall, weight 45 kg) older female had subcutaneous haemorrhage in the left arm, axillary, shoulder and lateral thorax regions (figure 1A). Autopsy disclosed a left axillary haematoma in the centre of the haemorrhagic area (figure 1B). Despite intensive efforts, the source of the haemorrhage could not be identified in the axillary haematoma, but axillary artery haemorrhage was carefully excluded.

We confirmed the presence of puncture marks on the shunt vessels in the left cubital dent and the lower brachium, lying 3 cm, 5 cm and 6 cm above the lowest mark, with the absence of overt surrounding haemorrhage below the area 6 cm above the highest mark (figure 1C). The widespread light purple coloration in the left forearms, except for the shunt area, lacked substantial subcutaneous haemorrhage, suggesting previous haemorrhage (figure 1C). These findings suggested that the axillary haemorrhage was not derived from the shunt vessels.

Figure 1 (A) Haemorrhage in the left arms and lateral thorax. (B) Left axillary hematoma, expanding to the lateral thorax and upper brachium. (C) Shunt vessels with puncture marks (arrows) without circumferential haemorrhage. (D) Atherosclerotic stenosis of a shunt vessel (Elastic-Masson staining, $\times 20$).

Histology

Histological examination of the shunt vessels demonstrated severe and irregular intimal thickening with disruption of elastic fibrils and luminal stenosis (figure 1D). The heart (448 g) showed concentric hypertrophy, with $<50\%$ stenosis in the left descending coronary artery and no particular histological abnormality. The polycystic kidney with severe atrophy (left 38 g, right 55 g) showed intensive glomerular sclerosis and focal lymphocyte infiltration, but no crescent formation (figure 2A). There were no characteristic MPA lesions in small arteries, arterioles, veins, venules and capillaries (figure 2A). The lungs (left 244 g, right 222 g) presented with anaemia and a microscopic focus of fungal infection, but no findings of pre-existing alveolar hemorrhage or MPA lesions (figure 2B). There was neither gastrointestinal haemorrhage nor brain abnormality. There were no particular lesions or damage in other organs.

OUTCOME AND FOLLOW-UP

(An autopsy case).

DISCUSSION

The older female with a history of MPA died from haemorrhage due to ruptured axillary vessels. From the axillary haematoma, the haemorrhage expanded to the left lateral thorax and upper brachium, but not to the shunt vessels. The distribution of the haemorrhage and the lack of overt haemorrhage around the shunt vessels with puncture marks suggested that the haemorrhage was derived from the axillary

small vessels. Rupture or aneurysm of the axillary artery and its visible branches was excluded. We were unable to identify the source of haemorrhage from small axillary vessels, which is a well-known difficulty at autopsy. However, there was no doubt that the axillary haematoma contained small vessels, a potential source of haemorrhage, wrapped with heavy coagula. Steroid therapy may have diminished both pulmonary and renal MPA lesions (figure 2).

This is the first report of axillary vessel haemorrhage related to haemodialysis or MPA. Spontaneous haemorrhage related to haemodialysis has been described in the inferior thyroid artery,⁴ but not in the axillary or other vessels. It is surprising that axillary haemorrhage related to haemodialysis has not been reported previously, given the spatial proximity of the brachial and axillary vessels. In MPA patients, there are only two reports of spontaneous haemorrhage from the left gastric artery⁵ and femoral artery.⁶ In non-MPA cases, three cases have been documented with haemorrhage from the axillary vessels: one that occurred 3 days after axillary catheterisation,⁷ another with a non-traumatic cause,⁸ and a third due to non-traumatic shoulder dislocation.⁹ Shinzato *et al* suggested that since the axillary vein lacks the structure to cover the bleeding site, haematoma can expand from the lateral thorax.⁷ Empirically, pathologists are aware of the susceptibility of older patients to haemorrhage, but this has not been formally described in the literature. In our case, the renal and pulmonary vessels lacked MPA or other ANCA-related polyangiitis lesions and successful steroid treatment might have concealed vascular lesions but promoted fragility.

Figure 2 (A) H&E staining of lung ($\times 100$). Neither MPA-related lesions in pulmonary arterioles, venules and capillaries (insert, $\times 200$) nor signs of alveolar haemorrhage are apparent. (B) Elastica-Masson staining of kidney ($\times 100$) showing sclerotic glomeruli without crescent formation. MPA-related lesions were not found in the arterioles (red arrow), venules (yellow arrow) and capillaries.

The causality of haemodialysis for the axillary haemorrhage was addressed for clarification in the medicolegal autopsy. Due to the severe atherosclerotic stenosis, the flow in the shunt vessels may have been reduced by a posture change or an unidentified change related to haemodialysis. Such a problem would have prompted the physician to perform repeated punctures. The axillary haemorrhage might have misled the physician into performing brachial compression for haemostasis. This practice and an unidentified change related to haemodialysis might have raised the pressure of small venous or artery vessels in the axilla, thereby inducing rupture in the background of vascular fragility and haemorrhagic tendency. Additionally, aspirin (daily) and heparin (during haemodialysis) may have promoted haemorrhage.

Learning points

- This is the first report of death due to haemorrhage resulting from a ruptured axillary vessel related to haemodialysis or MPA. As such, it constitutes a warning regarding performance of haemodialysis for patients with this condition.
- Axillary small vessels may become fragile due to MPA, ageing, atherosclerosis and steroid therapy; and aspirin and heparin may promote haemorrhage.
- Reduced shunt flow related to haemodialysis may prompt brachial punctures, with resulting axillary haemorrhage that can be misleading in suggesting the need for brachial compression for haemostasis. This can lead to increased axillary vessel pressure and subsequent rupture.

Competing interests None.

Patient consent Obtained.

REFERENCES

1. **Davies DJ**, Moran JE, Niall JF, *et al*. Segmental necrotising glomerulonephritis with antineutrophil antibody: possible arbovirus aetiology? *Br Med J (Clin Res Ed)* 1982;**285**:606.
2. **Kallenberg CG**, Brouwer E, Weening JJ, *et al*. Anti-neutrophil cytoplasmic antibodies: current diagnostic and pathophysiological potential. *Kidney Int* 1994;**46**:1–15.
3. **Bonsib SM**. Polyarteritis nodosa. *Semin Diagn Pathol* 2001;**18**:14–23.
4. **Tsai SF**, Hung SW, Wu MJ, *et al*. Spontaneous rupture of inferior thyroid artery in a uremic patient on maintenance hemodialysis. *Intern Med* 2011;**50**:1271–2.
5. **Yahata K**, Okamoto C, Imamaki H, *et al*. Fatal hemoperitoneum due to rupture of the left gastric artery in a patient with microscopic polyangiitis. *Clin Exp Nephrol* 2009;**13**:512–7.
6. **Abe M**, Okada K, Maruyama N, *et al*. A case of femoral hemorrhage in a patient with microscopic polyangiitis with low levels of myeloperoxidase-antineutrophil cytoplasmic autoantibody. *Clin Exp Nephrol* 2011;**15**:414–8.
7. **Shinzato T**, Fukui M, Kooguchi K, *et al*. Hemorrhagic shock 3 days after catheterization from the axillary vein. *J Anesth* 2010;**24**:290–2.
8. **Jazayeri S**, Tatou E, Cheynel N, *et al*. A spontaneous rupture of the external iliac vein revealed as a phlegmasia cerulea dolens with acute lower limb ischemia: Case report and review of the literature. *J Vasc Surg* 2002;**35**:999–1002.
9. **Subbarao JV**, Chintam R, Rao MS, *et al*. Non traumatic dislocation of shoulder with rupture of axillary vessel branch in a paraplegic patient: a case report. *J Am Paraplegia Soc* 1990;**13**:15–7.

This pdf has been created automatically from the final edited text and images.

Copyright 2012 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.

Please cite this article as follows (you will need to access the article online to obtain the date of publication).

Unuma K, Uozaki H, Kuroda R, Uemura K, Yoshida K. Death from axillary haemorrhage during haemodialysis in a patient with a history of microscopic polyangiitis. *BMJ Case Reports* 2012;10.1136/bcr.11.2011.5194, Published XXX

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