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CASE REPORT - NEUROSURGERY GENERAL

Thromboembolic infarction caused by an unknown patent foramen ovale 30 years after VA shunt insertion: a case report and review of the literature

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Abstract

Background Ventriculoatrial shunt (VA) insertion is one of the possible surgical procedures to treat hydrocephalus. However, it is also associated with several complications such as obstruction and shunt infection as well as life-threatening complications like intraatrial thrombus or thrombosis on the distal catheter. In this case report, we share a rare case of a patient with a VA shunt, who was admitted to our hospital with a stroke.

Case description A 56-year-old female patient with suspected acute stroke was admitted to the stroke unit. CT and MRI scans showed multiple cerebral infarctions in both hemispheres. The transesophageal echocardiography (TEE) showed at the tip of the VA shunt catheter, which was implanted about 30 years ago due to aqueduct stenosis, also a thrombotic formation as the reason of stroke. Interestingly, the tip of the catheter was not in the right atrium as expected, but in the left atrium. Further evaluation showed a patent foramen ovale (PFO), through which the catheter migrated from the right to the left side. At first, conservative treatment with anticoagulation was started with the aim to dissolve the thrombotic formation; however, a control TEE showed an unchanged mass at the catheter tip. Therefore, a ventriculoperitoneal shunt was implanted and the proximal shunt catheter was removed with an additional closure of the PFO by our heart surgeons. Postoperatively, the patient was discharged 10 days later in good condition to a rehabilitation center.

Conclusions Thromboembolic events due to a PFO are rare but possible life-threatening complication after VA shunt insertion. Therefore, preoperative cardiac diagnostic might be clinically relevant prior to a VA shunt implantation to avoid such complications.

Keywords Ventriculoatrial shunt · Unknown PFO · Migrated catheter tip · Stroke

Introduction

Hydrocephalus is a common neurosurgical disease, which can occur at any age. The treatment depends on the etiology of hydrocephalus; however, the most commonly performed procedure is the shunt implantation. Previously, several methods had been carried out, such as ventriculopleural, ventriculoureteral, or ventriculogastric shunt with a high rate of complication [16]. Among them, two surgical methods, ventriculoperitoneal (VP) and ventriculoatrial (VA) shunt implantation, have been shown to be the most effective ones with low complication rate [4, 21].

To date, the most performed and preferable shunt implantation is the VP shunt [6–8, 11, 14, 15]. There are several reasons for this, such as the ease in placement and ease of revision of it. Several studies reported and compared those methods, but there was no significant difference in terms of perioperative or postoperative complication. Both methods were associated with infections, shunt obstruction, shunt dysfunction, or intracerebral/subdural hemorrhage [1, 3, 17, 19]. Particularly for VA shunt, some studies reported about life-threatening

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complications such as cardiopulmonary complications like thromboembolism especially at the right atrium, vena cava thrombosis, jugular thrombosis, thrombosis on the distal catheter, pulmonary hypertension, or infective endocarditis [2, 6, 9]. Despite those possible complications, VA shunt is still an inevitable alternative to VP shunt in certain circumstance [2].

It is worth mentioning that there is no obligatory preoperative cardiac diagnostic in the clinical routine prior to VA shunt implantation. However, there is no evidence that a PFO is a contraindication for VA shunts. To date, this matter is not sufficiently described in the literature.

Therefore, here, we present an unusual case of a female patient with a VA shunt and an unknown PFO. Our patient presented with a sudden occipital headache on the left side, visual changes, and slurred speech. In 1989, she was treated with a VA shunt insertion because of hydrocephalus due to aqueductal stenosis.

Case report

We present a case of a 56-year-old female with a sudden occipital headache on the left side, visual changes, and slurred speech. With a suspected acute stroke, the patient was admitted to the stroke unit. A computer-assisted tomography (CT) and magnetic resonance imaging (MRI) were performed, in which embolic infarctions of the posterior cerebral artery (PCA) on the left side, superior cerebellar artery (SCA) on the right side, and posterior inferior cerebellar artery (PICA) on the left side were detected. Furthermore, a shunt catheter was also seen in the right occipital horn, which was implanted due to aqueduct stenosis about 30 years ago.

In the following days, stroke diagnostic was performed. Electrocardiography and duplex sonography were not conclusive for the etiology of stroke; however, transesophageal echocardiogram (TEE) showed an approximately 7-mm mass with suspected thrombotic materials at the tip of the distal shunt. Surprisingly, the tip of the distal shunt catheter was not as expected in the right atrium, but in the left atrium (Fig. 1). Additionally, an unknown PFO was described. It was obvious that the distal shunt catheter moved over the years from the initial right atrium through an unknown PFO into the left atrium. Therefore, we started anticoagulation therapy with low-molecular-weight heparin with the aim to prevent thromboembolic complication as well as resolve the thrombotic materials. The anti-Xa-level was regularly controlled to adjust the effective dose of heparin. A second CT scan did not show any new infarct area as well as no significant difference of ventricular width compared with the initial CT scan.

After 7 days, once more, a 3D-TEE was performed to evaluate the development of the thrombotic mass under the anticoagulation treatment. However, it showed an unchanged formation size at the tip of the shunt and it was clear to see that the mass on the distal catheter was a flattening thrombosis. Due to the high risk of cerebral embolism/stroke with the unchanged TEE finding, an interdisciplinary surgical approach with heart surgeons was chosen. Prior to the heart surgery, the distal part of the ventriculoatrial shunt was disconnected from its pump and a new VP shunt implanted on the right side. The correct placement was controlled via CT scan and X-ray of the shunt system. After 10 days, the patient underwent an open-heart surgery over a standard median sternotomy. The distal shunt catheter with an attached fluctuate mass was removed from the heart, and the persistent foramen ovale was closed (Fig. 2).

The patient recovered well from both operations. Accordingly, we were able to send the patient to a rehabilitation clinic in good clinical status with some remaining visual impairment. After 8 weeks, our patient came to our neurosurgical department for a follow-up checkup. She reported that she is in a good general condition with persisting right homonymous quadrant hemianopia. In summary, she clearly recovered from both operative interventions.

Discussion

Here, we report about a rare but life-threatening complication of a VA shunt caused by the distal tip of the catheter which moved from the right into the left atrium through an unknown PFO.

VA shunt is a proven alternative procedure to a VP shunt for the treatment of hydrocephalus. Nevertheless, it also inherits several complications such as obstruction, infection, pulmonary hypertension, distal or proximal catheter malfunction, and thromboembolic events [12]. One of the most feared complications is cardiopulmonary life-threatening complications. Lundar et al. [13] described that the latency period of cardiopulmonary complications ranges between 10 to 20 years after VA shunt insertion. Since extraneous material has a thrombogenic characteristic in case of direct contact to the blood system, thrombosis in the right or left atrium, jugular thrombosis, or vena cava thrombosis can occur and cause life-threatening complications as described in several studies [6, 9, 18].

To the best of our knowledge, several case reports referred to thromboembolic events caused by the shunt catheter placed in the right atrium; however, complications caused by migrated shunt catheter has been understudied. Indeed, there was one case from 2009 reporting a distal rest tip of the catheter (1 ft), which migrated over the years through a PFO into the left atrium, causing life-threatening complication. However, to date, there were no cases about a migrated functioning VA shunt catheter reported.

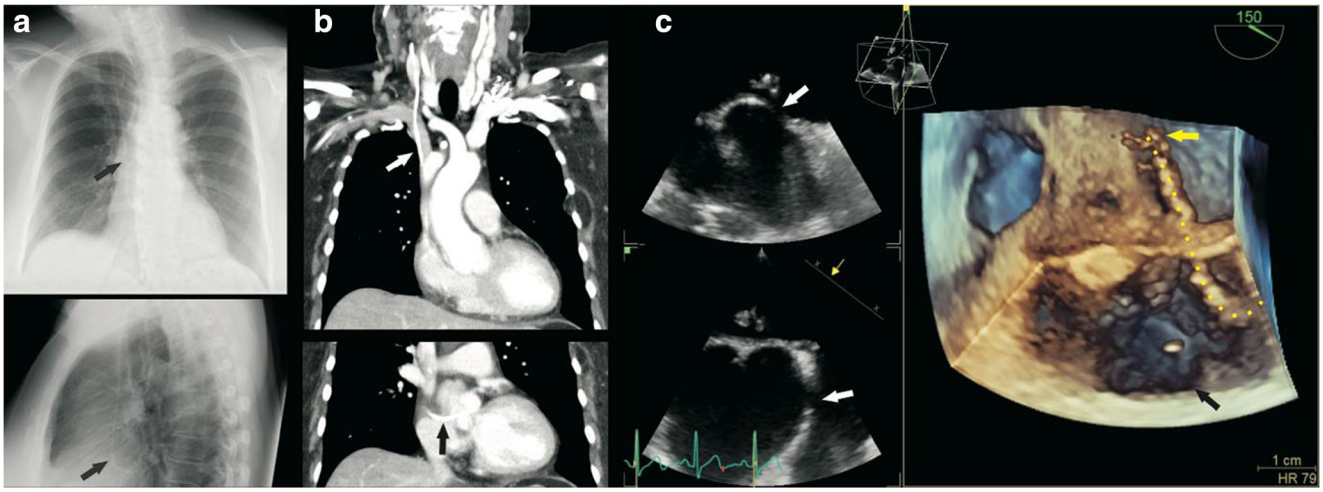

Fig. 1 Series of chest X-ray, chest CT scans, and images of 3D-transesophageal echocardiography course of the shunt catheter over the right atrium in the left atrium. **a** Black arrow. **b** White and black arrow. **c** On the left side: (white arrow) PFO; on the right side: (black arrow) right atrium, (yellow arrow) tip of the shunt catheter in the left atrium

The frequency of a PFO in the population reported in autopsy studies was 15 to 35% [5]. Hagen et al. [5] described that the size of PFO tended to increase with increasing age. They noted an increasing size, from a mean of 3.4 mm based on the first decade of life to 5.8 mm in the 10th decade of life, whereas the conventional VA shunt catheter has a diameter of approximately 1.1 mm. Thus, at an early age, patients with an unknown/known PFO have an increased risk for a repositioning of the catheter tip. Additionally, through the accumulation of comorbidities like pulmonary hypertension, cardiovascular diseases, and dilative cardiomyopathy, the preload of the right atrium could be increased over the years causing latent PFO to become a manifest PFO inducing the shunt catheter movement. Although this complication is quite seldom, it seems to be important to perform an additional preoperative diagnostic to exclude a relevant PFO before VA shunt placement to avoid rare but relevant life-threating complications.

Another alternative to prevent thromboembolic events in patients with VA shunt might be an anticoagulation treatment. It is important to mention that in our case thromboembolic complications occurred 30 years after catheter implantation. Anticoagulation is widely used in patients with extraneous material in the blood system to avoid thromboembolic complications. [10]. Mostly, the anticoagulation could be paused in the course through the epithelialization of those materials reducing the thromboembolic risk.

However, in the case of VA shunt, the proximal catheter is located in the right atrium, floating in the blood system resulting in the absence of an epithelialization. Therefore, thrombus formation could occur at any time, even after 30 years as described in our case. Additionally, prophylactic antithrombotic treatment could be considered in the future to prevent those thromboembolic complications in patients with a PFO where VA shunt is absolutely indicated. On the other hand, if there is a contraindication of anticoagulation

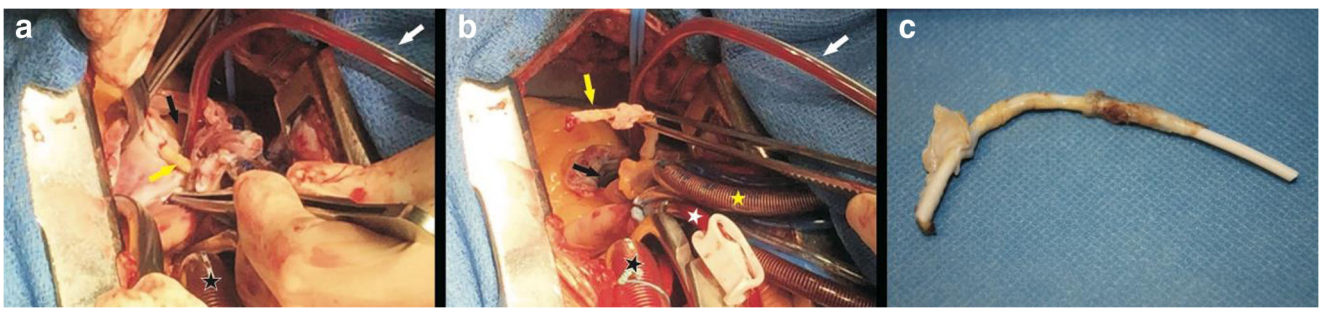

Fig. 2 Intraoperative resection of the distal catheter. **a** (Yellow arrow) distal shunt catheter, (black arrow) opened right atrium, and (white arrow) suction (black star) aortic root cannula; **b** (yellow arrow) distal shunt catheter with thrombotic formation, (black arrow) right atrium, (black star) aortic root cannula, (white arrow) suction, (yellow stars) venous cannulas, and (white star) aortic root suction; and **c** removed distal shunt catheter with thrombotic formations

treatment, closure of PFO could be considered prior to a VA shunt implantation in the future.

Conclusion

Thromboembolic events due to a PFO are rare but possible life-threatening complications after VA shunt insertion. Additionally, preoperative cardiac diagnostic in order to exclude PFO might be clinically relevant prior to a VA shunt implantation to avoid such complications. Patients with obligatory VA shunt and manifested PFO, anticoagulation therapy, or closure of PFO prior to a VA shunt could be considered.

Compliance with ethical standards

The informed consent was obtained from the patient.

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