ESC European Society of Cardiology

European Heart Journal - Case Reports (2019) **3**, 1–4
doi:10.1093/ehjcr/ytz109

CASE REPORT
Arrhythmias/electrophysiology

Acute fluctuating neurological deficits after pulmonary vein isolation: unmasking a rare complication due to spontaneous spinal subdural bleeding: a case report

Giacomo Maria Cioffi *, François Regoli, Giulio Conte, and Angelo Auricchio

Department of Cardiology, Fondazione Cardiocentro Ticino, Via Tesserete 48, CH-6900 Lugano, Switzerland

Received 10 January 2019; first decision 4 February 2019; accepted 17 June 2019; online publish-ahead-of-print 5 July 2019

Background Pulmonary vein isolation (PVI) is becoming the therapy of choice for symptomatic paroxysmal drug-refractory atrial fibrillation (AF). The most frequently reported complications are vascular complications (1.4%). Bleeding complications of the central nervous system have rarely been described. We report a case of spontaneous spinal bleed after PVI.

Case summary A 68-year-old woman with a 2-year history of highly symptomatic paroxysmal AF (EHRA 3) was referred for a PVI redo procedure. A high-density mapping showed pulmonary vein reconnection of all pulmonary veins successfully isolated by radiofrequency ablation. During the entire procedure, the patient had sinus rhythm with an ACT around 300 s. No intraprocedural and peri-procedural complications occurred. Four hours after haemostasis, the anticoagulation clotting time (ACT) was 110 s and rivaroxaban (20 mg) was reinitiated. In the following hours, the patient developed fluctuating neurological lower limb symptoms. A lumbar magnetic resonance imaging showed a subdural spinal haematic collection with an associated epidural component from L3 to S2 exerting compression over the dural sheath. A conservative treatment approach was adopted with progressive recovery of sensorial and motor deficits. After 5 months, the patient still presented residual lower limb motor deficits necessitating the support of a walking stick.

Discussion We describe the first case of a spontaneous spinal bleeding following PVI. Given the gradual diffusion of PVI to treat AF in more clinically complex patients with a larger range of comorbidities, particular consideration should be given to seek predisposing bleeding factors in order to assess the risk for neurological complications.

Keywords Pulmonary vein isolation • Atrial fibrillation • Case report • Spinal bleeding • Haematomyelia • Anticoagulation

Learning points

- Catheter ablation has a growing role in the management of patients with atrial fibrillation and a growing number of patients with multiple comorbidities are being treated. These are fragile patients and may present high bleeding risk.
- Post-pulmonary vein isolation neurological complications are very rare events and potentially life-threatening.
- Spinal cord bleeding can have insidious and misleading presentation with fluctuating central or peripheral neurological symptoms.

* Corresponding author. Tel: +41 (0) 91 805 33 47, Fax: +41 (0) 91 805 31 67, Email: giacomo.cioffi@swissonline.ch

Handling Editor: Tor Biering-Sørensen

Peer-reviewers: Rami Riziq Yousef Abumuaileq and Habib Khan

Compliance Editor: Rahul Mukherjee

Supplementary Material Editor: Vishal Shahil Mehta

Introduction

Pulmonary vein isolation (PVI) is becoming the therapy of choice for symptomatic paroxysmal drug-refractory atrial fibrillation (AF). The most frequently reported complications are vascular complications (1.4%) in a setting of overall low complication rate (<2.9%).[3] Bleeding complications of the central nervous system (CNS) have rarely been described. We report a case of spontaneous spinal bleeding after PVI not reported in the literature so far.

Timeline

Timeline	Events
7 months before presentation	First pulmonary vein isolation (PVI) procedure.
3 months before presentation	New severely symptomatic atrial fibrillation episodes.
Day 1	PVI redo procedure performed.
4 h later	Restarting oral anticoagulation (Rivaroxaban).
6–9 h later	Beginning of neurological fluctuating deficits. Thoracic, chest, and cerebral angio-computed tomography scan was performed.
12 h later	Worsening symptoms. A cervical, dorsal and lumbar native magnetic resonance imaging was performed with evidence of subdural spinal haematic collection with an associated epidural component from L3 to S2.
24 h later	Elevated surgical risk. Conservative approach.
Day 6	Clinical stability with minimal improvement.
Day 9	Discharge from cardiology department to neurological rehabilitation centre.
5 months after discharge	Still minor sensorial and motor deficits. Need of a walking stick.

Case presentation

A 68-year-old woman with a 2-year history of highly symptomatic drug-refractory paroxysmal AF (EHRA 3), previously treated with a previous PVI procedure, was referred to our division for a PVI redo procedure. The patient (height 160 cm, weight 69 kg) presented a history of two-vessel coronary artery disease treated with percutaneous revascularization, a CHA_2DS_2-VASc score of 4, and HAS-BLED score of 2. She had a history of hypertension, hypothyroidism, and a congenital ostium secundum atrial septal defect with a left-right spontaneous shunt. The patient presented also moderate kyphoscoliosis of the vertebral column, most prominent at the lumbar level. She was treated with rivaroxaban 20 mg/day, aspirin, sotalol, thyroxin, furosemide/spironolactone medication, and pantoprazole. The pre-procedural blood values, including routine coagulation indices were all normal.

The anticoagulation therapy was interrupted 36 h before the procedure and a transoesophageal echocardiogram performed on the same day of the procedure excluded left atrial appendage thrombi. The procedure was performed under general anaesthesia and vascular access was obtained through the right femoral vein. High-density mapping was performed during sinus rhythm and showed pulmonary vein reconnection of all pulmonary veins. A radiofrequency ablation catheter (Thermocool Smarttouch, Inc., Irvine, CA, USA) was performed; it showed a reconnection of three pulmonary veins. A mapping procedure obtained 20 min after the last ablation point showed a persistent deconnection of all pulmonary veins. Atrial programmed stimulation as well as repeated high-rate pacing bursts did not induce AF. During the entire procedure, the patient had a sinus rhythm, remained stable with vital signs within range of normality; ACT was stable around 300 s with a final procedural value of 259 s. No reversion of anticoagulation was performed and no intraprocedural and peri-procedural complications occurred. The patient was then carefully transferred to the ward bed with the aid of a sliding mat; a mandatory 4 h bed rest and immobilization was prescribed.

At the time of femoral vein haemostasis (4 h after PVI completion), the ACT was 110 s; rivaroxaban (20 mg) was then administered. In the following hours, the patient developed right gluteal and bilateral lumbar pain, right lower limb paraesthesia, and motor deficits below the knee [patellar and achillean hyporeflexia; iliopsoas, quadriceps, ankle dorsiflexors, plantar flexors, and long toe extensors motor deficits (MMT 2)]. A psoas sign was also present. The neurological symptoms fluctuated presenting an on–off pattern intermittently affecting either the right or left lower limb; there was the suspicion for either a transient central or peripheral involvement. A complete thoracic, chest, and cerebral angio-computed tomography scan was performed and acute retroperitoneal or cerebral bleeding were excluded.

Furthermore, a cervical, dorsal, and lumbar native magnetic resonance imaging was then performed. This examination showed a subdural spinal haematic collection with an associated epidural component from L3 to S2 (8×11 mm in the axial plane, see *Figure 1*; 65×11 mm in the sagittal plane, see *Figure 2*). The subdural spinal haematoma exerted compression over the dural sheath, abolishing the liquoral representation around the cauda equina lateral right nerve roots within L3–L4 and L4–L5 and within L5–S1 mainly in the posterior bilateral location.

After consultation with neurologists and neurosurgeons, a 'wait-and-see' treatment approach by withholding anticoagulation therapy and patient immobilization was taken. Indeed, the operative risk of subsequent lower limb paralysis associated to the surgical approach was estimated too high and exceeding possible benefit. In the following 9 days, a neurological rehabilitation and sub-therapeutic anticoagulation regimen was initiated. There was progressive recovery of sensorial and motor deficits. After 5 months though, the patient still presented residual lower limb motor deficits necessitating the support of a walking stick.

Discussion

To our knowledge, this is the first case of spontaneous spinal subdural bleeding observed after a PVI procedure.

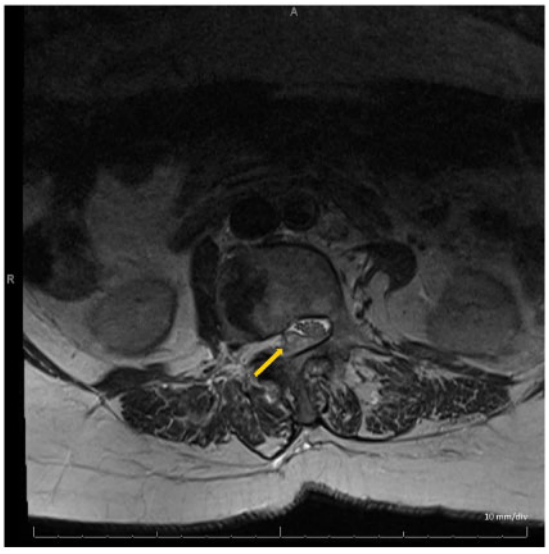

Figure 1 Subdural spinal haematic collection 8 x 11 mm, axial plane.

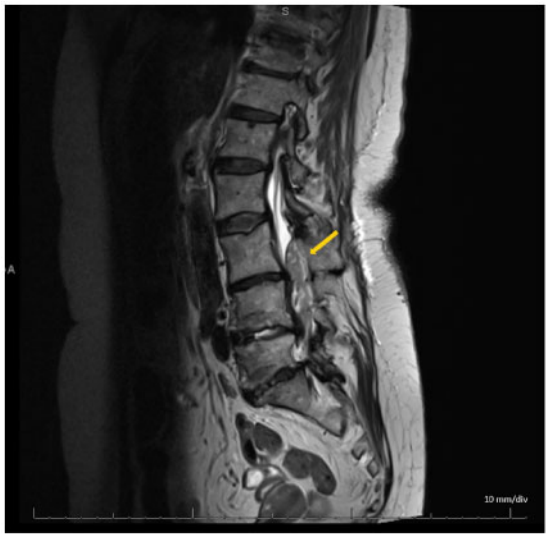

Figure 2 Subdural spinal haematic collection 65 x 11 mm, sagittal plane.

Although this complication has never been reported following PVI, it emphasizes the need of careful evaluation of multimorbidity patients and of possible muscle-skeletal changes beyond the pathologies included in the HAS-BLED score.

Oral anticoagulation was interrupted 36 h before the procedure; this was in agreement with the 2017 HRS/EHRA/APHRS/SOLAECE recommendations[12], when the procedure was performed. Subsequently, recommendations by scientific societies[13] changed into uninterrupted DOACs at the time of PVI. Although speculative, during uninterrupted DOACs (in cases when CHA_2DS_2-VASc ≥3, availability of cardiac imaging assessment at procedural time) or interrupted within 12–24 h, the bleeding risk in a patient could even be higher than that assessed by the HAS-BLED score. Indeed, anatomic conditions may increase the bleeding risk and are not part of the HAS-BLED score.

Vascular complications are the most frequent procedure-related adverse events following PVI procedure. These include generic haematoma of the groin, retroperitoneal haematoma, pseudoaneurysm, arteriovenous fistula, and hemothorax (subclavian or internal jugular venous access) with an incidence in the range of 1–2%[3]. Peri-procedure anticoagulation regimen seems to be an important factor in the occurrence of these complications.

Intracranial severe spontaneous bleeding complications are rare after PVI (incidence of 0.3[1]). However, as described in the present case, such complications may cause invalidating and irreversible body injury which may be particularly limitative in more compromised patients. As observed in the present case, even careful procedural planning with guidelines recommendations of peri-procedural anticoagulation management did not prevent a severe CNS bleeding adverse event.[2–3]

Spinal cord bleeding is a unique condition with significant disabling consequences. It is classified based on the primary location of bleeding into intramedullary (haematomyelia), subarachnoid haemorrhage (SAH), subdural haemorrhage, and epidural haemorrhage. It requires emergency investigation and treatment.[4–6] Based on the existing literature spinal vascular malformations such as intradural arteriovenous malformations are the most common cause of atraumatic intramedullary spinal cord haemorrhage. Additional causative factors include warfarin or heparin anticoagulation, hereditary or acquired bleeding disorders, primary spinal cord tumours, spinal cord metastases, or a delayed complication of spinal radiation[7–11].

Onset may vary with sudden, severe back or neck pain and sometimes radicular or with rapidly progressive flaccid quadri/biparesis, with subsequent ventilatory failure reported in cases where bleeding involves the cervical and superior dorsal spine. Magnetic resonance imaging with and without gadolinium is the gold standard neurological imaging modality. There are no clinical trials to guide the management of acute intramedullary spinal cord haemorrhage, and subsequent treatment is usually directed towards the underlying cause. Early surgical treatment is always indicated when the patient's neurological status progressively deteriorates with severe central complications (such as ventilatory failure) but also conservative treatment is possible, whenever neurological impairment is minimal and self-limiting.

Conclusion

In conclusion, for the first time spinal subdural bleeding after PVI is reported. Careful peri-procedural management of anticoagulation therapy as well as assessment of patient bleeding risk are mandatory. Little is known on the predisposing factors that could identify which patients may be at risk of bleeding complications such as that reported in this case.

Lead author biography

Giacomo Maria Cioffi, 34, raised in Lugano (CH), works for Cardiocentro Ticino as resident in Cardiology since 2018, with special interest in Electrophysiology.

Supplementary material

Supplementary material is available at *European Heart Journal - Case Reports* online.

Slide sets: A fully edited slide set detailing this case and suitable for local presentation is available online as Supplementary data.

Consent: The author/s confirm that written consent for submission and publication of this case report including image(s) and associated text has been obtained from the patient in line with COPE guidance.

Conflict of interest: A. A. has been a consultant to Medtronic, Boston Scientific, Biosense Webster, and LivaNova and has received speakers' fees from Medtronic, Boston Scientific, and LivaNova. F. R. consultation and speaker fees Medtronic, Boston Scientific, LivaNova/Microport, Bayer, Abbott and Daichi Sankyo. G.C. and G.M.C. have declared no conflict of interest.

References

1. Kirchhof P, Haeusler KG, Blank B, De Bono J, Callans D, Elvan A, Fetsch T, Van Gelder IC, Gentlesk P, Grimaldi M, Hansen J, Hindricks G, Al-Khalidi HR, Massaro T, Mont L, Nielsen JC, Nölker G, Piccini JP, De Potter T, Scherr D, Schotten U, Themistoclakis S, Todd D, Vijgen J, Di Biase L. Apixaban in patients at risk of stroke undergoing atrial fibrillation ablation. *Eur Heart J* 2018;**39**:2942–2955.
2. Marrouche NF, Brachmann J, Andresen D, Siebels J, Boersma L, Jordaens L, Merkely B, Pokushalov E, Sanders P, Proff J, Schunkert H, Christ H, Vogt J, Bänsch D. Catheter ablation for atrial fibrillation with heart failure. *N Engl J Med* 2018;**378**:417–427.
3. Hoyt H, Bhonsale A, Chilukuri K, Alhumaid F, Needleman M, Edwards D, Govil A, Nazarian S, Cheng A, Henrikson CA, Sinha S, Marine JE, Berger R, Calkins H, Spragg DD. Complications arising from catheter ablation of atrial fibrillation: temporal trends and predictors. *Heart Rhythm* 2011;**8**:1869–1874.
4. Brinkmeier-Theofanopoulou M, Tzamalis P, Wehrkamp-Richter S, Radzewitz A, Merkel M, Schymik G, van Mark G, Bramlage P, Schmitt C, Luik A. Periprocedural anticoagulation during left atrial ablation: interrupted and uninterrupted vitamin K-antagonists or uninterrupted novel anticoagulants. *BMC Cardiovasc Disord* 2018;**18**:71.
5. Aldhoon B, Wichterle D, Peichl P, Čihák R, Kautzner J. Complications of catheter ablation for atrial fibrillation in a high-volume centre with the use of intracardiac echocardiography. *Europace* 2013;**15**:24–32.
6. Palomaki A, Lehtola H, Kiviniemi T, Mustonen P, Hartikainen J, Nuotio I, Ylitalo A, Hartikainen P, Airaksinen KEJ. Spontaneous and traumatic intracranial bleeds in patients with atrial fibrillation. *Eur Heart J* 2017;**38**:ehx504.P4568.
7. Shaban A, Moritani T, Al Kasabx S, Sheharyarx A, Limayex KS, Adams HP Jr. Spinal cord hemorrhage. *J Stroke Cerebrovasc Dis* 2018;**27**:1435–1446.
8. Leep Hunderfund AN, Wijdicks EF. Intramedullary spinal cord hemorrhage (hematomyelia). *Rev Neurol Dis* 2009;**6**:E54–E61.
9. Pegat B, Drapier S, Morandi X, Edan G. Spinal cord hemorrhage in a patient with neurosarcoidosis on long term corticosteroid therapy: case report. *BMC Neurol* 2015;**15**:123.
10. Helmy A, Mellor G. Spontaneous cervical cord haemorrhage: an unusual presentation. *Emerg Med J* 2007;**24**:e16.
11. Kim JS, Lee SH. Spontaneous spinal subarachnoid hemorrhage with spontaneous resolution. *J Korean Neurosurg Soc* 2009;**45**:253–255.
12. Calkins H, Hindricks G, Cappato R, Kim YH, Saad EB, Aguinaga L, Akar JG, Badhwar V, Brugada J, Camm J, Chen PS, Chen SA, Chung MK, Nielsen JC, Curtis AB, Davies DW, Day JD, d'Avila A, de Groot NMSN, Di Biase L, Duytschaever M, Edgerton JR, Ellenbogen KA, Ellinor PT, Ernst S, Fenelon G, Gerstenfeld EP, Haines DE, Haissaguerre M, Helm RH, Hylek E, Jackman WM, Jalife J, Kalman JM, Kautzner J, Kottkamp H, Kuck KH, Kumagai K, Lee R, Lewalter T, Lindsay BD, Macle L, Mansour M, Marchlinski FE, Michaud GF, Nakagawa H, Natale A, Nattel S, Okumura K, Packer D, Pokushalov E, Reynolds MR, Sanders P, Scanavacca M, Schilling R, Tondo C, Tsao H-M, Verma A, Wilber DJ, Yamane T. 2017 HRS/EHRA/ECAS/APHRS/SOLAECE expert consensus statement on catheter and surgical ablation of atrial fibrillation. *Heart Rhythm* 2017;**14**:e275–e444.
13. Steffel J, Verhamme P, Potpara TS, Albaladejo P, Antz M, Desteghe L, Haeusler KG, Oldgren J, Reinecke H, Roldan-Schilling V, Rowell N, Sinnaeve P, Collins R, Camm AJ, Heidbüchel H. The 2018 European Heart Rhythm Association Practical Guide on the use of non-vitamin K antagonist oral anticoagulants in patients with atrial fibrillation. *Eur Heart J* 2018;**39**:1330–1393.