

A Stroke and a Strange Intrapulmonary Shunt

Joana Laranjeira Correia, Gonçalo R. M. Ferreira, Vanda Neto, João Fiúza, António Costa, Miguel Correia

Centro Hospitalar Tondela-Viseu, Viseu, Portugal

ARTICLE INFO

Article history:

Submitted: 13. 11. 2022

Accepted: 19. 12. 2022

Available online: 21. 8. 2023

Klíčová slova:

Arteriovenózní malformace

Cévní mozková příhoda

Paradoxní embolie

Zobrazovací metody pro vyšetření srdece

Keywords:

Arteriovenous malformations

Cardiac imaging techniques

Paradoxical embolism

Stroke

SOUHRN

Spektrum plicních arteriovenózních malformací (pulmonary arteriovenous malformation, PAVM) se pohybuje v rozmezí od mikroskopických lézí až po izolované nebo četné velké abnormální zkraty mezi plicnicí a plicní žilou. Mezi hlavní komplikace PAVM, jež jsou výsledkem intrapulmonálního zkratu, patří cévní mozková příhoda, mozkový absces a hypoxemie. Paradoxní embolie je považována za jednu z hlavních příčin cévních mozkových příhod u mladých pacientů; ve vzácných případech k ní může dojít i na úrovni plic. Popisujeme případ původně zdravé 44leté ženy s intrapulmonálním zkratem prokázaným jícnovým echokardiogramem během vyšetření kryptogenní cévní mozkové příhody. Ani použité pokročilé zobrazovací metody nezjistily žádné arteriovenózní malformace.

© 2023, ČKS.

ABSTRACT

Pulmonary arteriovenous malformations (PAVMs) spectrum extends from microscopic lesions to single or multiple large abnormal pulmonary artery to pulmonary venous connections. The main complications of PAVMs result from intrapulmonary shunt and include stroke, brain abscess, and hypoxemia. Paradoxical embolism is considered one of the main causes in young stroke patients and rarely, paradoxical embolism can also develop at pulmonary level. We describe a case of a previously healthy 44-year-old woman with the evidence of intrapulmonary shunt on transesophageal echocardiogram during the study of a cryptogenic stroke. However, no arteriovenous malformations were detected in spite of using advanced imaging techniques.

Introduction

Pulmonary arteriovenous malformations (PAVMs) spectrum extends from microscopic lesions to single or multiple large abnormal pulmonary artery to pulmonary venous connections.¹ The main complications of PAVMs result from intrapulmonary shunt and include stroke, brain abscess, and hypoxemia.²

Paradoxical embolism is considered one of the main causes in young stroke patients and rarely paradoxical embolism can also develop at pulmonary level.^{3,4} We describe a case of a previously healthy 44-year-old woman with the evidence of intrapulmonary shunt on transesophageal echocardiogram during the study of a cryptogenic stroke. However, no arteriovenous malformations were detected in spite of using advanced imaging techniques.

Case description

A 44-year-old female patient was referred to the ecocardiography laboratory to perform a transesophageal study

in the context of cryptogenic stroke investigation. There were no visible intracardiac shunts, so we performed an agitated saline study ("bubble study") after the Valsalva manouever, in order to exclude a patent foramen ovale. Immediately after injection we saw adequate opacification of the right atrium with concomitant left deviation of the interauricular septum, but no evidence of contrast passage to the left cavities. Nevertheless, in later cardiac cycles (>4 cycles after injection), suddenly microbubbles could be seen in the left cavities (Fig. 1). This type of abnormality raised the possibility of an intrapulmonary shunt. To prove this hypothesis, we performed injections of agitated saline with direct visualization of the different pulmonary veins. After finding an adequate plane to visualize the left pulmonary veins (Fig. 2) and using agitated saline, it is possible to see microbubbles in the left superior pulmonary vein, in transit to the atrium (Fig. 3 and Video 1). The same findings were not reproduced with visualization of the right pulmonary veins. These findings are compatible with an intrapulmonary shunt. However, when the computed tomography pulmonary angiography was performed no intrapulmonary shunt was identified.

Address: Joana Maria Laranjeira Correia, Av. Rei Dom Duarte, 3504-509 Viseu, Portugal, e-mail: joanalaranjeira1116@gmail.com

DOI: 10.33678/cor.2022.135

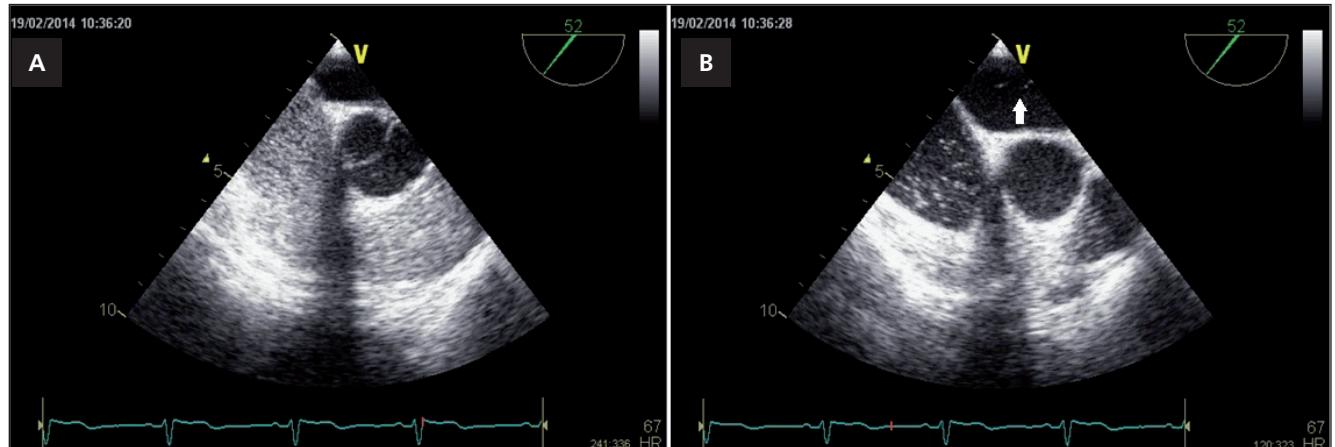


Fig. 1 – TEE, Midesophageal aortic short axis view, centered on the interauricular septum. (A) Immediately after injection of agitated saline, adequate opacification of the right atrium is seen, but without any passage of microbubbles to the left cavities. (B) Later, more than 4 heartbeats after injection we suddenly detect presence of microbubbles on the left atrium (white arrow).

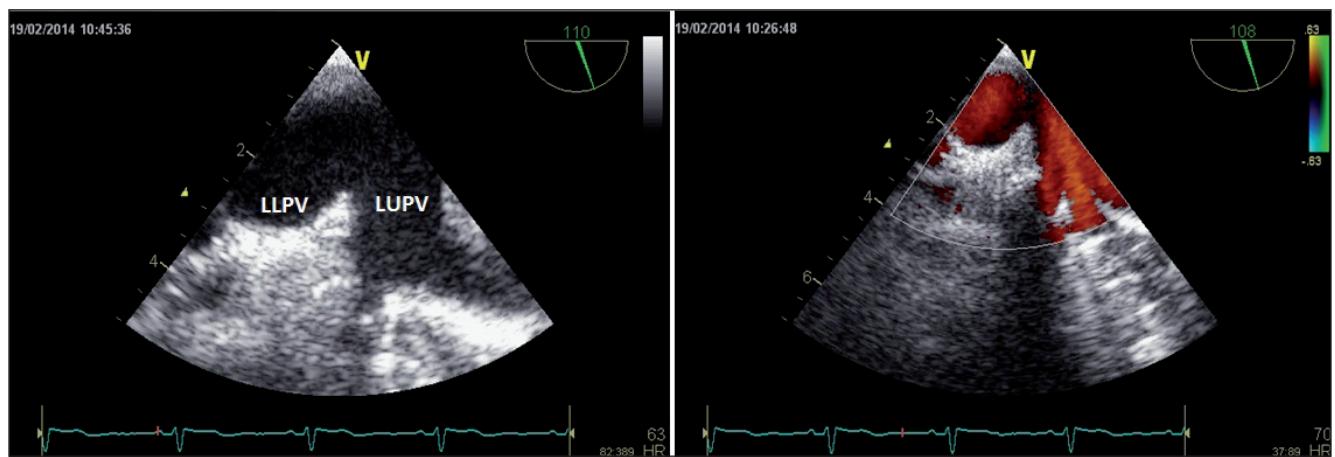


Fig. 2 – TEE, left pulmonary veins view, illustrating the pathway of the left upper pulmonary vein (LUPV) and left lower pulmonary vein (LLPV).

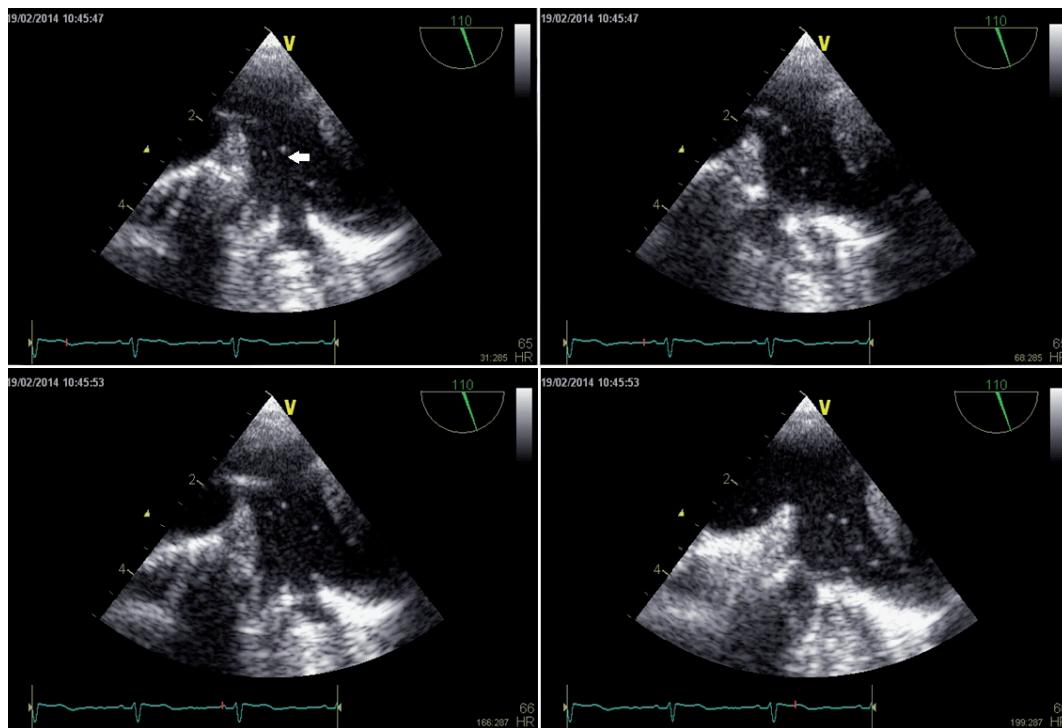


Fig. 3 – TEE, left pulmonary veins view, after injection of agitated saline. It is possible to see microbubbles in transit to the left atrium, especially in the LUPV (white arrow), here demonstrated in multiple heartbeats.

Discussion/Conclusion

Intrapulmonary shunt is commonly demonstrated by contrast echocardiogram when bubbles from agitated saline are visualized in the left atrium within 3–6 beats after being noted in the right side of the heart, as we present in this case report. Bubbles are not normally observed in the absence of vascular dilatation because lung capillaries act as filters.^{5–8}

There are a few case reports with the evidence of intrapulmonary shunts on echocardiographic study, that are not identifiable in spite of using advanced imaging techniques.^{6,9} The possible diagnosis is microvascular pulmonary arteriovenous malformations (PAVM) that can be diagnosed with lung biopsy if diffuse disease.^{1,9} On other hand, an intrapulmonary shunt with dimensions smaller than the acquisition cuts may not be detected on CT.

This case illustrates the embolic outcome of intrapulmonary shunt, due to paradoxical embolization, which demonstrates the importance of a systematized approach and etiological research.

Conflict of interest

No conflict of interest.

Funding

This research did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors.

Supplementary video is available online.

References

1. Cartin-Ceba R, Swanson KL, Krowka MJ. Pulmonary arteriovenous malformations. *Chest* 2013;144:1033–1044.
2. Shovlin CL. Pulmonary arteriovenous malformations. *Am J Respir Crit Care Med* 2014;190:1217–1228.
3. Wozniak L, Mielczarek M, Sabiniewicz R. Paradoxical brain embolism in a young man: is it only a patent foramen ovale? *Neurol Neurochir Pol* 2015;49:61–64.
4. Abushora MY, Bhatia N, Alnabki Z, et al. Intrapulmonary shunt is a potentially unrecognized cause of ischemic stroke and transient ischemic attack. *J Am Soc Echocardiogr* 2013;26:683–690.
5. Felix A, Alcantara ML. To Close or not to Close PFOs in Cryptogenic Stroke, an Evolving Question. *Int J Cardiovasc Sci* 2020;33:318–320.
6. Khabbaza JE, Krasuski RA, Tonelli AR. Intrapulmonary shunt confirmed by intracardiac echocardiography in the diagnosis of hepatopulmonary syndrome. *Hepatology (Baltimore)* 2013;58:1514–1515.
7. Soliman OI, Geleijnse ML, Meijboom FJ, et al. The use of contrast echocardiography for the detection of cardiac shunts. *Eur J Echocardiogr* 2007;8:S2–S12.
8. Kerut EK, Bourgeois B, Serio J, Nanda NC. Intrapulmonary shunts and its implications for assessment in stroke and divers with type II decompression illness. *Echocardiography* 2014;31:3–4.
9. Nosib S, Watt K, Penz E, Fenton M. A 58-year-old woman with hypoxia, hypoxaemia, a hole in the heart and a ... herring! Intracardiac or extracardiac shunt? That is the question! *BMJ Case Rep* 2015;2015:bcr2015211975.
10. Erudilho E, Marchini JFM, Ghandour MS, et al. Percutaneous embolization of pulmonary arteriovenous fistulae in patients with hereditary hemorrhagic telangiectasia. *Rev Bras Cardiol Invasiva* 2013;21:85–88.
11. White RI Jr. Pulmonary arteriovenous malformations: how do I embolize? *Tech Vasc Interv Radiol* 2007;10:283–290.