Jorge A. Lazareff Warwick Peacock Langston Holly Jon Ver Halen Anthony Wong Charles Olmstead

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J. A. Lazareff (⊠) · W. Peacock L. Holly · J. Ver Halen · A. Wong C. Olmstead Division of Neurosurgery, UCLA School of Medicine, Box 957039, Los Angeles, CA 90095-7039, USA e-mail: lazareff@surgery.medsch.ucla.edu Tel.: +1-310-206-6677 Fax: +1-310-794-2147

Introduction

Patients who require multiple shunt revisions (MSR) are well known to pediatric neurosurgeons. Any large series of hydrocephalic patients allows the inference that each patient will have between 1.6 and 3.6 shunt revisions [9, 15]. Numerous studies have been conducted to identify the factor/s responsible for ventricular shunt malfunction [7, 10–12], and very valuable information has been obtained about the relevance of age, etiology and surgeon's experi-

Abstract Ventricular shunts that require multiple revisions are familiar to pediatric neurosurgeons. We conducted a retrospective study to determine whether patients who require repeated shunt revisions represent a particular cohort within shunted hydrocephalic children. The clinical records of 244 children who had undergone shunt procedures between January 1990 and January 1996 were examined. They were divided into group 1: children with no shunt failure (n=136), group 2: children with one shunt revision (n=52), group 3: children with 2 or 3 shunt revisions (n=34), and group 4: patients who had 4 or more shunt revisions (n=22). Patients in groups 3 and 4 accounted for 54.8% of the total of 531 shunt procedures. Etiology of hydrocephalus, nature of the dysfunction, CSF characteristics, and variables related to the surgical procedure were analyzed for each group. We observed a progressive shortening of the intervals between

revisions as the numbers of surgeries increased, indicating that shunts that tended to fail repeatedly did so sooner than those that did not. A Kaplan-Meier shunt survival curve showed that group 2 had a slower rate of failure than either group 3 $(\chi^2 = 7.13, P < 0.01)$ or group 4 $(\chi^2 = 4.76, P < 0.05)$. The etiologies of the hydrocephalus were not randomly distributed among the four groups ($\chi^2 = 81.4$, P < 0.001); there was a predominance of congenital conditions in group 1. Repeated shunt revisions were associated with a progressive increase in the concentration of monocytes in the CSF (Kruskal-Wallis, P<0.05). Our data suggest that multiple shunt revisions constitute a phenomenon that may be caused by specific, still unidentified, biological factors.

Key words Hydrocephalus · Shunt failure · Monocyte · Ventricular catheter

ence to the success of a shunt procedure. However, no attempt has been made to distinguish between patients who have had a single shunt revision and those with multiple revisions.

When the issue of multiple shunt failure is addressed it can be argued that it results from the incidental repetition of the same factors are responsible for single shunt failure. On the other hand, considering that repeated shunt failure seems to be a persistent problem, it is reasonable to hypothesize that there are distinctive patient characteristics that lead to multiple shunt revisions.

Multiple shunt failures: an analysis of relevant factors

In the following retrospective study of 244 patients who had shunted hydrocephalus we identified those who suffered three or more revisions. We asked whether there were unique clinical problems differing from those encountered by patients who had functioning shunts or required only one shunt revision.

Patients and methods

All pediatric patients who had a ventricular shunt placed for hydrocephalus of various etiologies at UCLA Medical Center between January 1990 and January 1996 were identified. In those 244 patients 531 shunt-related surgical procedures were performed.

The patients were grouped as follows: Group 1, patients who did not require a shunt revision; group 2, patients who required only one shunt revision; group 3, patients who had 2–3 revisions; group 4, patients who required 4 or more shunt revisions. For the purpose of this communication, groups 3 and 4 are considered to have had MSR.

For all 244 patients the following data were retrospectively collected from their charts and operative records: age at the time of surgery, gender, etiology of hydrocephalus, surgical procedure(s), duration of surgical procedure(s), surgical team, number of persons circulating in the operating room (OR) during the surgery, characteristics of the CSF collected at every surgical procedure, and the type of shunt device. In cases that required revision of the shunt the age at each procedure, the time to failure and the cause of the malfunction were determined.

The etiologies of the hydrocephalus were grouped as follows: congenital conditions (included aqueductal stenosis, Dandy-Walker, arachnoid cyst, encephalocele and meningomyelocele), tumor, intraventricular hemorrhage (IVH), prior hemispherectomy (for intractable epilepsy), and meningitis. The duration of the surgical procedure was grouped into three categories: <1 h, 1-2 h, >2 h. The chief surgeon was either a full-time pediatric neurosurgeon or a nonpediatric neurosurgeon. The residents who participated in the surgeries were recorded as either senior or junior, according to the stage of their training at the time of the surgeries. The number of persons circulating was grouped as follows: two, three, four, and five or more persons. Four surgery start times were defined; early morning (6:30 a.m. to 8:59 a.m.), late morning (9:00 a.m. to 11:59 a.m.), afternoon (noon till 5 p.m.) and late schedule (5 p.m. till 6:29 a.m.).

For statistical analysis the data collected were cross-tabulated and χ^2 tests were performed. The data relating to the composition of CSF were analyzed with a Kruskal-Wallis test. Calculations of comparative time to shunt failure in groups 2, 3 and 4 were evaluated using the Kaplan-Meier nonparametric survival statistics. In all cases alpha was set at P < 0.05.

Results

Of all patients who underwent ventricular shunt surgery, 55.7% (n=136, group 1) did not require shunt revision during the time frame analyzed; 21.3% (n=52, group 2) had a single revision; 13.9% (n=34, group 3) had two to three revisions, and 9% (n=22, group 4) required four or more shunt revisions. The mean age was 3.6 years (range 1 day to 18 years) in group 1, 4.6 years (range 1 day to 18 years) in group 2, 4.7 years (range 1 day to 16.8 years) in group 3, and 5.3 years (range 1 day to 16 years) in group 4.

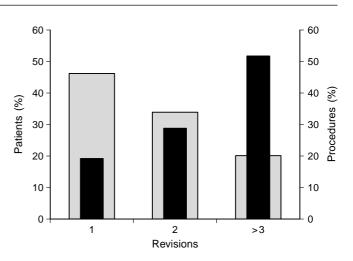


Fig. 1 Percentage of patients (*open bars*) and procedures (*filled bars*) for the four groups in this study

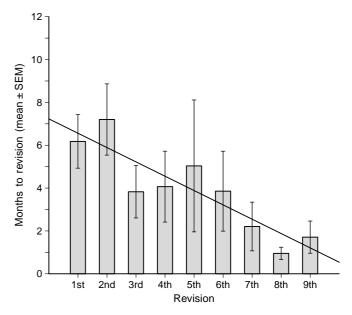


Fig. 2 Months (\pm SEM) to revision for successive failures in patients in the MSR group. There was a significantly negative linear relationship between subsequent shunts and the time to failure (Spearman rs=-0.850; P<0.01)

The distribution of the surgical procedures by group was as follows: group 1, 136; group 2, 104; group 3, 111; group 4, 180. As shown in Fig. 1, patients in the MSR group accounted for 54.8% of all surgical procedures, with an average of 3.26 procedures per patient in group 3 and 8.18 procedures per patient in group 4.

We next analyzed the rate of shunt failure as a function of revision number. As shown in Fig. 2, the interval between revisions became progressively shorter as the number of surgeries increased, indicating that shunts that tended to fail repeatedly did so sooner than those that did not. The Kaplan-Meier shunt survival curves are shown in

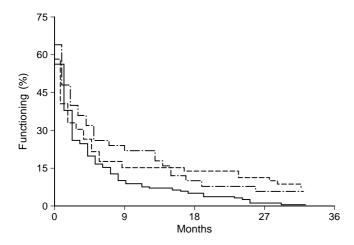


Fig. 3 Survival curves for the first 3 years for the three groups that required revisions. *Solid* 4 or more revisions, *dot*, *dash* 2–3 revisions, *dash* 1 revision. The single-revision group (2) had significantly slower rates of failure than either group 3 (χ^2 =7.13, *P*<0.01) or group 4 (χ^2 =4.76, *P*<0.05). There were no significant differences between groups 3 and 4 (χ^2 =0.081, *P*~0.50)

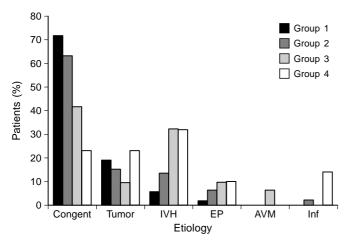


Fig. 4 Percentage distribution of the etiologies for the four groups in this study (*Congent* aqueductal stenosis, meningomyelocele, Dandy Walker; *IVH* intraventricular hemorrhage; *EP* prior hemispherectomy for intractable epilepsy; *AVM* arterial venous malformation; *Inf* prior meningitis)

Fig. 3. The single-revision group (2) showed significantly lower rates of failure than either group 3 ($\chi^2 = 7.13$, P < 0.01) or group 4 ($\chi^2 = 4.76$, P < 0.05). There were no significant differences between groups 3 and 4 ($\chi^2 = 0.081$, $P \sim 0.50$).

The etiologies of the hydrocephalus were not randomly distributed among the four groups ($\chi^2 = 81.4$, P < 0.001). As shown in Fig. 4, congenitally occurring meningomyelocele and aqueductal stenosis were highest in the zero or one revision groups, while IVH, tumor, and infections (postmeningitis) were proportionally more frequent in group 4 patients. All but one of the tumors in the MSR

	Group 1	Group 2	Group 3	Group 4
Surgeon				
Pediatric neurosurgeon	105	78	77	160
Nonpediatric neurosurgeon	31	26	34	20
Circulating personnel ^a				
2	37	33	41	52
3	29	25	35	40
4	41	28	17	54
5 or more	29	18	18	34
Start time				
First case	51	29	28	47
Second case	31	25	25	40
Afternoon	40	34	37	69
Evening	14	16	21	24
Duration				
1 h or shorter	30	22	43	46
1 h to 2 h	64	38	37	60
2 h or longer	42	44	31	74

^a In operating room during surgery

group were supratentorial. A total of 20 patients had shunt infections. All of them were in the MSR groups: 8 in group 3, and 12 in group 4. Groups 3 and 4 had the same infection rate, 8%. Five children in group 3 and only 1 of the group 4 patients had their shunts infected at the first surgical procedure. The other patients in group 4 had their shunts infected at the time of 4th or 7th procedure. The overall infection rate of our four groups was 4.3%. The bacteria responsible were *Staphylococcus* epidermidis, *S. aureus, Escherichia coli* and *Pseudomonas aeruginosa*.

A total of 127 malfunctions were attributed to ventricular catheters, 43 to peritoneal catheters, and 73 to the valve systems. There was a statistically significant difference between proximal malfunction and the other two categories (χ^2 =52.7944; P<0.001). The most common cause of malfunction in the ventricular and peritoneal catheters was obstruction, followed by suboptimal placement and disconnection. With regard to the valve system, insufficient drainage and fracture-leakage were the most commonly encountered problems. Three types of shunt systems were used; distal slit valve, Delta, and PS Medical. There were no statistically significant differences between the type of valve and the frequency or nature of the shunt malfunction. There were 5 ventriculoatrial shunts and 4 ventriculopleural shunts, all in group 4 patients. The indication for extraperitoneal shunting was the same in all cases: a nonabsorbing peritoneum. Table 1 summarizes the analyses of factors directly related to the surgical procedure. None of these factors was significantly associated with an increased risk of shunt malfunction.

There were no significant differences among the four groups in the concentration of protein, lymphocytes or eosinophils in the CSF (data not shown). In contrast, repeated

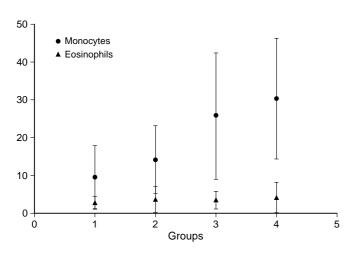


Fig. 5 Mean±SD of monocytes and eosinophiles in the CSF of the four groups of this study. Values are percentages of white cells. A progressive increase in monocyte concentration is observed. For monocytes a significant difference between groups is observed (Kruskal-Wallis P < 0.05)

shunt revisions were associated with a progressive increase in the concentration of monocytes in the CSF (Kruskal-Wallis P < 0.05; Fig. 5).

Discussion

A small proportion of hydrocephalic children required the largest number of shunt procedures at our institution. Interestingly, after the second revision the time span between revisions shortened progressively. These observations suggest that factors outside the surgical procedure itself, and possibly related to an ongoing reactive process involving the brain parenchyma, may be responsible for the significantly shorter survival rate in the MSR group.

In the majority of patients in the MSR group hydrocephalus was secondary to diverse pathologies, among which tumors and intraventricular hemorrhage clearly predominated. That children with tumor and hydrocephalus are likely to present shunt obstruction is well known [14], and this is probably due to the particular distribution of supratentorial pediatric brain tumors that usually develop adjacent to the lateral ventricles. In our series, most of the MSRassociated tumors were supratentorial, in contrast to the situation of patients in group 1, who had a larger number of tumors that caused hydrocephalus by invading the IV ventricle. Therefore, it is reasonable to suggest that the anatomical integrity of the lateral ventricles is the principal factor in the success of shunt surgery in a patient with a brain tumor. Neonatal IVH was a significant factor in shunt malfunction, which in part can be ascribed to age [10]; nonetheless, it does not explain the repetition of failures also observed as the patients grew older. Finally, children shunted after hemispherectomy for medically intractable seizures also belonged to the MSR group.

It appears that the common denominator for the cohort of MSR patients is a postnatal supratentorial brain injury. Past experimental evidence shows that an interaction between abnormal periventricular astrocytes and CSF takes place after the ependymal wall is disrupted by the hydrocephalus [3-5]. Therefore, is reasonable to hypothesize that when the histological reactive process elicited by the primary pathology interacts with the ventricular catheter the chances of shunt obstruction are increased. The inflammatory nature of this process will not be altered by replacement of the catheter; on the contrary, the procedure may trigger a set of events that will cause repeated shunt obstructions with a shorter time span between episodes. We are aware that only a prospective study analyzing the components of the tissue that obstruct the catheter will prove or disprove our assumption. Nevertheless, it is worth noting that some observations reported in our series are in keeping with the above hypothesis. First, as previously reported [7], a significant proportion of shunt failures were due to obstruction of the ventricular catheter. Traditionally the choroid plexus is considered the culprit in the case of this complication [6], but it can be argued that after three or more revisions the remnant choroid tissue in the vicinity of the foramen of Monro may not be sufficient to obstruct the catheter; more importantly, reactive glia, ependyma or "debris" was identified as the main component of the catheter plug in a large number of cases [13]. Second, parallel to the number of revisions there is a progressive increase, albeit mostly within the normal range, in the monocyte concentration in the CSF; considering the physiological role of the monocytes, this finding is suggestive of a continuous inflammatory process elicited by the original pathology and perpetuated by repeated shunt revisions. This inflammatory process was specific for only one cell type, not affecting the concentration of lymphocyte or eosinophils; the latter remained below the level considered relevant for shunt infection [15]. At our institution we place the ventricular catheter through a posterior parietal burr hole, and although some authors have advocated the advantage of the coronal approach to prevent catheter obstruction [11], the issue is still not resolved [1, 2].

Of relevance for distal shunt obstruction in the MSR group were two children whose hydrocephalus was a consequence of fungal meningitis. The clinical management of these patients was difficult. In view of the length of the treatment we could not place an external ventricular drain, and although at the time of shunt placement the CSF had close to normal characteristics, the peritoneum and the pleura reacted to the intratechal presence of the histoplasm or to the fluconazole that was injected into the Rickham reservoir of the shunt system. Fortunately the infection has been controlled and neither child had impaired cognitive delay; their current VA shunts are working adequately with no evidence of systemic consequences. When trying to identify the characteristics of the multiple failure group, we followed previous studies of shunt malfunction that analyze the relevance of factors that are not directly related to hydrocephalus. Under that light, the surgeon's experience and a series of events that may hamper its performance were scrutinized. The surgeon's experience is considered relevant to shunt outcome in adult hydrocephalic patients [8], but was not a factor in a pediatric series [10]. Our results seem to support the view that experienced pediatric neurosurgeons are not of pivotal importance for the outcome of shunt surgery. Nevertheless, it has to be taken into consideration that the surgeons grouped in the other group from the two pediatric neurosurgeons are faculty members of our institution. The larger number of surgeries in group 4 that were performed by one of the two experienced pediatric neurosurgeons may correlate with the intrinsic difficulty of those cases. The other variables that were analyzed, such as time of the day when the surgery was performed and number of OR personnel present at the time of the procedure, may be significant for the incidence of shunt infection.

Our data support the impression that repeated obstruction of the ventricular catheter is not a serendipitous phenomenon. Enhanced understanding of the biological processes that take place in the brain parenchyma of a patient with secondary hydrocephalus will favor the prevention of multiple shunt failures.

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