CASE REPORT

Spontaneous adrenal haemorrhage after catheter ablation of supraventricular tachycardia

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Accepted 10 September 2015

SUMMARY

Catheter ablation is established as a first-line therapy for most patients with recurrent supraventricular tachycardia (SVT), with high success rates and very low complication rates. A 60-year-old woman developed severe right flank pain following straightforward catheter ablation for SVT. This was caused by a spontaneous right adrenal haemorrhage, which, after much delay, was eventually recognised as the cause of her symptoms. Adrenal haematomas are rare and, to the best of our knowledge, this is the first reported case of spontaneous adrenal haemorrhage occurring after any interventional cardiac procedure. Clinicians should be aware of this rare but potentially serious complication and consider it as a differential diagnosis in any patient with severe flank pain following interventional cardiac procedures, to prevent delays in diagnosis.

BACKGROUND

Catheter ablation is established as a first-line therapy for most patients with recurrent supraventricular tachycardia and considered a class IB indication in the most recent international guidelines.¹ Clinical success rates are high (generally in excess of 96%) while procedural complications are very low. In a prospective registry of 1197 patients with atrioventricular nodal re-entrant tachycardia (AVNRT) who underwent catheter ablation in 1998, complications were reported in only 25 (2%) patients (atrioventricular nodal injury in 15 patients, haematoma in 6 patients, venous thrombosis in 2 patients and pneumothoraces in 2 patients). More contemporary single centre series have reported even lower major complication rates of 0.8% (4 of 524 patients), which consisted of one pericardial effusion, two femoral pseudoaneurysms and a single case of pulmonary oedema.² In this article, we detail a previously unreported complication with potentially serious clinical consequences following routine catheter ablation, which clinicians should be cognisant of to avoid, first, delays in diagnosis and second, subjecting patients to unnecessary investigations.

CASE PRESENTATION

A 60-year-old Chinese woman with recurrent palpitations refractory to verapamil and β -blockers was referred to the arrhythmia clinic. Her symptoms correlated with 12-lead ECG documentation of a narrow complex, short RP tachycardia, which could occasionally be terminated by vagal manoeuvres. Her medical history was notable only for

mechanical back pain and her only regular medication was verapamil.

Therefore, she consented to be admitted electively for electrophysiological studies and ablation. The procedure was performed under conscious sedation with midazolam and fentanyl. Venous access was attained using two and three 6 Fr sheaths in the right and left femoral veins, respectively, through which quadripolar diagnostic EP catheters were positioned at the high right atrium, His bundle and right ventricular apex, and a decapolar catheter was placed within the coronary sinus. Catheter insertion was performed under fluoroscopic guidance, without any difficulty or resistance. Electrophysiological studies confirmed dual atrioventricular node physiology and excluded the presence of an accessory pathway. A narrow complex tachycardia with a tachycardia cycle length of 300 ms, and simultaneous atrial and ventricular activation (VA time 9 ms) were readily and reproducibly inducible. Ventricular overdrive pacing during tachycardia yielded a ventricle-atrial-his-ventricle (VAHV) response with a corrected post-pacing interval minus tachycardia cycle length (PPI-TCL) of 227 ms, confirming the diagnosis of typical AVNRT. Slow pathway modification was performed using a solid tipped 4 mm ablation catheter, with slow pathway block as the end point, which persisted after 20 min of waiting time. All catheters and sheaths were removed and haemostasis was achieved by manual compression of both femoral veins. No anticoagulant was given throughout the procedure.

However, 20 min following the procedure in the recovery area, the patient developed sudden onset of excruciating right flank pain (rated by the patient as 10 of 10 in severity), which remained refractory to paracetamol and tramadol. Physical examination revealed stable blood pressure of 110/70 mm Hg, sinus tachycardia of 105 bpm and a soft, non-tender abdomen. There were no visible cutaneous changes over the right flank. A broad list of differential diagnoses, largely unrelated to the ablation procedure, was considered, and included retroperitoneal bleeding, pyelonephritis, renal calculus and hepatobiliary disease.

INVESTIGATIONS

Bedside echocardiography revealed a structurally normal heart and excluded pericardial bleeding. Laboratory testing showed a slight decrease in haemoglobin (post-ablation 12.6 compared with 13.9 g/dL preoperatively) and marked leucocytosis ($15.3 \times 10^9/L$ with predominant neutrophilia). Serum

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To cite: Kristanto W, Chan PF, Yeong SS, et al. *BMJ Case Rep* Published online: [please include Day Month Year] doi:10.1136/bcr-2015-209754

electrolytes, glucose, creatinine, liver enzymes, amylase, pH, blood gas analysis and urinalysis were normal. An urgent CT of the abdomen and pelvis was performed to exclude vascular injury with resultant intra-abdominal bleeding. The liver, pancreas, spleen and both kidneys appeared normal without any mass lesions. Gallstones were visible within a non-distended, thin-walled gallbladder. Free fluid was absent from the peritoneum and retroperitoneal bleeding was excluded. However, a nodule measuring 3.4 cm×2.4 cm (figure 1) was seen within the right adrenal gland. The left adrenal gland appeared normal. The right adrenal mass was thought to be an incidentaloma or haematoma. The clinical relevance of this finding was not fully appreciated at this point in time.

OUTCOME AND FOLLOW-UP

Despite the patient requiring intravenous opiates to achieve pain relief, her haemodynamic status remained stable and serial full blood counts did not suggest continuing haemorrhage.

Biochemical testing including serum cortisol, adrenocorticotrophic hormone, catecholamines, aldosterone and dehydroepiandrosterone, to exclude a hormonally active adrenal tumour, was performed and all measurements were within normal limits. The possibility of spontaneous adrenal haemorrhage being the cause of her symptoms was considered during multidisciplinary discussions among the cardiac electrophysiology (EP), radiology and endocrinology teams. The likelihood was thought to be low, as such a complication had not been reported following any percutaneous cardiac interventions, and it seemed unlikely that such a small haematoma could account for the patient's disproportionately severe pain. Even if adrenal haemorrhage was the correct diagnosis, the prognosis for the patient was considered to be good, as there were no signs of continuing haemorrhage or adrenal insufficiency. The consensus among the teams was to undertake further evaluations in the search for an alternative explanation for the patient's severe pain. A general surgical consult was sought. A working diagnosis of cholecystitis was made on the basis of persistent right flank pain, leucocytosis and presence of gallstones. The patient continued to require intravenous opiates, including morphine, for pain relief and was started on intravenous antibiotics for the next 4 days. On further review by the hepatobiliary surgeons, the diagnosis of cholecystitis was felt to be unlikely. An alternative but equally unconvincing diagnosis of gastro-oesophageal reflux was made and proton pump inhibitor therapy was suggested.

After extensive investigations over 4 days, which excluded other differential diagnoses, we concluded that the right spontaneous adrenal haemorrhage was the most probable cause of the patient's severe postoperative pain. The patient was discharged well and pain-free. A repeat CT scan was performed 2 months later, which demonstrated a much smaller, heterogeneous, non-enhancing right adrenal gland (figure 2). Taken in conjunction, these radiological findings were highly supportive

Figure 1 CT showing a right adrenal haematoma measuring 3.4 cm×2.4 cm (white arrows); (A) transverse plane and (B) coronal plane.

Figure 2 Contrast CT scan showing resolution and shrinkage of the right adrenal haematoma 2 months later (white arrows); (A) transverse plane and (B) coronal plane.

of an evolving spontaneous right adrenal haematoma, which was finally recognised as the sole cause of the patient's severe postoperative flank pain.

The patient remained well and free from palpitations during more than 1 year of follow-up.

DISCUSSION

To the best of our knowledge, this is the first case of spontaneous unilateral adrenal haemorrhage following any interventional cardiac procedure, including electrophysiological studies and catheter ablation.

Adrenal haemorrhage is rare and has been associated with trauma, severe stress caused by surgery, sepsis and hypotension, anticoagulation, and haematological disorders such as thrombocytopenia, pregnancy and tumours.³ In a recent review of all 133 cases of haemorrhagic adrenal masses reported in the literature up to 2010, 20% were malignant in the form of adrenocortical cancers or metastases.⁴ Another 50% were due to pheochromocytomas, while other benign tumours such as myelolipomas and adrenal adenomas accounted for another 15%. Haematomas were found in 17% of cases.

The most common presentation of unilateral adrenal haemorrhage is with flank pain, as described in our patient. A potential sequel of adrenal haemorrhage is persistent bleeding, which may necessitate further intervention such as blood transfusion, endovascular embolisation of the bleeding adrenal artery and even surgical adrenalectomy. Adrenal haemorrhages are unilateral in 95% of cases, but in rare cases of bilateral adrenal haemorrhage, there is the additional life-threatening risk of adrenal insufficiency, which will require supportive therapy with intravenous fluid and steroid replacement. It is therefore not surprising that, in autopsy series of patients with adrenal haemorrhages compared to clinical case series, the proportion of cases with bilateral haemorrhages was much greater, by as much as 73%.⁵

The adrenal glands have a rich arterial supply provided by three arteries, in contrast to their limited venous drainage, which is critically dependent on a single vein.⁵ In situations of physiological stress such as during the preoperative and perioperative period, blood flow to the adrenal glands and cortisol production can increase several fold. Consequently, increased adrenal blood flow could lead to venous congestion, culminating in haemorrhagic necrosis, as seen in our patient. The electrophysiological studies and ablation were performed under conscious sedation and therefore the patient was comfortable throughout the procedure.

However, it is possible that she may have been anxious in the days leading up to her admission. It would be speculative to comment on whether better preprocedural counselling or a short course of anxiolytics in such patients would reduce the occurrence of such a rare complication.

Learning points

- ▶ Spontaneous adrenal haemorrhage is rare.
- ▶ With ever increasing numbers of patients undergoing interventional cardiac procedures, clinicians should consider adrenal haemorrhage as a differential diagnosis in patients reporting severe postprocedural flank pain.
- ▶ A small adrenal haemorrhage can cause severe flank pain.
- ▶ An adrenal haematoma can be visualised on CT scan but definitive diagnosis requires serial imaging confirming evolution or resolution of the mass.
- ▶ Potential life-threatening sequelae include persistent bleeding and acute adrenal insufficiency.

Contributors All the authors were involved in patient care, drafting of the manuscript, the literature search and approval of the final manuscript, and agree to be accountable for the accuracy and integrity of this submission. PK came up with the idea, and is the guarantor.

Competing interests None declared.

Patient consent Obtained.

Provenance and peer review Not commissioned; externally peer reviewed.

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