

A mathematical model of survival in a newly inserted ventricular shunt

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Object. The object of this study was to mathematically model the prognosis of a newly inserted shunt in pediatric or adult patients with hydrocephalus.

Methods. A structured search was performed of the English-language literature for case series reporting shunt failure, patient mortality, and shunt removal rates after shunt insertion. A metaanalytic model was constructed to pool data from multiple studies and to predict the outcome of a shunt after insertion. Separate models were used to predict shunt survival rates for children (patients < 17 years old) and adults.

Results. Shunt survival rates in children and adults were calculated for 1 year (64.2 and 80.1%, respectively), 5 years (49.4 and 60.2%, respectively), and the median (4.9 and 7.3 years, respectively). The longer-term rates predicted by the model agree closely with those reported in the literature.

Conclusions. This model gives a comprehensive view of the fate of a shunt for hydrocephalus after insertion. The advantages of this model compared with Kaplan–Meier survival curves are discussed. The model used in this study may provide useful prognostic information and aid in the early evaluation of new shunt designs and techniques. (DOI: 10.3171/PED-07/12/448)

KEY WORDS • cerebrospinal fluid shunt • hydrocephalus • pediatric neurosurgery • shunt failure • shunt malfunction

ALTHOUGH ventricular shunts have been remarkably effective in treating hydrocephalus, their rate of failure remains quite high. Yet it is difficult to predict the fate of the average patient in whom a shunt is placed. There are several causes for shunt failure, including under- and overdrainage, mechanical blockage, valve failure, disconnection, and infection. Follow-up durations vary in published studies of ventricular shunts. Many case series are hampered by incomplete follow-up, because not all patients return to the same institution in which shunt insertion occurred. Shunt-related complications occur at varying rates in different series, in part due to different shunt devices, surgical practices, and definitions of complications. Rates of complication and death also vary according to patient age and the diseases causing the hydrocephalus. Some patients never require shunt revision, whereas others need several revisions in a single year. Thus, despite experience with many thousands of shunt procedures, physicians find it difficult to find reliable predictions when counseling patients and their families on the prognoses of newly implanted shunts.

The purpose of this report is to simulate the outcome of a recently placed shunt over time in the average child or adult with hydrocephalus. These projections are compared with measurements reported in the literature.

Clinical Material and Methods

We performed a structured literature search of English-language articles in the MEDLINE database for the years between 1950 and 2007 (March), using the key words “hydrocephalus,” along with various combinations of the key words “treatment,” “shunt” (“shunts,” “shunted,” “shunting”), “complications,” “mortality,” and “death.” This search was supplemented with additional references from the bibliographies of articles previously read by the authors and by using the “Related Articles” feature of PubMed. We excluded animal studies, editorials, case reports, letters to the editor, reviews, and studies duplicating data from previous reports. This process yielded 1993 publications, which were reviewed for data relating to rates of shunt complication, removal, and replacement, or patient mortality rates. In the 126 case series used in this study, one or more of these rates were reported or could be calculated from the data given.

We abstracted estimates of the probability of patient death, shunt failure, and shunt removal. Data for children (Table 1) and adults (Table 2) were pooled separately. Following the convention in the literature, we used the age of 17 years as the separation between childhood and adulthood. The reported point estimates of pooled data represent

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TABLE 1

*Abstracted estimates of the probability of death, shunt failure, and shunt removal in pooled data for children**

Category	No. of Patients	Probability	95% CI	Reference No.
mortality rate at yr 1	5,574	0.0410	0.0355–0.0465	1,4,23,52,54,61,62,99,125,130
annual mortality rate after yr 1	2,236	0.0141	0.0086–0.0251	
shunt failure rate at yr 1	18,213	0.3128	0.2579–0.3678	1–3,8,9,11,20,21,23,24,26–28,31,33,35,39–42,44,46,
annual shunt failure rate after yr 1	7,150	0.0454	0.0307–0.0600	47,49,50,57,59,62,63,68,74,76,77,85,87,88,93,96– 98,101,103,107–110,124,130,132,136
annual shunt removal rate	1,976	0.0039	0.0014–0.0064	33,51,56,134

* Rate = perioperative + annual rate. Abbreviation: CI = confidence interval.

variance-weighted means and were tested for heterogeneity.^{38,67} The rate of shunt removal in adults is so low that it is considered zero for modeling purposes.^{51,71} All probabilities that vary over time (such as mortality rates) were corrected for rates in 2000, the latest year for which accurate data are available.

We calculated the rate of shunt failures in the first year and annual failure rates for subsequent years, where available. In studies not listing delayed complication rates explicitly, we estimated these rates from the number of delayed shunt failures and the mean number of patient-years beyond 1 year on follow-up (for each study, the former number divided by the latter equals the mean annual failure rate). In reports employing Kaplan–Meier survival curves, calculating the mean annual failure rate usually involved manually calculating the number of subjects at risk for shunt failure and the number of shunt failures at each step in the published curves.

We used Markov modeling techniques¹¹⁶ to simulate the course of patients undergoing shunt insertion. These techniques enabled us to follow the progress of a simulated cohort of patients. The possible pathways and outcomes after shunt insertion are shown in Fig. 1. During any Markov model cycle (in this case, 1 year), a patient in whom a shunt is placed may die. If he or she survives, his or her shunt may continue to work normally, fail, or be removed for arrested hydrocephalus. The probability of a shunt or patient transitioning from one state to another is determined by the pooled estimates in Tables 1 and 2. During each cycle, these probabilities are applied to patients in each state, and new percentages are calculated. The model runs until the proportion of functioning shunts approaches zero. A multistate, time-dependent, transition decision model was constructed using the TreeAge Pro 2006 software package (TreeAge

Software). This transition decision model was run separately for children and adults, because of their different transition probabilities.

Results

The state of a shunt inserted into a child, predicted by our model at any point in time, is illustrated in Figure 2. Failure and mortality rates are highest in the first year, and decrease thereafter. Figure 3 shows the same categories for shunt insertions in adults. Even though patient deaths are greater in adults with shunt insertions, shunts in adults fail more slowly and tend to survive longer than those in children. Median shunt survival times, as well as 1- and 5-year shunt survival rates, agree closely with estimates from the literature.

Discussion

Our model illustrates the relative brevity of shunt survival, both in children and adults. Shunt failure is the greatest contributor to loss of a functioning shunt. In addition, the underlying illnesses and the risk of perioperative complications during shunt insertion contribute to patient mortality rates.

A number of case series have examined shunt longevity and yielded varied results. Our model yields a 1-year shunt survival rate in children of just over 64%, a rate that is well within the range reported in most case series. Median shunt survival predicted by our model is approximately 5 years in children; estimates in the literature range from less than 1 year to more than seven, varying with children's ages, hydrocephalus origins, and other factors.^{2,23,27,31,35,39,60,62,74,75,88,93,101,108,110,136} Although new shunts have a somewhat longer

TABLE 2

*Abstracted estimates of the probability of death, shunt failure, and shunt removal in pooled data for adults**

Category	No. of Patients	Probability	95% CI	Reference No.
mortality rate at yr 1†	7809	0.0365	0.0269–0.0461	10,12,13,16,18,19,22,30,43,48,58,69,70,72,73,82,90, 99,100,102,105,106,112–114,117,123,127,128, 131,133,135
annual mortality rate after yr 1	384	0.0166	0.0070–0.0260	6,16,83,92,95,113
shunt failure rate at yr 1	3879	0.1625	0.1109–0.2139	5,7,10,12,13,15–19,22,25,29,30,32,34,37,43,45,48,
annual shunt failure rate after yr 1	1687	0.0522	0.0296–0.0749	50,53,55,58,64–66,68–70,72,73,78–82,84,89–91, 94,95,102,104–106,111,112,114,115,117,118,120, 121,123,126,129,131,133,135,137

* Shunt removal rate probability is effectively 0 in adults.

† Rate = perioperative + annual rate.

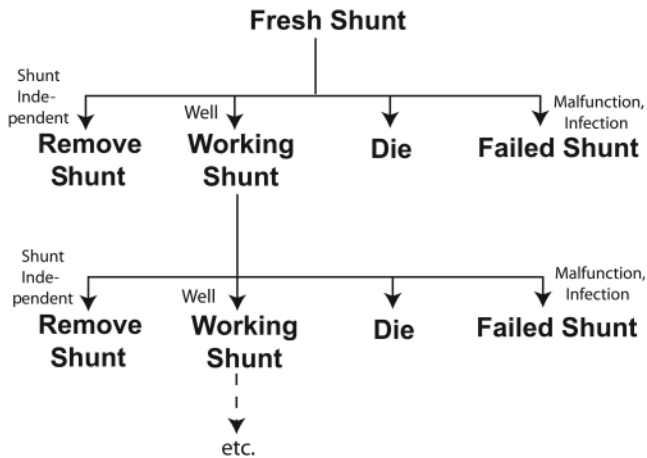


FIG. 1. Diagram of the possible states and transitions of a shunt and the patient in the Markov model. During any 1-year cycle, a newly inserted shunt may fail, be removed, or continue working. The patient may die of natural or shunt-related causes. This cycle continues in the Markov model until no working shunts are left.

survival rate in adults, the disparity of rates among case series is also quite large.

Many of the studies cited employ Kaplan–Meier curves to calculate shunt survival. Although use of these curves is a generally accepted approach, it has severe limitations.¹⁴ The number of patients in most of the trials is relatively small and the hydrocephalus origins vary. For example, some studies include only neonates or infants, whereas others do not include children with brain tumors; these factors may alter measured shunt survival rates. It is not always clear which events lead to study censorship (exclusion from later analysis times), shunt failure, shunt removal, loss or withdrawal from follow-up, or patient death.^{36,119} There are additional technical issues, and the Kaplan–Meier approach

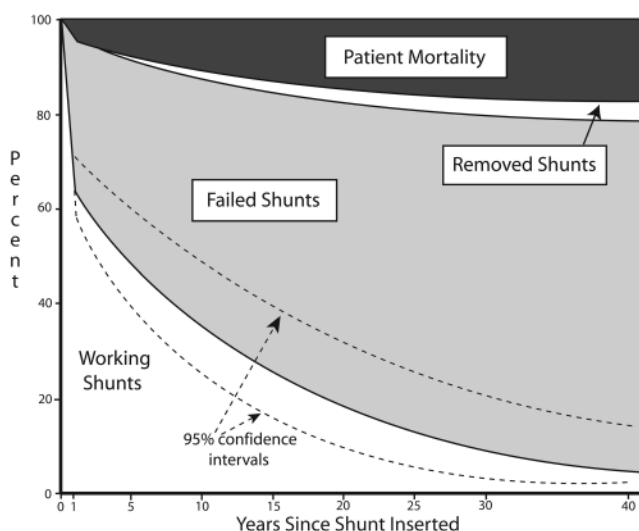


FIG. 2. Graph of the predicted survival rates of a newly inserted shunt in a child. At each point in time, the graph illustrates the relative proportion of shunts that remain functional (with 95% CIs), fail, or are removed. Also shown are the percentages of patients treated using shunts, who later died.

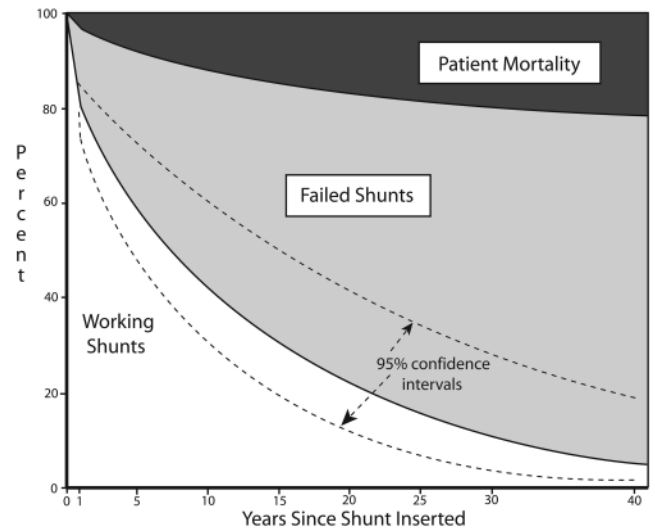


FIG. 3. Graph of the predicted survival rates of a newly inserted shunt in an adult. At each point in time, the graph shows the relative proportion of shunts that remain functional (with 95% CIs) or that fail. Also shown are the percentages of patients treated using shunts, who later died.

makes several assumptions that may render it inappropriate for tracking shunt survival.^{86,122} These other issues include the dwindling number of patients in the study over time, rendering the technique least certain and hence least useful in predicting long-term events. Other shortcomings of the Kaplan–Meier technique include potential biases caused by handling competing risks (other than, as in this study, shunt failure), difficulty timing failures, or censored events. Survival rates cannot be pooled from multiple studies using Kaplan–Meier methodology.

The Markov model is dynamic and can project events into the future.¹¹⁶ This model can serve as a guide for explaining shunt prognosis to patients and families before surgery. It can also be applied as a realistic baseline against which to compare the results of pilot studies in new shunt devices, experimental techniques, and other potential advances in shunt preservation. Investigators can get a sense of whether their preliminary results are superior to standard shunt survival before investing considerable time and effort on multicenter trials. This model is limited, as are all such mathematical approaches, by the quality of the underlying data. Pooling so many case series drawn from different times, geographic locales, shunt techniques, and patient populations has a homogenizing effect and minimizes differences.

Conclusions

Although many factors influence shunt survival, it is possible to model the prognosis of the average shunt inserted into a child or adult and to predict future behavior of the shunt. The results calculated for our models are well within the ranges cited in the literature.

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