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ARTICLE

Surgical technique of retrograde ventricle-sinus shunt is an option for the treatment of hydrocephalus in infants after surgical repair of myelomeningocele

A derivação ventrículo-sinusal retrógrada é uma opção para o tratamento de hidrocefalia em lactentes após correção de mielomeningocele

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ABSTRACT

Introduction: Treatment of hydrocephalus is accomplished primarily through a ventricular-peritoneal shunt (VPS). This study aims to describe the application of retrograde ventricle-sinus shunt (RVSS) in patients with hydrocephalus after surgical treatment of myelomeningocele. **Method:** A prospective, randomized and controlled pilot study. We consecutively enrolled 9 patients with hydrocephalus after surgical repair of myelomeningocele from January 2010 to January 2012. These patients underwent elective RVSS or VPS. Five underwent RVSS and 4 underwent VPS. Patients were followed for one year with quarterly evaluations and application of transcranial Doppler. **Results:** RVSS group showed outcomes similar to those of VPS group. Doppler revealed significant improvement when comparing preoperative to postoperative period. RVSS group had significantly higher cephalic perimeter than VPS group. Neuropsychomotor development, complications and subjective outcomes did not differ between groups. **Conclusion:** RVSS shunt is viable; it is an alternative option for the treatment of hydrocephalus.

Keywords: neurosurgery, hydrocephalus, shunt, myelomeningocele.

RESUMO

O tratamento da hidrocefalia é realizado principalmente através de uma derivação ventrículo-peritoneal (DVP). Nosso objetivo é descrever a aplicação da derivação ventrículo-sinusal retrógrada (DVSR) em pacientes com hidrocefalia após o tratamento cirúrgico de mielomeningocele. **Método:** Estudo prospectivo, randomizado e controlado. Selecionados consecutivamente 9 pacientes com hidrocefalia após correção cirúrgica de mielomeningocele de janeiro de 2010 a janeiro de 2012. Eles foram submetidos à DVSR ou DVP. Cinco foram submetidos à DVSR e 4 à DVP. Foram seguidos por 1 ano com realização trimestral de avaliações e aplicação do Doppler transcraniano. **Resultados:** O grupo DVSR apresentou desfechos semelhantes ao grupo DVP. O Doppler mostrou melhora significativa quando comparado o pré-operatório com o pós-operatório. O grupo DVSR apresentou perímetro cefálico significativamente maior que o grupo DVP. O desenvolvimento neuropsicomotor e complicações não diferiram entre os grupos. **Conclusão:** A derivação ventrículo-sinusal retrógrada é viável; ela é uma opção para o tratamento de hidrocefalia.

Palavras-chave: neurocirurgia, derivações liquóricas, mielomeningocele, hidrocefalia.

Myelomeningocele is a neural tube defect in neurulation embrionary stage by the third week of life. Even in locations with high living standard and supplementation of folic acid, its incidence may reach 1-2 per 1000 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[1,2,3].

Treatment consists in cerebrospinal fluid (CSF) shunts with ventricular endoscopy or ventricular catheter and drainage to another body cavity. In the case of hydrocephalus

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secondary to myelomeningocele, both methods have considerable complications and limited therapeutic success[4,5,6,7].

Ventricle-peritoneal shunt (VPS) is the preferred option, although has a revision rate of more than 70% of patients in life and until 30% in the first year. An important complication is shunt infection, which may occur in up to 5% of the cases. Another drawback is siphoning phenomenon, leading to overdrainage and subdural effusions[8,9,10,11].

Neuroendoscopy, on the other hand, has increased in experience and indications. Endoscopic third ventriculostomy (ETV), which consists in fenestrating third ventricle and communicating with interpeduncular and pre-pontine cisterns, is the most applied ventricular endoscopic procedure. However, in patients with myelomeningocele, ventricular anatomy is notoriously variable, with patterns that hinder endoscopy and increase operative risks[8,9,10,11].

Thus, alternatives are needed to treat hydrocephalus in these patients efficiently and safely. Retrograde-ventricular sinus shunt (RVSS), developed by El-Shafei, shunts lateral ventricle to superior sagittal sinus (SSS) in the opposite direction of venous blood flow and is a feasible and rational alternative for the treatment of hydrocephalus, minimizing siphon effect[12,13,14,15,16,17,18,19,20,21,22,23,24,25,26].

This study aims to describe initial clinical results of RVSS in hydrocephalus after surgical repair of myelomeningocele.

METHOD

Study design

It is an intervention, prospective, randomized and controlled study. The allocation was parallel with ratio of 2: 2.

Eligibility criteria

We consecutively enrolled patients diagnosed with hydrocephalus after surgical repair of myelomeningocele from January 2010 to January 2012, users of Hospital das Clinicas, University of São Paulo. They were screened after postnatal surgical treatment for myelomeningocele. These patients underwent elective RVSS or VPS.

Inclusion criteria

Inclusion criteria were acceptance from parents to participate in research; myelomeningocele patients in the postoperative period, without infectious complications and without prior ventricular shunts who developed progression of hydrocephalus after first month of life, but under 6 months old. Infants performed magnetic resonance imaging (MR) with vascular study showing patency of SSS.

Exclusion criteria

Exclusion criteria: other types of congenital hydrocephalus, previous infectious complications or previous ventricular shunts, refusal of parents and guardians to participate in research; MR with SSS thrombosis signals.

Ethical adherence

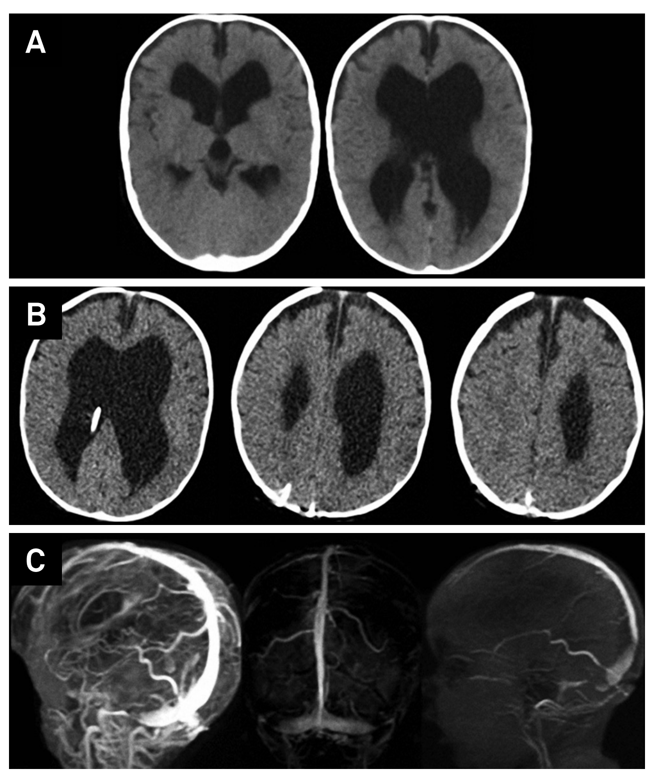
Project and informed consent form were approved without restrictions by the Ethics Committee in Research of HCFMUSP, under number 0178/09.

RVSS technique (Figures 1 and 2)

Surgical technique involves 2 cranial burr holes in the same arcuate incision in the scalp of parietal region. One burr hole in posterior parietal bone (Frazier point) and the other in the middle third of the sagittal suture. Small opening is made in the dura mater of the parietal incision and the lateral ventricle is punctured and then small opening is made in sagittal sinus and the catheter is inserted approximately 2 cm against the direction of blood flow. Catheter used to perform shunt was the one in PS Medical valve® (Medtronic) material, which is already in routine use in Hospital das Clínicas[12,13,14,15,16,17,18,19,20,21,22,23,24,25,26].

Sample (Table 1)

Nine patients were included in the study for a period of two years. 3 were allocated to the control group (VPS) and 6 patients in RVSS group. Allocation was made by randomization in the operating room. 5 of the 6 patients allocated to RVSS underwent surgical treatment. Patient 6 had surgery



CT: computed tomography; MR: magnetic resonance; SSS: superior sagital sinus.

Figure1. Image study. (A) Pre operative tomography revealing ventriculomegaly. (B) CT after RVSS. (C) MR revealing pervious SSS.

converted to VPS due to intraoperative inaccurate identification of SSS and bleeding. 3 patients initially allocated to VPS group underwent surgery without complications.

VPS group was then composed of four patients, three males (75%) and 1 female (25%). Average age was 3 months. All patients had lumbosacral myelomeningocele. Two of the 3 patients (66%) had club foot. The degree of strength in the lower limbs ranged from 2 to 5. CP ranged from 40 to 45 cm. Bregmatic fontanelle was hypertense, with 2+/4+ in 3 patients and 3+/4+ in 1 patient. Neuropsychomotor development (NPMD) was adequate for the age group in 3 patients and delayed in 1. Evans index ranged from 0.47 to 0.50.

RVSS group consisted of 5 patients, 4 male (80%) and 1 female (20%). Average age was 5 months. All patients had lumbosacral myelomeningocele. 3 of 5 patients (60%) had club foot. The degree of strength in the lower limbs ranged from 0 to 5. Cephalic perimeter (CP) ranged from 44 to 47 cm. Bregmatic fontanelle was hypertensive, with 2+/4+ in 4 patients and 3+/4+ in 1 patient. NPMD was adequate for the age group in 3 patients and delayed in 2. Evans index ranged from 0.46 to 0.50.

Follow-up

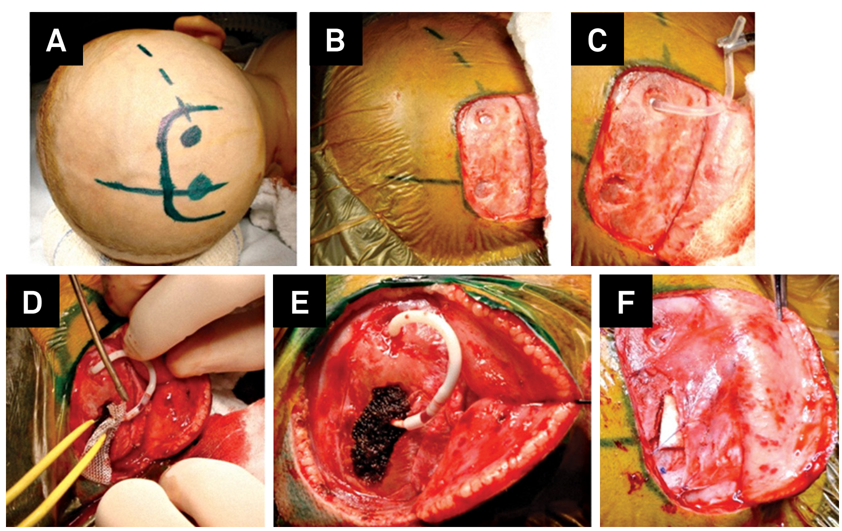
After surgery, patients were followed with pre-established consultations at 3 months, 6 months, 9 months and 1 year. In these consultations, data were evaluated and tabulated for:

CP (3 months, 6 months, 9 months and 1 year); Bregmatic fontanelle(3 months, 6 months, 9 months and 1 year); Transcranial doppler (TD) (3 months and 1 year[54,55,56,57]): mean velocity of blood flow in right and left middle cerebral artery (MCA), basilar artery (BA) and SSS; resistence index in MCA and BA; pulsatility index in MCA and BA; *Computed tomography (CT) – Evans Index (3 months and 1 year); NPMD - (3 months, 6 months, 9 months and 1 year); Complications and shunt revisions; Outcomes focused on patients and caregivers.*

Outcome

Primary: transcranial Doppler measurements, including mean velocity in right and left MCA, resistence index and pulsatility index.

Secondary: evaluation of cephalic perimeter, Evans ratio, fontanelle, neuropsychomotor development, complications and outcomes focused on patients and caregivers.



SSS: superior sagital sinus.

Figure 2. Phases of surgical technique. (A) Positioning and arcuate incision. (B) Opening with medial and lateral burr holes. (C) and (D) Catheterization of lateral ventricle and SSS. (E) Hemostasis. (F) Closure with pericranium and skin closure.

Table 1. Pre operative patient data.

Group	Patient	Gender	Age (months)	MM site	Club foot	Strength	CP (cm)	Fontanelle	NPMD	Evans
RVSS	1 - MFB	M	6	Lumbossacral	Yes	2	45	Hypertense 2+/4+	Adequate	0.47
RVSS	2 – ENM	M	6	Lumbossacral	No	5	47	Hypertense 3+/4+	Delayed	0.50
RVSS	3 – JPM	M	4	Lumbossacral	Yes	0	46	Hypertense 2+/4+	Delayed	0.48
RVSS	4 – RSJ	F	3	Lumbossacral	Yes	3	44	Hypertense 2+/4+	Adequate	0.46
RVSS	5 – SBS	M	6	Lumbossacral	No	5	46	Hypertense 2+/4+	Adequate	0.48
VPS	6 – LGS	M	1	Lumbossacral	No	5	40	Hypertense 2+/4+	Adequate	0.50
VPS	7 - CGG	M	3	Lumbossacral	Yes	5	44	Hypertense 2+/4+	Adequate	0.47
VPS	8 – AEA	M	2	Lumbossacral	Yes	3	45	Hypertense 3+/4+	Adequate	0.48
VPS	9 - KAP	F	6	Lumbossacral	No	2	43	Hypertense 2+/4+	Delayed	0.49

RVSS: retrograde ventricule-sinus shunt; VPS: ventricular-peritoneal shunt; CP: cephalic perimeter; NPMD: neuropsychomotor development.

Statistics

In this study, numerical data are presented as mean ± standard deviation (SD) or as median with range where appropriate. Categorical data are presented as percentages. When comparing the groups, the significance level is considered when $p < 0.05$. To determine the distribution of our data, the Kolmogorov-Smirnov test was used. To evaluate categorical variables, chi-square tests were used and Mc'Nemar, respectively, as appropriate. To evaluate ordinal variables, t-Student, Wilcoxon and Mann-Whitney tests were applied when applicable.

RESULTS

There were no statistically significant differences between VPS and RVSS groups ($p > 0.05$).

Follow-up

Cephalic perimeter (CP)

Figures 3 and 4 show CP before surgery and after surgery. Average perimeter of VPS group patients (42.5 cm) was statistically lower than RVSS group (46.8 cm) in the late follow-up.

Patients undergoing RVSS showed change in the CP curve, not abruptly reducing CP, but making it grow at a physiological rate (Figure 3).

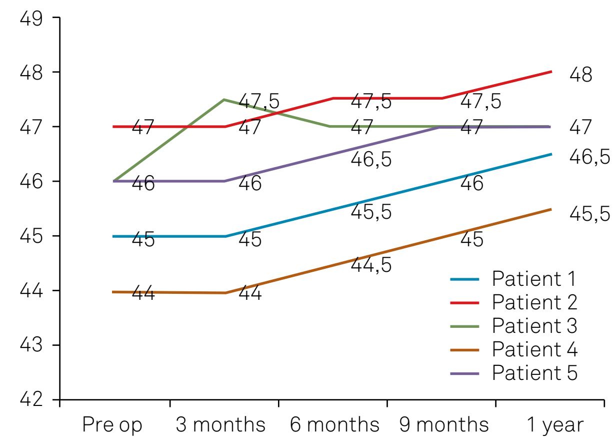
Patient 3 showed increased head circumference curve even after RVSS. Transcranial Doppler showed high resistance, pulsatility and maintained speeds, suggesting non-functioning of the system. A revision surgery found a non-functioning catheter, without thrombus.

Patients undergoing VPS changed CP percentile, with CP abrupt reduction after surgery and return to growth (Figure 4).

Bregmatic fontanelle

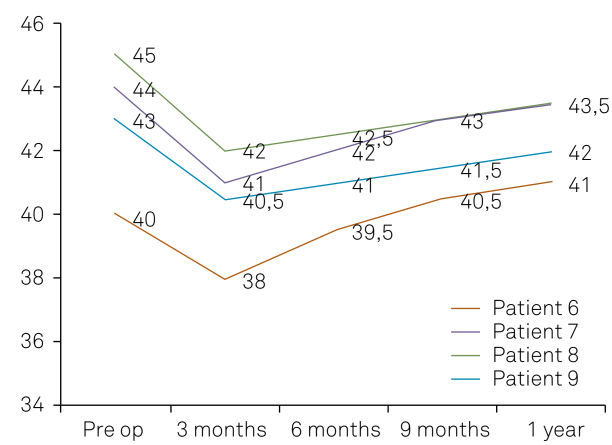
Table 2 shows the evolution of the bregmatic fontanelle characteristics.

Fontanelle characteristics preoperatively did not differ between patients. Postoperatively, there were normotensive patients in RVSS group. In VPS group, fontanelle oscilated between normotensive and mild hypotension. The difference did not reach statistical difference.



RVSS: retrograde-ventricular sinus shunt; CP: computed tomography.

Figure 3. Cephalic perimeter (cm) curve in RVSS group.There was no decrease in CP.



VPS: ventricular-peritoneal shunt; CP: computed tomography.

Figure 4. Cephalic perimeter curve (cm) in VPS group.There was immediate decrease in CP, followed by late growth of skull.

Table 2. Fontanelle pattern during follow-up.

Group	Patient	Fontanelle				
		Pre-Op	3 Months	6 Months	9 Months	1 Year
RVSS	1	Hypertense 2+/4+	Normal	Normal	Closed	Closed
RVSS	2	Hypertense 3+/4+	Normal	Normal	Closed	Closed
RVSS	3	Hypertense 2+/4+	Hypertense 2+/4+	Normal	Normal	Closed
RVSS	4	Hypertense 2+/4+	Hypertense +/4+	Normal	Normal	Closed
RVSS	5	Hypertense 2+/4+	Normal	Normal	Closed	Closed
VPS	6	Hypertense 2+/4+	Normal	Normal	Hypotense (+/4+)	Closed
VPS	7	Hypertense 2+/4+	Normal	Normal	Hypotense (+/4+)	Closed
VPS	8	Hypertense 3+/4+	Normal	Normal	Hypotense (+/4+)	Closed
VPS	9	Hypertense 2+/4+	Normal	Normal	Closed	Closed

RVSS: retrograde ventricule-sinus shunt; VPS: ventricular-peritoneal shunt.

Table 3. Transcranial Doppler. Mean velocity, pulsatility and resistence index analysis in studied vessels.

Patient	Pre Op						Post Op						1 Year					
	MV – R MCA	MV – L MCA	MV BA	MV SSS	PI	RI	MV – R MCA	MV – L MCA	MV BA	MV SSS	PI	RI	MV – R MCA	MV – L MCA	MV BA	MV SSS	PI	RI
1	70	61	43	45	1.1	0.75	80	84	54	40	0.91	0.6	80	83	53	40	0.86	0.62
2	45	48	32	36	1.32	0.71	59	58	35	39	0.9	0.6	62	64	38	41	0.89	0.61
3	53	60	23	30	1.50	0.91	54	56	24	28	1.5	0.94	70	78	45	36	0.94	0.6
4	51	56	36	35	1.5	0.75	66	62	43	40	1.15	0.65	65	64	46	40	1.12	0.64
5	49	54	34	33	1.4	0.7	68	64	45	41	1.1	0.65	70	68	47	42	1.1	0.62
6	47	50	33	37	1.25	0.75	60	58	35	40	0.95	0.6	62	65	39	42	0.86	0.61
7	54	58	27	32	1.45	0.85	70	72	38	34	1.0	0.63	71	73	40	35	0.96	0.6
8	46	49	24	32	1.3	0.8	62	63	30	33	0.95	0.7	64	62	34	36	0.9	0.67
9	65	61	38	45	1.4	0.75	85	87	40	38	1.05	0.62	84	79	43	41	1.1	0.68

MV – R MCA: Mean velocity (cm/s) in right middle cerebral artery; MV – L MCA: Mean velocity in left middle cerebral artery; MV BA: Mean velocity in basilar artery; MV SSS: Mean velocity in superior sagittal sinus; PI: pulsatility index; RI: resistance index.

Transcranial doppler

Table 3 describes Doppler data of patients before and after surgery. Preoperative transcranial Doppler did not differ between VPS and RVSS groups. In both groups, except for patient 3 (discussed above), there was an increase in mean flow velocity, decreased pulsatility index and decreased resistance index in all vessels analyzed, these data maintained inlate post operative (1 year). There was no significant difference between groups.

Computed tomography

Table 4 shows the evolution of Evans index in follow-up. In analogy to what happened with CP, patients undergoing RVSS presented slight reduction in Evans index. In VPS group, there was a postoperative reduction of ventricular size greater than in RVSS group, reaching statistical significance.

Neuropsychomotor development

In the preoperative period, three patients had delayed psychomotor development for age, patients 2, 3 and 9, two from RVSS group and 1 from VPS group. After surgical treatment, profile of patients persisted. Patients with adequate development continued adequacy and patients with developmental delay have evolved, but keeping lagging behind the average age.

During follow-up work, all patients received rehabilitation support in specialized centers with urological, orthopedic and physiatric monitoring.

Complications and shunt revision

Patient 3 had head circumference curve rising even after RVSS. Transcranial Doppler showed resistance, pulsatility and maintained speeds, suggesting system dysfunction. A revision surgery found non-functioning catheter, without the presence of thrombus.

Patient 6 had a surgery converted for VPSdue to intraoperative inaccurate identification of SSS and bleeding. In VPS

Table 4. Evans Index during follow-up.

Group	Patient	Pre-Op	3 Months	1 Year
RVSS	1	0.47	0.46	0.45
RVSS	2	0.50	0.49	0.47
RVSS	3	0.48	0.48	0.47
RVSS	4	0.46	0.46	0.46
RVSS	5	0.48	0.47	0.47
VPS	6	0.50	0.50	0.49
VPS	7	0.47	0.43	0.41
VPS	8	0.48	0.43	0.42
VPS	9	0.49	0.45	0.43

RVSS: retrograde ventricule-sinus shunt; VPS: ventricular-peritoneal shunt.

group, there were no complications during follow-up. No collections, infection or dysfunction signals.

Outcomes focused on patients and caregivers

All caregivers reported improvement in the aspects of patient care, feeling optimistic about the surgery, with no difference from one group to another. They reported satisfaction with procedure, unrepentant and would undergo treatment again if they could choose.

According to caregivers, most of major change and impact aspects were the decrease in head growth, decreased irritability and improvement after surgery.

DISCUSSION

Little has changed over the past decades in terms of shunt revision rate. Numerous series show that over 70% of shunt patients require long-term review and 54% may need four or more revisions, with all additional risks, costs and negative outcomes[4,5,6,7,8,9,10]. ETV also has known limitations, especially in patients with myelomeningocele and Chiari type II[4,5,6,7,8,9,10,11].

RVSS proposes a safe and more physiological option, using less prosthetic material. Additionally, only addresses the cranial region. Besides that, procedure uses internal jugular vein function as a physiological anti-siphon system. In theory, it can be used in any hydrocephalus etiology, at any age. Seven case series with a total of 265 patients undergoing ventricular sinus shunt for hydrocephalus were published since the introduction of technique. In Egypt, El Shafei reported a success rate of over 80%[12,13,14,15,16,17,18,19,20,21,22,23,24,25,26].

El-Shafei et al pioneered by performing puncture in retrograde direction to the direction of blood flow, using the impact pressure of the blood flow in the sinuses of the dura to maintain the CSF pressure greater than the pressure within the dural regardless of changes posture or intrathoracic pressure. They also pioneered use of TD[12,13,14,15,16,17,18,19,20,21,22,23,24,25,26,27,28,29,30]. Toma et al, in a recent review, showed that, in 265 patients, there was no sinus thrombosis, air embolism, uncontrollable operative bleeding or nephritis associated with shunt[25].

In our sample, transcranial Doppler revealed improvement in patterns similarly in both treatment groups. Head circumference, tomography and fontanelle were characteristic in each group. In RVSS group, there was no abrupt reduction in head circumference. CP assumed a physiologic contour but remained high. In VPS group, CP reduced abruptly after surgery, with subsequent regrowth. Similarly, fontanelle in RVSS group remained normotensive, meaning cranial normotension. In VPS group, they became normotensive or hypotensive, especially in older children and may infer a siphoning component in older children with a more upright posture.

In 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.

Caregivers opinions were unanimous in accepting the surgery and confirm its benefits. However, it is difficult to establish a clear distinction between the clinical improvement due to aging and consequent development of children and clinical improvement as a result of treatment.

Some limitations of the study should be mentioned. Initially, our number of patients, although enough to draw up a pilot study, is small for us to make more robust inferences. We believe that our number was lower than planned due to strict inclusion criteria. In addition, our assessment of psychomotor development was brief and lacks objective data and neuropsychological tests. We must also remark that our follow up was of 1 year and should be larger.

We applied in our protocol computed tomography (CT) instead of transfontanellar ultrasound (US) to evaluate Evans index. Although US is cheaper and avoids using radiation, we preferred to apply CT in accordance to protocol proposed by El Shafei et al.

Additionally, RVSS is still relatively unknown in neurosurgical community, often feared due to manipulation of a potentially hazardous structure, superior sagittal sinus. Because of this, few centers have performed this technique even experimentally and thus the learning curve in our country is still early despite the simple technical concept.

In summary, clinical evaluation and Doppler data showed that both treatment modalities were effective, with different CP standards postoperatively. We believe that RVSS can be a viable and safe alternative for the treatment of hydrocephalus in selected cases. Future increase in number of patients treated might disclose further results.

In conclusion, surgical technique of RVSS is feasible. Clinical results are comparable with VPS, being a viable alternative in the treatment of hydrocephalus, especially in cases after myelomenigocele repair.

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