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Assessment of the Factors Influencing the Survival of the Ventricular Shunt in Infantile Hydrocephalus

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Abstract: *The purpose of the present research is to perform a retrospective analysis of 242 ventricular shunts implanted in childhood due to infantile hydrocephalus, submitted to a long-term follow-up (27.84 ± 6.13 years) and investigate the factors influencing the survival of the shunt, applying modern mathematical approaches. The contingent consists of 242 patients who were shunted due to infantile hydrocephalus in childhood, 55% of whom were under 6 months old when the shunt was implanted. For the entire follow-up period, patients underwent 375 revisions (average 1.55 revisions per shunt), 38% are without shunt failure, with only one revision – 26%, with two – 14%. The remaining 22% have 3 or more revisions (4.58 shunt revisions) and they make 65% of all revisions. 68.5% of all revisions are due to mechanical complications; to functional (inadequate drainage) – 7.5%. 9.1% of the patients had inflammatory complications. We found that the age at shunting (under 3 months – the worst trend) and the interval to the first revision (over 6 months – a smaller number of revisions) are of the biggest importance for the number of the following revisions. If the patient survives one year after shunting without failure, the risk of such is under 50% during the whole shunt existence.*

Keywords: *Infantile hydrocephalus, Shunt complications, Shunt survival, Pediatric neurosurgery, InterCriteria analysis.*

Introduction

Hydrocephalus, which is a heterogeneous disease characterized by abnormal dilatation of the ventricles of the brain as a result of various etiologies [19], remains one of the most common pathologies treated in pediatric neurosurgical departments [4]. The pathophysiological mechanism is not fully understood, and ventriculostomy and shunting have been used to treat this condition, although shunting appears to be superior in terms of clinical improvement, so for more than half a century, shunts have been used as the main method of treatment throughout the world [6]. In Bulgaria, the first valve was implanted in 1966, but their mass usage started after 1980. The Clinic of Neurosurgery at the University Hospital "St. Ivan Rilski" – Sofia is the first successor of the first clinical neurosurgical hospital in Bulgaria and, respectively, the clinic with the longest history and experience in the treatment of infantile hydrocephalus. A small number of pediatric neurosurgeons have been performing the implantations and revisions of shunts. This reduces the differences in surgical behaviour and is one of the advantages of the monocentric study.

Initially, the right atrium was used primarily as a drainage site for ventricular cerebrospinal fluid. In unison with the trends [5] in the period around 1990 we gradually went to drainage in the intraperitoneal area, as a type of surgery with mild ongoing complications. Ventriculoatrial shunts are now implanted only in cases of problems in the peritoneal cavity (hyporesorbtion, adhesions, inflammatory processes). Another direction in shunt evolution is the usage of different valve types, as the manufacturers' and the neurosurgeons' intention is to lower the percentage of functional complications. This way after using slit valves we went to valves with fixed pressure, flow-control valves, anti-siphon device valves (ASD), and in recent years – programmable valves and valves with antibiotic impregnated catheters.

Although ventriculoperitoneal shunt (VPS) placement is one of the most common neurosurgical procedures and despite improvements in valves, VPS placement complications are common and many shunt revisions are expected throughout life [5, 11, 16]. According to the literature, failure rate of VPS varies from 1 in 10 to a quarter of all primary shunts in the first year after placement [4], with the reported rate of shunt failure and subsequent revisions reaching 85% throughout the life of a shunt. It is the subsequent surgical interventions that account for the significant economic, medical, patient (dissatisfaction) and social costs of treating hydrocephalus.

Purpose: To perform a retrospective analysis of 242 ventricular shunts implanted in childhood due to infantile hydrocephalus with long-term follow-up and to investigate factors influencing shunt survival by applying different approaches, e.g., InterCriteria Analysis (ICrA), descriptive analysis, ANOVA, Kaplan-Meier survival analysis.

Materials and methods

The Clinic of Neurosurgery at the University Hospital "St. Ivan Rilski" – Sofia, where the present study was conducted, is the oldest specialized and largest neurosurgical clinic in Bulgaria. For the 20 years (1984-2003) 459 valves had been implanted in this clinic for treating hydrocephalus with different etiology (infantile hydrocephalus – 242; tumours and hydrocephalus – 124; DWS and dysraphic syndrome – 53; porencephalic cysts – 16; arachnoid cysts – 14; hydrocephalus with subdural leakages – 10). The authors retrospectively reviewed the medical records of all patients with infantile hydrocephalus.

Inclusion criteria

Patients with infantile hydrocephalus, up to 18 years of age, regardless of gender, with infantile hydrocephalus treated with a ventriculoperitoneal shunt. The later age of those operated for the first time after one year is due to either a late diagnosis (in the first half of the period 1984-2003 the imaging diagnosis in the country was with fewer devices per capita) or step-progressive form of hydrocephalus, which manifests clinically later.

Exclusion criteria

Patients over 18 years of age. Patients with lumboperitoneal anastomosis due to their small number and specific complications.

The follow-up was long-term with a mean duration of 27.84 ± 6.13 years (mean ± standard deviation).

In the present study, in addition to the established statistical procedures (e.g., descriptive analysis, ANOVA, Kaplan-Meier survival analysis), a relatively new approach of InterCriteria Analysis for discovering interrelationships between the criteria was applied [2, 12].

Since ICrA is based on intragroup relations (relations between the evaluations of any two objects by one criterion are matched to those by another criterion), it allows the use of analogous data from different sources. In the case of medical data, such as the subject of this study, data from different laboratories/hospital units can be analysed with ICrA without the need for unification of reference values and methodology. This makes ICrA extremely suitable for detecting regularities between criteria, in this case – consonance and dissonance between factors affecting the long-term survival of ventricular shunts.

Results and discussion

Data collection

The contingent of patients included all children with implanted valves in the above period (57% – male and 43% – female). The average age of the patients studied was 2 years and 7 months, with the youngest patient at shunting at 0.8 months and the oldest one – at 18 years. The percentage distribution of patients according to their age at the initial shunt implantation is as follows: up to 1 month – 5%; from 1 to 3 months – 28%; from 3 to 6 months – 22%; from 6 months to 1 year – 13%; from 1 to 3 years – 12%; over 3 years – 20%.

Surgical outcome

In patients shunted at the age under 3 months, the percentage of shunts without revisions is the smallest and we observe the worst trend of first shunt failure (without statistically significant difference $p > 0.05$).

Table 1 presents the results of applying ICrA to the patient data (age at shunting, number of revisions, interval to first revision, age at first revision, interval between first and second revision and age at second revision) to establish which of the studied indicators are decisive for a large number (more than 3) of revisions. A strong negative consonance (according to [1]) was found between the number of revisions and the age at bypass, age at first and age at second revision, i.e., a greater number of revisions are performed in patients shunted at an early age, as well as in those who required a first and/or second revision at a younger age.

Table 1. Results from ICrA

	Number of revisions	**Age at implantation**	**Age at I revision**	**Interval to I revision**	**Age at II revision**	**Interval I-II revision**	**Age at III revision**	**Number of revisions**
Number of revisions	$\langle 1.0; 0.0\rangle$	$\langle 0.66; 0.34\rangle$	$\langle 0.17; 0.81\rangle$	$\langle 0.66; 0.34\rangle$	$\langle 0.08; 0.91\rangle$	$\langle 0.68; 0.3\rangle$	$\langle 0.08; 0.91\rangle$	$\langle 0.66; 0.32\rangle$
Age at implantation	$\langle 0.66; 0.34\rangle$	$\langle 1.0; 0.0\rangle$	$\langle 0.4; 0.58\rangle$	$\langle 0.45; 0.53\rangle$	$\langle 0.26; 0.72\rangle$	$\langle 0.49; 0.49\rangle$	$\langle 0.26; 0.72\rangle$	$\langle 0.43; 0.55\rangle$
Age at 1 revision	$\langle 0.17; 0.81\rangle$	$\langle 0.4; 0.58\rangle$	$\langle 1.0; 0.0\rangle$	$\langle 0.4; 0.6\rangle$	$\langle 0.87; 0.13\rangle$	$\langle 0.13; 0.87\rangle$	$\langle 0.87; 0.13\rangle$	$\langle 0.15; 0.85\rangle$
Interval to 1 revision	$\langle 0.66; 0.34\rangle$	$\langle 0.45; 0.53\rangle$	$\langle 0.4; 0.6\rangle$	$\langle 1.0; 0.0\rangle$	$\langle 0.26; 0.74\rangle$	$\langle 0.51; 0.49\rangle$	$\langle 0.26; 0.74\rangle$	$\langle 0.45; 0.55\rangle$
Age at 2 revision	$\langle 0.08; 0.91\rangle$	$\langle 0.26; 0.72\rangle$	$\langle 0.87; 0.13\rangle$	$\langle 0.26; 0.74\rangle$	$\langle 1.0; 0.0\rangle$	$\langle 0.23; 0.77\rangle$	$\langle 1.0; 0.0\rangle$	$\langle 0.28; 0.72\rangle$
Interval 1-2 revision	$\langle 0.68; 0.3\rangle$	$\langle 0.49; 0.49\rangle$	$\langle 0.13; 0.87\rangle$	$\langle 0.51; 0.49\rangle$	$\langle 0.23; 0.77\rangle$	$\langle 1.0; 0.0\rangle$	$\langle 0.23; 0.77\rangle$	$\langle 0.72; 0.28\rangle$
Age at 3 revision	$\langle 0.08; 0.91\rangle$	$\langle 0.26; 0.72\rangle$	$\langle 0.87; 0.13\rangle$	$\langle 0.26; 0.74\rangle$	$\langle 1.0; 0.0\rangle$	$\langle 0.23; 0.77\rangle$	$\langle 1.0; 0.0\rangle$	$\langle 0.28; 0.72\rangle$
Interval 2-3 revision	$\langle 0.66; 0.32\rangle$	$\langle 0.43; 0.55\rangle$	$\langle 0.15; 0.85\rangle$	$\langle 0.45; 0.55\rangle$	$\langle 0.28; 0.72\rangle$	$\langle 0.72; 0.28\rangle$	$\langle 0.28; 0.72\rangle$	$\langle 1.0; 0.0\rangle$

During the follow-up period, 242 patients underwent 375 revisions (an average of 1.55 per patient). 91 patients (38%) remained without shunt failure, and the number of reoperations is detailed in Fig. 1. As it can be seen, with more than 7 revisions there are only 5 patients (2% of the total number), but 15.5% of all revisions are due to them. More than 1/3 of the patients are only with the initial operation and 1/4 are with one revision.

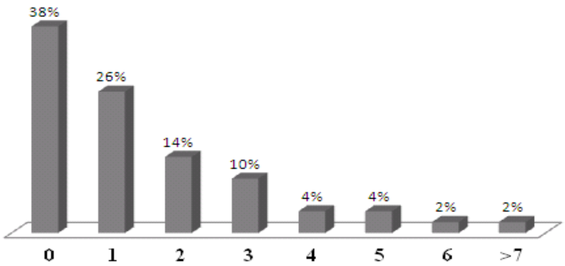

Fig. 1 Percentage of the groups of patients without and with different numbers of revisions

The predominant number of shunts with revisions undergo 1 or 2 revisions (40%), including distal catheter extension revisions. In this study, the criteria “larger number of revisions” means 3 and more revisions per shunt. In this series, 22% of the shunts are with “3 or more revisions” and they account for 65% of all revisions. In this group, each patient underwent 4.58 revisions on average, while in the whole contingent one revised shunt has 2.48 revisions on average. Almost half of the patients (47%) in the cohort are re-operated in the first 6 months after shunting, while in the group of patients with 1 or 2 revisions, this percentage is only 22%.

The average interval to the first revision is 48 months (mean 22 months; minimum 1 day; maximum 260 months). The average interval to the first revision according to the number of revisions is:

- for the group of patients with one revision – 69.1 months;
- for the group of patients with two revisions – 46.42 months;
- for the group of patients with three and more revisions – 24.42 months.

First shunt failure in the group with 3 or more revisions occurs earlier compared to the other two groups, which is statistically reliable ($p < 0.05$).

Shunt complications

According to the type of the different complications in the analysed material we divided them to:

- ✓ Mechanical complications, most frequently expressed in obstruction of ventricular or distal catheter, or of the valve mechanism. They make 68.5% of all 375 revisions. In our cohort, they are distributed as follows: peritoneal obstruction – in 20% of all complications, proximal shunt obstruction – 17% and atrial obstruction – 13%.
- ✓ More rarely (3%) we have disconnection, valve migration, intraventricular hemorrhage, and skin problems.
- ✓ Extremely rare complications. We observed only 2 cases of extremely rare complications (0.5%) – 1 urethra perforation and 1 large intestine perforation with penetration through the penis and the anus.
- ✓ Functional complications, due to inadequate momentary or permanent drainage, different than the one in a normally functioning shunt system, are the cause for 28 revisions (7.5%).
- ✓ Inflammatory complications were observed in 22 (9.1%) of 242 patients and were the cause of 77 (20.5%) of all 375 revisions. Over 50% of infections are in the first 2 months after shunting, and only 2 patients were diagnosed more than a year after surgery (1 sepsis after 8 years and 7 months and 1 sepsis after 1 year and 1 month).

In the last two cases, we categorize the infections as “late”. In 5 patients the inflammatory manifestations recurred.

Shunt survival

The largest percentage (25%) of the patients is revised in the first year (Fig. 2).

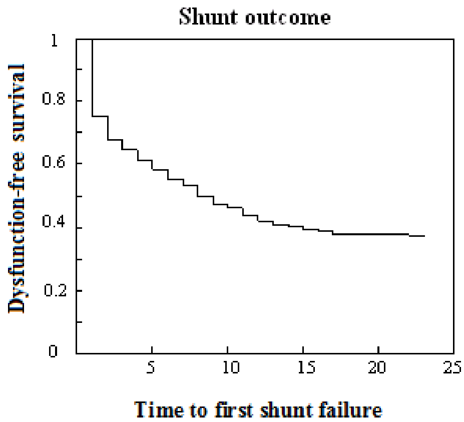

Fig. 2 Kaplan-Meier curve for shunt survival after implantation: on the x-axis – time to first shunt failure; on the y-axis – dysfunction-free survival.

As seen from Fig. 2 1/4 of the patients undergo first shunt failure during the first year. After the third year, we observe an even percentage of “failed” shunts. After the 17-th year, the curve becomes parallel to the abscissa.

Only half of the remaining patients undergo revisions during the follow-up, i.e., if a patient survives the first 12 months after implantation without failure, the risk of such is under 50% for the whole shunt existence. As shown in Fig. 2, during the subsequent years of follow-up no significant difference in the percentage of the average first shunt failure (2-3%) is observed. In conclusion, we observed a significantly higher number of revisions during the first decade after implantation compared to the second decade ($p < 0.01$).

The groups “patients with only 2 revisions” and “patients with 3 and more revisions” were compared by applying ANOVA analysis (Table 2), where age and intervals are given in months.

Table 2. Comparing the behaviour (average age and average intervals) of the two groups “patients with only 2 revisions” and “patients with 3 and more revisions”

	Patients with only 2 revisions ***N* = 34**	**Patients with 3 and more revisions** ***N* = 53**	***p***
The average age at implantation	31.15	31.86	0.9498
The average age at the first revision	77.56	55.65	0.1165
Average interval to the first revision	46.42	23.79	0.0120
The average age at the second revision	128.86	71.74	0.0005
The average interval between first and second revision	51.29	16.09	0.0001

The average age at the first revision and the interval next to it, as well as the interval between the first and second revision, are statistically shorter for the group “patients with 3 and more revisions” ($p < 0.01$).

Discussion

Despite continuous attempts to reduce the incidence of ventriculoperitoneal shunt complications, such as improved sterile techniques, antibiotic-impregnated catheters, and programmable valves, their failure remains a major problem, often leading to multiple and expensive hospital admissions [17]. Despite this, long-term outcomes have rarely been reported in patients with pediatric hydrocephalus [10]. The present study aims to analyse retrospectively the causes of complications as well as subsequent revisions.

Data collection

Due to the purity (etiological homogeneity) of the studied group, we can make much more significant and certain conclusions, because this eliminates the influence of different etiological factors (dysraphism, tumours, cysts, etc.). Observations are only on cases of infantile hydrocephalus, manifested in the first year or later in late diagnosis or step-progressive hydrocephalus.

Most of them (55%) are shunted at ages up to 6 months. In the youngest patients (up to 3 months) the percentage of shunts without revisions is the smallest, but there is no statistically significant difference ($p > 0.05$) between the number of complications in them and children shunted over the age of 3 months. This group of patients also shows the worst trend of first shunt failure ($p > 0.05$). ICrA showed that younger age at shunting, as well as younger age at first and second revision, were factors for a higher number of subsequent revisions. Other authors [3] also report young age as a risk factor for bad shunt survival.

Surgical outcome

In the studied cohort, 38% of the shunts remained unaudited and 26% – with only one revision. The “bad reputation” of shunts is due to a limited number of patients, namely patients with “3 or more revisions”. They make up 1/5 of the shunts, but they account for 2/3 of all revisions. This trend is especially pronounced in 5 of the patients with more than 7 revisions, who underwent 15% of all revisions in the cohort. In our opinion, this reinstates shunt as a good way to treat hydrocephalus. There are also articles in the literature with a significantly higher share of children who have undergone numerous revisions [12].

In our study, we found that the shorter the interval to the first revision is, the sooner and the shorter the intervals to the following revisions are. We find similar observations in other authors [20]. The group of patients with interval between implantation and first revision over 6 months is with better shunt survival than the group of patients with interval between implantation and first revision under 6 months, which has some prognostic value.

Shunt complications

Mechanical complications cause 68.5% of all revisions. Other authors have similar data [7] and report 60% mechanical complications in their long-term follow-up studies. Even in short-term follow-up (up to 3 years) they make up to 38% of the revisions [13, 18].

Disconnection, valve migration, intraventricular haemorrhage and skin problems are rarer. According to Di Rocco et al. [7] complications related to migration make 8.8% of all mechanical complications, while disconnection – 1.4%.

Extremely rare complications – hollow organs penetration in this series is 0.5%, which correlates with the literature [8]. Most often these are the intestines (0.1% of all complications in CSF shunt systems) [9, 21]. Next are the bladder [4], stomach, liver, scrotum [15] and others [8]. Mortality among these complications reaches 15% [9].

Functional complications lead to slit ventricle, secondary craniosynostosis, subdural collection, isolated ventricles and are the reason for 7.5% of the revisions. This percentage is close to the one in the literature – 10% [3]. According to other authors [14] hyperdrainage occurs in much wider ranges – 3.7÷30%.

Inflammatory complications accompany the implantation of a shunt, which is a foreign body in the organism. Although all companies try to change and improve the design and the material of the valves, inflammatory complications are still one of the most problematic complications. Despite surgeons' attempts to reduce their frequency, infections most often affect 8.5-15% of patients [3, 9, 22].

Shunt survival

During our study, we formulated the following factors for shunt survival:

- factors for good shunt survival in the group are:
  - percentage of shunts without revisions,
  - percentage of shunts with only one revision;
- factor for bad shunt survival in the group – percentage of shunts with 3 and more revisions.

The shunt survival rate in the study during the first 12 months (75%) was comparable to literature data on the subject [22]. In general, the percentage of shunts with no revisions (39%) is relatively high, which is due, on the one hand, to the homogeneity of the studied material (infantile hydrocephalus only) and, on the other hand, to the neurosurgical experience.
The operations were performed in a specialized centre by a small number of pediatric neurosurgeons, performing 25-50 major shunt procedures and revisions per year. This emphasizes the importance of neurosurgical experience in a relatively simple intervention.

Conclusion

The present study covers a period of almost three decades. It follows up an etiologically homogeneous group of patients – children, shunted for infantile hydrocephalus in a single neurosurgery centre. Age at implantation, as a factor, shows that the youngest patients (shunted under 3 months) have the worst survival trend, have the worst survival trend, which is confirmed by both correlation and InterCriteria analyses. In this group, the percentage of shunts without revisions is the smallest. These patients are the ones who undergo shunt failure first. The analysis of the interval to the first revision shows that patients revised after more than
6 months after implantation have better survival. If a patient survives the first 12 months after shunting without failure, the risk of such is under 50% for the whole shunt's existence.

Our long-standing experience and shunt observation showed that the relatively small group of patients with "3 and more revisions" makes 2/3 of all revisions and it gives shunt bad repute. Exactly this group deserves more careful and deep analysis to reduce it.

Our follow-up results show that for most of the patients with infantile hydrocephalus ventricular shunts are a good treatment method.

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