Case series of ventriculopleural shunts in adults: a single-center experience

Claudia Craven, MSc, MRCS, Hasan Asif, BSc, MBBS, Amna Farrukh, MBBS, Flavia Somavilla, MBBS, Ahmed K. Toma, MD, FRCS(SN), and Laurence Watkins, MA, FRCS(SN)

Victor Horsley Department of Neurosurgery, National Hospital for Neurology and Neurosurgery, Queen Square, London, United Kingdom

OBJECTIVE The peritoneal cavity is widely used as the destination of choice for cerebrospinal fluid shunts. Various alternative sites have been used, particularly in the presence of certain contraindications. The pleural cavity has been used; however, a paucity of evidence details ventriculopleural (VPL) shunt survival, complication, and revision rates in adults. The aim of this study was to present a single center’s experience with VPL shunts, identifying complication, revision, and survival rates.

METHODS A single-center, retrospective case series analysis was conducted for VPL shunt insertions and revisions over a period of 5 years. Demographic as well as clinical data were collected. Ventriculopleural shunt survival was assessed using Kaplan-Meier curves and the log rank (Cox-Mantel) test.

RESULTS Twenty-two VPL shunts were inserted in 19 patients. Median survival of the VPL shunts was 14 months. Pathological indication for the VPL shunt did not significantly affect survival. A total of 10 complications was observed: 2 infections, 2 cases of overdrainage, 2 obstructions, 1 distal catheter retraction, 2 symptomatic pleural effusions, and 1 asymptomatic pleural effusion.

CONCLUSIONS Ventriculopleural shunting is a safe and viable second-line procedure for cases in which ventriculoperitoneal shunts are unsuitable. While VPL shunts have a high revision rate, their complication rate is comparable to that of VP shunts. Ventriculopleural shunt survival can be improved by careful patient selection and the implementation of a combination of valves with antisiphon devices.

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KEY WORDS ventriculoperitoneal; ventriculoatrial; ventriculopleural; cerebrospinal fluid; idiopathic intracranial hypertension; hydrocephalus

Ventriculoperitoneal (VP) shunting is the conventional cerebrospinal fluid (CSF) diversion procedure for the surgical management of hydrocephalus and disorders of CSF dynamics. However, there are occasions when the peritoneum is a suboptimal choice and an alternative distal site is necessary. Such circumstances include the presence of abdominal adhesions due to prior surgery, abdominal pathology, pregnancy, or recurrent complications from abdominal distal tubing. The common alternative distal sites include the cardiac atria, with a ventriculoatrial (VA) shunt, or the pleural space, with a ventriculopleural (VPL) shunt.

Pleural shunting of CSF was first described in the 20th century by Heile and was introduced in the management of hydrocephalus by Ransohoff in 1954. The shunt constitutes a surgically created passageway between a cerebral ventricle and the pleural cavity for the drainage of excess CSF.

Case reports of pleural effusions, pneumothorax, empyema, overdrainage, and other rare but serious complications are deterrents to the frequent use of VPL shunts. While historical literature describes high rates of pleural effusion (20%) following VPL shunt insertion, contemporary research describes VPL shunting as an effective and safe alternative to VP shunts in the pediatric population. In the adult population, however, no survival analysis of VPL shunts has been published.

Nowadays, VPL shunts are often inserted together
with an antisiphon valve to prevent overdrainage owing to negative intrathoracic pressures during inspiration. Such valves can be adjustable or nonadjustable. At our institution, adjustable valves are the preferred option as they enable an additional element of nonsurgical management.

While studies have demonstrated higher complication rates with VPL and VA shunts, there is, theoretically, greater potential for severe complication following VA shunt placement, including shunt nephritis, injury to major vessels, cardiac thrombi, and endocarditis. For this reason, at our institution, the VPL shunt is the preferred secondary choice over a VA shunt in circumstances in which a VP shunt cannot be placed. However, the decision to use a VPL or VA shunt also takes into account patient and surgeon preferences, as well as anatomical and pathological anomalies.

The group of patients referred to our unit is unusually complex, often having experienced multiple failed shunt attempts with treatment-refractory hydrocephalus. We report our experience with complications and factors that influence the survival of VPL shunts inserted in adults.

Methods

Study Design and Participants

A single-center retrospective analysis was conducted for VPL shunt insertions and revisions over a period of 5 years (July 2010–July 2015). All patients had been referred to our tertiary care unit following multiple failed VP shunt insertions or complex comorbidities prohibiting routine VP shunt insertion. Inclusion criteria included an age over 18 years at the time of VPL shunt insertion. Demographic data including age, sex, diagnosis, and number of previous CSF diversions (non-VPL shunts) were collected. Clinical data on indication, valve type used, complications, and number of VPL shunt revisions were also recorded.

Ventriculopleural Shunt Placement and Technique

General anesthesia is induced and patients are positioned supine with their head turned 45° on a surgical horseshoe headrest. A wedge is placed under the ipsilateral shoulder. The cranial phase of the operation is identical to that for VP shunt insertion. A 3-cm incision is made in the chest wall at the midclavicular line and second anterior intercostal space. Pectoralis major and intercostal muscles are split, exposing the parietal pleura, taking care to avoid any neurovascular structures. The distal catheter is subcutaneously tunneled from the cranial incision to the chest wall incision. A small hole is made into the parietal pleura, and the distal end of the catheter is cut to 15 cm. The distal catheter is then introduced into the pleural cavity and secured with 2-0 Ethibond Excel (polybutylate-coated braided polyester, Ethicon, Johnson and Johnson Medical Ltd.) under positive pressure ventilation (to reduce the risk of pneumothorax). Muscular closure is performed with 2-0 Vicryl Plus (fast absorbing polyglactin 910, Ethicon, Johnson and Johnson Medical Ltd.) and is followed by continuous closure of subcutaneous tissue and skin. All patients undergo postoperative lateral skull radiography and anteroposterior and lateral chest radiography to confirm catheter location. Since the pleural pressures are lower than those in the peritoneum, we routinely set the valve at a higher pressure setting (for example, proGAV, Miethke GmbH & Co. KG; set at 10 cm H2O rather than the 5 cm H2O for a VP shunt).

Statistical Analysis

The end point criterion for shunt failure was defined as the point at which the VPL shunt was revised. Ventriculopleural shunt survival was assessed using Kaplan-Meier curves and the log rank (Cox-Mantel) test. All statistical tests were performed on GraphPad Prism 6.0c.

Results

Patient Characteristics

Nineteen patients were identified (4 males and 15 females) as having VPL shunts inserted between 2010 and 2015. One patient required bilateral VPL shunts, and 2 patients had their VPL shunt removed and a new one inserted on the contralateral side, totaling the insertion of 22 new VPL shunts. Mean age (± standard deviation) at the last follow-up with a VPL shunt in situ was 37.9 ± 15.4 years. The mean follow-up among all 19 patients since the first VPL shunt insertion was 3.07 ± 1.31 years (1.17–4.92 years).

Five patients had a diagnosis of idiopathic intracranial hypertension (IIH), 13 had hydrocephalus (5 had hydrocephalus secondary to a tumor, 5 secondary to subarachnoid hemorrhage, and 3 with congenital hydrocephalus), and 1 had CSF disequilibrium (demonstrating both low and high intracranial pressure on monitoring).

Shunt Details

Indication for VPL Shunt

The predominant indication for a VPL shunt was adhesions due to multiple previous abdominal surgeries (11 patients [57.9%]). Other indications included planned significant abdominal surgery (2 patients [10.5%]), suspicion of poor peritoneal absorption (fluid collection, 3 patients [15.8%]), multiple episodes of distal tubing obstruction (1 patient [5.3%]), and abdominal pain (2 patients [10.5%]).

Primary Versus Secondary Procedure

Three patients had primary VPL shunts and 16 underwent VPL shunt insertion as a secondary procedure. Those with a primary VPL had preexisting abdominal pathology (adhesions or pain from prior abdominal surgeries). Previous CSF diversion surgeries had failed in the secondary VPL shunt group, whose members had undergone a mean of 4.06 ± 3.13 prior VP shunt operations. This same group had fewer revisions of their VPL shunts in the 5-year study period, undergoing a mean of 2.43 ± 1.86 operations after VPL shunt insertion.

Shunt Valve Type

Eighteen VPL shunts involved the insertion of adjustable valves: 7 proGAV, 7 proSA, 2 proGAV + proSA (Miethke GmbH & Co. KG), and 2 Certas Plus programmable valves (Codman, DePuy Synthes). Four VPL shunts were fitted with fixed-pressure CSF flow control nonadjustable...
valves (Medtronic Ltd.). All devices had antisiphon components integrated with the valve.

Shunt Follow-Up

Overall, the median follow-up for all new VPL shunts in the defined study period was 12 months (range 1–59 months) up to the last follow-up point (July 1, 2015). The median time to the first VPL shunt failure was 7.50 months (range 1–23 months).

Shunt Survival Analysis

Overall

Median survival of VPL shunts was 14 months (range 1–23 months). At 23 months, 12 (54.5%) of the 22 shunts had primary revision (10) or were removed (2) (Fig. 1A). Ten VPL shunts required no further intervention. Of the 10 shunts with primary revision, 7 required secondary revision, 3 required tertiary revision, and none required more than 3 revisions. Ventriculopleural shunt survival improved with subsequent revisions, with a median survival of 7 months (range 1–33 months) after a first revision, and subsequent revisions having a survival of at least 13 months (range 1–47 months; Fig. 1B and C).

By Indication

Ventriculopleural shunts inserted to manage hydrocephalus secondary to subarachnoid hemorrhage and tumors (14 shunts) had a median survival of 18.5 months (range 1–59 months). Median VPL shunt survival among patients with IIH (7 shunts) was 7 months (range 1–33 months). There was no significant difference on comparisons of median survival.

By Valve Type

The combination of proGA V + proSA demonstrated a greater percentage of survival than any adjustable valve alone. The proGA V valve demonstrated a median survival of 10 months, the Certas 8.5 months, the proSA 4 months; and of the 2 shunts with proGA V + proSA in place, neither failed at a follow-up of 33 months. Of the nonadjustable fixed pressure valves, 1 failed at 4 months and 3 remained in situ at 24 months’ follow-up. There was no significant difference on comparisons of median survival.

Shunt Complications

Ten complications were reported following 22 VPL shunt insertions: 2 infections, 1 retraction, 2 obstructions (1 distal and 1 proximal), 3 pleural effusions (2 of which were symptomatic), and 2 cases with symptomatic overdrainage (confirmed by ICP monitoring). In addition to these complications, 2 shunts had no symptomatic benefit to the patient. There were no mortalities from complications or shunt dysfunction (Fig. 2). Both patients with symptomatic pleural effusions presented with pleuritic chest pain. One also had associated shortness of breath, but neither patient appeared septic.
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Complications reported post-VPL shunt insertion, including those reported postrevision.

On plain anteroposterior radiography, both effusions were moderate (4 and 9 cm, respectively) and one required aspiration. The aspirate was sterile, with inflammatory cells only and no evidence of infection. Both patients required surgical revision of their shunts; in 1 case the shunt was converted to a VP shunt, and in the other case the distal shunt end was externalized and the shunt was eventually replaced. Neither required further revision at the time of follow-up.

Discussion

Ventriculopleural shunts are an alternative option to the VP shunt, when the peritoneum is unsuitable due to abdominal pathology, abdominal adhesions, or recurrent complications with abdominal distal tubing.27

Ventriculoperitoneal shunts are exposed to neutral intraabdominal pressure (IAP), mildly raised IAP in morbid obesity and pregnancy, or raised IAP in peritonitis, ileus, pancreatitis, and trauma.3,13,15,31,34 In contrast to the peritoneum, the pleural cavity has negative pressure.3 Thus, for the VPL shunt, the pathophysiology, associated complications, and surgical considerations—including valve choice—are inherently unique.

Ventriculopleural shunts are anecdotally associated with increased pleural complications; however, a paucity of evidence details survival, complication, and revision rates for VPL shunts in adults since the introduction of adjustable valves. We present a single institution’s experience using VPL shunts mostly as a secondary procedure.

Study Limitations

The complication rate in our study is higher than those in other studies owing to selection bias given our acceptance of the complex hydrocephalus cases referred to our institution.17,32 This group of patients predominately underwent VPL shunt placement as a secondary procedure or for comorbidities prohibiting routine VA or VP shunt placement. Our study’s outcomes may not be entirely attributable to the use of VPL drainage but may be multifactorial due to the overall algorithm used in decision making as well as individual characteristics of each patient and his or her pathology.

Survival Analysis

This is the first study presenting Kaplan-Meier survival analysis for VPL shunts in adults. The median survival of VPL shunts was 14 months, after which time 54.5% were revised or removed (follow-up range 1–59 months). Comparatively, the reported failure rates for a VP shunt range from 23.0% to 46.3%.12,25 There were no cases of significant morbidity or mortality associated with VPL shunt insertion in our study.

Those who had a VPL shunt as a secondary procedure had fewer returns to the operating theater during the 5-year study period (2.43 ± 1.86 VPL shunt operations), in comparison with the number while non-VPL shunts were in place (4.06 ± 3.13 prior VP shunt operations).

The incidence of revision following VPL shunt insertion was 45.4% (compared with 32.5% for VP shunts in adults).23 The median time to primary VPL shunt failure in adults in this cohort was 7.50 months, comparatively worse than that in the pediatric population at 10 months.22

Pathological indication for a VPL shunt did not significantly affect the duration of shunt survival (p = 0.6041). This is notable because one may have expected IIH patients to have more shunt failures because of greater pressures and increased body mass index, making for more challenging surgery. Excisions of brain tumors have been associated with early VP shunt malfunction.33 In contrast, the few cases of tumor-associated hydrocephalus requiring a VPL shunt in our study did not have a significantly worse outcome than those with other pathologies (p > 0.05). This nonsignificant finding is probably attributable to the sample size of each pathological group.

Complications of VPL Shunts

The traditional suspect, pleural effusion, does not appear to be solely responsible for the relatively high VPL shunt revision rate in this study.14 Twenty-two shunts were inserted, 10 of which had complications. There were only 2 symptomatic pleural effusions and no cases of pneumothorax. There were 5 mechanical complications: 2 overdrainage, 1 proximal obstruction, 1 distal obstruction, and 1 retraction.

The finding that pleural complications are not primarily responsible for the high revision rates of VPL shunts concurs with data in the few studies investigating complications in adults (Table 1).17,32 Megison and Benzel demonstrated a low proportion (4.55%) of revisions due to pleural complications.17

Two patients, both with IIH, underwent shunt revision due to unresolved symptoms. In light of this finding, screening for coexisting primary headaches and optimizing their medical management may be particularly important when selecting patients for VPL shunt insertion, espe-
cially in light of the strong association between coexisting primary headaches and IIH (68%).

In light of the dynamic negative pressures exerted on VPL shunts, it was interesting to compare various valve devices and their outcomes. The 2 cases with proGA V + proSA demonstrated a lower shunt failure rate. While this finding was nonsignificant, there is established evidence that the addition of antisiphon devices can prevent complications such as overdrainage in VPL shunts.5,15,23

At our center we routinely use adjustable valves with antigravity components. Our choice of valve is not based on pathology; however, the choice of the opening pressure setting is determined by shunt destination. For VPL shunts we start with an opening pressure of 10 cm H2O, rather than the 5 cm H2O for VP shunts. This is a preemp
tive measure to prevent postural overdrainage and pleural effusions. Given that 20.0% of shunt revisions were for pleural effusions (and 20% of complications were due to symptomatic pleural effusions), we hypothesize that a higher standard opening pressure ought to be used in VPL shunts.

**Literature Review**

A few studies describe survival and complication rates for other distal shunt sites in the adult population. Such sites include the aforementioned cardiac atria and peritoneal cavity and the less commonly used venous circulation (retrograde ventriculo-venous including sinuses and jugular veins) and cholecystic, uteretic, and vesicular sites.1,2,4,7,11,26,28 Each distal site has its own peculiar complications in addition to the heterogeneous structural risks due to catheter migration.4,10,28,30,35

Ventriculoatrial shunts have associated morbidity (of heterogeneous severity) secondary to complications including shunt nephritis, shunt infection, pulmonary embolism,
endocarditis, and cardiac and pulmonary thrombi.9,10,21,33,36,38
An alternative second-line option is VPL shunting.

The evidence on survival, revision, and complication rates for these alternative shunts is scarce because the shunts are infrequently used in adults. The majority of outcome data are based on the pediatric population, which may not accurately correlate with the observed complication and shunt survival rates in adults. An example of adult results contrasting with pediatric results includes the finding that there was no significant difference in complication rates between VA and VP shunts.9 Moreover, we observed no cardiopulmonary complications. There is heterogeneity in outcome and reporting experiences with these rarer shunts in adults (Table 2). It is possible that the VA shunt is superior to the VPL shunt, and a comparative study of both insertion techniques would be an interesting avenue to pursue.

Our literature review of adult shunts suggests that the complication rate seen in VPL shunts is similar to those seen with VP shunts. The complication rate of 45.4% following VPL shunt insertion in this study compares favorably to rates observed by McGovern et al., who reported complications for VP and VA shunts as 47.1% and 43.2%, respectively.9 Overall, the contemporary body of evidence suggests that distal catheter sites in adults are understudied; therefore, many may be quite reasonable alternatives to the VP shunt.

Conclusions

Ventriculopleural shunting is a safe and viable second-line procedure for adult cases in which VP shunts are unsuitable. While VPL shunts have a high revision rate, their complication rate is comparable to that of VP shunts. Ventriculopleural shunt survival may be improved by careful patient selection and the implementation of a combination of valves with antisiphon devices.

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Disclosures
Mr. Watkins has received honoraria from and served on advisory boards for Medtronic, Codman, and B Braun.

Author Contributions
Conception and design: Toma, Watkins. Acquisition of data: Craven, Farrukh, Somavilla. Analysis and interpretation of data: Craven, Asif, Toma. Drafting the article: Craven, Asif, Farrukh, Somavilla. Critically revising the article: Craven, Asif, Toma, Watkins. Reviewed submitted version of manuscript: Craven, Toma, Watkins. Approved the final version of the manuscript on behalf of all authors: Craven. Statistical analysis: Craven, Asif. Study supervision: Toma, Watkins.

Supplemental Information
Previous Presentations
Portions of this work were presented in abstract form at Hydrocephalus 2014, the Sixth Meeting of the International Society for Hydrocephalus and Cerebrospinal Fluid Disorders held in Bristol, United Kingdom, on September 6–8, 2014.

Correspondence
Claudia Craven, The National Hospital for Neurology and Neurosurgery, Victor Horsley Department of Neurosurgery, Queen Square, London WC1N 3BG, United Kingdom. email: claudia.craven@gmail.com.