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VENTRICULO-PLEURAL SHUNT – AN ALTERNATIVE TREATMENT FOR OBSTRUCTIVE HYDROCEPHALUS

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Introduction. Hydrocephalus represents a major issue in the pathology of neurosurgery. For several decades ventriculo-peritoneal shunt (VP) has been the golden standard treatment, however the multiple associated complications have raised awareness of the necessity for more safe and adjustable procedures. Ventriculo-pleural shunt (VPLS) represents an alternative treatment when the classical VP is not applicable. Literature presents a paucity of research on the outcome and complications of this shunt procedure. The aim of this case presentation is to highlight the efficacy of VPLS as an alternative treatment.

Case presentation. A 30-year-old patient with secondary hydrocephalus and alteration of the neurological status has been admitted to the hospital. His historical medical reports included congenital infectious hydrocephalus at 6 months and multiple attempts of VP placement and ventricular-cardiac drainages. This year he underwent right external ventricular-jugular drainage as both internal jugular veins were ligated and the amount of CSF could no longer support the tubing. After 2 weeks of good functionality, his external drainage was blocked and decompensation of hydrocephalus was demonstrated on the CT. Consequently, the patient underwent VPLS placement under a combined team of neurosurgery and thoracic surgery. Postoperatively, the evolution was good and the control CT shows the placement of the tube in the right pleura with a minimum collection of CSF.

Conclusion. The outcome of this case report suggest the efficacy of this alternative treatment in hydrocephalus. Further prospective studies will be required to support the outcome of this procedure on large cohorts.

Keywords: Obstructive hydrocephalus, Ventriculo-pleural shunt, CT Scan, MRI 3T.

INTRODUCTION

Hydrocephalus has a broad pathogenesis and multiple facets of manifestation. It was defined precisely as an active distension of the ventricular system as a consequence of an inadequate CSF passage occurring at some point from the ventricular system within the brain to its point of absorption.[1] Prevalence of the hydrocephalus varies with the study, yet it is considered to be encountered at 1.1 per 1000 infants.[2] Hydrocephalus aetiology includes a wide plethora of conditions namely infections (rubella, *Toxoplasma gondii*), tumours, aqueduct obstruction and Myelomeningocele.[3–5] Ventriculo-peritoneal shunt (VP) has been in the last decades the golden standard treatment for hydrocephalus, primarily because of the large capacity of the peritoneum for the resorption of the CSF.[6] However, in patient presenting multiple shunt revision, abdominal surgeries or obesity, VP practice has been replaced with other shunt alternatives.

Ventriculo-pleural shunt (VPLS) has been described in 1914 and introduced in the medical practice in 1954 by Ransohoff[7], especially as an alternative to the peritoneal or atrial shunts when thrombosis or infections hinder the normal passage of CSF via the other shunts. Reduced familiarity

associated with pleural and cardiac complications such as effusion, tachycardia or cardiac tamponade with heart failure made this technique to be used as the ultimate treatment of hydrocephalus.[8,9] In this article we present the safety and efficacy of VPLS reported in a patient who have undergone multiple shunt revisions.

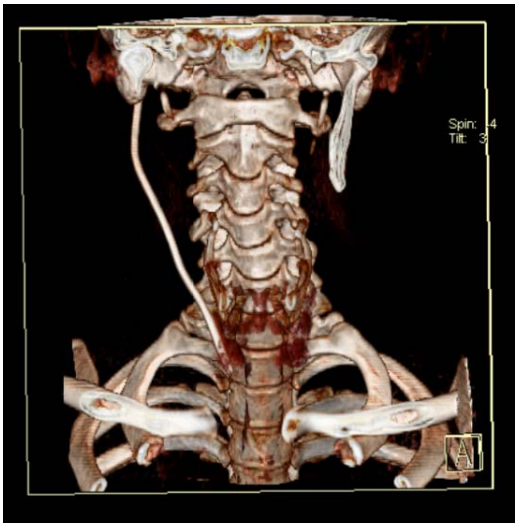

Figure 1. CT 3D: External ventriculo-jugular shunt with low-pressure valve.

CASE PRESENTATION

A-30-year old male patient with secondary hydrocephalus has been admitted to the hospital for neurological status alteration including dizziness, headaches, gait dysfunction and GCS 15. His past medical reports included congenital post-infectious hydrocephalus diagnosed at 6 months followed by multiple VP placement and revisions, ventricular-cardiac drainage and both internal jugular veins ligatured in the past few years.

After admission, paraclinical investigations were performed and MRI 3T at the cranial level demonstrated obstructive hydrocephalus with two non-functional VP, aqueduct stenosis and periventricular demyelination. Abdominal CT Scan showed a voluminous fluid collection at the median and para-median mesentery and the distal catheter within the cavity. Consequently, the first approach was the revision of the shunt and change of the Miethke valve with a Delta low-pressure valve. However, the tubular system didn't function so the entire VP was removed and a right ventriculo-jugular drainage system was used taking in consideration that the internal jugular vein was ligatured – Figure 1.

Intraoperative, shunt placement was verified with C-ARM imaging and the peritoneal cyst was evacuated. Postoperative, patient general state was favourable, afebrile, with improvement of the neurological status confirmed on CT.

At two weeks after surgery, the patient came back to the hospital, presenting a right cervical fluid collection, painless, with no sign of infection. Cerebral CT showed tetraventricular hydrocephalus with small oedema spaces around ventricles – Figure 2 and cervical CT demonstrated right cervical cyst 3,8/2,3 cm – Figure 3.

Figure 2. Cranial CT: Large ventricles demonstrating decompensation of the hydrocephalus with non-functional external ventriculo-jugular shunt.

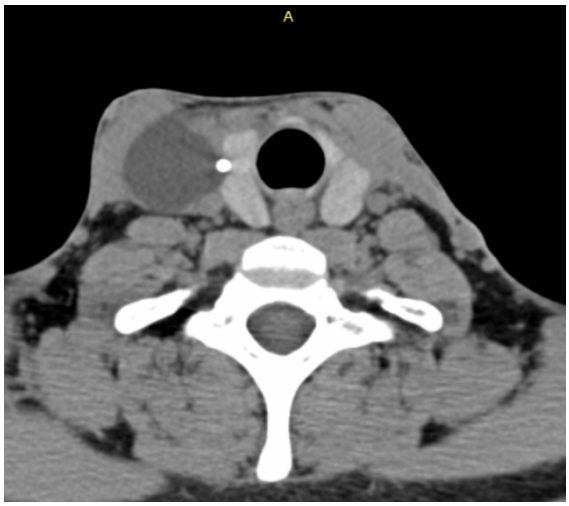

Figure 3. Preoperative CT: right cervical cyst at the insertion of the external shunt.

External jugular shunt was no more functional. The following surgery involved evacuation of the cervical cyst and implementation of ventriculo-pleural shunt on the right side – Figure 4.

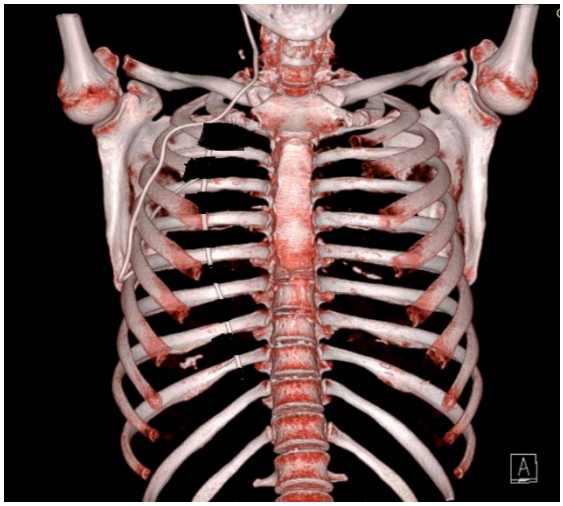

Figure 4. CT 3D: Right ventriculo-pleural shunt with low-pressure valve.

General anaesthesia was induced with the patient in supine position. Incision at the cranial level was similar to any other shunt placement. Specific to the VPLS was the skin incision at the 5th intercostal space at the anterior axillary line with careful stratum-by-stratum dissection of the subcutaneous and muscular layers in order to avoid any injury of the neurovascular tracts.

Parietal pleura was visible and respiratory pulmonary movement were distinguished. With a tunneler the distal catheter was moved under the skin from the cranial level and placed in the pleural cavity and secured in order to avoid any migration. On the postoperative CT, it was demonstrated little right pleural effusion due to CSF accumulation – Figure 5. At 8 weeks postoperative, patient presents neurological status improved and normal gait recovered.

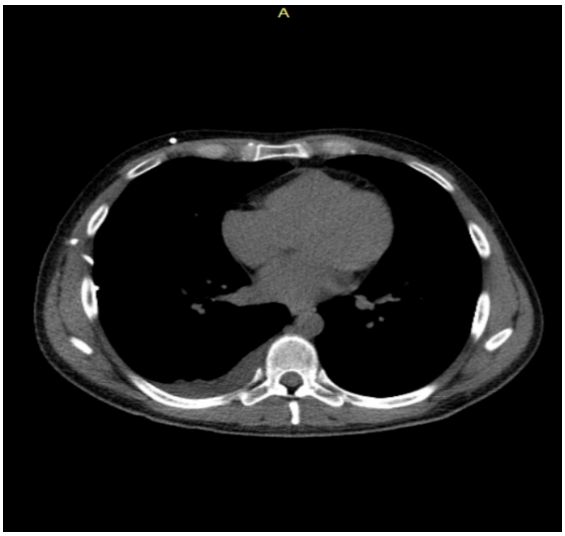

Figure 5. Postoperative thoracic CT: right pleural effusion with shunt placement at the intercostal space.

DISCUSSIONS

Hydrocephalus treatment is broadly divided in shunt treatment and ventriculocisternostomy (VCS). The most common used shunt is VP. More than 75% of the patients have improved their condition after shunting which made this procedure to be the golden standard treatment for hydrocephalus. [10] Reduction of hydrocephalus could be obtained also with ventriculo-atrial shunt (VA) used when VP is no longer functional. However, complications such as infections, atrial thrombosis, endocarditis are disadvantages which make this type of shunt.[11] Functional failure of VP could arise from overdrainage, underdrainage or slit ventricles syndrome that could lead to subdural hematoma and abnormal size of the ventricles[12].

Ventriculo-pleural shunt represents an alternative procedure to the classical VP. Success rate represented as the functionality of the shunt after 1 year after surgery reported in the literature of the past 20 years varies from 43%–70% and pleural effusion varies from 0%–30% [13, 14] depending on the age of the patient. It seems like infants are prone to develop effusion more often than the adults due to the small pleural capacity of CSF absorption. Also, seniors and infants may develop an inflammatory reaction with an increase in the production of pleural liquid leading to excessive accumulation and reduction in the absorptive capacity[15,16.]

The most frequent complication of VPSL is overdrainage leading to pleural effusion The negative pressure within the pleura exerts a "siphon effect" on the shunt leading to an excessive accumulation of CSF, which can be mitigated by the use of anti-siphon devices.[17] Valve system is another factor that lead to an accumulation of CSF. In an adjustable shunt system, the opening pressure can be non-invasive transcutaneous adjusted from high pressure to low pressure. Patient with this programmable shunt system reported less postoperative complications which necessitate shunt revision, less morbidity associated with secondary operations and were supposed to pay less for their care services.[18] Subsequently, pleural effusion is one complication related to shunt malfunction, yet some studies consider it not to be the primary factor leading to the high revisions rate of VPLS.[19,20] We consider that asymptomatic pleural effusion can be treated conservatively, however large pleural effusion is an indication for shunt revisions or replacement.[21] Other complications include migration of VPLS between the ribs and the pleura or trans-diaphragmatic that could happen once the patient

started to grow. Coiling of the catheter could happen if it is too long[22].

An alternative to the shunt placement is VCS which represent an endoscopic fenestration of the third floor ventricle and subarachnoid space. Fenestration can be sharp or blunt with semi-sharp device or with the endoscope itself, with a balloon assisted catheter, by coagulation with monopolar or by laser incision penetration[23]. Indication of this procedure is first of all non-communicating hydrocephalus[1]. Absolute contraindications include communicating hydrocephalus when CSF flows unimpeded as the obstruction is positioned down to the ventricle system and the impossibility to manipulate the endoscope into the ventricles which is usually encountered after chronic shunting when ventricles are reduced in volume[24]. VCS has been associated with lower complications, varying from 4.1–9.3%, comparing to the VP – 23–48% – which has been associated with ventriculitis, shunt abscess, obstruction or migration of the catheter[25–30]. Successful rate varies from study-to-study representing the functionality of the stoma up to several months, even years, usually being around 60–80% and is dependent on age and aetiology[31,32]. Lower success rate have usually been reported in case of infection or haemorrhage (10%) comparing to aqueduct stenosis (100%)[33].

CONCLUSIONS

Ventriculo-pleural shunting represents a safe and efficient procedure in the treatment of hydrocephalus. We reported the case of a patient with multiple intervention for CSF drainage, whose eventually therapy was VPLS. In literature, reduced surgical experience and associated pleural complications made this shunt to be considered as an alternative therapy to the classical VP. Adjustable valve system leads to reduced overdrainage of CSF and less postoperative complications. Other surgical treatment represented by ETV are reliable therapies in hydrocephalus, but indication must be respected. All in all, VPLS surgery should be performed in a multidisciplinary team formed by neurosurgeons and thoracic surgeons.

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Authors declare that there is no conflict of interests.

ABBREVIATIONS

CSF – Cerebrospinal fluid
VP – Ventriculo-peritoneal shunt
VPLS – Ventriculo-pleural shunt
VCS – Ventriculocisternostomy
VA – Ventriculo-atrial shunt
CT – Computer Tomography
MRI – Magnetic Resolution Imaging

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