Risk Factors for Malfunction of Ventriculoperitoneal Shunts Performed by Medical Residents in Children: An **Exploratory Study**

Fatores de risco para disfunção de derivações ventrículoperitoneais realizadas por médicos residentes em crianças: Estudo exploratório

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Abstract

Introduction Ventriculoperitoneal shunts (VPSs) are common neurosurgical procedures, and in educational centers, they are often performed by residents. However, shunts have high rates of malfunction due to obstruction and infection, especially in pediatric patients. Monitoring the outcomes of shunts performed by trainee neurosurgeons is important to incorporate optimal practices and avoid complications. **Methods** In the present study, we analyzed the malfunction rates of VPSs performed in children by residents as well as the risk factors for shunt malfunction.

Results The study included 37 patients aged between 0 and 1.93 years old at the time

Keywords

- ventriculoperitoneal shunts
- ► hydrocephalus
- ► shunt malfunction
- medical resident
- ► teaching hospital
- ► ventriculostomy

of surgery. Congenital hydrocephalus was observed in 70.3% of the patients, while 29.7% showed acquired hydrocephalus. The malfunction rate was 54.1%, and the median time to dysfunction was 28 days. Infections occurred in 16.2% of the cases. Cerebrospinal fluid leukocyte number and glucose content sampled at the time of shunt insertion were significantly different between the groups (p = 0.013 and p = 0.007, respectively), but did not have a predictive value for shunt malfunction. In a multivariate analysis, the etiology of hydrocephalus (acquired) and the academic semester (1st) in which the surgery was performed were independently associated with lower shunt survival (p = 0.009 and p = 0.026, respectively).

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Conclusion Ventriculoperitoneal shunts performed in children by medical residents were at a higher risk of malfunction depending on the etiology of hydrocephalus and the academic semester in which the surgery was performed.

Resumo

Introdução As derivações ventrículo-peritoneais (DVPs) são procedimentos neurocirúrgicos comuns e, em centros educacionais, muitas vezes são realizados por residentes. No entanto, os shunts apresentam altas taxas de mau funcionamento devido a obstrução e infecção, especialmente em pacientes pediátricos. O monitoramento dos resultados das válvulas realizadas por neurocirurgiões em treinamento é importante para incorporar as práticas ideais e evitar complicações.

Métodos No presente estudo, analisamos as taxas de mau funcionamento de DVPs realizados em crianças por residentes, assim como os fatores de risco para mau funcionamento da válvula.

Resultados O estudo incluiu 37 pacientes com idades entre 0 e 1,93 anos na época da cirurgia. Hidrocefalia congênita foi observada em 70,3% dos pacientes, enquanto 29,7% apresentaram hidrocefalia adquirida. A taxa de disfunção foi de 54,1% e o tempo médio para disfunção foi de 28 dias. Infecções ocorreram em 16,2% dos casos. O número de leucócitos do líquido cefalorraquidiano e o conteúdo de glicose coletados no momento da inserção da válvula foram significativamente diferentes entre os grupos (p = 0,013 e p = 0,007, respectivamente), mas não tiveram um valor preditivo para o mau funcionamento da válvula. Em uma análise multivariada, a etiologia da hidrocefalia (adquirida) e o semestre letivo (1°) em que a cirurgia foi realizada foram independentemente associados a menor sobrevida do shunt (p = 0,009 e p = 0,026, respectivamente).

Conclusão: Derivações ventrículo-peritoneais realizadas em crianças por médicos residentes apresentaram maior risco de mau funcionamento dependendo da etiologia da hidrocefalia e do semestre letivo no qual a cirurgia foi realizada.

Palavras-chave

- derivação ventrículoperitoneal
- ► hidrocefalia
- disfunção de derivação respectivamente).
 ventrículo-peritoneal Conclusão: Deriv
- ► hospital de ensino
- ► ventriculostomia

Introduction

Hydrocephalus is an important cause of neurological disability and can accompany several other conditions, such as infections, neoplasms, cerebrovascular diseases, and trauma. Its prevalence is estimated to be of 85 per 100,000 individuals, while the prevalence is higher among children (88 per 100,000 individuals), especially due to congenital malformations and neonatal complications related to prematurity.¹

The treatment of hydrocephalus is surgical, and even though endoscopic third ventriculostomy has yielded significant advances in the management of this condition (particularly in cases of obstructive hydrocephalus), ventriculoperitoneal shunts (VPSs) remain the most common treatment option.^{2,3} Ventriculoperitoneal shunts are effective for most cases; however, the rates of VPS malfunction are very high, and can increase up to 84.5%.^{4–6}

Several classifications depending on the site of the problem and on the presence of infection have been proposed to categorize VPS malfunctions. Indeed, VPS malfunctions are associated with worse outcomes, and a single malfunction has a high predictive value for further malfunctions, necessitating several surgical procedures throughout the lifetime of the patient. Thus, a thorough understanding of the risk factors for VPS malfunction is essential to optimize the clinical follow-up and surveillance and facilitate early detection of malfunction and, ultimately, provide better care and improved prognosis.

Although guidelines and recommendations for shunt implantation emphasize that the procedure should be performed by an experienced neurosurgeon, shunt procedures are performed by residents at many centers, especially in teaching hospitals in low- and middle-income countries (LMICs), as well as in developed countries. This dilemma between patient safety and surgical education has made it important to monitor the surgical results of procedures performed by trainee neurosurgeons. In the present study, we aimed to analyze the malfunction rates of VPSs performed in children by residents as well as the risk factors for shunt revision.

Methods

This was a retrospective cohort study of a case series of patients attending the Pediatric Neurosurgery Outpatient Clinic at the Hospital das Clínicas da Faculdade de Medicina de Botucatu of the Universidade Estdual de São Paulo, Botucatu, state of São Paulo, Brazil. This is a university hospital located in the center-west region of the state of

São Paulo in Brazil, and it is the referral center for up to 2 million patients in 68 cities. The study protocol was approved by the local Institutional Review Board.

Using electronic medical registries, we recovered the data of patients who had undergone VPS procedures at this center. The surgical procedures were performed by 2nd-year medical residents under the supervision of experienced staff. We included all children born between 2013 and 2018 who were diagnosed with hydrocephalus and were treated with VPS. The children were routinely followed up at 1, 3, 6, and 12 months postoperatively, and then annually. The exclusion criteria were surgeries performed by nonresident neurosurgeons, patients lost to follow-up, and previous endoscopic third ventriculostomy.

The independent variables were age at VPS insertion, previous use of an external ventricle drain (EVD), cause of hydrocephalus, the semester in which the scholar was at the time of the surgery (since medical residency begins every March, the 1st academic semester was defined as March to September, and the 2nd semester from October to February), and the cerebrospinal fluid (CSF) parameters from the intraoperative sampling. The primary outcome was shunt malfunction and time for its occurrence. The secondary outcome was the cause of malfunction (obstruction or infection) and the microorganisms isolated from the cases that presented with infections.

For the statistical analysis, the distribution of the data was assessed using the Kolmogorov-Smirnov test. Comparisons between groups were performed using the Mann-Whitney test. Correlations were tested using the Spearman test. The chi-squared and the Fisher exact tests were used to compare categorical data. Multivariate analysis with Cox regression curves was used to analyze shunt survival with adjustments for covariates. Receiver operating characteristic (ROC) curves were generated to identify the predictive values of CSF parameters on shunt revision. For all tests, the level of statistical significance was set at 5%. Statistical analyses were performed using IBM SPSS Statistics for MacBook, version 24 (IBM Corp., Armonk, NY, USA).

Results

We evaluated the data from 37 patients (21 boys and 16 girls) aged 3.57 \pm 1.44 years old. The mean follow-up duration was of 765.05 days (\sim 2 years). The age of the patients at the time of surgery ranged from 0 to 1.93 years old (median: 2 months

Table 1 Causes of shunt malfunction

Malfunction mechanism	n (%)
Catheter obstruction	7 (35%)
Shunt infection	6 (30%)
Catheter misplacement	3 (15%)
Wound dehiscence	2 (10%)
CSF hyperdrainage	1 (5%)
Catheter migration	1 (5%)

Abbreviation: CSF, cerebrospinal fluid.

Among 37 patients, 20 required a shunt revision.

old). In total, 59.5% of the surgeries (n = 22) were performed in the 1st academic semester. Six children (16.2%) had received a prior EVD. Among the cases of congenital hydrocephalus (n = 26; 70.3%), 21 were caused by malformations of the central nervous system (such as aqueduct stenosis and myelomeningocele), and 5 were related to congenital infections (toxoplasmosis and cytomegalovirus). Among the cases of acquired hydrocephalus (n = 11; 29.7%), 9 were caused by peri-intraventricular hemorrhage (PIVH) of prematurity, and 2 were attributed to neonatal meningitis.

The overall rate of shunt revision was 54.1%, and the median time for revision was 28 days (interquartile range = 418.5 days). The most common causes of shunt revision were catheter obstruction and shunt infection (**-Table 1**). Most of the microorganisms related to infection belonged to the Staphylococcus genus (4 out of 6). The overall infection rate was 16.2%. There was no difference in the time for shunt review between cases requiring revision due to infectious and noninfectious causes (mean: 297.33 versus 278.3 days, respectively, p = 0.494). In the CSF parameter data collected at the time of ventricular catheterization, patients with acquired hydrocephalus had higher levels of leukocytes (p = 0.013) and lower levels of glucose (p = 0.007) than those with congenital hydrocephalus (- Table 2). The children with and without shunt revision showed no difference in median age at the time of shunt insertion (64.0 versus 65.0%, respectively; p = 0.964). Similarly, none of the CSF parameters were significantly different between patients who required shunt revision and those did not.

In the univariate analysis, the semester in which shunt insertion was performed was not associated with the rate of shunt revision: the rate was 63.6% for procedures performed

Table 2 Cerebrospinal fluid parameters according to the etiology of hydrocephalus

CSF parameter	Acquired hydrocephalus (n = 11)	Congenital hydrocephalus ($n = 26$)	p-value
Leukocytes (median [IQR])	2.00 (8.00]	0.0 (0.00]	0.013
Protein mg/dl (median [IQR])	87.00 (404.00]	63.0 (98.00]	0.201
Red cells (median [IQR])	12.0 (70.00]	5.0 (55.00]	0.586
Glucose mg/dl (mean \pm SD)	26.81 ± 8.76	37.43 ± 10.68	0.007
Lactate mmol/L (mean \pm SD)	2.33 ± 0.61	2.08 ± 1.00	0.529

Abbreviations: CSF, cerebrospinal fluid; IQR, interquartile range; SD, standard deviation.

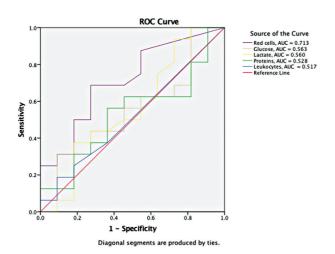


Fig. 1 Receiver operating characteristic curves for cerebrospinal fluid parameters. Red blood cells showed a higher area under the curve, but none of the parameters were statistically associated with the risk of shunt revision.

in the 1st semester and 40.0% for those performed in the 2nd semester ($p\!=\!0.157$). All 6 patients who had previously received an EVD required shunt revision, while 45.2% of those without an EVD required shunt revision ($p\!=\!0.022$). Similarly, the rate of shunt revision was higher among patients with acquired hydrocephalus than among those with congenital hydrocephalus (81.8 versus 42.3%; $p\!=\!0.036$). Since the rate of previous EVD usage was higher among patients with acquired hydrocephalus (45.5 versus 3.8%; $p\!=\!0.005$), further multivariate analysis was necessary for covariate adjustment.

The ROC curves of CSF parameters for predicting shunt revision did not yield significant cutoff values. For red blood cell count, the area under the curve (AUC) was 0.713 (p = 0.064). The other parameters had an AUC between 0.5 and 0.6 (p > 0.6), as shown in **Figure 1**.

In the Cox regression analysis with covariate adjustment, age at the time of surgery was not associated with a higher risk of shunt revision (Exp [B] = 0.998; 95% confidence interval [CI]: 0.994–1.002). However, the semester in which the surgery was performed showed a significant difference: surgeries performed in the 1st semester had a higher risk than those in the 2nd semester (odds ratio [OR] = 3.145; 95% CI: 1.149–8.612; p = 0.026). Patients with congenital hydrocephalus were also at a lower risk for shunt revision than those with acquired hydrocephalus (OR = 0.303; 95%CI: 0.124–0.741; p = 0.009). The differences are shown in **Figure 2**.

Discussion

Ventriculoperitoneal shunts are common neurosurgical procedures and one of the first surgical procedures learned by trainee neurosurgeons. Despite the relatively easy technique, they carry non-negligible risks for complications. For pediatric patients, these risks are even higher, especially the risk of infection. Therefore, close monitoring of the surgical results is of utmost importance for quality surveillance and improvement.

The rate of revision in our study was quite high (54.1%) and it was comparable to the higher rates reported in the literature.^{13,14} Nevertheless, the infection rate (16.2%) was not as high. However, this finding must be analyzed with care, since we considered the first malfunction as the primary outcome. Patients who required a revision were at a higher risk of new subsequent revisions, since a single revision *per se* is recognized as an important risk factor for new revisions either due to obstruction or to infection.^{15–18} Long-term follow-up assessments show that up to 84.5% of the patients

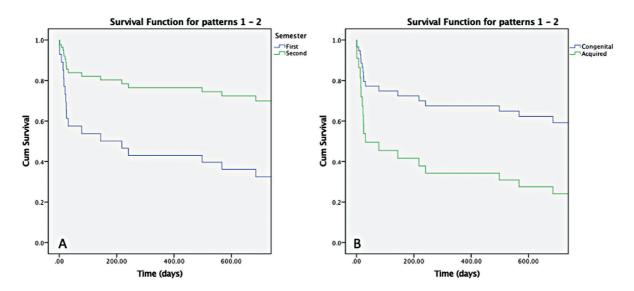


Fig. 2 Cumulative survivals of shunts performed in the 1st or 2nd semester (A, p = 0.026) and for cases of congenital or acquired hydrocephalus (B, p = 0.009). In all cases, most revisions occurred within the first 30 days. Shunts implanted in the 1st semester and in cases of acquired hydrocephalus had lower survival rates.

require at least 1 shunt revision, with high mortality rates directly associated with infection episodes.^{5,8}

Regarding the causes of hydrocephalus, we found that most cases involved congenital etiologies and that the proportion of acquired diseases was lower. A multicenter study on VPS infections also reported a higher proportion of congenital malformations as the leading etiology. 17 In our study, the main cause of acquired hydrocephalus was prematurity-related peri-intraventricular hemorrhage, which may be the reason why some CSF parameters (leukocyte number and glucose levels) were different at the time of shunt insertion. However, these parameters were not significantly different between patients who underwent shunt revision and those who did not. The CSF parameters also could not predict shunt malfunction, even though the number of red cells showed a higher AUC, which is expected, because patients with peri-intraventricular hemorrhages have higher rates of shunt malfunction. 19 Studies with larger sample sizes could provide more insights into the role of CSF red cells in predicting shunt malfunction regardless of the etiology of hydrocephalus.

The use of EVDs was associated with acquired hydrocephalus, since peri-intraventricular hemorrhage and neonatal meningitis often require a temporary EVD. These patients most often required shunt revision, which is consistent with the literature: peri-intraventricular hemorrhage is an important risk factor for VPS malfunction. In addition, prematurity itself is another independent risk factor for VPS malfunction. Therefore, premature children with peri-intraventricular hemorrhages should be closely monitored after shunt implantation, especially in the 1st postoperative month – when the infections and obstructions typically occur.

In our study, VPS implantation in the 1st academic semester was associated with a higher risk of revision, which indicates the effect of learning curves on the surgical outcomes of VPSs, as demonstrated previously.²¹ Among the 20 cases of shunt malfunction, 5 (25%) could be attributed to low surgical experience (catheter misplacement and wound dehiscence). The "July effect" has been identified as an important factor related to complications of surgeries performed in the beginning of the training of new staff. Early resident transition may be responsible for this phenomenon. However, several studies have not demonstrated this finding, which could be attributed to good resident training with sufficient guidance and support.²²⁻²⁵ When present, this effect is generally very small. ^{26,27} Nevertheless, these studies were conducted in high-income countries, and there is a lack of evidence regarding the equivalent "July effect" among neurosurgical trainees in low- and middle-income countries (LMICs). In this regard, a recent survey on the perceptions of LMIC neurosurgery residents of their educational programs highlighted concerns regarding inadequate exposure to subspecialties, exhausting work hours, and inconsistent supervision.²⁸ Additional studies are needed to evaluate the extent to which the lack of ideal supervision interferes with surgical outcomes, even though this was not the case in our setting.

Patient safety is of utmost importance, and all attempts should be made to accomplish it. However, balancing patient safety and the training of young surgeons is a constant challenge, especially in centers of academic education. Regarding VPSs, there is a clear dichotomy: the best practice guidelines advocate for these procedures to be performed by senior surgeons; however, most academic centers reserve these procedures for the early training years, given the high technical complexity of other neurosurgical procedures that should be acquired.

The limitations of our study included the small sample size and the lack of comparison with shunt procedures performed by graduate neurosurgeons. Studies addressing infection should have a strong series, especially if infectionrelated factors are considered in the analysis. Also, the demonstration of higher rates of complications related to the performing residents would demand a control group composed of graduate neurosurgeons. However, as an exploratory study, the present results may be an eye-opener for future Brazilian networks and multicenter collaborations aiming to both understand the learning curve of our residents and to implement best standardized practices throughout the country. In addition, since the primary outcome was the first shunt malfunction and the cases were subsequently censored, we did not evaluate the cumulative effect of a single shunt malfunction on repeated malfunctions and infections, which has been demonstrated in other studies. Moreover, long-term follow-up evaluations could provide additional data. Despite these limitations, our study reinforces the data on worse outcomes of VPSs performed in children with peri-intraventricular hemorrhage. Furthermore, our study adds new knowledge by demonstrating an equivalent "July effect" related to neurosurgery training for VPSs in children.

In conclusion, VPSs performed in children by medical residents were at a higher risk of malfunction, depending on the etiology of hydrocephalus and on the academic semester in which the surgery was performed.

Ethical Statement

The present retrospective study was approved by our institutional review board (IRB No. 2.533.607/2018). The parents of the patients signed informed consent forms.

Data Availability Statement

All data generated or analyzed during the present study can be retrieved upon request to the corresponding author.

Conflict of Interests:

The authors have no conflict of interests to declare.

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