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Review Article

The efficacy of cystoperitoneal shunting for the surgical management of intracranial arachnoid cysts in the elderly: A systematic review of the literature

Joseph Merola[1], Susruta Manivannan[2], Setthasorn Ooi[3], Wen Li Chia[3], Milan Makwana[1], Jozsef Lang[1], Paul Leach[1], Malik J. Zaben[4]

[1]Department of Neurosciences, University Hospital of Wales, Cardiff, [2]Department of Neurosurgery, Southampton General Hospital, Southampton General Hospital, Southampton, [3]Institute of Psychological Medicine and Clinical Neurosciences, School of Medicine, Cardiff University, University Hospital of Wales, [4]Department of Neurosurgery, Cardiff and Vale University Health Board, Cardiff, United Kingdom.

E-mail: Joseph Merola - merolaj@doctors.org.uk; Susruta Manivannan - manivannansusruta@gmail.com; Setthasorn Ooi - ooisz@cardiff.ac.uk; Wen Li Chia - chiaw@cardiff.ac.uk; Milan Makwana - milanmakwana@hotmail.com; Jozsef Lang - jozsef.lang@wales.nhs.uk; Paul Leach - paul.leach2@wales.nhs.uk; *Malik J. Zaben - zabenm@cardiff.ac.uk

***Corresponding author:**
Malik J. Zaben,
Department of Neurosurgery,
Cardiff and Vale University
Health Board, Cardiff,
United Kingdom.

zabenm@cardiff.ac.uk

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ABSTRACT

Background: Intracranial arachnoid cysts (AC) are benign, cerebrospinal fluid filled spaces within the arachnoid layer of the meninges. Neurosurgical intervention in children and young adults has been extensively studied, but the optimal strategy in the elderly remains unclear. Therefore, we performed a single center retrospective study combined with a systematic review of the literature to compare cystoperitoneal (CP) shunting with other surgical approaches in the elderly cohort.

Methods: Retrospective neurosurgical database search between January 2005 and December 2018, and systematic review of the literature using PRISMA guidelines were performed. Inclusion criteria: Age 60 years or older, radiological diagnosis of intracranial AC, neurosurgical intervention, and neuroradiological (NOG score)/clinical outcome (COG score). Data from both sources were pooled and statistically analyzed.

Results: Our literature search yielded 12 studies (34 patients), which were pooled with our institutional data (13 patients). CP shunts (7 patients; 15%), cyst fenestration (28 patients; 60%) and cyst marsupialisation/resection (10 patients; 21%) were the commonest approaches. Average duration of follow-up was 23.6, 26.9, and 9.5 months for each approach, respectively. There was no statistically significant association between choice of surgical intervention and NOG score ($P = 0.417$), COG score ($P = 0.601$), or complication rate ($P = 0.955$). However, CP shunting had the lowest complication rate, with only one patient developing chronic subdural haematoma.

Conclusion: CP shunting is a safe and effective surgical treatment strategy for ACs in the elderly. It has similar clinical and radiological outcomes but superior risk profile when compared with other approaches. We advocate CP shunting as first line neurosurgical intervention for the management of intracranial ACs in the elderly.

Keywords: Cystoperitoneal shunt, Endoscopic fenestration, Intracranial arachnoid cysts, Marsupialisation

INTRODUCTION

Arachnoid cysts are benign, cerebrospinal fluid (CSF)-filled spaces within the arachnoid layer of the meninges. They have a predilection for the middle cranial fossa but can appear in the anterior

and posterior cranial fossae and more rarely within the sella or parenchyma.[59] They are commonly first encountered incidentally, resulting in an estimated prevalence of 1.1–2.3% in adults and 2.6% in children.[43,55]

Individuals may remain asymptomatic for their entire life despite the apparent expansive behaviour of arachnoid cysts which can, in some instances, cause mass effect on surrounding structures. Symptomatic lesions tend to present with signs and symptoms of raised intracranial pressure, focal neurological deficit, or cortical irritation resulting in seizure activity. The patients may also suffer with neuropsychiatric disturbances, which have been shown to improve following cyst decompression.[18] Several studies have examined the clinical features and management of arachnoid cysts in the pediatric cohort. While the underlying aetiology of arachnoid cysts in children is more likely congenital,[25] in adults there are alternative plausible aetiologies for example traumatic brain injury.[18] Within studies exploring arachnoid cysts in adults, there is a paucity of data regarding management in elderly patients. However, a recent Swedish population-based study reported a prevalence as high as 2.3% in the elderly.[43]

The general consensus among neurosurgeons dictates that surgical decompression is recommended for symptomatic cysts and can lead to improved quality of life.[37] However, there are a several surgical options including needle aspiration, cystoperitoneal (CP) shunting, endoscopic fenestration (into basal cisterns, subarachnoid space or ventricles), or craniotomy for cystectomy or marsupialization.[7,38,40,61] Despite considerable evidence regarding surgical treatment in children and young adults, the optimal surgical strategy in the elderly remains unclear. In this study, we combine our retrospective data with a systematic review of the literature to clarify the role of CP shunting in elderly patients with symptomatic intracranial arachnoid cysts.

MATERIALS AND METHODS

Institutional data

A retrospective review of our local neurosurgical database between January 2005 and December 2018 was performed to identify patients fulfilling the following criteria: (1) age more than or equal to 60 years; (2) radiological diagnosis of arachnoid cyst or histological diagnosis if available; (3) surgical management of arachnoid cyst. The following data were extracted: patient demographics, clinical presentation, anatomical location of cyst, cyst size and volume, Galassi score (if located in the middle cranial fossa), choice of surgical intervention, post-operative complications, clinical/neuroradiological outcomes, and duration of follow-up. Clinical (COG) neuroradiological outcomes (NOG) at follow-up were defined in accordance with the classification system designed by Helland and Wester (2007) [Table 1].[21]

Table 1: Helland and Wester (2007)[12] Outcome classification for surgically treated arachnoid cysts.

COG 1	Symptom resolved	NOG 1	Cyst no longer visible
COG 2	Symptom reduced	NOG 2	Fluid volume still visible but<50%
COG 3	Symptom unchanged	NOG 3	Fluid volume still visible but>50%
COG 4	Symptom worse	NOG 4	No change

NOG: Neuroimaging outcome groups, COG: Clinical outcome group

Systematic review of the literature

Systematic review of the literature was performed in accordance with Preferred Reporting Items for Systematic Reviews and Meta-Analyses guidelines.[36] The protocol for this systematic review was registered on PROSPERO (CRD42020180628).

Search strategy

A multi-database (PubMed, Embase, Web of Science) literature search between January 1980 and May 2020 was performed by authors SO and WC. Conflict of opinion was settled by PL and MZ. Varying combinations of the following search terms were used: arachnoid cyst, cystoperitoneal shunt, endoscopic fenestration, and marsupialization. Only articles in English were included in the study. The bibliographies of identified papers were examined to identify any further relevant articles. All titles and abstracts were reviewed. Eligible studies were included if they satisfied the following criteria: 1) diagnosis of intracranial arachnoid cysts; 2) patient age equal to or >60 years; 3) reported outcomes of surgical intervention including, but not limited to, CP shunt, endoscopic fenestration, and marsupialisation. Any uncertainties were discussed with the authors JM/MZ.

Data extraction/analysis

Data were extracted from selected papers and included patient demographics, clinical presentation, anatomical location of cyst, cyst size and volume, Galassi score (if located in the middle cranial fossa), choice of surgical intervention, post-operative complications, clinical/neuroradiological outcomes, and duration of follow-up. Critical appraisal of included studies, and risk of bias was analysed by SM, MM, and JL using an adapted version of the risk of bias in non-randomised studies of interventions assessment tool.[48]

Statistical analysis

Baseline characteristics (patient demographics, clinical presentation, cyst location, size, volume) were summarized

using descriptive statistics. Continuous variables were reported as means with standard deviation. Outcome data (NOG, COG, complication rates) were analyzed using the Chi-square statistic given the non-parametric distribution of our data. For this purpose, NOG/COG was dichotomized into satisfactory outcomes defined as outcomes 1–2, and unsatisfactory defined as 3–4.

RESULTS

Retrospective analysis of our institutional data yielded a total of 13 patients within the elderly cohort that underwent surgical management of arachnoid cysts. Average age was 66 years, and the majority of patients were female (n = 10; 76.9%). Cyst locations included frontal (n = 4; 30.8%), posterior fossa (n = 4; 30.8%), sellar (n = 2; 15.4%), intracerebral (n = 1; 7.7%), interhemispheric (n = 7.7%), and middle cranial fossa (n = 1; 7.7%). Cysts were either left sided (n = 6; 46.2%), right sided (n = 2; 15.3%), or located in the midline (n = 5; 38.5%). Four patients (30.7%) underwent CP shunt insertion, 6 underwent cyst fenestration (46.2%) – five were through craniotomy and one through endoscope, 2 patients had cyst marsupialisation (15.4%), and 1 patient (7.7%) underwent a mix of endoscopic fenestration and CP shunt insertion [Table 2].

Following systematic review of the literature, a total of 12 articles were eligible for inclusion, consisting of: 4 observational cohort studies, 4 prospective cohort studies, 3 non-randomised comparative studies, and 1 retrospective study. The majority of include studies were at a moderate risk of bias (n = 9; 75%) [Table 3]. A total of 34 patients were isolated from included studies [Appendix 1 for individual details] and pooled with our institutional data for further analysis, yielding a total of 47 patients. Average age of included patients was 67 years (range 60–94 years), and the majority of patients were female (n = 25; 53%). The most common location was within the middle cranial fossa (n = 19; 40%), with a mode of 3 for Galassi score – 4 were Galassi 1, 4 were Galassi 2 and 5 were Galassi 3 (6 were unstated). The remainder was constituted of frontal (n = 8; 17%), posterior fossa (n = 6; 13%), sellar/suprasellar (n = 5; 11%), interhemispheric fissure (n = 4; 9%), intra- or para-ventricular (n = 4; 9%), and intracerebral (n = 1; 2%) regions. The cyst dimensions were quantified for nine patients (19%) with an average volume of 21.3 cm^3 (range 7.2–65.4 cm^3).

CP shunting was performed on 7 patients (15%), whilst the majority of patients underwent cyst fenestration (n = 28, 60%). Cyst fenestration was specified as being performed endoscopically (n = 3) or via craniotomy (n = 4) in 7 patients, whilst the remainder were unspecified. Of the remainder, craniotomy and cyst resection or marsupialisation (n = 10; 21%), ventriculoperitoneal shunt insertion (n = 1; 2%), or combined endoscopic fenestration and CP shunting (n = 1; 2%) were performed. The final two patients were excluded from further analysis. The average follow-up duration was 17.8 months (range 0–96 months).

The clinical and radiological outcomes were analyzed according to Helland and Wester (2007) classification[21] [Table 1]. Only one complication was reported in the CP shunting group: A chronic subdural haematoma requiring burr hole drainage over a 3-year follow-up period. Complications in the fenestration group (n = 5, 18%) included an acute subdural hematoma, CSF leak and meningitis, subdural hygroma, and 2 cyst recurrences requiring re-treatment. Complications in the cyst resection/marsupialization group (n = 2, 20%) included wound dehiscence requiring washout and antibiotics and recurrence of cyst requiring repeat surgery. Average duration of follow-up was 23.6, 26.9, and 9.5 months for CP shunting, cyst fenestration, and cyst resection groups

Table 2: Baseline characteristics of patients who underwent surgery at our institution.

S. No.	Age	Sex	Location	Side	Volume (cm^3)	Surgery type
1.	69	F	Frontal	L		CP Shunt
2.	78	F	Frontal	L		CP Shunt
3.	60	F	Frontal	L	52.6	CP Shunt
4.	61	F	Frontal	L		CP Shunt
5.	60	F	IC-O	L		Marsupialisation
6.	64	F	Interhemispheric	M	26.3	Fenestration-C
7.	64	F	PF	R	9.8	Fenestration-C
8.	65	F	PF	R	9.8	Fenestration-C
9.	67	F	PF	M	20.2	Fenestration-C
10.	60	M	PF	M	65.4	Fenestration-C
11.	70	F	Sellar	M	4.7	Fenestration-E
12.	79	M	Sellar	M	2.6	Marsupialization
13.	63	M	MCF	L	22.3	Fenestration–E+CP shunt

MCF: Middle cranial fossa, IC-O: Intracerebral occipital, PF: Posterior fossa, L: Left, R: Right, M: Midline, CP: Cystoperitoneal Fenestration, C: craniotomy and fenestration, E: Endoscopic fenestration

Table 3: Summary of risk of bias assessment as per ROBINS-I criteria.

S. No.	Study	Confounding	Selection	Intervention classification	Deviation from intervention	Missing data	Measurement of outcome	Selection of reported result	Overall
1.	Gjerde *et al.* 2013	Low	Low	Low	Low	Low	Low	Moderate	Moderate
2.	Isaksen *et al.* 2013	Low	Low	Low	Low	Moderate	Low	Moderate	Moderate
3.	Khan 2012	Low	Low	Low	Low	Critical	Low	Serious	Critical
4.	Maher and Goumnerova 2011	Low	Low	Low	Low	Moderate	Moderate	Low	Moderate
5.	Oertel *et al.* 2009	Low	Low	Low	Low	Low	Moderate	Low	Moderate
6.	Shim *et al.* 2012	Low	Low	Low	Low	Low	Moderate	Low	Moderate
7.	Shou *et al.* 2015	Low	Low	Serious	Low	Low	Critical	Serious	Critical
8.	Tabakow *et al.* 2019	Low	Low	Low	Low	Low	Moderate	Low	Moderate
9.	Tunes *et al.* 2013	Low	Low	Low	Low	Moderate	Moderate	Low	Moderate
10.	Wester 1996	Low	Low	Low	Low	Low	Low	Moderate	Moderate
11.	Choi 2011	Low	Low	Serious	Low	Low	Serious	Moderate	Serious
12.	Cilluffo 1983	Low	Low	Low	Low	Low	Moderate	Low	Moderate

ROBINS-I: Risk of bias in non-randomised studies of interventions

respectively. Overall, there were no statistically significant associations between NOG score (P = 0.417), COG score (P = 0.601), or complication rate (P = 0.955) and surgical approach [Tables 4 and 5].

Cyst resection/marsupialisation had the lowest complication rate at 10% but with the shortest follow-up duration, while fenestration had the highest complication rate at 18% with the longest follow-up duration.

Table 4: Outcomes categorized according to surgical treatment method.

Surgical treatment	NOG score				COG score			
	1	2	3	4	1	2	3	4
Cystoperitoneal shunt	2	4	1	0	4	2	1	0
Fenestration	1	16	7	1	11	12	0	1
Resection	4	3	1	0	4	4	1	0

NOG: Neuroradiological outcomes, P=0.417., COG=clinical outcomes, P=0.601.

Table 5: Surgical complication rates and average follow-up time P=0.955.

Surgical treatment	Complication (%)	Follow-up (months)
Cystoperitoneal shunt	1/7 (14)	23.6
Fenestration	5/28 (18)	26.9
Resection	2/10 (20)	9.5

DISCUSSION

It is generally agreed that symptomatic intracranial arachnoid cysts require surgical treatment to alleviate symptoms and prevent progression to permanent neurological deficit. Surgical treatment options include needle aspiration, cyst fenestration, cyst resection/marsupialization, or CP shunting. The optimal strategy remains enigmatic among the neurosurgical community. To date, several studies examine evidence for optimal management in children and young adults, but not in the elderly. Our findings demonstrate that CP shunting is an effective first line management option for management of arachnoid cysts in the elderly.

CP shunting is generally considered safe and effective with relatively low rates of morbidity and mortality. Our findings demonstrate no significant difference in outcome when comparing this approach with fenestration or surgical resection. From studies included in our systematic review, only one patient that underwent CP shunting suffered a complication. This patient developed a chronic subdural hematoma over a 3-year follow-up period and required burr hole drainage. There were, however, no reports of repeat surgery for the arachnoid cyst itself. In contrast, complications

of cyst fenestration or resection were more directly related to the procedure. Although development of subdural collections following cyst fenestration is a recognized neurosurgical complication in younger patients, we could not identify any studies reporting this in the elderly. This could be due to underreporting of conservatively managed subdural collections or a true difference in risk of this complication in the elderly. For example, a study by Tunes *et al*. (2014) reported outcomes from seventeen patients surgically treated for temporal arachnoid cysts, of which four patients were included in our review. Whilst the development of chronic subdural hematoma was reported in three patients, the age of the respective patients was not specified.[54]

Another concern following treatment of arachnoid cysts is recurrence or failure requiring repeat surgery. Our results demonstrate that repeat surgery was not required in any patients that underwent CP shunting during an average follow-up 23.6 months. However, three patients that required repeat surgery due to recurrence of symptoms had initially undergone cyst fenestration or resection, with an average follow-up 35 months. Further studies are required to elucidate whether this remains true over longer periods of follow-up. A recent review by Hall *et al*. (2019) reported repeat surgery rates of 24.4% following endoscopic cyst fenestration, and 14.7% after microsurgically treated patients. Only three young patients underwent CP shunting, and one required further surgery due to a chronic subdural hematoma that was also present pre-operatively.[17]

Although shunt dependence is a post-surgical concern following CP shunting,[29] this was not demonstrated in our review. Included studies did not consistently report choice of valve, but in our cohort we used either low (n = 3) or medium (n = 1) fixed pressure valves. The average follow-up of the patients from our department was 15.3 months, and thereafter, no patient was re-admitted with symptoms or signs suggestive of shunt dependence. Nonetheless, advancements in shunt valve technology provide several solutions to this potential complication.

CP shunting of arachnoid cysts has been shown to have similar outcomes in the more widely studied younger population.[1] In an elderly population, CP shunting is equally effective at achieving satisfactory outcomes with a lesser degree of associated morbidity. Indeed, shunt-associated morbidity such as shunt infection or dependence appears less prevalent in this age group, demonstrated by the BASICS study finding of only 1% of shunt revisions due to infection in the elderly.[32] This is likely to decrease further with advancements in shunt valve technology, neuro-navigation, and reduced life-time risk. A shunt is also likely to be the first choice when there is associated hydrocephalus. Furthermore, the inherent risks of having a shunt that are considered in a younger population, such as commitment to a lifetime of shunt dependence or future employment impediments, are arguably less of an issue in the elderly.

Study limitations

The authors acknowledge limitations of this review. The review of our institutional cohort was retrospective, sample size is small and there is variation in follow-up time. The systematic review included only non-randomised studies and bias assessment indicated moderate to critical bias. Larger randomized trials are needed to address this especially in the context of an ageing population.

CONCLUSION

CP shunting is a safe and effective first line surgical management strategy for symptomatic intracranial arachnoid cysts in the elderly population. Similar clinical and radiological outcomes are demonstrated when compared with cyst fenestration or resection, but a superior risk profile and reduced surgical burden. Considering its less invasive nature, CP shunt may therefore be considered the first line surgical treatment of intracranial ACs.

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Declaration of patient consent

Patient's consent not required as patients identity is not disclosed or compromised.

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Conflicts of interest

There are no conflicts of interest.

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APPENDIX

Appendix 1: Literature review.

Anderson	1966	Case report
Little	1973	Case series
Benedetti *et al.*	1977	Case series
Dyck	1977	Case report
Handa	1977	Case report
LaCour	1978	Case report
Leo	1979	Case series
Harter *et al.*	1980	Case report
Tsuda	1980	Case series
Spaziante	1981	Case series
Tsuda	1981	Case series
Modesti	1982	Case series
Harada	1983	Case report
Korosue	1983	Case series
Cilluffo	1983	Observational cohort
Clavel	1985	Case series
Nakagawa	1985	Case report
Kurokawa	1986	Case report
Spaziente *et al.*	1986	Case report
Lee	1987	Case report
Wiener	1987	Case series
Wiener	1987	Case series
Kohn	1989	Case series
Sakai	1989	Case report
Iwama	1991	Case report
Shirakawa *et al.*	1991	Case report
Yamakawa	1991	Case report
Hirohata *et al.*	1992	Case series
Minamori	1992	Case report
Yanaka	1992	Case report
Rivieres *et al.*	1993	Case series
Servadei	1993	Case series
Caruso *et al.*	1994	Case report
Watanabe *et al.*	1994	Case series
Koga	1995	Case report
McAndrew	1995	Case report
Wester	1996	Observational cohort
Takeuchi *et al.*	1996	Case report
Paladino	1998	Case series
Huang *et al.*	1999	case report
Kim	1999	Case series
Miyamoto *et al.*	1999	Case series
Patankar	1999	Case report
Ryu	1999	Case report
Saeki	1999	Case report
Sandberg	1999	Case report
Lutcherath	2000	Case series
Sommer	2000	Case report
Stowe	2000	Case series
Takai *et al.*	2000	Case report
Kollias	2001	Case series
Martin	2001	Case report
Thompson	2001	Case report
Weil	2001	Case report
Hishikawa *et al.*	2002	Case series
Marcoux *et al.*	2002	Case report
Pena	2002	Case report
Schoner	2002	Case report
Topsakal	2002	Case report
Yamasaki *et al.*	2002	Case report
Tohma	2004	Case report
Yasuda	2005	Case report
Mitsos	2006	Case report
Ramtahal	2006	Case report
Strojnik	2006	Case report
Thorat	2006	Case report
Tucker	2006	Case report
Chhabra *et al.*	2007	Case report
Kotil *et al.*	2007	Case report
Ohnishi	2007	Case report
Sivaraman	2008	Case report
Wester	2008	Case report
Oertel *et al.*	2009	Prospective cohort
Suzuki *et al.*	2009	Case report
Chen *et al.*	2010	Case report
Kim	2010	Case series
Lwu	2010	Case report
Marin	2010	Case report
Mattox	2010	Case report
Oertel *et al.*	2010	
Maher and Goumnerova	2011	Non-randomised trial
Park *et al.*	2011	Case report
Funaki *et al.*	2012	Case report
Lindvall *et al.*	2012	Case report
McLaughlin	2012	Technical note
Shim *et al.*	2012	Prospective cohort
Tahir	2012	Case report
Gaberel *et al.*	2012	Case report
Verhoeven	2012	Case report
Zheng *et al.*	2012	Case report
Choi *et al.*	2013	Observational cohort
Khan	2013	Retrospective review
Graillon *et al.*	2013	Case report
Idris *et al.*	2013	Case report
Isaksen *et al.*	2013	Non-randomised trial
Gjerde *et al.*	2013	Observational cohort
Zanini	2013	Case report
Tunes *et al.*	2014	Non-randomised trial
Oyama	2014	Technical note
Tarantino *et al.*	2014	Case report
Balani *et al.*	2015	Case report
Hayashi *et al.*	2015	Case report
Shou *et al.*	2015	Prospective cohort
Ogawa *et al.*	2015	Case report
Ruiz-Juretschke	2015	Case report

(Contd...)

(Contd...)

Appendix 1: *(Continued).*

Dawkins *et al.*	2016	Case report
Feletti *et al.*	2016	Case report
Sugimoto *et al.*	2016	Case report
Bucuk *et al.*	2017	Case report
Di Gaeta *et al.*	2017	Case report
Hendrix *et al.*	2017	Case report
Kirschenbaum	2017	Case report
Mormont *et al.*	2017	Case report
Olvera-Castro	2017	Case report
Corona-Ruiz *et al.*	2018	Case report
Qin	2018	Case report
Cova *et al.*	2019	Case report
de Oliveira *et al.*	2019	Case report
Tabakow *et al.*	2019	Prospective cohort
Kim	2019	Case report
Masaki *et al.*	2019	Case series
Wahl *et al.*	2019	Case report
Kimura	2020	Case series
Literature review[2-6,8-16,19,20,22-24,26-28,30,31,33-35,38,39,41,42,44-47,49-54,56-58,60,62]		