

Revisiting Retrograde Ventriculosinus Shunt as an Alternative for Treating Hydrocephalus in Children

Revisitando a derivação ventriculosinusal retrógrada como uma alternativa para tratamento de hidrocefalia em crianças

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Abstract

Introduction Retrograde ventriculosinus shunt (RVSS) is a useful option in the daily routine of neurosurgeons dealing with hydrodynamics. The objective of this manuscript is to review the main data about RVSS.

Methods We performed a critical review. The keywords used were *hydrocephalus*, *shunt*, *venous sinus*, *ventriculosinus shunt*, *retrograde ventriculosinus shunt*, and *sagittal sinus*. The search was performed in the Medline (Pubmed) and EMBASE databases.

Results Van Canneyt et al (2008) and Pinto et al (2016) performed experimental studies confirming the effectiveness of RVSS. El Shafei et al (1985, 1987, 2001) authors reported several cases treated with efficacy and few complications. Oliveira et al (2015, 2016) compared RVSS and ventriculoperitoneal shunt (VPS) in the treatment of hydrocephalus after myelomeningocele repair in infants, with similar functional results. Oliveira et al (2015, 2016) also described the applicability of RVSS in situations when VPS is not feasible, or when the peritoneum is not useful.

Discussion Retrograde ventriculosinus shunt is a safe and more physiological option, which requires the use of less prosthetic material. It is feasible and applicable. Especially in children, it generates a normotensive state after shunting, allowing centrifugal head growth, once there is no intracranial hypotension due to overdrainage, which may reflect in long-term better psychomotor development.

Conclusions The surgical technique of RVSS is feasible. The clinical results are comparable with those of the VPS.

Keywords

- hydrocephalus
- ventriculosinus shunt
- treatment
- review

Resumo

Introdução A derivação ventriculosinusal retrógrada (DVSR) deve ser uma opção na rotina de neurocirurgias que trabalham com hidrodinâmica. O objetivo deste estudo é revisar os principais dados sobre DVSR.

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Métodos Realizamos uma revisão crítica. As palavras-chave usadas foram *hydrocephalus shunt*, *venous sinus*, *ventriculosinus shunt*, *retrograde ventriculosinus shunt*, e *sagittal sinus*. A busca foi feita nas bases de dados Medline (Pubmed) e EMBASE.

Resultados Van Canneyt et al (2008) e Pinto et al (2016) realizaram estudos experimentais confirmando a eficiência do DVSR. El Shafei et al (1985, 1987, 2001) relataram vários casos tratados com eficiência e poucas complicações. Oliveira et al (2015, 2016) compararam DVSR e derivação ventriculoperitoneal (DVP) no tratamento de hidrocefalia após reparo de mielomeningocele em crianças, obtendo resultados funcionais similares. Oliveira et al (2015, 2016) também descreveram a aplicabilidade do DVSR em situações em que o DVP não é possível, ou o peritônio é hostil.

Discussão A DVSR é uma opção segura e mais fisiológica, que requer menos material protético; é factível e aplicável. Particularmente em crianças, gera um estado normotenso após o *shunt*, permitindo crescimento cefálico centrífugo, uma vez que não há mais hipotensão intracraniana devido à hiperdrenagem, o que pode refletir em melhor desenvolvimento psicomotor em longo prazo.

Conclusões A técnica cirúrgica DVSR é viável. Os resultados clínicos se são comparáveis à DVP.

Palavras-chave

- hidrocefalia
- *shunt* ventrículo-sinusal
- tratamento
- revisão

Introduction

Treating hydrocephalus has always been challenging, especially in children. One century ago, there were no feasible options to treat hydrocephalus, and the natural history of the disease was death by intracranial hypertension or progressive enlargement of the cranial vault in infants.¹⁻⁷

The Twentieth Century witnessed a dramatic change in the comprehension of the disease, the surgical apparatus and the development of materials to enable shunting of the cerebrospinal fluid (CSF) from the ventricles to other sterile body cavities. From neuroendoscopy to shunt valve technologies, we could follow significant changes in the paradigms of the treatment of hydrocephalus.^{2,8-27}

However, since the 1990s, a few improvements changed the learning curves and the outcome of the hydrocephalus treatment. Currently, ventriculoperitoneal shunt (VPS) is the mainstay for treatment, as well as endoscopic third ventriculostomy (ETV), which is advocated in specific cases. Over the past decades, shunt revision rates became stable, reaching over 70% of cases in a lifetime. Endoscopic third ventriculostomy also has known limitations, especially in patients with myelomeningocele and Chiari type II.^{5-7,28}

Ventriculosinus shunt (VSS), specifically when performed in a direction opposite to the blood flow, as described by Ismail El Shafei in the 1980's, is a useful option in the daily routine of neurosurgeons dealing with hydrodynamics. Although recent experiences with VSS are scarce, we believe they should be discussed once more and linked to the classic studies by El Shafei.^{6,7,29-31}

The objective of this manuscript is to discuss new clinical data about retrograde ventriculosinus shunt (RVSS) and link them to the pertinent literature.

Methods

We performed a critical review to address the application, advantages and disadvantages of RVSS to treat hydrocephalus, especially in children. The keywords used were *hydrocephalus*, *shunt*, *venous sinus*, *ventriculosinus shunt*, *retrograde ventriculosinus shunt*, and *sagittal sinus*. The search was performed in the Medline (Pubmed) and EMBASE databases.

All manuscripts were included. A review of the retrieved references was also performed to find crossed references. Any references known by the authors were also included.

Results

Since Toma et al⁵ published a full review about VSS in 2010, only 5 manuscripts updated the subject, 1 by El-Shafei et al, and 4 by Oliveira et al and Pinto et al. In the present article, we discuss the evolution of the technique and its current results.^{6,8,10,22,25,28,32-34}

Review

Gartner, in 1896, suggested that the most physiological way to treat hydrocephalus would be a connection of the ventricles with the venous blood system of the head and neck. Portnoy proposed that the perfect system to treat hydrocephalus would be the one that allowed for CSF drainage, could keep intracranial pressure (ICP) normal, and would not cause complications.^{3,5,14,20,26,27}

Ventriculoperitoneal shunt (VPS) is the preferred option to treat hydrocephalus, even though it has a revision rate of more than 70% of patients throughout their lifetime, and of 30% of patients in the first year. An important complication is the siphoning phenomenon, which leads to over drainage and subdural effusions.^{35,36}

Siphoning leads to over drainage of the intracranial CSF into the distal site when the patient with VPS is in the orthostatic position. To solve this problem, anti-siphoning devices (ASDs) were added to the VPS line, increasing the pressure of the drainage system as the patient raises the head, thus decreasing the drainage of the CSF. Some of them include gravitational devices, which change the pressure of the whole liquid column according to a range of head positions, allowing different grades of additional pressure. However, such solutions did not eliminate the hydrodynamic phenomenon of siphoning, and significantly increased the cost of the prosthesis.^{8-18,29,32-44}

In the beginning of the XXI century, Aschoff stated that there were more than 300 shunt systems available; however, none could reach what was proposed by Gartner and Portnoy. Several sites to perform the shunt were also tried, from the gallbladder to the bladder, but the peritoneal cavity was the site in which the procedure reached higher accuracy and effectiveness.⁵

In the 1940's, Ingraham induced hydrocephalus in dogs and performed treatment with an anterograde ventriculo-jugular shunt; however, the distal end of the catheter obstructed due to siphoning. When positioning the catheter against the blood flow, the catheter flow would be laminar, and there would be no turbulence zone, which would prevent blood clotting and catheter obstruction.¹⁴

This hemodynamic principle was demonstrated in the studies by El Shafei and El-Rifaii. Another principle that was also demonstrated by these authors was the fact that the internal jugular vein, which collapses in the orthostatic position, has the capacity to work as a physiological anti-siphoning system. Thus, to use this natural property, any ventriculo-venous shunt catheter should have the distal extremity located proximally to the internal jugular vein, in the opposite direction of the blood stream. Thus, there is an improvement in hemodynamics with the removal of the regions of turbulence, and the avoidance of back flow and thrombosis (►Fig. 1).^{8,29,32-34,40-44}

RVSS Technique and Variations

El-Shafei et al pioneered the technique by performing a puncture in the retrograde direction of the blood flow in the superior sagittal sinus (SSS) using the pressure impact of the blood flow in the dural sinuses to maintain the CSF pressure greater than the dural sinus pressure, regardless of changes in posture or intrathoracic pressure (►Fig. 2). These authors modified their technique later by adding a valve of very low pressure, recognizing that, in the event of a reduction in intracranial pressure (ICP) due to lumbar puncture or sudden change of heart output, some retrograde flow of venous blood to the distal catheter could hinder the shunt. They also broke new ground by using transcranial Doppler (TCD) data to indirectly evaluate cerebral hydrodynamics through the cerebral blood flow velocities.^{8,29,32-34,40-44}

The surgical technique as described by El-Shafei et al involves two cranial burr holes in the same arcuate incision in the scalp on the parietal region. One burr hole is in the posterior parietal bone (Frazier point), and the other, in the middle third of the sagittal sinus. A small opening is made in the dura mater of the parietal incision, the lateral ventricle is

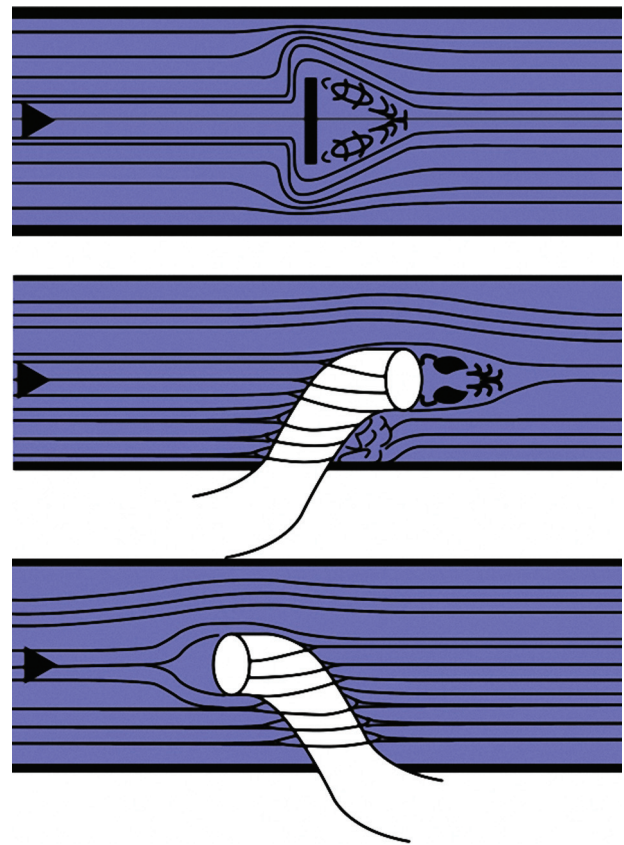


Fig. 1 In the upstream region of the obstacle, there will be an impact zone, whereas in the downstream region, there will be a weak zone.

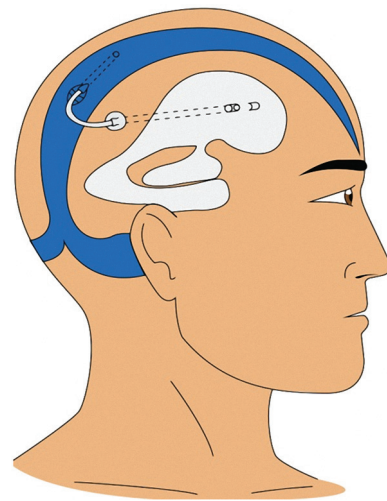


Fig. 2 RVSS design.

punctured, and then, a small opening is made in the sagittal sinus, and the catheter is inserted ~ 2 cm against the direction of the blood flow (►Fig. 3). El-Shafei et al published the results with the use of a single catheter and with the association of a very low pressure gradient valve (just to add another mechanism to avoid back flow).^{8,29,32-34,40-44}

In the small but increasing experience of our group, we have adopted the classic technique by El-Shafei et al with a valveless

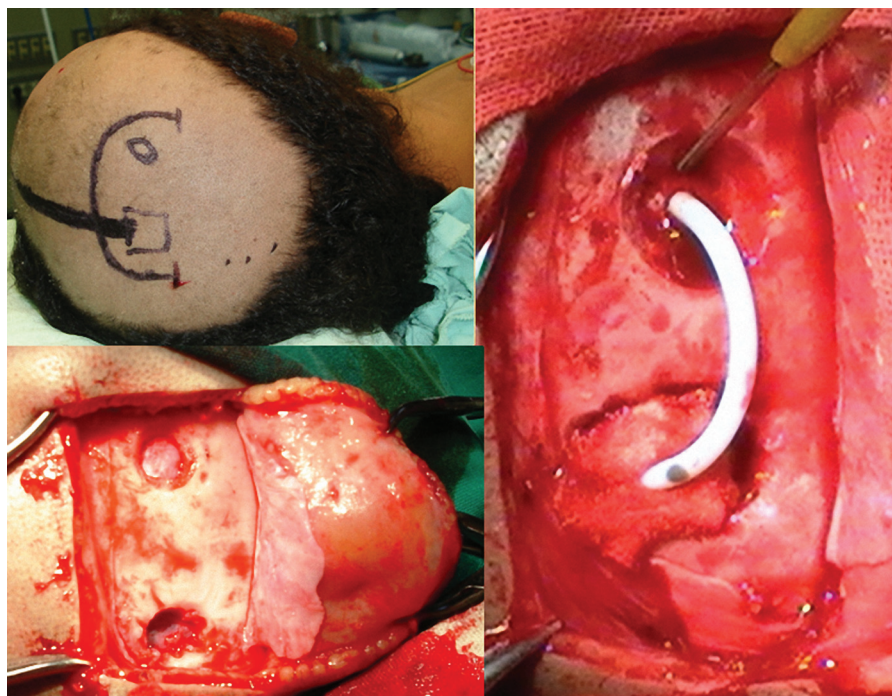


Fig. 3 Phases of the surgical technique. Positioning and arcuate incision. Opening with medial and lateral burr holes. Catheterization of lateral ventricle and superior sagittal sinus. Hemostasis. Closure with pericranium and skin closure.

catheter. However, in adult patients, we have changed landmarks for ventricular and superior sagittal sinus (SSS) puncture. We used a frontal incision to enable the puncture in Kocher's point, and another incision over the SSS to allow for 1 burr hole in the middle third of the sagittal sinus, ~ 3 cm behind the bregma. The lateral ventricle is punctured, and then a small opening is made in the SSS, and the catheter is inserted ~ 2 cm against the direction of the blood flow. The catheter used to perform the shunt is, by convenience, the PS Medical valve (Medtronic, Dublin, Ireland) ventricular catheter, which is already in routine use at our institution. The ventricular

catheter is the only one applied without the use of a valve system (► **Fig. 4**).^{10,21,22}

Another potential option designed by our group, but with limited application until now, is the fragmentation of the catheter in two parts: one catheter for the ventricular puncture, and another for the SSS puncture. Then, the margin of one catheter has a connector to allow the connection of both catheters. The last variation with no clinical applications described in the literature is a combination of two catheters of different diameters connected as described before (► **Fig. 5**). Using this rationale, it would be possible

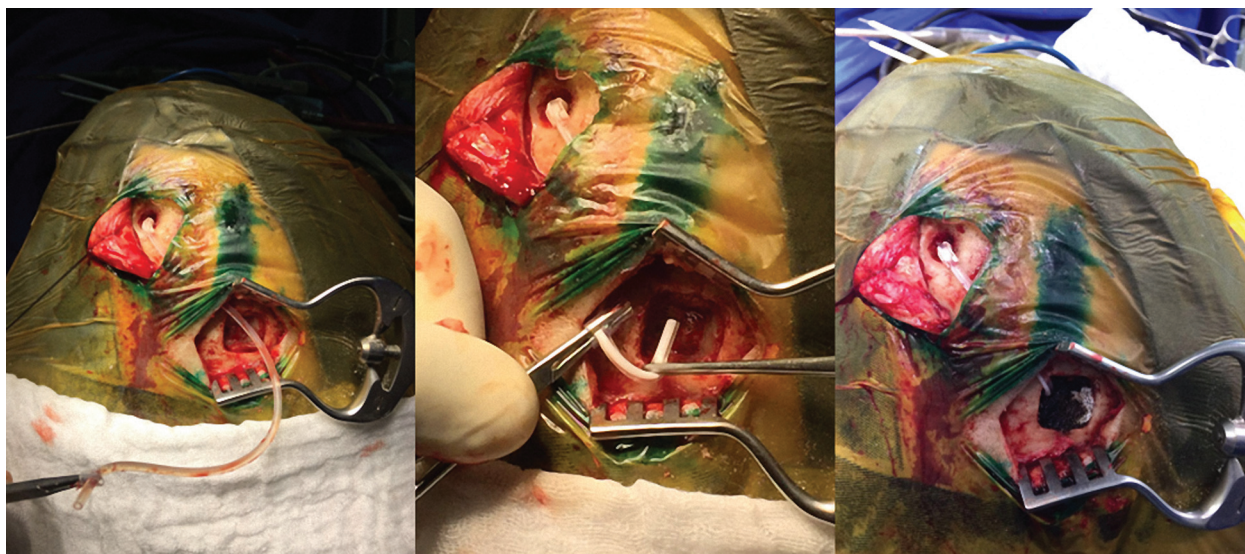


Fig. 4 Retrograde ventriculosinus shunt after failed ventriculoperitoneal shunt. Left ventricular puncture followed by burr hole over the SSS and puncture.

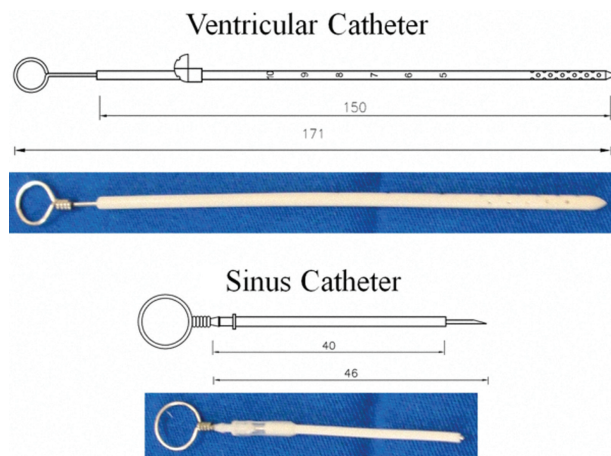


Fig. 5 Option for retrograde ventriculosinus shunt catheter, with two catheters of different diameters connected.

to increase or decrease flow resistance and, therefore, the risk of back flow. However, these ideas should be further discussed in experimental and clinical bases.^{10,21,22}

Experimental Basis for RVSS

Van Canneyt et al performed an experimental study to analyze RVSS. Firstly, an experimental model of the cerebral ventricles, the arachnoid villi, the cortical veins, and the SSS was built. Secondly, a numerical model of the cortical veins and the SSS was built. No over drainage was found in the antegrade or the retrograde positions of the shunt. Blood reflow was only found while mimicking lumbar puncture or changes in position with the experimental model (lowering the intracranial pressure or increasing the sinus pressure rapidly). These authors suggested that the main advantages of this method include short length of derivation and prevention of siphoning. After an experimental analysis, they concluded that when the catheter is placed in an antegrade manner, the pressure at the catheter tip is 3.3 Pa, which is lower than that of the SSS. In the backward position, the catheter tip pressure was 16.7 Pa, serving as a protection against overdrainage.^{6,12–15,25,26}

Pinto et al proposed and designed an animal experimental model for RVSS using mongrel dogs and inducing hydrocephalus with kaolin. They stated that the RVSS technique was feasible and even simple, not requiring a large learning curve.²⁵

In the dogs with the RVSS model, they could successfully induce hydrocephalus and its clinical features. The surgical procedure finished with no remarks, however they failed to predict that the external diameter of the catheter used (2.5 mm) would prevent the normal blood flow through the SSS, which also has a 2.5 mm caliber, resulting in cerebral venous infarction due to venous blood stasis and drainage blocking of the hypertensive CSF accumulated in the ventricles (–Fig. 6). Thus, they proposed changing the external diameter of the catheter to adapt to canine anatomy and proportions.²⁵

Clinical Basis for RVSS

El Shafei is surely the most important name in the experimental and clinical development of RVSSs. The first article he published (along with another author) in 2001 included 56

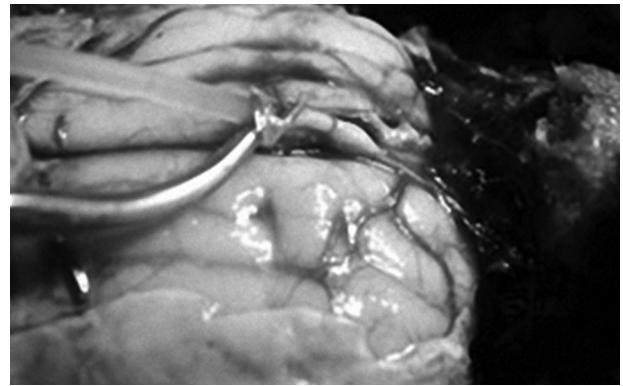


Fig. 6 Catheterization of the superior sagittal sinus (SSS) in a canine model of retrograde ventriculosinus shunt. The catheter had almost the same diameter as the SSS, so there was venous blockage and infarction.

patients and was followed by a second article with other authors in 2005, which included 54 patients, producing a total of 110 patients reviewed retrospectively; 99 patients had valveless catheters, and 11 had catheters with low pressure valves.^{8–13,29,32–34,40–44}

In 2010, they published the last experience. During the past 40 years, 229 RVSSs were implanted; 219 patients (95.63%) benefited from the shunt, and only 2 of them needed a single shunt revision. The manifestations of increased intracranial pressure disappeared, different degrees of regression of the ventriculomegaly occurred in patients with closed craniums, but not in infants and young children with open craniums, and there were no problems related to improper CSF drainage or venous thrombosis. The follow-up period is of up to 40 years (mean 9 years 8 months).^{8–13,29,32–34,40–44}

El-Shafei showed that, in children, the clinical improvement occurred immediately after shunt insertion with normotensive fontanelle and disappearance of the hairy scalp engorged veins, with gradual relief of eye signals. The psychomotor development of the patients was satisfactory.^{8–12,29,32–34,40–44}

Paul C. Sharkey, in 1965, published his experience with 4 pediatric patients. Fourteen years later, in 1979, Cecil J. Hash published a series of 36 procedures. And Wen reported 15 cases of obstructive hydrocephalus in 1965, and then changed his technique in 52 patients in the pediatric age group, and published the results in 1982.⁵

Børgesen, Gjerris and Agerlin also applied the technique. The first series was published in 2001. The second article was published in 2004 with two extra authors, and included the follow-up results of 45 patients from Denmark, Italy and Norway.^{1,5,45}

Most series treated all kinds of hydrocephalus in children and adults of all age groups. However, cases of spina bifida were excluded. All authors reported clinical and radiological improvement. The clinical findings, such as headache, nausea, vomiting and papilledema changed after the surgery.⁵

The follow-up period and shunt revisions were variable. The revision rate varied between 11.8% and 37.5%. Most patients had a shorter follow-up period. In healthy young children, brain growth causes an increase in the thickness of the cerebral



Fig. 7 Post-operative aspect of a surgically repaired myelomeningocele.

mantle and head circumference, while the ventricular size remains stable, that is, in normal situations, the strength of the brain wraps is lower than the CSF pressure, allowing centrifugal growth.¹

The last review on the subject showed that among 265 patients, there were no reports of sinus thrombosis, air embolism, uncontrollable operative bleeding or nephritis associated with the shunt.⁵

RVSS versus VPS in Hydrocephalus after Myelomeningocele Repair

Myelomeningocele is a neural tube defect in neurulation embryonic stage by the third week of life. Its incidence may

reach 1–2 per 1,000 live births. Even with all advances in intrauterine and postnatal management, up to 90% of these patients may present hydrocephalus in different degrees, requiring treatment (► **Fig. 7**). Ventriculoperitoneal shunt and ETV are classic options, but revisions rates are high in VPS; ETV still has a limited success in cases of Chiari II, especially in younger infants.^{7,16,23,46}

Oliveira et al compared RVSS and VPS in a randomized trial with preliminary results. A total of 9 patients were included in the study for a period of 2 years. Four patients were allocated to the control group (VPS), and 5 patients to the RVSS group. Transcranial Doppler was used to analyze indirect hydrodynamic patterns, as reported previously by El-Shafei.^{27,47,48}

The follow-up included evaluations of the cephalic perimeter (CP), bregmatic fontanelle, TCD, computed tomography, neuropsychomotor development, complications and shunt revision, and outcomes focused on patients and caregivers.²²

The cephalic perimeter, the tomography and the fontanelle were characteristic in each group. In the RVSS group, there was no abrupt reduction in head circumference after surgery. The CP assumed a physiologic contour, but remained high. In control group, the CP reduced abruptly after surgery, with subsequent regrowth. Similarly, the fontanelle in the RVSS group remained normotensive, meaning cranial normotension. In the control group, they became normotensive or hypotensive, especially in older children, and could imply a siphoning component in older children with a more upright posture (► **Figs. 8 and 9**).²²

The TCD showed significant improvements in cerebral hemodynamics parameters after treatment in both groups,

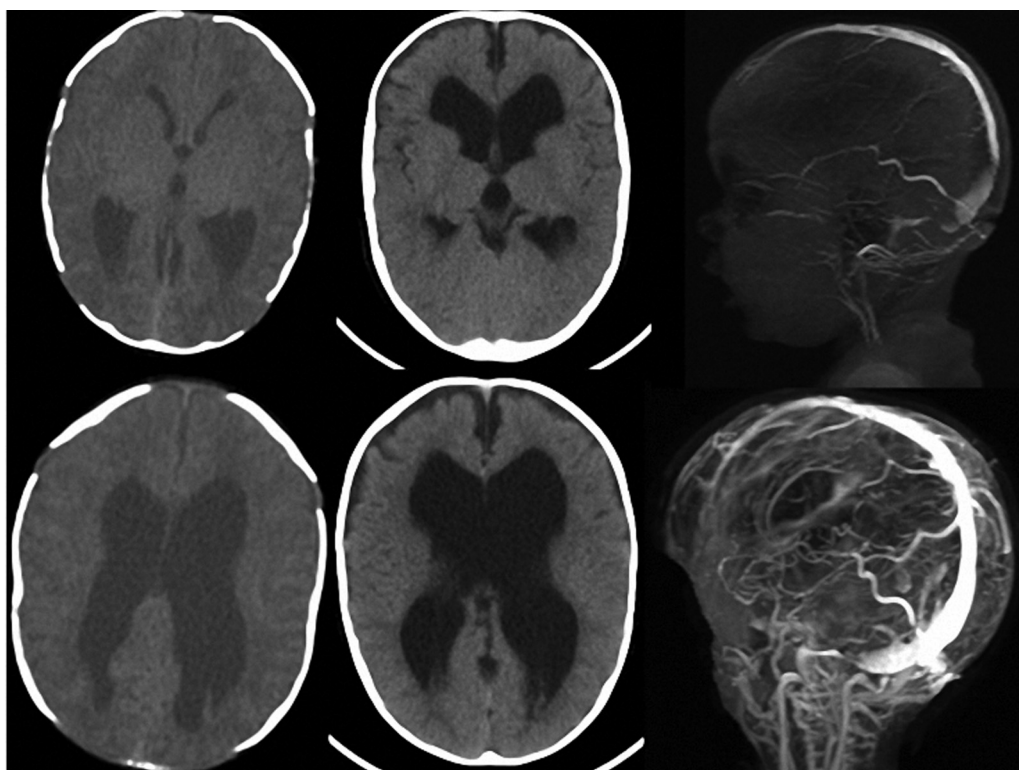


Fig. 8 Pre-operative evaluation before retrograde ventriculosinus shunt. Normal ventricular size at the age in months. Hydrocephalus at the age of 6 months. Normal sinus anatomy.

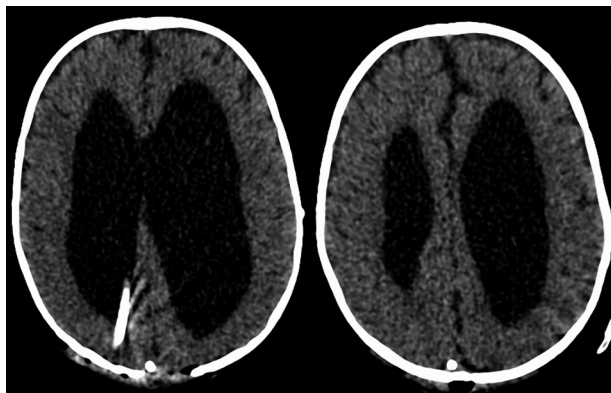


Fig. 9 Postoperative retrograde ventriculosinus shunt evaluation. There is no clear decrease in ventricular size.

with decreased velocity of intracranial vessels and a decrease in the pulsatility and resistance indexes (→ **Figs. 10** and **11**). There were no differences between VPS and RVSS. The TCD associated with clinical and physical examinations proved to be a safe and appropriate tool to assess dysfunction of RVSS.^{6,7,22,30,31}

In the RVSS group, there was need for one revision and one conversion to VPS. In VPS group there was no revision or complications. Psychomotor development after surgery was similar in both groups. However, further evaluation is needed with specific neuropsychological tests which can detect subtle changes.²²

RVSS and Failed VPS

Peritoneal cavity alterations are one of the most frequent causes of VPS failure, due to the formation of primary or secondary adhesions, fibrosis, ascites, cysts, pseudocysts and peritonitis.¹⁰ Although there are other second options, like ventriculopleural and ventriculoatrial, they may be useful, but they are potentially harmful.¹⁰

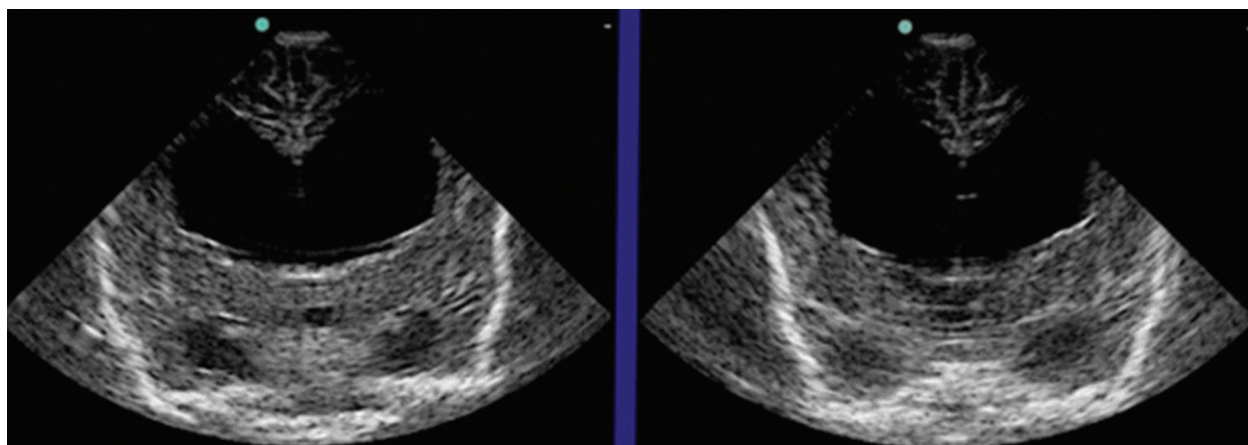


Fig. 10 Duplex evaluating ventricular size. There is no clear ventricular size decrease before (left) and after (right) retrograde ventriculosinus shunt.

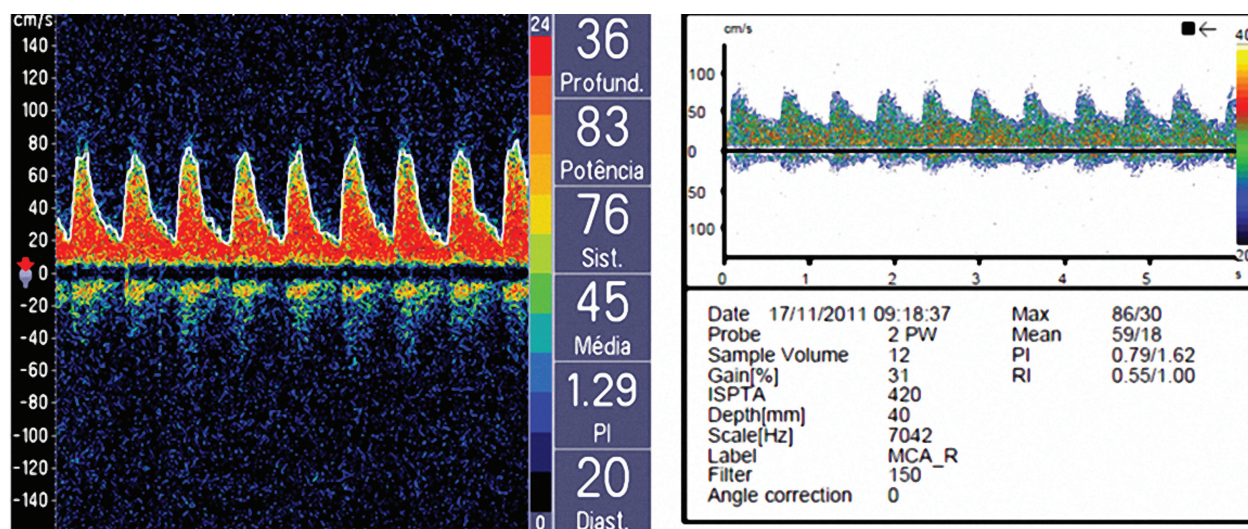


Fig. 11 Transcranial Doppler (TCD) of a retrograde ventriculosinus shunt (RVSS) patient. Pre-operative and postoperative TCD. Postoperative mean velocities in middle cerebral artery (MCA) decreased after RVSS, and there was a decrease in pulsatility index (PI).

Oliveira et al described three cases of RVSS applied in situations when VPS is not feasible, or the peritoneum is not useful. In one case, there was refractory ascites; in another, a peritonitis, and in the last, pseudocyst formation. In all three cases, the patients were adults and tolerated RVSS to treat the underlying hydrocephalus. One patient needed revision surgery to reposition the catheter, which was not inside the SSS. Although applied in adults, this experience also sheds light over the potential use of RVSS in children with a hostile peritoneum.¹⁰

Discussion

Retrograde ventriculosinus shunt is a safe and more physiological option that requires the use of less prosthetic material. It is feasible and applicable, as long as there is pervious SSS to allow drainage of excessive CSF. It uses a natural anti-siphon device (internal jugular vein), and enables CSF drainage inside the venous system. Manipulation of the SSS is safer than usually believed, and one important matter is that the dural opening for the ventricular puncture should be just enough to catheterize the ventricle and avoid creating CSF leakage.^{8-24,29,32-34,40-44}

The main advantages are the use of less prosthetic material, the safety and efficacy, respecting hydrodynamics and avoiding over drainage. The main disadvantages are: the necessary learning curve and the possibility of retrograde flow if a lumbar puncture is performed (which can be solved if a low-pressure valve is added to the shunt system). Complications like dysfunction and infection are not common, and overdrainage is an unlikely event.^{8,29,32-34,40-44}

In RVSS, although there is no immediate and marked ventricular size decrease (which is common in VPS), there is stable ventricular size with improved hemodynamics and hydrodynamics revealed by a decrease in the pulsatility index (PI) after surgery, which is maintained in the late follow-up. In patients with increased PI there was also clinical impairment, which was indicative of the need of a revision surgery.^{8,29,32-34,40-44,49}

Especially in children, RVSS generates a normotensive state after shunting, allowing centrifugal head growth, once there is no intracranial hypotension due to over drainage, which may reflect in long-term better psychomotor development. Additionally, there is no need to operate the abdomen, avoiding complications of disabsortive peritoneum.^{12-24,49}

In our sample, TCD was used successfully for diagnostic and follow-up evaluation of hemodynamics and hydrodynamics in the pre- and postoperative phases of RVSS. It was technically feasible in all patients, correlated closely with other clinical and imaging parameters, and was sensitive for the identification of system malfunction.⁴⁹

Conclusions

The surgical technique of RVSS is feasible. The clinical results are comparable with those of VPS. To date, we believe it should be performed especially after myelomeningocele

repair in young infants, and as an alternative to failed VPS after multiple revisions.

Conflicts of Interest

The authors have no conflicts of interest to report.

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