

Stridor and Horner's syndrome, weeks after attempted right subclavian vein cannulation

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ABSTRACT

A 23-year-old woman presented with renal failure resulting from polycystic kidney disease (PKD) aggravated by tubulo-interstitial nephritis. Emergency haemodialysis was planned, and cannulation of the right subclavian vein was attempted, but failed. During this procedure, inadvertent arterial puncture occurred. Transient mild ischaemia of the right arm, and a transient Horner's syndrome were noted. Seven weeks later she presented with severe stridor with impending respiratory failure necessitating emergency intubation; the right-sided Horner's syndrome had recurred. CT imaging showed a large pseudo-aneurysm of the brachiocephalic artery resulting in severe compression of the trachea. Using a prosthetic graft, the operation for the pseudo-aneurysm was successful; there were mild neurological sequelae. Although her family history was negative, autosomal dominant PKD should be considered, and we discuss the possible role of a pre-existing PKD-associated aneurysm.

INTRODUCTION

Mechanical complications may occur during attempted cannulation of central veins.^{1,2} One of these adverse events is inadvertent arterial puncture with subsequent potentially fatal injury to the subclavian artery.^{3,4} Bleeding may result in acute-onset airway obstruction,⁵ but also late onset sequelae, such as complications from false aneurysms of the vertebral artery and thyrocervical trunk have been reported.^{6,7} Horner's syndrome has been reported after internal jugular vein puncture.^{8,9} We report both sequelae (i.e. Horner's syndrome and airways obstruction) resulting

from pseudo-aneurysm formation of the brachiocephalic artery after attempted cannulation of the right subclavian vein.

CASE REPORT

A 23-year-old woman presented to the nephrology division of our hospital with malaise and a skin rash. Her previous medical history was remarkable for recurrent pyelonephritis and polycystic kidneys. She had been treated by her family physician for a urinary tract infection with amoxicillin-clavulanic acid; a urinary specimen grew *E. coli* >10⁶ colony-forming units per ml. Dysuria had subsided and her urinary output was unchanged. Apart from the skin rash, and the palpable kidneys, and slight overweight, physical examination was unremarkable. Her blood chemistry showed mild anaemia (Hb after rehydration, 5.7 mmol/l) and renal failure (urea 115 mmol/l, serum creatinine 1810 µmol/l). Arterial blood gas analysis showed pH 7.33, *p*aO₂ 16.3 kPa, *p*aCO₂ 2.5 kPa, HCO₃⁻ 10 mmol/l. She was admitted to hospital and haemodialysis was planned. During attempted insertion of a haemodialysis catheter into the right subclavian vein, inadvertent arterial puncture occurred. This was followed by transient ischaemia of the right arm, and an incomplete right-sided Horner's syndrome was observed, with ptosis and miosis of the right eye; she also had a hoarse voice. The procedure was converted to cannulation of the femoral vein, followed by haemodialysis. A renal biopsy showed tubulo-interstitial nephritis, presumably resulting from an adverse effect of the antimicrobial therapy that was given for her urinary tract infection. The Horner's syndrome

subsided gradually but incompletely, and the hoarseness of her voice was no longer noticeable. Her renal failure subsided gradually after corticosteroid treatment was started (60 mg of prednisolone for four weeks, then gradually tapered) and she was discharged home after three weeks of admission.

One month after discharge – seven weeks after attempted cannulation of the right subclavian vein – she presented to the emergency department of our hospital with severe dyspnoea. She had noticed gradual swelling of her face, and gradually increasing dyspnoea over the last three days. On examination her face was cushingoid, and her neck was swollen; there was an audible stridor. Her right arm was oedematous with no arterial pulsations. She was admitted to the intensive care unit, and developed respiratory failure during the first night of observation, and had to be intubated and mechanically ventilated.

A portable chest radiograph showed a right paratracheal mass, with deviation of the trachea to the left, and compression of the tracheal lumen (*figure 1*). Contrast-enhanced computed tomography showed gross aneurysmatic dilatation of the brachiocephalic artery starting just above the artery's origin at the aortic arch, and extending beyond the right subclavian artery. The lumen of the subclavian artery could not be visualised (*figure 2*).



Figure 1

Posteroanterior chest radiograph taken in upright position, showing severe deviation of the trachea to the left (arrows), and severe narrowing of the tracheal lumen, extending from the thoracic inlet to the main carina

During the operation, through a mid-sternal approach with upward extension of the incision to the right, the brachiocephalic vein and artery and the right carotid artery were identified and mobilised; the brachiocephalic artery showed



Figure 2

Contrast-enhanced computed tomography showing a large haematoma (H) around a small contrast-enhanced lumen of the brachiocephalic artery (B); deviation and compression of the trachea (T); deviation of the superior caval vein (C), and of the oesophagus (E)

gross aneurysmatic dilatation. In order to explore the right carotid artery, the anterior scalenic muscle had to be sacrificed. After clipping the aneurysm was opened, and intimal dehiscence was noticed; there was no communication with the haematoma surrounding the lumen. Blood clots were evacuated, and fresh bleeding appeared to come from the apparently damaged right subclavian artery. A vascular prosthetic graft was used to bypass the brachiocephalic artery; the graft connected the aortic arch and the right carotid artery. A second prosthetic graft was used to connect the right subclavian artery to the first graft end-to-side. After surgery, pulsation of arteries was satisfactory, and the postoperative course was uneventful. Post-extubation, the right hemidiaphragm was partly elevated, probably due to pressure to the right phrenic nerve during surgery; and there was hoarseness of her voice that appeared to be due to right vocal cord paresis. A mild transient paresis of her left arm was believed to result from clamping of the right carotid artery during surgery.

DISCUSSION

Our patient sustained late-onset airways obstruction, and Horner's syndrome, resulting from pseudo-aneurysm formation, seven weeks after inadvertent puncture of the brachiocephalic artery. She also had an injury to the right subclavian artery that caused flow limitation to the right arm, but apparently, collateral circulation prevented her from developing ischaemic symptoms. Although she

complained of a swollen face, no clear superior caval vein compression resulted from the aneurysm. The right vocal cord dysfunction was diagnosed as a recurrent laryngeal nerve injury, and although this was believed to result from compression during surgery, compression resulting from the aneurysms cannot be ruled out.

We only found one case report in the non-English literature on false aneurysm formation of the brachiocephalic artery after attempted cannulation of the right subclavian vein,¹⁰ but a combination of this uncommon but life-threatening complication with Horner's syndrome has not been reported. False aneurysm of the subclavian artery with stridor and dysphagia resulting from tracheal and oesophageal obstruction has, however, been reported.^{11,12}

Could she have developed these problems because of a pre-existing aneurysm, associated with an inherited syndrome that caused autosomal dominant polycystic kidney disease (ADPKD) and aneurysm formation? Although she did not admit to having family members with ADPKD, most of these patients have mutations in one of the two genes – PKD 1 and 2¹³ – which code for the protein polycystin that is also expressed in the vascular wall,¹⁴ and some 10% of these individuals may develop arterial aneurysms.¹³ Aneurysms usually occur intracranially,¹⁵ but occasionally other vascular structures including carotid and vertebral arteries may be affected.¹⁶

In most cases of inadvertent arterial puncture during attempts to cannulate the internal jugular and subclavian veins, no adverse events are noted.² Our patient had severe azotaemia with subsequent thrombocytopathia, and impaired platelet function may have aggravated the arterial bleeding.^{1,3} Other identifiable risk factors were also present – obesity being one and, possibly, multiple attempts and failure to cannulate the subclavian vein.^{17,18} Experience is important^{1,2} but the attending physician's great experience did not prevent this complication from occurring. In our opinion, however, experience comes from learning, and the learning process is unavoidable; indeed, training and learning are paramount for any teaching hospital. Some authors advocate ultrasound guidance, but in one large prospective randomised controlled trial with a very experienced team who inserted central venous catheters, no benefit could be demonstrated regarding prevention of failures and complications.¹ Ultrasound guidance has been advocated for selected, high-risk cases.^{2,18} A recent meta-analysis of randomised studies including predominantly medical personnel with limited experience in inserting catheters showed, however, significant reduction in the complication rate when ultrasound guidance was used for cannulation of the subclavian and internal jugular veins.¹⁹

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