Analysis of Non-Ventriculoperitoneal Shunts at Red Cross War Memorial Children's Hospital

Master of Medicine in Neurosurgery

University of Cape Town

Supervisor: Prof JMN Enslin

Co-supervisor: Prof AA Figaji

MMed Candidate: Dr B De John

DECLARATION

I declare that the work contained here within is of my own accord, having been obtained in an independent manner. Furthermore, no part has, is being, or is to be submitted for another degree to any other University. I confirm that this work has not been reported or published prior to registration for my Master of Medicine in Neurosurgery.

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Name: Byron De John

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SUPERVISOR DECLARATION

I declare that this planned publication meets all requirements to be included in a dissertation, namely:

- The journal publishing this paper is accredited by the department of higher education and training and has been approved by the UCT Health Sciences Specialist Training Committee
- The candidate is the first author on the paper
- The candidate contributed the most to the paper
- The candidate developed the protocol and wrote the paper under supervision
- The candidate was involved in the analysis, presentation, and interpretation of results
- The other authors and their contributions to the paper are stated

Name: Prof JMN Enslin

Staff Number: 01429977

Signature:

Date: 2023/02/06

ACKNOWLEDGEMENTS, FORMAT, CONTRIBUTIONS

This thesis is in a publication ready format but has not yet been submitted for publication. I would like to acknowledge the support and input from my supervisors Professor JMN Enslin and Professor AA Figaji. I would like to thank Prof U Rohlwink for her contribution towards the protocol and the data analysis. I would like to acknowledge the hard-working team of doctors, nurses, support staff and researchers at Red Cross Children's Hospital for their contribution towards the betterment of paediatric care, especially within Neurosurgery.

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1. Instructions to Authors from World Neurosurgery Journal

- **Article structure:**
    - Subdivision - unnumbered sections
        - Divide your article into clearly defined sections. Each subsection is given a brief heading. Each heading should appear on its own separate line. Subsections should be used as much as possible when cross- referencing text: refer to the subsection by heading as opposed to simply 'the text'.
    - Introduction
        - State the objectives of the work and provide an adequate background, avoiding a detailed literature survey or a summary of the results.
    - Material and methods
        - Provide sufficient details to allow the work to be reproduced by an independent researcher. Methods that are already published should be summarized, and indicated by a reference. If quoting directly from a previously published method, use quotation marks and also cite the source. Any modifications to existing methods should also be described.
    - Results
        - Results should be clear and concise.
    - Discussion
        - This should explore the significance of the results of the work, not repeat them. A combined Results and Discussion section is often appropriate. Avoid extensive citations and discussion of published literature.
    - Conclusions
        - The main conclusions of the study may be presented in a short Conclusions section, which may stand alone or form a subsection of a Discussion or Results and Discussion section.
    - Appendices
        - If there is more than one appendix, they should be identified as A, B, etc. Formulae and equations in appendices should be given separate numbering: Eq.

(A.1), Eq. (A.2), etc.; in a subsequent appendix, Eq. (B.1) and so on. Similarly for tables and figures: Table A.1; Fig. A.1, etc.

- **Abstract:**
  - A concise and factual abstract is required. The abstract should state briefly the purpose of the research, the principal results and major conclusions. An abstract is often presented separately from the article, so it must be able to stand alone. For this reason, References should be avoided, but if essential, then cite the author(s) and year(s). Also, non-standard or uncommon abbreviations should be avoided, but if essential they must be defined at their first mention in the abstract itself.
  - Abstracts should be 250 words, maximum.
  - Original Articles require a structured abstract with the following headings: Objective (or Background), Methods, Results, Conclusions.
- **References**
  - Citation in text
    - Please ensure that every reference cited in the text is also present in the reference list (and vice versa). Any references cited in the abstract must be given in full. Unpublished results and personal communications are not recommended in the reference list, but may be mentioned in the text. If these references are included in the reference list they should follow the standard reference style of the journal and should include a substitution of the publication date with either 'Unpublished results' or 'Personal communication'. Citation of a reference as 'in press' implies that the item has been accepted for publication.
  - Reference links
    - Increased discoverability of research and high quality peer review are ensured by online links to the sources cited. In order to allow us to create links to abstracting and indexing services, such as Scopus, Crossref and PubMed, please ensure that data provided in the references are correct. Please note that incorrect surnames, journal/book titles, publication year and pagination may prevent link creation. When copying references, please be careful as they may already contain errors. Use of the DOI is highly encouraged.

- Reference style
  - Text: Indicate references by (consecutive) superscript arabic numerals in the order in which they appear in the text. The numerals are to be used outside periods and commas, inside colons and semicolons. For further detail and examples you are referred to the AMA Manual of Style, A Guide for Authors and Editors, 11th Edition.
  - List: Number the references in the list in the order in which they appear in the text.

2. Data capture instrument(s)

Data collection form Headings

Patient name	Folder number	DOB	Age	Sex	Aeitology of HCP	Comorbid conditions	No. of previous shunt Procedures

Reason for VP shunt failure	Date of non-VPS insertion	Route of Non-VPS procedure	Indication for insertion

Brand of shunt used	Ultrasound guided Technique (Y/N	Other relevant surgical procedure information	Immediate surgical complications

Short-term complications	Long-term Complications	No. admissions since insertion non-VPS	Revision of non-VPS (Y/N

Time to Revision	Reason for failure of non-VPS	Revision Procedure

3. Ethics approval letter and other required approvals

i. University of Cape Town Human Research Ethics Committee Approval

UNIVERSITY OF CAPE TOWN
Faculty of Health Sciences
Human Research Ethics Committee

Room G50- Old Main Building
Groote Schuur Hospital
Observatory 7925
Telephone [021] 406 6492
Email: hrec-enquiries@uct.ac.za
Website: www.health.uct.ac.za/fhs/research/humanethics/forms

22 June 2020

HREC REF:317/2020

Prof N Enslin
Department of Neurosurgery
J-Floor, OMB
Email: johannes.enslin@uct.ac.za
Student: dejohn.byron@gmail.com

Dear Prof Enslin

PROJECT TITLE: RETROSPECTIVE REVIEW OF NON-VENTRICULO-PERITONEAL-SHUNTS AT RED CROSS CHILDREN'S HOSPITAL-MMED CANDIDATE-DR BYRON DE JOHN.

Thank you for submitting your study to the Faculty of Health Sciences Human Research Ethics Committee (HREC) for review.

It is a pleasure to inform you that the HREC has **formally approved** the above-mentioned study.

This approval is subject to strict adherence to the HREC recommendations regarding research involving human participants during COVID -19, dated 17 March 2020.

Approval is granted for one year until the 30 June 2021.

Please submit a progress form, using the standardised Annual Report Form if the study continues beyond the approval period. Please submit a Standard Closure form if the study is completed within the approval period.
(Forms can be found on our website: www.health.uct.ac.za/fhs/research/humanethics/forms)

The HREC acknowledge that the student: Dr Byron De John will also be involved in this study.

Please note that the ongoing ethical conduct of the study remains the responsibility of the principal investigator.

Please note that for all studies approved by the HREC, the principal investigator **must** obtain appropriate institutional approval, where necessary, before the research may occur.

Please quote the HREC reference number in all your correspondence.

Yours sincerely

PROFESSOR M BLOCKMAN
CHAIRPERSON, FHS HUMAN RESEARCH ETHICS COMMITTEE

HREC 317/2020sa

ii. Department of Surgery Research Committee Approval

UNIVERSITY OF CAPE TOWN

Department of Surgery
Departmental Research Committee
Dr Timothy Pennel
D24 Office, Groote Schuur Hospital
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South Africa
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26 May 2020

Dr B De John

Department of Surgery
University of Cape Town

Dear Dr De John

RE: Project 2020/072

PROJECT TITLE: Retrospective Review Of Non-Ventriculo-Peritoneal Shunts At Red Cross Children's Hospital

The above protocol has been reviewed by the Department of Surgery Research Committee. I am pleased to inform you that the committee approved the scientific merit of the study, and endorse the protocol for submission to the relevant ethics committee.

Although this letter serves as confirmation that the above protocol has successfully passed through the surgical DRC, respective ethics committees still require DRC chair signature before submission.

Please use the above project number in all future correspondence,

Yours sincerely

DR TIMOTHY PENNEL
CHAIR: SURGICAL DRC

DR MARITZ LAUBSCHER
CHAIR: PROTOCOL REVIEW COMMITTEE

LIST OF TABLES

Table 1 Demographics and Baseline Characteristics

		Ventriculoatrial		Ventriculopleural		Ventriculovesical		Ventriculocholecystic		Total	
		n	%	n	%	n	%	n	%	n	%
Route of Non-VPS procedure		25	42,4%	32	54,2%	1	1,7%	1	1.7%	59	100,0%
Sex	Male	9	56,3%	16	59,3%					25	58,1%
	Female	7	43,8%	11	40,7%					18	41,9%
Median Age in Years at first CSF Diversion (Range)		0,4	(0-1,5)	0,6	(0-5,0)	0,8		0,8		0,6	(0-5,0)
Mean Age in Years at first CSF Diversion (+/-SD)		0,5	(0,4)	1,0	(1,2)	0,8		0,8		0,8	(1,0)
Median Age Years at Non-VP Shunt (Range)		2,9	(0,3-14,9)	5,3	(0,5-13,4)	3,6		3,8		4,0	(0,3-14,9)
Mean Age in Years at Non-VP Shunt (+/-SD)		5,2	(4,6)	5,9	(4,1)	3,6		3,8		5,5	(4,3)
Median Number Previous Shunt Procedures (Range)		6,0	(2-28)	4,5	(2-17)	21,0		25,0		5,0	(2-28)
Mean Number Previous Shunt Procedures (+/-SD)		7,7	(6,2)	6,2	(4,4)	21,0		25,0		7,4	(6,0)
Multiple Medical Comorbidity	No	7	43,8%	15	55,6%	1	100,0%	1	100,0%	22	51,2%
	Yes	9	56,3%	12	44,4%					21	48,8%
Aeitology of HCP	Idiopathic	6	37,5%	9	33,3%					15	34,9%
	Myelomeningocele	5	31,3%	4	14,8%					9	20,9%
	Post-infectious	1	6,3%	4	14,8%	1	100,0%	1	100,0%	5	11,6%
	4th Ventricular Outflow Obstruction	2	12,5%	3	11,1%					5	11,6%
	TBM			4	14,8%					4	9,3%
	Intraventricular Haemorrhage	1	6,3%	2	7,4%					3	7,0%
	Tumour related			1	3,7%					1	2,3%
	Aqueductal Stenosis	1	6,3%							1	2,3%
	Total	16	100,0%	27	100,0%	1	100,0%	1	100,0%	43	100,0%

Table 2 Indication for Non-Ventriculoperitoneal Shunt Insertion

		Ventriculoatrial		Ventriculopleural		Ventriculovesical		Ventriculocholecystic		Total	
		n	%	n	%	n	%	n	%	n	%
Indication Non-VPS	Abdominal Pseudocyst	8	32,0%	11	34,4%					19	32,2%
	Intra-abdominal Sepsis	6	24,0%	7	21,9%					13	22,0%
	Significant Abdominal Adhesions	3	12,0%	2	6,3%	1	100,0%	1	100,0%	7	11,9%
	Bowel/bladder perforation/erosion	2	8,0%	4	12,5%					6	10,2%
	Abdominal Surgery	2	8,0%	3	9,4%					5	8,5%
	VPL Shunt Dysfunction	1	4,0%	3	9,4%	1	100,0%			5	8,5%
	Pleural Effusion related to VPL Shunt	3	12,0%							3	5,1%
	VA Shunt Dysfunction	2	8,0%	1	3,1%					3	5,1%
	Multiple Failed VPS (Concern of peritoneal malabsorption)	1	4,0%							1	1,7%
	Bowel Injury	1	4,0%	1	3,1%					2	3,4%
	TB abdomen			1	3,1%					1	1,7%
	Vent-Vesical Shunt Dysfunction							1	100,0%	1	1,7%
	Vent-GB Shunt Dysfunction	1	4,0%							1	1,7%

Table 3 Shunt Survival and Reason for Failure

		Ventriculoatrial		Ventriculopleural		Ventriculo-vesical		Ventriculo-cholecystic		Total	
		n	%	n	%	n	%	n	%	n	%
Route of Non-VPS procedure		25	42,4%	32	54,2%	1	1,7%	1	1,7%	59	
Lost to Follow Up		3	12,0%	3	9,4%					6	10,2%
Revision of non-VPS (Y/N)	No	12	54,5%	10	34,5%					22	37,3%
	Yes	10	45,5%	19	65,5%	1	100,0%	1	100,0%	31	52,5%
Median Follow Up Time Years		1,9	(0-8,5)	4,2	(0,3-10,0)					2,7	(0-10,0)
Mean Follow Up Time Years (+/- SD)		2,6	(2,3)	4,4	(3,0)					3,5	(2,8)
Median Shunt Survival Time months (range)		13,5	(0-67)	5	(0-118)					6,0	(0-118)
Mean Shunt Survival Time months (+/- SD)		19,3	(21,1)	21,9	(33,5)					20,0	(28,3)
Shunt Survival at	3 months	16	72,7%	18	62,1%					34	100,0%
	6 months	13	59,1%	14	48,3%					27	79,4%
	12 months	11	50,0%	10	34,5%					21	61,8%
	18 months	11	50,0%	9	31,0%					20	58,8%
Reason Shunt Failure	Shunt Blockage	6	60,0%	5	26,3%	1	100,0%	1	100,0%	13	41,9%
	Pleural Effusion			7	36,8%					7	22,6%
	Shunt Sepsis			4	21,1%					4	12,9%
	Displaced Distal Catheter	1	10,0%	3	15,8%					4	12,9%
	Shunt Disconnection	1	10,0%	1	5,3%					2	6,5%
	Intracranial Sepsis			1	5,3%					1	3,2%
	Endocarditis	1	10,0%							1	3,2%
	Shunt Nephritis	1	10,0%							1	3,2%
	Unsuccessful Shunt Insert	1	10,0%							1	3,2%
	Pleural Empyema			1	5,3%					1	3,2%
Immediate Revision Procedure	External Ventricular Drain	3	30,0%	9	47,4%	1	100,0%	1	100,0%	14	45,2%
	Ventriculo-peritoneal Shunt	6	60,0%	6	31,6%					12	38,7%
	Ventriculo-Pleural Shunt	1	10,0%	3	15,8%					4	12,9%
	Ventriculo-Vesical Shunt			1	5,3%					1	3,2%
	Total	10	100,0%	19	100,0%	1	100,0%	1	100,0%	31	100,0%

Table 4 Complications Non-Ventriculoperitoneal Shunts

		Ventriculoatrial		Ventriculopleural	
		n	% of total	n	% of total
Immediate Surgical Complications	Non-significant arrythmia	3	12,0%		
	Failed Seldinger Technique	3	12,0%		
	Deep Atrial Insertion	1	4,0%		
	Arterial Puncture	1	4,0%		
	Minor Blood Loss	1	4,0%		
	Surgical Emphysema			1	3,1%
	Total	9	36,0%	1	3,1%
Short Term Complications (30 Days)	Asymptomatic Pleural Effusion			6	18,8%
	Symptomatic Pleural Effusion			5	15,6%
	Pneumothorax (No intervention)			2	6,3%
	Displaced Distal Catheter			2	6,3%
	Ventriculitis			1	3,1%
	Subdural Hygroma	1	4,0%	1	3,1%
	Deep Catheter in Atrium	2	8,0%		
	Superficial Wound Infection	1	4,0%		
	Total	4	16,0%	17	53,1%
Long-term	Shunt Sepsis			3	9,4%
	Symptomatic Pleural Effusion			2	6,3%
	Asymptomatic Pleural Effusion			1	3,1%
	Empyema			1	3,1%
	Displaced Distal Catheter			1	3,1%
	Death			1	3,1%
	Subdural Hygroma	1	4,0%	2	6,3%
	Shunt Disconnection	1	4,0%	1	3,1%
	Endocarditis	1	4,0%		
	Shunt Nephritis	1	4,0%		
	Total	4	16,0%	12	37,5%

Table 5 Surgical Related Factors

		Ventriculoatrial		Ventriculopleural		Ventriculo-vesical		Ventriculo-cholecystic		Total	
		n	%	n	%	n	%	n	%	n	%
Brand of shunt used	OSVII	5	20.0%	2	6.3%					7	11.9%
	Bactiseal	16	64.0%	17	53.1%	1	100.0%	1	100.0%	35	59.3%
	Miethke	1	4.0%							1	1.7%
	Not Stated	3	12.0%	11	34.4%					14	23.7%
	Essential			2	6.3%					2	3.4%
Ultrasound guided Technique (Y/N)	No	2	8.0%							2	3.4%
	Yes	23	92.0%							23	39.0%
	N/A			32	100.0%	1	100.0%	1	100.0%	34	57.6%
Formal Open Cut Down	No	21	84.0%							21	35.6%
	Yes	4	16.0%							4	6.8%
	N/A			32	100.0%	1	100.0%	1	100.0%	34	57.6%
Xray Screening Intra-op	No	18	72.0%							18	30.5%
	Yes	7	28.0%							7	11.9%
	N/A			32	100.0%	1	100.0%	1	100.0%	34	57.6%
Antibiotics Post-op	Yes	23	92.0%	24	75,0%	1	100.0%	1	100.0%	49	84.5%
	Not Stated	2	8.0%	8	25,0%					9	15.5%

List of Figures

Figure 1: Non-Ventriculoperitoneal Shunt Survival Over Time (as Percentage)

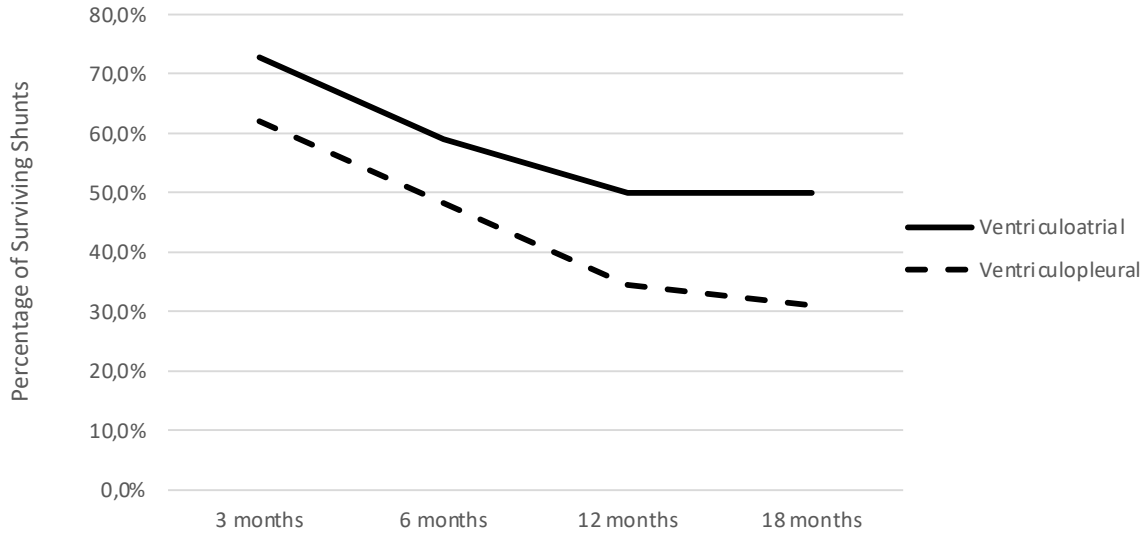
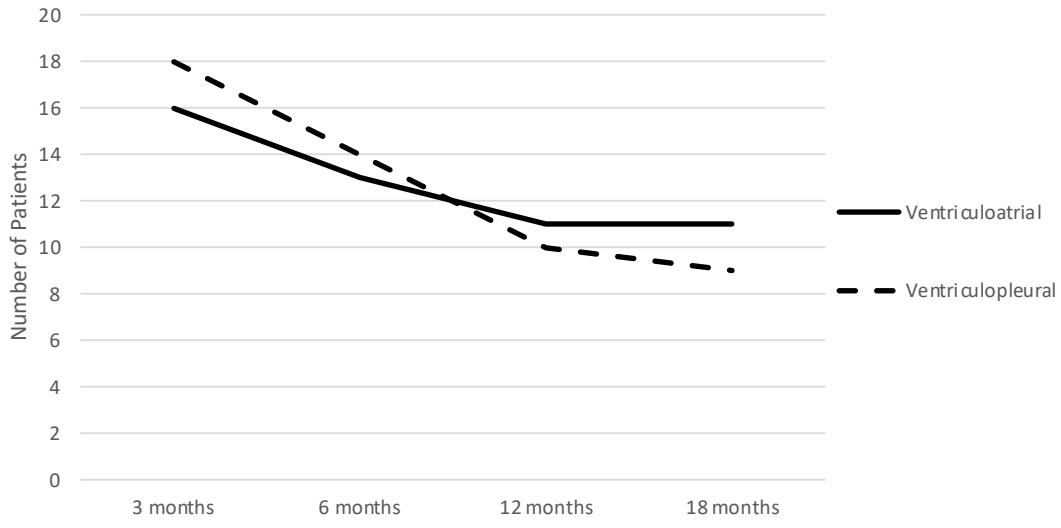


Figure 2: Non-Ventriculoperitoneal Shunt Survival Over Time (Absolute Count)



LIST OF ABBREVIATIONS

CSF - Cerebrospinal fluid

ECG - Electrocardiogram

ETV - Endoscopic third ventriculostomy

EVD - External ventricular drain

HCP – Hydrocephalus

PTB - Pulmonary tuberculosis

RCCH - Red Cross War Memorial Children's Hospital

TBM – TB meningitis

VA Shunt – Ventriculoatrial shunt

VC Shunt – Ventriculocholecystic

VP Shunt - Ventriculoperitoneal shunt

VPL Shunt – Ventriculopleural shunt

VV Shunt – Ventriculovesical shunt

ABSTRACT

Background: At Red Cross War Memorial Children's Hospital (RCCH) it is the preferred practice to use non-ventriculoperitoneal (non-VP) shunts when the peritoneum is ineffective or contra-indicated for cerebrospinal fluid (CSF) diversion, and when endoscopy is not an option. The objective of this study is to evaluate the clinical course of patients having undergone these procedures.

Method: A single centre retrospective review at RCCH wherein forty-three children with a total of 59 episodes of non-VP shunt placement over a 12-year period were identified for inclusion.

Results: Twenty-five ventriculoatrial (VA) and 32 ventriculopleural (VPL) shunts were analysed with a median age at insertion of 2,9 (0,3-14,9) and 5,3 years (0,5-13,4) respectively. The median number of previous shunt procedures prior to VA or VPL shunt insertion was 6,0 (2-28) versus 4,5 (2-17) respectively. Three VA (12,0%) and three VPL (9,4%) shunt patients were lost to follow up. Of those remaining, 10 VA shunts (45,5%) compared to 19 (65,5%) VPL shunts required revision. One ventriculovesical and one ventriculocholecystic shunt were placed in the same patient after 21 and 25 shunt related procedures respectively, and both were revised within 3-weeks of insertion. Median shunt survival was 8 months longer for the VA compared to the VPL shunts, being 13,5 (0-67) and 5 months (0-118) respectively. Complications for VA shunts were low, with the overall shunt sepsis rate in the VA group at 4% (n=1) compared to 15,6% (n=5) in the VPL group.

Conclusion: Our findings support that VA and VPL shunts are acceptable second-line options in an already compromised group of patients where safe treatment options are limited, provided attention is paid to the technical details specific to their placement.

MANUSCRIPT

Introduction

Hydrocephalus (HCP) causes death and disability by increased pressure due to cranial accumulation of cerebrospinal fluid (CSF). [1] It represents a major public health concern globally: Dewan et al., estimated nearly 400,000 new cases of paediatric hydrocephalus annually worldwide, 180,000 of which occur in Africa. [2, 3]

Historically, CSF diversion procedures have been met with varying degrees of success and 36 diversion sites have been reported.[4-6] First attempts by Ferguson in 1898 involved CSF drainage from the lumbar theca to the peritoneum with a silver wire. In 1908 Payr introduced shunting from the ventricle directly into the sagittal sinus and jugular veins, whereas Kausch utilised a rubber conduit and drained CSF into the peritoneal cavity. [7, 8] Heile, in 1914, drained CSF to the pleural space and later in 1925 developed the first ventriculoureteral shunt. [8] In 1952 the first implantable ventriculoatrial (VA) shunt system was developed by Frank Nulsen and Eugen Spitz.[4, 5, 7-9] With the invention of silicone catheters and one-way valves by John Holter in 1956, these shunt systems could better withstand long-term mechanical stresses.[9] Following this, insertion of these systems via the venous route became popular, predominantly into the right atrium. Ransohoff in 1954 returned to the practice of ventriculopleural (VPL) shunting, but this was limited by the development of pleural effusions and shunt obstruction.[8, 10]

By the late 1960's Ames, Raimondi, and Matsumoto popularised the ventriculoperitoneal (VP) shunt technique, utilising the improved silicone devices, and by the 1970's VA shunts became infrequent. This was largely due to the ease of VP shunt insertion and revision rather than long-term efficacy. [5, 8, 10, 11] VP shunts, additionally, required fewer revisions following anatomical growth and had less potential for severe complications.[5, 9, 11, 12]. Consequently, at most centres, modern neurosurgeons have much less experience with placement of non-peritoneal shunts.

At Red Cross War Memorial Children's Hospital (RCCH) it has been the preferred practice to use non-VP shunts when the peritoneum has been deemed ineffective or is contra-indicated for CSF diversion, and where endoscopic third ventriculostomy (ETV) is not an option or has failed. Relative contra-indications to peritoneal placement include peritonitis, pancreatitis, ascites,

traumatic abdominal injuries, and significant adhesions following previous abdominal pathology or surgery.[4] At out institution VPL shunts were the first of the second-line options. Hoffman et al., and Jones et al., reported on their use of VPL shunts, supporting them as a safe alternative with better tolerance and fewer problematic pleural effusions in children over the age of 4 years. [10, 13] Over time, however, we have raised concerns about VPL shunt survival in our setting due to the high pulmonary tuberculosis (PTB) rate, reported to be 737 per 100 000 population.[14] Other concerns include the perceived high complication and poor survival rates of these systems in our context. We therefore have migrated towards VA shunt placement in this group of patients.

The objective of this study is to evaluate our current practice and outcomes of second-line CSF shunt placement when VP Shunt placement or endoscopy is not possible or feasible. We aimed to compare this to the literature and historical context of paediatric VA and VPL shunting.

Methodology

Study Population

This is a single centre retrospective review at the RCCH Neurosurgical Division, Cape Town, South Africa. All children under the age of 13 years (exceptions to this discussed below) who underwent non-VP shunt placement at RCCH between the 1st of January 2009 and the 31st December 2020 were included. Patients were identified from prospectively maintained databases, and their medical and operative records were reviewed. We excluded patients in whom a non-VP shunt had been inserted at another facility.

Shunt Technique

Most VA shunts were inserted by one of the authors (JE), utilising a percutaneous, ultrasound-guided, Seldinger-technique with fluoroscopy to guide shunt placement. Where this was not possible a formal open cut-down procedure was performed. More recently, however, distal catheter insertion length was calculated by measuring the distance from the planned skin incision to the sternal angle (angle of Louis) in order to limit radiation exposure and improve procedural workflow. This, together with chest lead electrocardiogram (ECG) monitoring, aimed to ensure catheter placement in the distal third of the superior vena cava. Catheter selection was typically an

antibiotic-impregnated proximal and distal catheter with a medium pressure Medtronic Atlas® valve.

Placement site for the VPL shunts was surgeon-dependent, with incision at the right-sided 5th intercostal space in the mid-axillary line being the preferred practice. Surgeon allocation was less specific. Blunt dissection to the pleura was followed by pleural opening and insertion of a shortened distal catheter, under valsalva manoeuvre.

Post-operative chest X-rays were routinely utilised in both VA and VPL shunt procedures to confirm catheter position and to exclude complications.

There were only single attempts at ventriculocholecystic (VC) and ventriculovesical (VV) shunts, both of which occurred in the same patient after all other avenues were exhausted. These were performed as an open surgical procedure using an antibiotic-impregnated catheter with the assistance of paediatric surgery and urology teams respectively.

Clinical and Surgical Variables

Variables collected included: demographic information (age; sex; timing and number of CSF diversion procedures); aetiology of hydrocephalus; comorbid conditions; evidence of previous tuberculosis infection; indication for non-VP shunt insertion; procedure related factors; surgical outcomes (time to shunt failure; reason for failure; immediate-, short- and long-term complications; immediate revision procedure following failure); and time of follow up.

Definitions

Typically, children are only managed until 12 years of age at RCCH, after which they are transferred to the adult division at another hospital. Exceptions to this are patients with significant comorbidities and small habitus who warrant ongoing treatment by a paediatric multidisciplinary team until the age of 18 years at RCCH. We divided complications into immediate, short and long-term. Immediate complications occurred at the time of surgery; short-term complications within 30 days of surgery; and long-term complications after 30 days from surgery. Patients were “lost to follow-up” if they did not attend their 3-month follow-up appointment.

Data Analysis

Data was described by measures of central tendency, and univariate and multivariate analysis was conducted through Intel SPSS software®.

Ethical Considerations

Ethical approval was granted by the University of Cape Town Human Research Ethics Committee (REF: 317/2020). All data was anonymised and ethical considerations were maintained throughout.

Results

Between 2009 and 2020 a total of 44 eligible patients were identified. A single patient was excluded because of insertion of a VPL shunt at another institution, leaving 43 patients with non-VP shunts, and 59 episodes of non-VP shunt insertion procedures amongst them.

Baseline Characteristics

A total of 25 VA shunts (42,4%), 32 VPL shunts (54,2%), 1 VV (1,7%) and 1 VC (1,7%) shunt were inserted in those 43 patients. 25 patients were male and 18 female. Nine patients (56,3%) in the VA shunt group and 12 patients (44,4%) in the VPL group had multiple medical co-morbidities (see **table 1**). Most hydrocephalus cases were of unknown aetiology (15 patients (34,9%)), followed by myelomeningocele in 9 patients (20,9%), post-infectious hydrocephalus in 5 patients (11,6%), and tuberculous hydrocephalus in 4 patients (9,3%). The patient who underwent both the VV and VC shunt procedures had post-infectious hydrocephalus.

Preceding Treatment

The median age at first CSF diversion procedure (VP shunt) was 0,4 years (0-1,5 years) for VA shunts versus 0,6 years (0-5,0 years) for VPL shunts. The median age at insertion of a VA shunt was 2,9 years (0,3-14,9 years), and 5,3 years (0,5-13,4 years) for VPL shunts. The median number of previous shunt procedures prior to VA shunt insertion was 6,0 (2-28) versus 4,5 (2-17) for VPL shunts. VV and VC shunts were placed in the same patient after 21 and 25 shunt related procedures respectively. Most indications for non-VPS insertion were related to abdominal pathology (see **table 2**) with 32,2% of shunts inserted due to the presence of abdominal pseudocysts (suspected low-grade infection), 22% due to proven intra-abdominal sepsis, 11,9% due to abdominal

adhesions and CSF malabsorption, 10,2% for hollow viscus perforations/erosions, and 3,4% for iatrogenic bowel injury. One (1,7%) non-VP shunt was inserted due to abdominal tuberculosis. The remaining indications included multiple failed VP shunts with concern of peritoneal malabsorption of CSF (1,7%), or failed other non-VP shunts and persistence of the above-mentioned scenarios (17,0%).

Table 1 Demographics and Baseline Characteristics

		Ventriculoatrial		Ventriculopleural		Ventriculovesical		Ventriculocholecystic		Total	
		n	%	n	%	n	%	n	%	n	%
Route of Non-VPS procedure		25	42,4%	32	54,2%	1	1,7%	1	1.7%	59	100,0%
Sex	Male	9	56,3%	16	59,3%					25	58,1%
	Female	7	43,8%	11	40,7%					18	41,9%
Median Age in Years at first CSF Diversion (Range)		0,4	(0-1,5)	0,6	(0-5,0)	0,8		0,8		0,6	(0-5,0)
Mean Age in Years at first CSF Diversion (+/-SD)		0,5	(0,4)	1,0	(1,2)	0,8		0,8		0,8	(1,0)
Median Age Years at Non-VP Shunt (Range)		2,9	(0,3-14,9)	5,3	(0,5-13,4)	3,6		3,8		4,0	(0,3-14,9)
Mean Age in Years at Non-VP Shunt (+/-SD)		5,2	(4,6)	5,9	(4,1)	3,6		3,8		5,5	(4,3)
Median Number Previous Shunt Procedures (Range)		6,0	(2-28)	4,5	(2-17)	21,0		25,0		5,0	(2-28)
Mean Number Previous Shunt Procedures (+/-SD)		7,7	(6,2)	6,2	(4,4)	21,0		25,0		7,4	(6,0)
Multiple Medical Comorbidity	No	7	43,8%	15	55,6%	1	100,0%	1	100,0%	22	51,2%
	Yes	9	56,3%	12	44,4%					21	48,8%
Aeitology of HCP	Idiopathic	6	37,5%	9	33,3%					15	34,9%
	Myelomeningocele	5	31,3%	4	14,8%					9	20,9%
	Post-infectious	1	6,3%	4	14,8%	1	100,0%	1	100,0%	5	11,6%
	4th Ventricular Outflow Obstruction	2	12,5%	3	11,1%					5	11,6%
	TBM			4	14,8%					4	9,3%
	Intraventricular Haemorrhage	1	6,3%	2	7,4%					3	7,0%
	Tumour related			1	3,7%					1	2,3%
	Aqueductal Stenosis	1	6,3%							1	2,3%
	Total	16	100,0%	27	100,0%	1	100,0%	1	100,0%	43	100,0%

Table 2 Indication for Non-Ventriculoperitoneal Shunt Insertion

		Ventriculoatrial		Ventriculopleural		Ventriculovesical		Ventriculocholecystic		Total	
		n	%	n	%	n	%	n	%	n	%
Indication Non-VPS	Abdominal Pseudocyst	8	32,0%	11	34,4%					19	32,2%
	Intra-abdominal Sepsis	6	24,0%	7	21,9%					13	22,0%
	Significant Abdominal Adhesions	3	12,0%	2	6,3%	1	100,0%	1	100,0%	7	11,9%
	Bowel/bladder perforation/erosion	2	8,0%	4	12,5%					6	10,2%
	Abdominal Surgery	2	8,0%	3	9,4%					5	8,5%
	VPL Shunt Dysfunction	1	4,0%	3	9,4%	1	100,0%			5	8,5%
	Pleural Effusion related to VPL Shunt	3	12,0%							3	5,1%
	VA Shunt Dysfunction	2	8,0%	1	3,1%					3	5,1%
	Multiple Failed VPS (Concern of peritoneal malabsorption)	1	4,0%							1	1,7%
	Bowel Injury	1	4,0%	1	3,1%					2	3,4%
	TB abdomen			1	3,1%					1	1,7%
	Vent-Vesical Shunt Dysfunction							1	100,0%	1	1,7%
	Vent-GB Shunt Dysfunction	1	4,0%							1	1,7%

Shunt Survival and Failure

Follow-up time: Of the 25 VA shunts, 3 (12,0%) were lost to follow up (patients from other provinces). In the VPL group, 3 (9,4%) were lost to follow up. The median time of follow up was 1,9 years (0-8,5 years) for the VA shunt group and 4,2 years (0,3-10,0 years) for the VPL group.

Revision rate: Of the 22 VA shunt patients with follow up, 10 (45,5%) required revision of their VA shunt (see **table 3**). Nineteen VPL patients (65,5%) required revision. Both VV and VC shunts were revised within 3 weeks of placing them due to malabsorption of CSF and hydrocephalus.

Shunt survival: Median shunt survival for the VA shunts was 13,5 months (range 0-67) and 5 months (range 0-118) for VPL shunts. Of the 22 VA shunts included, survival rates at 3, 6, 12, 18 months were 72,7%; 59,1%; 50,0% and 50,0% respectively. In comparison shunt survival in the 29 VPL shunts was 62,1%; 48,3%; 34,5%; and 31,0% at 3, 6, 12 and 18 months respectively (see **figure 1 and 2**). The VV shunt and VC shunt survived 4 and 21 days respectively, before shunt malfunction became evident.

Table 3 Shunt Survival and Reason for Failure

		Ventriculoatrial		Ventriculopleural		Ventriculo-vesical		Ventriculo-cholecystic		Total	
		n	%	n	%	n	%	n	%	n	%
Route of Non-VPS procedure		25	42,4%	32	54,2%	1	1,7%	1	1,7%	59	
Lost to Follow Up		3	12,0%	3	9,4%					6	10,2%
Revision of non-VPS (Y/N)	No	12	54,5%	10	34,5%					22	37,3%
	Yes	10	45,5%	19	65,5%	1	100,0%	1	100,0%	31	52,5%
Median Follow Up Time Years		1,9	(0-8,5)	4,2	(0,3-10,0)					2,7	(0-10,0)
Mean Follow Up Time Years (+/- SD)		2,6	(2,3)	4,4	(3,0)					3,5	(2,8)
Median Shunt Survival Time months (range)		13,5	(0-67)	5	(0-118)					6,0	(0-118)
Mean Shunt Survival Time months (+/- SD)		19,3	(21,1)	21,9	(33,5)					20,0	(28,3)
Shunt Survival at	3 months	16	72,7%	18	62,1%					34	100,0%
	6 months	13	59,1%	14	48,3%					27	79,4%
	12 months	11	50,0%	10	34,5%					21	61,8%
	18 months	11	50,0%	9	31,0%					20	58,8%
Reason Shunt Failure	Shunt Blockage	6	60,0%	5	26,3%	1	100,0%	1	100,0%	13	41,9%
	Pleural Effusion			7	36,8%					7	22,6%
	Shunt Sepsis			4	21,1%					4	12,9%
	Displaced Distal Catheter	1	10,0%	3	15,8%					4	12,9%
	Shunt Disconnection	1	10,0%	1	5,3%					2	6,5%
	Intracranial Sepsis			1	5,3%					1	3,2%
	Endocarditis	1	10,0%							1	3,2%
	Shunt Nephritis	1	10,0%							1	3,2%
	Unsuccessful Shunt Insert	1	10,0%							1	3,2%
	Pleural Empyema			1	5,3%					1	3,2%
Immediate Revision Procedure	External Ventricular Drain	3	30,0%	9	47,4%	1	100,0%	1	100,0%	14	45,2%
	Ventriculo-peritoneal Shunt	6	60,0%	6	31,6%					12	38,7%
	Ventriculo-Pleural Shunt	1	10,0%	3	15,8%					4	12,9%
	Ventriculo-Vesical Shunt			1	5,3%					1	3,2%
	Total	10	100,0%	19	100,0%	1	100,0%	1	100,0%	31	100,0%

Figure 1: Non-Ventriculoperitoneal Shunt Survival Over Time (as Percentage)

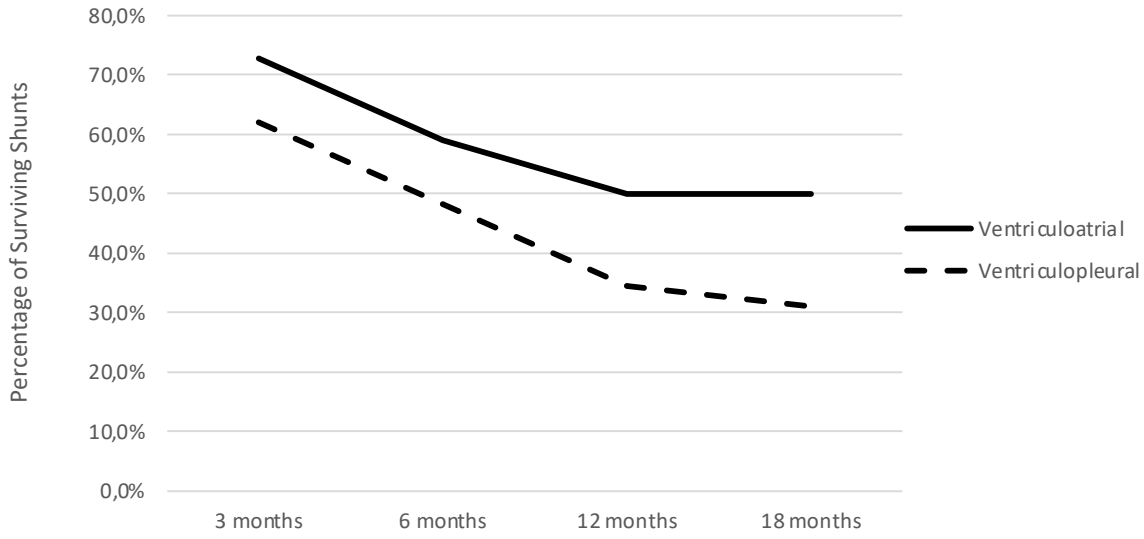
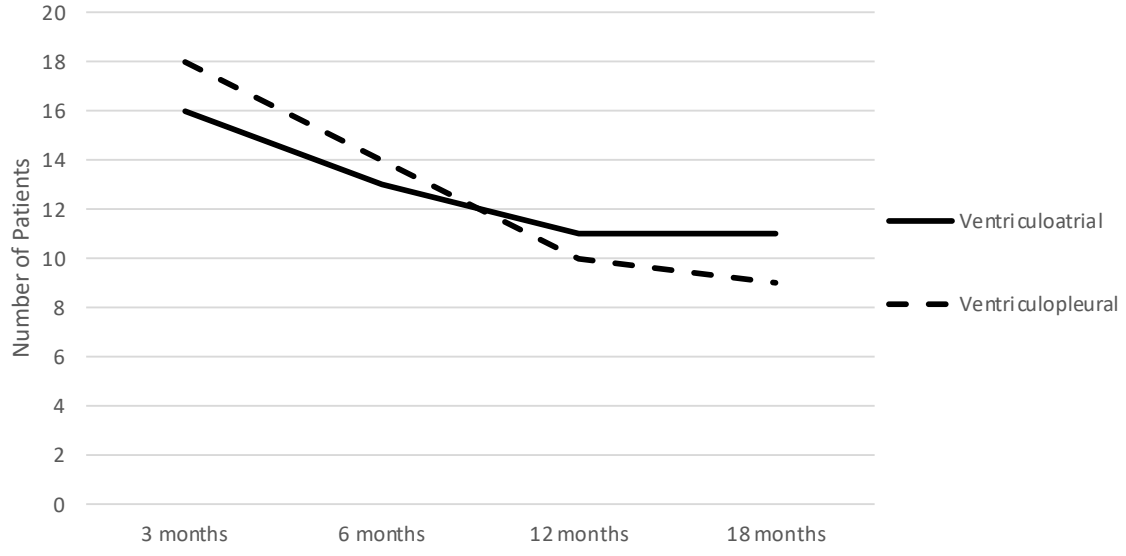


Figure 2: Non-Ventriculoperitoneal Shunt Survival Over Time (Absolute Count)



In the failed VA shunts, shunt blockage accounted for 6 cases (60,0%). Other causes included 1 shunt disconnection (10,0%), 1 displacement of the distal catheter (10,0%), and 1 endocarditis with shunt nephritis (10,0%). Unsuccessful VA shunt insertion was noted in 1 case (10,0%) necessitating a VPL shunt insertion at the time of the attempted VA shunt surgery. Shunt failure in the VPL group was due to shunt blockage in 5 cases (26,3%), symptomatic pleural effusions in 7 (36,8%), shunt sepsis in 3 (15,8%), and displacement of the distal catheter in 3 (15,8%). There was 1 case (5,3%) of pleural empyema.

In cases requiring revision of their VA or VPL shunt, the immediate revision procedure was a VP shunt or EVD in 9 (90,0%) and 15 cases (79,0%) respectively. In the VA and VPL shunt revision group there was sustained management of HCP with VP shunts in 5 (50,0%) and 11 cases (57,9%), respectively. The median time to initial revision of the VA and VPL shunts in this group was 75 and 157 days, respectively. Fifty percent of VA (n=5) and 42,1% (n=8) of VPL shunt revision cases required ongoing management of HCP with non-VP shunts. The median time to initial revision in this group being 34 days and 16 days, respectively.

Complications

Immediate complications: These were more common in the VA shunt group with 9 events in total (see table 4). These were minor in nature and included non-significant, transient arrhythmia (n=3), deep atrial insertion (n=1), failed Seldinger technique requiring formal open cut down (n=3), and arterial puncture (n=1). In the VPL group there was 1 event of surgical emphysema that required no further intervention.

Short-term complications: These were more common in the VPL group and included symptomatic pleural effusion in 5 cases (15,6%) requiring shunt revision, asymptomatic pleural effusion in 6 cases (18.8%) requiring no intervention, and 1 shunt sepsis (3,1%). The VA shunt group had 2 cases of deep atrial insertion of the distal catheter on post op screening (8,0%), and no episodes of shunt sepsis.

Long-term complications: These were more common in the VPL group with 10 events in total, including 2 symptomatic pleural effusions, 1 pleural empyema, and 3 shunt sepsis events, one of which resulted in death. The death occurred in a palliative patient known with severe baseline disability who re-presented with shunt sepsis and demised shortly thereafter, prior to any further

intervention from Neurosurgery. In the VA shunt group, the long-term complications were fewer but significant, with 1 event of endocarditis with shunt nephritis which required VA shunt revision after appropriate temporary diversion and intravenous antibiotics. Of note the patient made a good recovery. Overall the shunt sepsis rate in the VA shunt group was 4% (n=1) and 15,6% (n=5) in the VPL group.

Operative Technique

Ultrasound assistance (Seldinger technique) was used in 92,0% (n=23) of the VA shunt insertions, with 16,0% (n=4) of the shunts requiring a formal open neck dissection. Intra-operative fluoroscopy was performed in 28,0% (n=7). Most cases (92,0%, n=23) received post-operative antibiotics for at least 24 hours.

Table 4 Complications Non-Ventriculoperitoneal Shunts

		Ventriculoatrial		Ventriculopleural	
		n	% of total	n	% of total
Immediate Surgical Complications	Non-significant arrythmia	3	12,0%		
	Failed Seldinger Technique	3	12,0%		
	Deep Atrial Insertion	1	4,0%		
	Arterial Puncture	1	4,0%		
	Minor Blood Loss	1	4,0%		
	Surgical Emphysema			1	3,1%
	Total	9	36,0%	1	3,1%
Short Term Complications (30 Days)	Asymptomatic Pleural Effusion			6	18,8%
	Symptomatic Pleural Effusion			5	15,6%
	Pneumothorax (No intervention)			2	6,3%
	Displaced Distal Catheter			2	6,3%
	Ventriculitis			1	3,1%
	Subdural Hygroma	1	4,0%	1	3,1%
	Deep Catheter in Atrium	2	8,0%		
	Superficial Wound Infection	1	4,0%		
	Total	4	16,0%	17	53,1%
Long-term	Shunt Sepsis			3	9,4%
	Symptomatic Pleural Effusion			2	6,3%
	Asymptomatic Pleural Effusion			1	3,1%
	Empyema			1	3,1%
	Displaced Distal Catheter			1	3,1%
	Death			1	3,1%
	Subdural Hygroma	1	4,0%	2	6,3%
	Shunt Disconnection	1	4,0%	1	3,1%
	Endocarditis	1	4,0%		
	Shunt Nephritis	1	4,0%		
	Total	4	16,0%	12	37,5%

Discussion

VP shunts remain the preferred CSF diversion procedure to manage hydrocephalus, with the use of non-VP shunts reserved for instances in which the peritoneum is contra-indicated or has failed previously, except where ETV is appropriate. [15, 16]

Efficacy of VA and VPL Shunts

The Shunt Design Trial and Hydrocephalus Research Network's studies demonstrate a shunt failure rate of up to 47% in the first 2 years. [17, 18] By comparison, the VA shunt failure rate of 45% (median follow up 1,9 years) in our current series, is not dissimilar. However, the VPL shunt group had a 62% failure rate (median follow up 4,2 years). Although the duration of follow-up for the latter group is longer, there appears to be a difference at 12 months (Figure 1). Although differences between the groups due to the non-randomized selection are unavoidable, it is notable that the VA shunted patients were younger and therefore a higher rate of shunt dysfunction may have been expected.

The lower shunt survival rate for VA and VPL shunts is expected as these procedures are typically performed as second-line measures within a very heterogenous subset of patients who already have had several shunt complications. The patient demographic (low- and middle-income) and spectrum of disease may contribute to this. Warf et al., reported that up to 60% of hydrocephalus in Africa is due to infection. [19] In our study 9% of hydrocephalus was related to myelomeningocele, 11,6% to post-infectious cases, and 9,3% to TBM-related hydrocephalus. It is likely that many of the unknown aetiology group were post-infectious cases. Additionally, the insertion of VA and VPL shunts are undertaken in complex patients as evidenced by the high rate (48,8%) of medical co-morbidities and the high number of previous shunt-related procedures in our sample. One child had 28 prior shunt procedures.

Direct comparison between VPL and VA shunts as a second-line option is sparse, and is compromised by small numbers, relatively short follow-up, and compounded by the greater complexity of these patients in recent series. [20] Much of the available literature derives from historical series where these systems were inserted as a primary procedure and may not be directly applicable because surgical and perioperative techniques have changed over the intervening years.

These earlier studies, however, highlight the efficacy of these systems in the absence of other compounding comorbidities. Keucher and Mealey in 1979 demonstrated similar mortality and infection rates for VA and VP shunts in 228 patients with infantile non-neoplastic hydrocephalus, although VA shunts had more revisions, and late complications were more frequent and severe. [12]

Vernet et al., from 1970 to 1991, reviewed 120 cases of infantile hydrocephalus who underwent VA shunting as a primary procedure. [21] With an average follow up of 11 years, they demonstrated no operative mortality and only 1 shunt-related death which was secondary to shunt nephritis. Their infection rate was 4.2% with an average revision rate of 2.2 per patient. Of note, 66% of revisions were for elective lengthening of the atrial catheter. [21] Due to this disadvantage they supported VP shunts over VA shunts as a primary procedure. [21] A Norwegian study of 128 children followed up children who received a VA shunt as a primary procedure between 1967 to 1970 over a 45-year period: 30% of shunts were revised within a year, and 73% within the first decade, with 26,3% of revisions done for elective lengthening of the catheter.[22] Rymarczuk et al. showed no difference in the survival of VA vs VP shunts, excluding elective lengthening procedures in the VA group. [23]

Of interest in the adult population with VA shunts, where fewer revisions due to growth are needed, Lam and Villemure (49 patients), and Al-Schameri et al. (255 patients) demonstrated similar infection and complication rates between VP and VA shunts. [24, 25] Both favoured VP shunts as a primary procedure due to ease of placement and less potential for a severe complication. [24]

Yavuz et al. studied VA shunts as a second-line option in 10 patients aged 5 to 13 years. They reported 3 revisions due to thrombosis, endocarditis and pulmonary embolus.[26] Clark et al., also studied 94 VA shunt insertions in 38 patients as a second-line intervention. They reported higher revision rates to ours, with shunt survival rates of 53%, 43% and 27% at 6, 12 and 24 months respectively, and an overall infection rate of 11%.[27] They concluded that the percutaneous ultrasound-guided technique was safe with a serious adverse event rate of only 2%.

We prefer to reserve the use of VPL shunts for children over the age of 4 years, due to concerns about pleural effusions where lung capacity and compliance may be reduced. Hoffman et al., had a similar approach; 12 (20%) of their 59 patients developed pleural effusions, 6 of which were under 11 months of age. Twenty-three of their patients required no revision. [10] Jones et al., in 52 VPL shunt patients (mean age of 8 years) reported 3 shunt infections, 4 catheter obstructions, 1 symptomatic pleural effusion, and 1 death from shunt malfunction. [13] Martiınez-Lage et al., in 6 patients (5 to 13 years) noted no revisions after a mean follow-up of 2,5 years. [28] In an adult population, where pleural effusion may be less concerning, Craven et al. in 2017 studied 22 VPL shunts and reported a median shunt survival of 14 months. [29]

In a recent review Forte et al., found similar results to ours in their VPL and VA shunt comparison.[20] In a series of 36 VA shunt and 18 VPL shunt insertions over 15 years, VA shunt survival was 60.6%, 51.5% and 36.4% at 3, 6 and 12 months respectively, while VPL shunt survival was 56.3%, 43.8% and 37.5% respectively.[20] Median time to shunt revision was 8,5 and 5,5 months for VA and VPL shunts respectively. We concur with their conclusions about the role of VA or VPL shunts as a second-line procedure. They advised consideration of VA over VPL shunt insertion in those under 5 years. [20] Rymarczuk et al. in their review of 85 VA shunt patients over a 13 year period, further agrees with the second-line role these shunt systems serve and demonstrated similar outcomes to those above. [23]

VA and VPL shunt use may afford time for the peritoneum to heal, allowing later re-introduction of a VP shunt. In our series VP shunting was undertaken for 50,0% of VA (n=5) and 57,9% (n=11) of VPL shunt revisions. Once the original insult contraindicating the peritoneum has resolved, a VP shunt can be reconsidered in the settings of non-VPS failure.

Current Technique of VA shunts

With our shifting focus from VPL to VA shunts, one of the objectives was to evaluate the VA shunt placement technique. The Seldinger technique (percutaneous guidewire assisted placement), first described in 1981, has become preferred.[30] Clark et al. described the assistance of ultrasound guidance and intra-operative fluoroscopy to confirm distal tip position. [27] More recently, Della Pepa et al., reported venous catheter insertion under ultrasound guidance with

ECG-guided distal tip positioning. [31] This technique utilises an electrode-integrated venous catheter and relies on predictable changes in the ECG p-wave trace as the atrium is approached. [31] This technique was similarly described by Muhammad et al. and by McCracken et al. [32, 33]

In our study 23 (92,0%) VA shunts were inserted with the Seldinger technique under ultrasound guidance, with intra-operative fluoroscopy performed in 7 (28,0%). We noted that a technique using patient measurements together with chest lead ECG monitoring, to aid in correct catheter placement within the lower third of the superior vena cava, is safe and effective. Deep IVC placement was seen in only 2 cases (neither of these cases were done with fluoroscopy nor measurement to the angle of Louis techniques, with rather an estimate of 10cm used instead by the surgeon); both these patients remained well and have not required revision as of 2022.

Complications

Short and long-term complications were more common in the VPL group, most of which (n=7/21,9%) were related to pleural effusions with 1 case of pleural empyema (3,1%). This compared to the cohort by Forte et al., which reported a rate of pleural effusions at 22,2%.[20] In our study the shunt sepsis rate for the VA and VPL shunt group was 4% (n=1) and 15,6% (n=5) respectively, compared to infection rates reported by Forte et al., of 13.8% and 5.6% for VA and VPL shunts. [20]

Reported complications for VPL shunts include pneumothorax, lung injury, ventilatory difficulties, pneumocephalus, tension hydrothorax and fibrothorax.[4] Small asymptomatic pleural effusions are also commonly described.[34] Reported VA shunt complications include catheter thrombosis, thrombo-embolism (including pulmonary emboli), vessel perforation, bacterial endocarditis, arrythmia, nephritis, pulmonary hypertension and cor pulmonale. [4, 11, 12, 35] Interestingly, Vandersteene et al., demonstrated a pro-coagulant effect of CSF which is attributable to coagulation proteins and tissue factor.[36] Generally CSF concentrations in the venous system are well below the critical threshold required; however in certain circumstances they may increase the risk of clot formation. [36] Shunt-nephritis was first described in 1965 and is typically thought to arise from infection with low virulence organisms, triggering an immune complex deposition at the glomerular basement membrane. [11, 35, 37] In our one case of shunt-nephritis, a VP shunt

was inserted after antibiotic treatment. At the time of this study this patient is still doing well, with no signs of recurrence and no long-term sequelae.

Limitations of this study include its retrospective nature, descriptive methodology, limited long-term follow-up and the small cohort, the size of which precluded a reliable comparative statistical analysis. Due to the more recent insertion of VA shunts a lower median follow up time was encountered, which may also contribute to the apparent higher survival rate of these shunt systems. Comparison to conventional VP shunt survival and complications is limited by the selection criteria. Larger studies with longer term follow-up are recommended in order to establish robust clinical guidelines. However, due to small numbers, a multi-centre approach is necessary.

Conclusion

Our findings are comparable to recent studies of similar design, and support the use of VA and VPL shunts as effective second-line CSF diversion procedures. Additionally, we have, at times, found these procedures to act as a useful interim measure, "buying time" until the peritoneum can be re-considered as a CSF diversion site. No shunt operation is without its risks, but it is clear that in an already compromised group of patients where safe treatment options are limited, VA and VPL shunts remain good alternative options and should not be discarded by the neurosurgeon. Trainees should be taught correct, safe surgical techniques to reduce complications.

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