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Comparing ventriculoatrial and ventriculopleural shunts in pediatric hydrocephalus: a Hydrocephalus Clinical Research Network study

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OBJECTIVE When the peritoneal cavity cannot serve as the distal shunt terminus, nonperitoneal shunts, typically terminating in the atrium or pleural space, are used. The comparative effectiveness of these two terminus options has not been evaluated. The authors directly compared shunt survival and complication rates for ventriculoatrial (VA) and ventriculopleural (VPI) shunts in a pediatric cohort.

METHODS The Hydrocephalus Clinical Research Network Core Data Project was used to identify children \leq 18 years of age who underwent either VA or VPI shunt insertion. The primary outcome was time to shunt failure. Secondary outcomes included distal site complications and frequency of shunt failure at 6, 12, and 24 months.

RESULTS The search criteria yielded 416 children from 14 centers with either a VA (n = 318) or VPI (n = 98) shunt, including those converted from ventriculoperitoneal shunts. Children with VA shunts had a lower median age at insertion (6.1 years vs 12.4 years, p < 0.001). Among those children with VA shunts, a hydrocephalus etiology of intraventricular hemorrhage (IVH) secondary to prematurity comprised a higher proportion (47.0% vs 31.2%) and myelomeningocele

ABBREVIATIONS DVT/PE = deep vein thrombosis/pulmonary embolism; HCRN = Hydrocephalus Clinical Research Network; IVH = intraventricular hemorrhage; VA = ventriculopatrial; VP = ventriculoperitoneal; VPI = ventriculopleural. SUBMITTED January 29, 2024. ACCEPTED May 15, 2024.

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comprised a lower proportion (17.8% vs 27.3%) (p = 0.024) compared with those with VPI shunts. At 24 months, there was a higher cumulative number of revisions for VA shunts (48.6% vs 38.9%, p = 0.038). When stratified by patient age at shunt insertion, VA shunts in children < 6 years had the lowest shunt survival rate (p < 0.001, log-rank test). After controlling for age and etiology, multivariable analysis did not find that shunt type (VA vs VPI) was predictive of time to shunt failure. No differences were found in the cumulative frequency of complications (VA 6.0% vs VPI 9.2%, p = 0.257), but there was a higher rate of pneumothorax in the VPI cohort (3.1% vs 0%, p = 0.013).

CONCLUSIONS Shunt survival was similar between VA and VPI shunts, although VA shunts are used more often, particularly in younger patients. Children < 6 years with VA shunts appeared to have the shortest shunt survival, which may be a result of the VA group having more cases of IVH secondary to prematurity; however, when age and etiology were included in a multivariable model, shunt location (atrium vs pleural space) was not associated with time to failure. The baseline differences between children treated with a VA versus a VPI shunt likely explain current practice patterns.

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KEYWORDS ventriculoatrial shunt; ventriculopleural shunt; pediatric; Hydrocephalus Clinical Research Network; shunt failure; outcomes

N some children who undergo CSF shunt insertion for the treatment of hydrocephalus, the peritoneal cavity, which is the most common distal shunt catheter placement site,^{1,2} may not be viable because of concerns for necrotizing enterocolitis, congenital gastrointestinal conditions, history of extensive abdominal surgery, adhesions due to peritoneal scarring, abdominal pseudocysts, intraperitoneal infections, and ascites.³⁻⁵ In these patients, nonperitoneal sites are necessary to accomplish successful CSF diversion. The optimal location for the shunt terminus in patients with a nonviable peritoneal cavity is not known. Gmeiner et al.⁶ evaluated 61 patients who received ventriculoatrial (VA) shunts and determined that the atrium is an appropriate alternative for children who require shunt placement. Oyon et al.⁷ and Christian et al.⁸ have provided evidence that ventriculopleural (VPI) shunts are another option when ventriculoperitoneal (VP) shunt insertion is not possible.

The complication profile for VPI shunts includes pleural effusions secondary to the smaller absorptive surface area, particularly in children < 10 years of age.⁸ Similarly, VPI shunts are not used in children with baseline lung disease or diminished lung capacity in the setting of severe spinal deformity. Complications for VA shunts include venous thrombosis requiring anticoagulant or antiplatelet administration, endocarditis, immune complex–mediated shunt nephritis, and the potential for bloodstream infection requiring shunt externalization or explantation. Additionally, VA shunts may require periodic distal revision or lengthening procedures to keep the tip within the atrium.⁸ VA shunts also commonly require the support of a pediatric surgeon to assist with obtaining access.

Shunt survival, revision rates, and complication profiles of the VA and VPI distal implantation sites have not been directly compared. Additionally, although risk factors for VP shunt failure are well known, they remain unknown for VPI and VA shunts. In this study, we compared cohorts of children with hydrocephalus and either a VA or a VPI shunt. We investigated whether either distal catheter location was associated with time to shunt failure (revision or infection) after adjusting for age and hydrocephalus etiology. We hypothesized that 1) there is no difference in shunt survival between these two common nonperitoneal shunt sites, and 2) differences in baseline factors influence the decision to place either a VA or a VPl shunt.

Methods

Patient Identification

Data were extracted from the prospective Hydrocephalus Clinical Research Network (HCRN) Core Data Project (registry) for all children with either a VA or a VPI shunt who were treated between April 2008 and January 2023 at 14 HCRN centers (Children's of Alabama, Birmingham, AL; Primary Children's Hospital, Salt Lake City, UT; Seattle Children's Hospital, Seattle, WA; Children's Hospital of Pittsburgh, Pittsburgh, PA; St. Louis Children's Hospital, St. Louis, MO; Texas Children's Hospital, Houston, TX; The Hospital for Sick Children, Toronto, ON, Canada; Monroe Carell Jr. Children's Hospital at Vanderbilt, Nashville, TN; British Columbia Children's Hospital, Vancouver, BC, Canada; Alberta Children's Hospital, Calgary, AB, Canada; Children's Hospital of Los Angeles, Los Angeles, CA; Children's Hospital Colorado, Aurora, CO; Nationwide Children's Hospital, Columbus, OH; and Johns Hopkins Children's Center, Baltimore, MD). The registry tracks all hydrocephalus-related shunt surgeries at each center beginning on the date the center joined HCRN. Institutional review board approval with a waiver of individual patient consent was obtained from each clinical site as well as the data coordinating center.9

Pediatric patients (\leq 18 years of age) with first-time VA or VPI shunt placement were included. We included children whose first shunt was a VA or VPI shunt and also children in the database who had at least one earlier peritoneal shunt that was converted to a VA or VPI shunt. Patients were excluded if they had a shunt in a nonatrial or nonpleural terminus.

Úsing the registry, we also identified a cohort of children with VP shunts who had at least one previous shunt revision and the same median number of shunt surgeries. This cohort was entered into a multivariable model to determine whether any shunt terminus was associated with failure when compared with our cohort of children with VA and VPl shunts. For this analysis, we included all procedures through January 14, 2023, allowing for at least 6 months of follow-up, unless a subject had a shunt failure or censor event.

Data Collection

Demographic characteristics collected included sex, race/ethnicity, and age (at initial VA or VPI shunt implantation), which was categorized into four groups (0-2, 3-5, -5)6-12, and > 12 years). Clinical factors included etiology of hydrocephalus (intraventricular hemorrhage [IVH] secondary to prematurity, myelomeningocele, aqueductal stenosis, or other etiology), number of previous shunt surgeries, time from last shunt operation to implant at nonperitoneal site, first shunt insertion versus conversion from VP shunt, use of an endoscope to place the ventricular catheter, imaging guidance, antisiphon device use, comorbid cardiac conditions, complex chronic conditions, presence of gastrostomy tube, adherence to HCRN protocol during nonperitoneal shunt insertion, whether previous shunt surgery was performed within 12 weeks, and complications before discharge.

Factors collected from the failure surgery included the location of malfunction (distal, proximal, both, or valve), the type of failure for the VA or VPI shunt (revision or infection), and the next shunt surgery terminus (peritoneal, pleural, atrial, or other).

Outcomes

The primary outcome for the study was time to shunt failure (years). Secondary outcomes included number of shunt revisions (any hydrocephalus-related surgical procedures, including multiple procedures per patient) by 6-, 12-, and 24-month time points; type of shunt failure (revision or infection); location of shunt failure (proximal or distal); and hydrocephalus-related perioperative complications before hospital discharge (ascites, cardiac arrest, CSF leak, deep vein thrombosis/pulmonary embolism [DVT/ PE], bacterial meningitis, hyponatremia, motor deficit, visual/ocular deficit, pneumonia, pneumothorax, IVH, subdural hematoma, pressure sore, pseudomeningocele, seizure, urinary tract infection, wound problem, and sepsis).

Statistical Analysis

The primary analysis compared time to first failure of VA and VPI shunts using Kaplan-Meier curves. Risk factors for VA or VPI shunt failure were assessed with a Cox proportional hazards model. Variables included in the model were those with p < 0.2 on univariable analysis or those identified as clinically important based on earlier studies (e.g., recent revision, use of endoscope for ventricular catheter placement, and cardiac comorbidity). The proportional hazards assumption was plausible. Interacting variables were tested. An interaction between age and VA or VPI shunt was included in the model.

Several secondary analyses were also performed. To identify factors associated with the decision to place either a VPl or a VA shunt, we compared demographic and baseline clinical and hydrocephalus-related variables. Descriptive statistics were used to summarize patient demographics and outcome measures and are reported as counts and percentages for categorical variables and as



FIG. 1. CONSORT diagram for the study.

the median [first quartile, third quartile] for continuous variables. Associations among continuous variables were assessed using a Wilcoxon rank-sum test. All categorical variables were compared using Fisher's exact test. Lastly, a multivariable analysis was performed to examine time to shunt failure; this analysis included the group of patients with VP shunts identified in the registry. All analyses were conducted using SAS 9.4 (SAS Institute). Significance levels were set at p < 0.05.

Results

At the time of the investigation, data for 11,969 shunt patients from 14 North American centers were available in the registry. Among them, data existed for 491 children (4.1%) with nonperitoneal shunts; 416 children within the network were included in the analysis as having a firsttime VA or VPI shunt either as the first-line shunt or after conversion from a VP shunt. Twenty-two subjects had censor events and were not included in the number of failures within 6 months because they lacked 6-month follow-up. Of these, 7 patients relocated out of network, 3 transitioned to adult care, and 12 died, including 6 within 30 days of surgery. Only one of the deaths was related to hydrocephalus. The CONSORT (Consolidated Standards of Reporting Trials) diagram is shown in Fig. 1.

Demographics

Overall in the HCRN, among children with nonperitoneal shunts, VA shunts were more common than VPI shunts (318 VA, 98 VPI) (Table 1). Children who received VA shunts had a lower median age at insertion (6.1 years vs 12.4 years, p < 0.001) and a lower proportion were \geq 10 years of age (32.4% vs 62.2%, p < 0.001) when compared with children with VPI shunts. When examining the age groupings, we found that most VPI shunts were placed in children \geq 6 years (77.6%) whereas 50.9% of VA shunts were placed in children \geq 6 years, resulting in a useful natural cutoff within our dataset. No significant differ-

Variable	Overall (n = 416)	VA (n = 318)	VPI (n = 98)	p Value
Cohort				0.001‡
Comprehensive*	155 (37.3)	132 (41.5)	23 (23.5)	
Noncomprehensive†	261 (62.7)	186 (58.5)	75 (76.5)	
Male sex	237 (57.0)	175 (55.0)	62 (63.3)	0.163‡
Age at time of procedure, yrs	7.8 [1.4, 13.1]	6.1 [1.0, 11.6]	12.4 [6.8, 14.9]	<0.001§
Age ≥10 yrs	164 (39.4)	103 (32.4)	61 (62.2)	<0.001‡
Age range at implant, yrs	, ,			<0.001§
0-2	134 (32.2)	121 (38.1)	13 (13.3)	
3–5	44 (10.6)	35 (11.0)	9 (9.2)	
6–12	110 (26.4)	87 (27.4)	23 (23.5)	
>12	128 (30.8)	75 (23.6)	53 (54.1)	
Age <6 yrs	178 (42.8)	156 (49.1)	22 (22.4)	<0.001‡
Race, collapsed	n = 376	n = 296	n = 80	>0.999‡
White	252 (67.0)	198 (66.9)	54 (67.5)	
Black or African American	108 (28.7)	85 (28.7)	23 (28.8)	
Other	16 (4.3)	13 (4.4)	3 (3.8)	
Ethnicity				0.582‡
Not Hispanic or Latino	327 (78.6)	259 (81.4)	68 (69.4)	· · ·
Hispanic or Latino	50 (12.0)	38 (11.9)	12 (12.2)	
Unknown or not reported	39 (9.4)	21 (6.6)	18 (18.4)	
Etiology of hydrocephalus	n = 358	n = 281	n = 77	0.024‡
IVH secondary to prematurity	156 (43.6)	132 (47.0)	24 (31.2)	
Myelomeningocele	71 (19.8)	50 (17.8)	21 (27.3)	
Aqueductal stenosis	19 (5.3)	17 (6.0)	2 (2.6)	
Other etiology	112 (31.3)	82 (29.2)	30 (39.0)	
Shunt procedure type	X /			0.291‡
Primary, 1st-time shunt placement	38 (9.1)	33 (10.4)	5 (5.1)	
Secondary shunt revisions, no infection	156 (37.5)	117 (36.8)	39 (39.8)	
Secondary shunt placement, infection	222 (53.4)	168 (52.8)	54 (55.1)	
Presence of antisiphon device	71/386 (18.4)	55/291 (18.9)	16/95 (16.8)	0.761‡
Time from previous shunt surgery, wks	8.0 [3.0, 33.0]	7.0 [3.0, 29.0]	10.0 [4.5, 43.5]	0.200§
Previous shunt surgery	n = 295	n = 227	n = 68	0.007‡
Primary	58 (19.7)	52 (22.9)	6 (8.8)	
Revision	189 (64.1)	135 (59.5)	54 (79.4)	
Infection	48 (16.3)	40 (17.6)	8 (11.8)	
No. of previous shunt surgeries in HCRN registry	1 [1, 2]	1 [0, 2]	2 [1, 2]	0.936§
CCCs				
Cardiovascular	63 (15.1)	53 (16.7)	10 (10.2)	0.147‡
Neuromuscular	162 (38.9)	116 (36.5)	46 (46.9)	0.075‡
Respiratory	70 (16.8)	60 (18.9)	10 (10.2)	0.046‡
Renal	17 (4.1)	12 (3.8)	5 (5.1)	0.563‡
Gastrointestinal	38 (9.1)	32 (10.1)	6 (6.1)	0.316‡
Hematology &/or immunodeficiency	7 (1.7)	5 (1.6)	2 (2.0)	0.670‡
Metabolic	9 (2.2)	7 (2.2)	2 (2.0)	>0.999‡
Congenital or genetic defect	60 (14.4)	43 (13.5)	17 (17.3)	0.411‡
Non-CNS malignancies	9 (2.2)	8 (2.5)	1 (1.0)	0.692‡
No. of CCCs	. ,	. ,	. ,	0.621‡
0	155 (37.3)	125 (39.3)	30 (30.6)	· ·
1	146 (35.1)	102 (32.1)	44 (44.9)	

TABLE 1. General characteristics of children with nonperitoneal distal shunt terminus

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TABLE 1. General characteristics of children with nonperitoneal distal shunt terminus

Variable	Overall (n = 416)	VA (n = 318)	VPI (n = 98)	p Value
No. of CCCs (continued)				
≥2	115 (27.6)	91 (28.6)	24 (24.5)	
Ultrasound	63 (15.1)	49 (15.4)	14 (14.3)	0.873‡
Stereotaxis	63 (15.1)	48 (15.1)	15 (15.3)	>0.999‡
Endoscopy	41 (9.9)	35 (11.0)	6 (6.1)	0.179‡
HCRN protocol followed during NPS insertion	115/158 (72.8)	92/115 (80.0)	23/43 (53.5)	0.001‡
Antibiotic catheters placed				
Proximal	163 (39.2)	123 (38.7)	40 (40.8)	0.724
Distal	221 (53.1)	151 (47.5)	70 (71.4)	<0.001
Distal & proximal	114 (27.4)	79 (24.8)	35 (35.7)	0.039

CCC = complex chronic condition; NPS = nonperitoneal shunt.

Values are reported as number of patients (%) or median [IQR] unless otherwise indicated.

* Subject's entire shunt history is known and within the HCRN registry.

† Entire shunt history is not within the HCRN registry, but at least one shunt surgery is within the registry.

‡ Cochran-Armitage trend test.

§ Fisher's exact test.

ences were seen in race or ethnicity with respect to shunt terminus.

Among children with VA shunts, those with a hydrocephalus etiology of IVH secondary to prematurity made up a higher proportion (47.0% vs 31.2%) and those with myelomeningocele a lower proportion (17.8% vs 27.3%) when compared with children with VPl shunts (p = 0.024). For 38 children, VA or VPl shunt placement was the primary (initial) shunt surgery for the patient. The median time from previous shunt surgery was similar (VA 7 weeks vs VPl 10 weeks, p = 0.2). For those in whom the VA or VPI shunt placement was not the initial shunt surgery, infection as the reason for previous shunt failure was higher in VA than in VPl shunt subjects (17.6% vs 11.8%, p = 0.007). There was no significant difference in the median number of previous shunt surgeries (VA 1 vs VPl 2, p = 0.936). No differences were seen in the presence of cardiovascular complex chronic conditions or the total number of complex chronic conditions between VA and VPl shunt subjects. Children who received VA shunts had a higher proportion of respiratory complex chronic conditions (18.9% vs 10.2%, p = 0.046).

No differences were seen between VA and VPI shunt surgery in the use of ultrasound (p = 0.873), stereotaxis (p > 0.999), or endoscopy (p = 0.179) for ventricular catheter placement. HCRN shunt protocols were followed in a higher proportion of children who received VA shunts (80% vs 53.5%, p = 0.001). Antibiotic-impregnated catheters were used more often in patients receiving VPI shunts (35.7% vs 24.8%, p = 0.039); further analysis demonstrated this was driven by disparities in distal (VPI 71.4% vs VA 47.5%, p < 0.001) rather than proximal (VPI 40.8% vs VA 38.7%, p = 0.724) antibiotic catheter use.

Complications occurred infrequently in both groups (VA 6.0% vs VPI 9.2%, p = 0.257) (Table 2). Specifically for VPI shunts, pneumothorax occurred in 3 patients (3.1%) and no instances of pneumonia were discovered. DVT/PE occurred in 1 child (0.3%) with a VA shunt, and sepsis occurred in 2 children (0.6%) with a VA shunt.

Shunt Survival

Univariable analysis of VA versus VPI shunt survival demonstrated that age < 6 years and etiology of hydrocephalus had unadjusted associations with shunt survival (Table 3). Kaplan-Meier curves stratified by age showed

TABLE 2. Complications for each cohort

Variable	VA (n = 318)	VPI (n = 98)	p Value
Complication occurred	19 (6.0)	9 (9.2)	0.257
Ascites	3 (0.9)	0 (0.0)	>0.999
Cardiac arrest	1 (0.3)	1 (1.0)	0.416
CSF leak	2 (0.6)	1 (1.0)	0.554
Minor CSF leak	1 (0.3)	0 (0.0)	>0.999
Major CSF leak	2 (0.6)	1 (1.0)	0.554
DVT/PE	1 (0.3)	0 (0.0)	>0.999
Documented bacterial menin-	2 (0.6)	0 (0.0)	>0.999
gitis, positive CSF culture			
Hyponatremia	1 (0.3)	1 (1.0)	0.416
Motor deficit	1 (0.3)	0 (0.0)	>0.999
Visual/ocular deficit	1 (0.3)	0 (0.0)	>0.999
Pneumonia	1 (0.3)	0 (0.0)	>0.999
Pneumothorax	0 (0.0)	3 (3.1)	0.013
IVH	3 (0.9)	0 (0.0)	>0.999
SDH	1 (0.3)	0 (0.0)	>0.999
Pressure sores	0 (0.0)	1 (1.0)	0.236
Pseudomeningocele	2 (0.6)	0 (0.0)	>0.999
Seizure	4 (1.3)	2 (2.0)	0.629
Sepsis	2 (0.6)	0 (0.0)	>0.999
UTI	1 (0.3)	0 (0.0)	>0.999
Wound problem	2 (0.6)	0 (0.0)	>0.999

SDH = subdural hematoma; UTI = urinary tract infection.

Values are reported as number of patients (%) unless otherwise indicated. Fisher's exact test was used for analysis.

Variable	HR (95% CI)	p Value
Type of shunt		0.101
VPI	0.77 (0.56–1.05)	
VA	Reference	
Age <6 yrs		<0.001
No	Reference	
Yes	1.60 (1.24–2.07)	
Etiology of hydrocephalus		0.046
Aqueductal stenosis	0.60 (0.32-1.11)	
Myelomeningocele	0.67 (0.46-0.97)	
Other etiology	0.71 (0.51-0.98)	
Post-IVH secondary to prematurity	Reference	
No. of CCCs		0.925
0	Reference	
1	0.95 (0.70-1.28)	
≥2	0.95 (0.69–1.31)	
Cardiovascular CCCs		0.091
No	Reference	
Yes	1.33 (0.96–1.85)	
Conversion to VA or VPI shunt oc- curred after infection		0.451
No	Reference	
Yes	0.91 (0.70–1.17)	

TABLE	3. Univa	ariable	proportiona	l hazards	models	for	shunt
failure	(all subj	ects)					

Results are based on univariable models.

that children < 6 years of age with VA shunts had significantly lower survival than other cohorts (Fig. 2), but survival of VPl shunts in children \geq 6 and < 6 years of age was similar. Multivariable regression analysis including age, type of shunt (atrial or pleural), and etiology did not reveal any significant predictors for time to shunt failure (Table 4).

We found no difference in the percentage of children who experienced shunt failure (yes or no) at 6 months (VA 34% vs VPl 32%, p = 0.700), 12 months (VA 39% vs VPl 35%, p = 0.349), and 24 months (VA 49% vs VPI 39%, p = 0.038) (Table 5). To assess the cumulative hydrocephalus surgery burden, we compared procedure counts (including multiple procedures per patient) over time. We found no difference at 6 and 12 months, but by 24 months of followup, more surgeries had been performed in the VA cohort. Most of the failures in both groups combined were revision (84%), with no difference between VA and VPI shunts. More than 58% of failures for both VA and VPI shunts were proximal, whereas 4% of the failures were distal (16% both proximal and distal, 22% unknown). After shunt failure, VPl shunts were reimplanted in the child's pleural space 41.7% of the time and in the abdominal cavity 27.1% of the time, whereas VA shunt failures were replaced into the child's atrium 62.8% of the time (p < 0.001) (Table 5).

Additional multivariable analysis including a cohort of patients from the registry with VP shunts indicated that VPl shunts have similar survival to VP shunts, with VA shunts demonstrating a 1.42 higher odds of failure (Table 6).

In this multicenter study using data from the HCRN registry, we directly compared the use of VA and VPI shunts, because the atrial and pleural spaces are the most common sites of nonperitoneal shunt terminus. Most children received VA shunts (3:1 in this report), and children who received first-time nonperitoneal shunts terminating in the atrium tended to be younger. There was no difference in the shunt revision burden at 6 and 12 months. At 24 months, more shunt revisions had been performed in subjects with VA shunts. Children < 6 years of age with VA shunts appeared to have shorter shunt survival than those \geq 6 years or those of any age with VPI shunts, but in a multivariable model, shunt type (VA vs VPI) was not associated with shunt survival.

It was previously suggested that young age influenced the decision to avoid VPI shunts. However, in our survival analysis, although the sample of children < 6 years of age with VPI shunts was small (n = 22), age did not appear to influence VPI shunt survival.

Hydrocephalus etiology differed between the cohorts in this study, with a higher proportion of children with myelomeningocele in the VPl cohort versus a higher proportion of premature children with IVH in the VA cohort. This may be secondary to a challenging neonatal abdominal environment in the premature cohort and the predilection for premature children to have concurrent lung dysfunction and bronchopulmonary dysplasia. Univariable modeling suggested that etiology may indeed influence failure of nonperitoneal shunts, but no independent association was found on multivariable analyses.

Among 14 HCRN centers, we identified and included 98 VPI subjects, with VA shunts being 3 times more common. Christian et al.⁸ suggested that the pleural space is not more widely used because of unfamiliarity with the procedure. In their study, 73 patients (43%) required shunt revision—most commonly because of proximal obstruction (44%)—which is similar to our finding (58.5%). Twenty-two children in their series required revision for a symptomatic pleural effusion, which may be mirrored in our results of failure of VPI shunts, where reimplantation back into the pleural space occurred 41.7% of the time and reimplantation in the abdominal cavity occurred 27.1% of the time.

The initial report of VPl cases was presented by Ransohoff¹⁰ in 1954. Hoffman et al.¹¹ reported on 59 patients with a revision rate of 61% and an infection rate of 19%, but follow-up was lacking in their report. To date, the literature on the use of VPl shunts in children is based on single-center reports. Oyon et al.⁷ demonstrated a 30% overall shunt survival with 19 of 27 VPl shunts requiring revisions. They did not reveal any risk factors for shunt failure, although patients who underwent an early revision tended to be younger. The incidence of pleural effusion in their series was 26%. Our multicenter experience represents a modern experience with VPl shunts. Since 2000, only five studies have reported on VPl shunt outcomes, two of which reported on pediatric patients.^{8,12–15}

Proponents of VPI shunts often cite thromboembolic and cardiopulmonary complications, as well as the potential for shunt nephritis, as significant complications of



FIG. 2. Stratified Kaplan-Meier survival curves comparing VA and VPI shunts in children < 6 and \geq 6 years of age. Figure is available in color online only.

VA shunts.^{3,6,16–19} A recent comparison of nonperitoneal shunts demonstrated a lower complication rate in VA compared with VPI shunts (4% vs 15.6%); complications in VA shunts included distal catheter displacement, shunt disconnection, endocarditis, and shunt nephritis.²⁰

In our cohort of children, however, we did not find a difference in the aggregate complication rates between VA and VPI shunts. Pneumothorax was diagnosed in 3 patients in the VPl cohort. Although granular data are not available on the need for anticoagulation and other non-shuntrelated treatment interventions, this study demonstrates an overall similar safety profile of VA and VPI shunts in the HCRN. It has also been suggested that VA shunts are not an attractive alternative in young children because of the potential need for lengthening to accommodate for growth over time.⁷ We found that at 2 years the proportion of VA shunts revised was higher than that of VPI shunts. Our survival analysis revealed that children < 6 years of age with VA shunts had a significantly lower shunt survival; this may have been secondary to the need for distal catheter lengthening, although the location of failure analysis does not reveal a disproportionately large proportion of distal VA failures.

Christian et al.⁸ found that age < 10 years was an independent risk factor for the development of pleural effusion; however, they did not report on risk factors for VPI shunt failure. Our univariable analysis demonstrated that age ≥ 6 years was protective against failure for nonperitoneal shunts. Although the concept of younger children being at higher risk for shunt failure is not novel,²¹ this cohort study of VA and VPl shunts confirms a previous finding that younger children are more prone to shunt failure, specifically those with VA shunts.

As part of an additional analysis, we found that VPI

TABLE 4. Multivariable proportional hazards models for shunt failure (all subjects)

Variable	HR (95% CI)	p Value
Etiology		
Post-IVH secondary to prematurity	Reference	0.058
Aqueductal stenosis	0.57 (0.30-1.07)	
Myelomeningocele	0.77 (0.52-1.12)	
Other	0.68 (0.49-0.94)	
VA shunt, compared w/ VPI shunt		
For age ≥6 yrs	0.67 (0.44-1.04)	0.072
For age <6 yrs	1.73 (0.93–3.22)	0.085

Results are based on multivariable models including age, type of shunt, and etiology.

TABLE 5. Cohort and shunt characteristics for failure after nonperitoneal shunt implantation

Variable	VA (n = 318)	VPI (n = 98)	p Value
No. of revisions/failures after			
implantation*			
w/in 6 mos	n = 299	n = 95	0.700†
0	197 (65.9)	65 (68.4)	
1	59 (19.7)	18 (18.9)	
2	25 (8.4)	6 (6.3)	
≥3	18 (6.0)	6 (6.3)	
w/in 12 mos	n = 291	n = 91	0.349†
0	178 (61.2)	59 (64.8)	
1	51 (17.5)	20 (22.0)	
2	35 (12.0)	3 (3.3)	
≥3	27 (9.3)	9 (9.9)	
w/in 24 mos	n = 284	n = 90	0.038†
0	146 (51.4)	55 (61.1)	
1	58 (20.4)	23 (25.6)	
2	42 (14.8)	2 (2.2)	
≥3	38 (13.4)	10 (11.1)	
Location of failure	n = 188	n = 48	0.228‡
Unknown	42 (22.3)	9 (18.8)	
Proximal	112 (59.6)	26 (54.2)	
Distal	6 (3.2)	4 (8.3)	
Both	28 (14.9)	9 (18.8)	
Shunt failure after converting to NPS	n = 188	n = 48	0.816‡
Revision	160 (85.1)	39 (81.3)	
Infection	26 (13.8)	7 (14.6)	
Other	2 (1.1)	2 (4.2)	
Next shunt surgery terminus	n = 188	n = 48	<0.001‡
VP	34 (18.1)	13 (27.1)	
VPI	7 (3.7)	20 (41.7)	
VA	118 (62.8)	7 (14.6)	
Other	1 (0.5)	0 (0.0)	
Unknown	28 (14.9)	8 (16.7)	

* Children were excluded from the number of revision summaries if they were censored in the registry for relocation out of network, death unrelated to hydrocephalus, or transitioning to adult care prior to the end of the time period in review.

† Cochran-Armitage trend test.

‡ Fisher's exact test.

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shunts have similar survival to VP shunts, with VA shunts demonstrating a 1.42 higher odds of failure; this finding suggests that VPI and VP shunts may have similar survival rates, both of which are better than that of VA shunts. As previously discussed, this is likely a function of age; in our comparison of children with a similar number of previous shunt surgeries and age \geq 6 years, VA, VPI, and VP shunts all had similar shunt survival.

Our analysis revealed that the utilization of antibiotic catheters was lower in the VA cohort. This was driven by distal rather than proximal catheter use patterns. Information about the specific type of catheter (traditional

TABLE 6. Multivariable proportional hazards model examining shunt failure

Variable	HR (95% CI)	p Value
Type of shunt		<0.001
VP	Reference	
VPI	1.34 (0.98–1.82)	
VA	1.42 (1.21–1.68)	
Age <6 yrs		0.374
No	Reference	
Yes	1.05 (0.94–1.18)	
Etiology of hydrocephalus		< 0.001
Aqueductal stenosis	0.71 (0.57–0.87)	
Myelomeningocele	0.68 (0.58-0.79)	
Other etiology	0.84 (0.74-0.95)	
IVH secondary to prematurity	Reference	

Results are based on multivariable models, adjusting for each of the predictors in the table.

VP shunt tubing vs type E catheters) is not available in the registry; however, we hypothesize that dedicated atrial catheters, which are not antibiotic impregnated, may have been utilized.

Limitations

The study is derived from a registry of patients treated surgically for hydrocephalus. The current cohort includes those entered in the registry prospectively, but some children are missing data from previous shunt surgeries. Although all children have at least one prior surgery reported in the registry, a significant proportion of the children included are derived from the noncomprehensive cohort, in which a subject's entire shunt surgery history may not be available.

Additional limitations include the disparity in cohort sample size. Although we adjusted for differences in baseline characteristics, specifically age and etiology, the cohort size disparity limits our ability to directly compare the two treatment types. Additionally, the low number of patients in the VPl cohort < 6 years of age limits our ability to explore outcomes and make meaningful comparisons.

All data were collected with protocols for fidelity, validation, and quality control. The HCRN includes centers in North America only, so the findings should be carefully interpreted with respect to practice patterns and protocols worldwide. As with any surgical study, the choice of nonperitoneal site is subject to the bias of the treating surgeon. Treatment centers may have an undetected influence over shunt terminus choice; in this study, 4 sites had a > 50%proportion of VPI shunts placed, but in aggregate the site frequencies are small and were not included in the statistical model. There is currently no protocol in place in the HCRN to influence terminus choice, which was the impetus for this investigation. Several variables, such as using pediatric surgery assistance, intraoperative technique (cutdown vs Seldinger), and intraoperative imaging adjuncts (fluoroscopy, echocardiography, and electrocardiography), are not recorded in the registry; these may represent important factors in assessing shunt survival and should be examined in future studies.

As mentioned previously, granular data were not available for major complications for VA shunts (need for anticoagulation or antiplatelet therapy, development of pulmonary hypertension, and stroke) or VPI shunts (treatment for pneumothorax and delayed symptomatic pleural effusions) and other non-shunt-related treatment interventions in the HCRN registry. Despite these limitations, this investigation represents the largest comparative study of VA versus VPl shunts and provides context in relation to VP shunt survival. Our analysis affirms that patients selected for atrial and pleural termini are fundamentally different; our goal was to explore those differences and report shunt survival outcomes for this specific population of children (those who cannot receive peritoneal shunts). It should be noted that the focus of the investigation was not to directly compare nonperitoneal and peritoneal shunts.

Conclusions

In this study comparing VA and VPl shunts, we found that VA shunts are used more often overall and are placed in younger patients, especially those who were premature and had IVH. VPl shunts were more commonly placed in older children with myelomeningocele. When stratified by age, children < 6 years with VA shunts had the shortest shunt survival, which may be explained by the different etiologies; however, shunt location (VA vs VPl) was not associated with shunt survival in the multivariable model that included age and etiology. No differences were found in the cumulative frequency of complications. These results support that both atrial and pleural sites are viable when the abdomen is not usable. Overall, the findings suggest that the baseline differences observed between the cohorts of children treated with a VA versus a VPl shunt, specifically with respect to age and etiology, likely account for current practice patterns.

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Dr. Limbrick reported being the CMO of Rhaeos Inc. outside the submitted work. Dr. Hauptman reported personal fees from Medtronic and BK Medical outside the submitted work. Dr. Jackson reported consulting fees from Integra LifeSciences outside the submitted work.

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Supplemental Information

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