CASE REPORT

Rare sequelae following ventriculoatrial shunt: Case report and review of literature

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ABSTRACT

Ventriculoatrial shunt (VA) is one of the oldest solutions for hydrocephalus. However over subsequent years various complication of VA shunt such as obstructions, malposition, shunt infections, cardiac complications such as endocarditis, traumatic perforation, heart failure, tricuspid regurgitation, intraatrial thrombus, and pulmonary hypertension are reported. Hence, VA shunt procedure has fallen into disrepute. Still VA shunt may be a good option in selected patients with hostile peritoneum. Newer placement strategies and monitoring methods have been put forward to reduce complication following VA shunt. In this case report, we share a rare case of endocarditis with tricuspid regurgitation following a migrated retained calcified shunt tube in the right ventricle of heart 30 years after of VA shunt that was successfully managed.

Key words: Migrated, retained, shunt, ventriculoatrial

Introduction

Ventriculoatrial (VA) shunt is one of the oldest solutions for hydrocephalus. VA shunt approaches may cause potentially life-threatening complications such as pulmonary artery hypertension, cardiac failure and atrial fibrillation.[1] Ben-Ami *et al*.[2] reported gram-positive bacteremia and shunt nephritis in a woman who had a VA shunt for 10 years.

Newer placement strategies have been put forward to reduce complication following VA shunt. Chuang *et al*.[3] reported the use of percutaneous placement with real-time transesophageal echocardiogram monitoring. Endovascular placement was put forward by Gonzalez *et al*.[4] In this case report, we share a rare case of endocarditis with tricuspid regurgitation following a migrated retained calcified shunt tube in the right ventricle of heart 30 years after of VA shunt that was successfully managed.

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Case Report

This is the story of a 41-year-old man. The story starts when the patient was 12 years old (way back in 1983) when he developed hydrocephalus as a sequalae of tuberculous meningitis. He was treated with anti-tuberculous therapy and right VA shunt which was popular at that time. He improved and was discharged.

In 1990, he developed VA shunt dysfunction. Computed tomography (CT) showed hydrocephalus. He was treated with left sided ventriculoperitoneal (VP) shunt. Rt VA shunt was left *in situ*. He improved and discharged.

He was asymptomatic until 2010, then he developed diplopia, ataxia, clubbing and CSF leak through neck wound. Examination revealed a fistulous track posterior to sternomastoid with CSF leaking on the right side. CT scan showed hydrocephalus [Figure 1].

Exploration on right side showed obstruction due to block distal to shunt chamber. The distal end of the proximal ventricular catheter was disconnected from the shunt chamber

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and connected externally to a sterile reservoir to serve as an external ventricular drainage, as we suspected shunt infection. Fistula healed promptly. Exploration was done on the right side to remove the ventricular catheter and revise the shunt. However, we found it was adherent to brain matter, and so the intracranial part was retained *in situ*. Distal end was not addressed due to its right atrial location, and distal end was tethered to the jugular vein. A left VP shunt revision was done. CT scan showed a reduction of ventricular size to normal following revision of left sided VP shunt.

On the postoperative day seven, he developed a high fever that lasted for 2 weeks, with an erythematous rash. Blood picture showed leukocytosis and elevated ESR. Ultrasound of the abdomen showed splenomegaly. CSF culture was sterile. Blood culture revealed enterococci sensitive to Vancomycin. An echocardiogram was done which showed multiple vegetations with shunt tube in right ventricle with tricuspid regurgitation [Figure 2a-c]. Thus, we planned for removal of the distal end of VA shunt through the neck. The incision was made parallel to sternomastoid [Figure 3]. Shunt tube was seen entering through the facial vein going into the internal jugular vein. We tried to remove the shunt tube but were found stuck. Fever persisted. Thus, we planned for sternotomy and removal of shunt after cardiothoracic consultation.

He was taken up for median sternotomy, right atriotomy, and retrieval of the shunt tube under cardiopulmonary bypass.

Intraoperative findings

The right atrium was opened. The entire length of the tube was seen calcified. Shunt tube was seen coiled around the tricuspid valve. The distal tip was seen going through the tricuspid valve and lying in proximity to pulmonary valve [Figure 4]. Tube was removed with difficulty. Shunt tip was sent for culture and sensitivity. Culture revealed enterococci sensitive to vancomycin. Thus, injection vancomycin was started and continued for 28 days. The patient improved clinically. There were no temperature fluctuations. Blood counts normalized. Repeat blood cultures were negative. An echocardiogram was done 6 weeks postoperatively which showed healed vegetations [Figure 5].

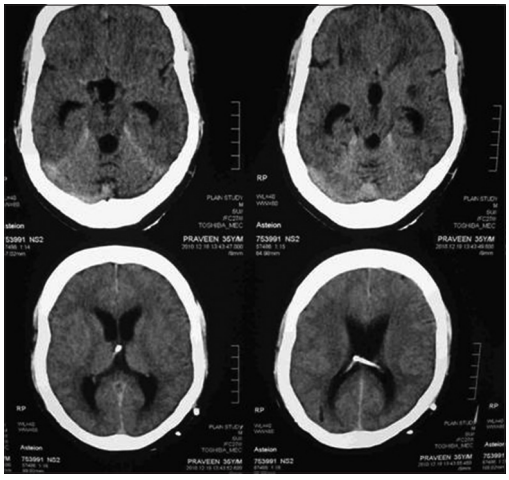

Figure 1: Computerized tomogram showing hydrocephalus following shunt dysfunction in 2010

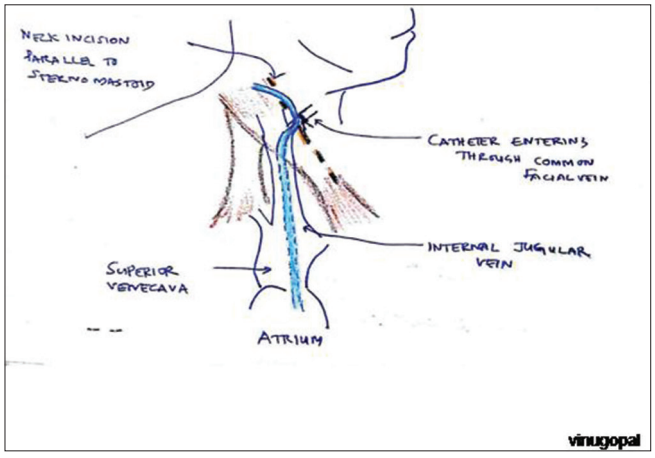

Figure 3: Schematic picture showing position of distal shunt tube

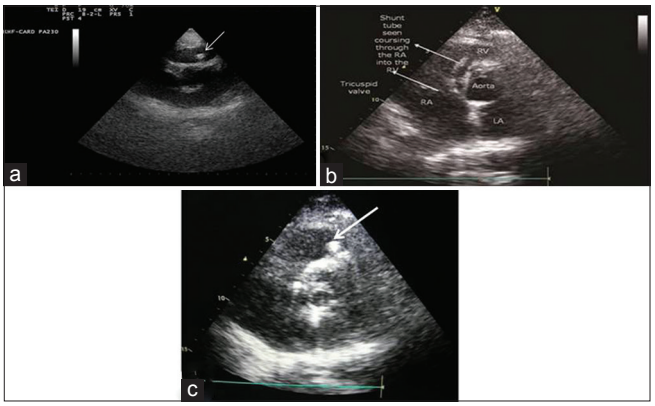

Figure 2: Echocardiogram (a) white arrow showing shunt tube in right atrium (b) shunt tube coursing from right atrium to right ventricle (c) white arrow showing vegetation adjacent to calcified shunt tube

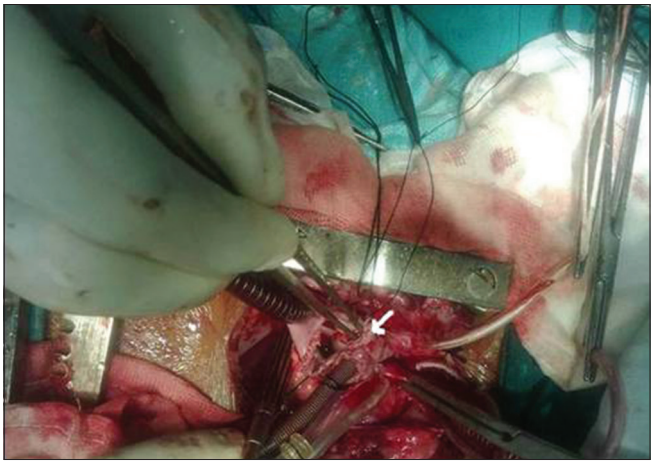

Figure 4: Intraoperative picture showing vegetation on thick tricuspid valve after removal of calcified shunt tube. White arrow showing thickened tricuspid valve

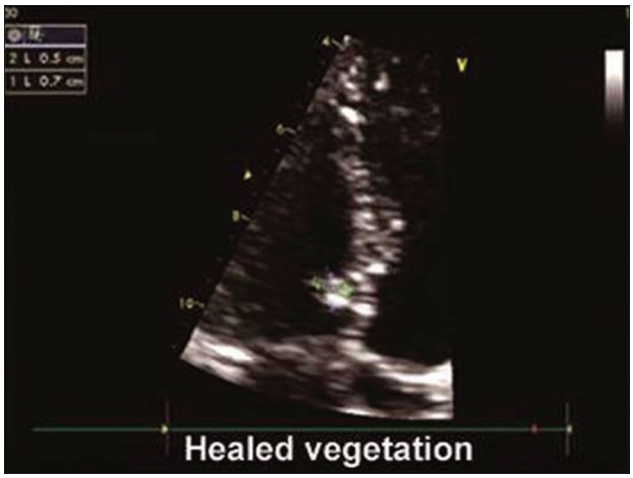

Figure 5: Echocardiogram showing healed vegetation

CT scan was repeated following the procedure, which showed complete resolution of hydrocephalus [Figure 6].

Discussion

VA shunts were used to drain cerebrospinal fluid from cerebral ventricles to the right atrium of the heart. After the introduction of Spitz-Holter valve in 1952, VA shunting became a treatment choice for hydrocephalus, especially in premature infants with necrotizing enterocolitis.[5] However, over subsequent years various complication of VA shunt were recognized. The concept of VP shunt came forward in late sixties with less long-term complications compared with VA shunts.[6,7] Thus, VA shunts became an outdated procedure following the introduction of VP shunt.

VA shunt may be undertaken in selected patients with hostile peritoneum. However, complications like obstruction, bacteremia, cardiac failure, pulmonary hypertension must be borne in mind.[8] Numerous life-threatening cardiac complications such as pulmonary artery hypertension, cardiac failure, and atrial fibrillation following VA shunt placement.[1,9-11] Natarajan and Mazhar[11] reported a case of a 57-year-old male patient who presented with new onset atrial fibrillation following VA shunt. They demonstrated catheter-related right heart complications like calcific tricuspid stenosis and dilated right atrium.[11] Chaw *et al*. reported infective endocarditis in one VA shunt case.[12]

The procedure of VA shunt involves placement of the distal end of shunt tube in right atrium through IJV or its major tributaries. Proper placement is identified by tip position at D6-D7 interspace with x ray. Placement of catheter is demonstrated in Figure 3. Recent advances in interventional neuroradiology and endovascular techniques had reduced the complication rates following VA shunt.[3,4] Metellus *et al*.[13] also reported that percutaneous placement of VA shunt with radiographic guidance improves the effectivity and safety of the technique.

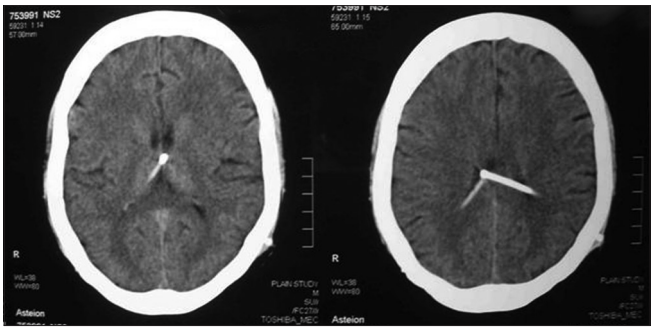

Figure 6: Computerized tomogram done on follow-up showing resolution of hydrocephalus

Conclusion

Even though there is a paucity of literature regarding VA shunt procedures, long-term complication is well documented. Even though, newer placement strategies and monitoring methods have been put forward to reduce complication VA shunt, the procedure has fallen into disrepute. VA shunt may be a good option in selected patients with hostile peritoneum. In this case report, we share a rare case of endocarditis with tricuspid regurgitation following a migrated retained calcified shunt tube in the right ventricle of heart 30 years after of VA shunt that was successfully managed.

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Conflicts of interest

There are no conflicts of interest.

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