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Comparison of posterior fossa decompression with or without duraplasty in children with Type I Chiari malformation

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Abstract

Purpose—Chiari Malformation Type I (CM1) is a common and often debilitating neurosurgical disease. Whether to treat CM1 patients with a traditional posterior fossa decompression with duraplasty (PFDD) or a less invasive extradural decompression (PFDO) is controversial. The purpose of this study was to compare clinical outcome and syrinx resolution between the two procedures.

Methods—We retrospectively reviewed the records of 36 patients treated with PFDD and 29 patients with PFDO between 2003 and 2011. We compared baseline demographic, clinical, and radiographic characteristics. The primary clinical outcome was the Chicago Chiari Outcome Scale (CCOS). The primary radiographic outcome was qualitative syrinx improvement or resolution.

Results—At baseline, age and sex distributions, radiographic characteristics, and presenting symptoms were similar in patients undergoing PFDD and PFDO. Patients undergoing PFDO had shorter surgical time (1.5 vs. 2.8 hours; $p < 0.001$) and length of hospital stay (2.1 compared to 3.3 days; $p < 0.001$). Cerebrospinal fluid-related complications were more common in patients receiving PFDD (7/36) than PFDO (0/29) (p=0.014). Clinical improvement, defined by the mean CCOS score, was comparable in patients receiving PFDO (14.7) and PFDD (14.6) (p=0.70). Among patients with postoperative syrinx imaging, 10/13 in the PFDD group improved or resolved, compared to 8/8 in the PFDO group (p=0.26).

Conclusions—Extradural decompression for CM1 produces comparable rates of clinical and radiographic improvement as the more-invasive decompression with duraplasty. Given the

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ETHICAL STANDARDS

The Washington University in St. Louis Institutional Review Board approved this study, and it was conducted in accordance with the Declaration of Helsinki and its later amendments. The Institutional Review Board waived the need for informed consent.

CONFLICT OF INTEREST

No authors have any conflict of interest.

increased morbidity and resource utilization associated with PFDD, PFDO should be considered an attractive first-line option for most CM1 patients.

Keywords

Pediatric neurosurgery; comparative effectiveness; outcomes research; suboccipital craniectomy; cerebrospinal fluid leak; pseudomeningocele

INTRODUCTION

Chiari malformation Type I (CM1) is a congenital deformity consisting of significant herniation of the cerebellar tonsils through the foramen magnum [1]. Radiographic evidence if CM1 is common, with a magnetic resonance imaging (MRI) prevalence in children close to 4% [2]. While some affected individuals are asymptomatic, crowding in the posterior fossa often leads to compression of the cerebellum and cervicomedullary junction, causing a variety of signs and symptoms, ranging from headache to brainstem compromise [3–5].

Surgical decompression for CM1 is a common pediatric neurosurgical procedure [6–9]. Traditionally, the procedure involves posterior fossa decompression and dural augmentation (PFDD) [6, 10–12]. However, increasingly some surgeons advocate an extradural, or osseous-only posterior fossa decompression (PFDO) for certain patients. This technique involves bone removal, lysis of epidural bands and/or thinning or splitting the dura, without a more invasive duraplasty [7, 11, 13–15].

Arguments supporting a less invasive PFDO over PFDD include shorter operative times and length of hospital stay, decreased pain and morbidity, and near-complete elimination of the risk of cerebrospinal fluid (CSF)-related complications [6, 8, 16]. Despite these advantages, some studies report PFDO is associated with higher rates of revision surgery for persistent disease. However, information regarding *symptomatic* outcome has been inconsistent, evaluated primarily using gestalt assessments, and no study has compared clinical outcomes using a validated outcome tool, such as the Chicago Chiari Outcome Scale (CCOS) [6, 13, 16–19]. Beyond questions of clinical improvement, many physicians are hesitant to perform PFDO in patients with syringomyelia [20], even though some evidence suggests rates of syrinx resolution are similar with both surgeries [6]. Given these areas of persistent controversy, the goals of this study are to compare long-term clinical and radiographic outcome in CM1 patients treated with PFDD and PFDO and to use this information to help guide future treatment decisions for patients with CM1.

METHODS

Patients

We retrospectively reviewed the records of 65 consecutive patients who underwent primary surgical decompression for symptomatic CM1 by one of two surgeons (M.D.S. and D.D.L.) from July 1, 2003 to March 30, 2011 at St. Louis Children’s Hospital (SLCH). All patients were under 18 years of age and diagnosed with CM1. We divided patients into two groups: one that underwent PFDD and a second that underwent a less-invasive PFDO. While treatment decisions varied by patient, the choice to perform PFDO most often resulted from

of a comprehensive shift in the senior author's practice in 2007. Thus, any bias in selecting patients for each procedure was minimized. However, given the observational nature of the study design, we compared preoperative clinical and radiographic characteristics of the two groups, assessing for any significant baseline differences.

Imaging

All patients underwent preoperative magnetic resonance imaging (MRI), which was either available for review or described in detail in the medical record. We defined a syrinx as a contiguous hyperintense fluid collection of at least 2mm in maximal anterior-posterior (AP) diameter suggesting fluid within the spinal cord If a syrinx was present, we noted the number of spanning levels and diameter. We compared these baseline parameters to the findings on the post-operative imaging.

Operative Procedure

We reviewed operative summaries to identify the surgical approach (PFDD or PFDO) and anesthesia records to determine the length of surgical time and estimated blood loss. Both surgeons applied the same surgical approach and technique. All patients were placed in the prone position with rigid head fixation or a padded horseshoe head holder. A midline incision was made extending from the inion to the upper cervical spine. After standard subperiosteal dissection from the occipital bone and upper cervical spine, a suboccipital craniectomy, extending from the foramen magnum to the inferior nuchal line, and C1 laminectomy were performed. For those patients who underwent PFDD, the dura was opened in a Y-shaped fashion. In order to restore normal CSF flow, arachnoid adhesions were released with sharp dissection and tonsillar coagulation or tonsillar resection, if indicated. A triangular patch of Durepair Dura Regeneration Matrix (TEI Biosciences, Medtronic Neurosurgery, Waltham, MA) or a pericranial graft was sutured for dural closure. In the PFDO group, the dura was not opened, and intraoperative ultrasound was used before and after lysis of the atlanto-occipital membrane to ensure adequate decompression. After meticulous hemostasis, the wound was closed in layers using interrupted suture.

Outcome Measures

All patients typically presented for routine follow-up evaluation two weeks after surgery, then subsequently at six, 12, and 24 months. *S*ymptomatic outcome was evaluated as close to 24 months as possible while still occurring between 12 and 36 months post-surgery. This time-frame was chosen to standardize follow-up timing among patients. The primary clinical outcome measure was the composite Chicago Chiari Outcome Scale (CCOS) score [18]. The CCOS is a standardized, recently validated[21] tool for evaluating clinical outcome in CM1 patients and includes categories for pain symptoms, non-pain symptoms, functionality, and complications, with a total possible score of 16. For those patients with syringes preoperatively, the primary radiographic outcome was postoperative qualitative syrinx improvement (diameter or length) or resolution. In addition, we noted significant complications, including cerebrospinal fluid (CSF)-related complications (aseptic meningitis or reoperation for repair of pseudomenginocele/CSF leak), wound infection, and osseous only procedure converted to duraplasty. The diagnosis of aseptic meningitis was based on

the following criteria: positive CSF studies on lumbar puncture (with negative cultures), fever, headache, and treatment response to steroids. Hospital charge data were based on average daily charges at SLCH, averaged across intensive care unit and hospital ward settings.

Statistical Analysis

All statistical analyses were done using SPSS version 17.0 (IBM, Armonk, NY). Continuous and ordinal variables were compared using Student's t tests and Mann-Whitney U tests based on assumptions of normality, and categorical variables were compared using Fisher's Exact Test. P values < 0.05 were considered significant.

RESULTS

65 patients underwent suboccipital craniectomy for CM1 during the study period. 36 patients underwent PFDD, whereas 29 underwent a less-invasive PFDO. Demographic information, baseline MRI characteristics, and rates of common presenting signs and symptoms are shown in Table 1. Age and sex distributions, along with radiographic characteristics were similar in both groups. The most common presenting symptom was headache (82%), frequently post-tussive in nature (18%). Sensory changes was the only sign or symptom that differed between the PFDD (39%) and PFDO groups (10%) (p=0.01).

Characteristics of initial surgical decompressions and postoperative complications are shown in Table 2. The average surgical time in the PFDD group was 2.8 hours compared to 1.5 hours in the PFDO group ($p < 0.001$). Mean length of hospital stay (3.3 compared to 2.1 days) was also significantly longer in the PFDD group ($p < 0.001$). Overall CSF-related complications were more common in the PFDD (19.4%) than PFDO (0%) group (p=0.014). Two patients in the PFDO group required repeat surgery with duraplasty for persistent disease, compared to one patient in the PFDD group (p=0.58).

25 patients (69%) in the PFDD group and 19 patients (66%) in the PFDO group had clinical outcome data available in the 12 to 36 month follow-up window (Table 3). The PFDD and PFDO groups experienced comparable clinical improvement (Mean (SD) CCOS score = 14.6 (1.2) for PFDD; 14.7 (1.1) for PFDO; p=0.70). There were also no significant differences in the CCOS component subscores, though there was a trend toward improved complications in the PFDO group (p=0.053).

Among patients with preoperative syringes, 13/15 (87%) in the PFDD group and 8/8 (100%) in the PFDO group had follow-up imaging available (p=0.53). Follow-up MRI characteristics are shown in Table 3. With regard to the primary radiographic endpoint, there was a no difference in syrinx resolution or improvement between groups (p=0.26). Among the eight patients with preoperative syringes in the PFDO group, all experienced qualitative syrinx improvement or resolution. By comparison, among the 13 patients with preoperative syringes in the PFDD group, 77% improved or resolved, while the remainder were stable. Both groups experienced similar reductions in the number of levels the syrinx spanned (p=0.20) and the maximum AP diameter (p=0.42).

DISCUSSION

This study compares the results of 29 children who underwent extradural posterior fossa decompression to 36 children who underwent a more invasive decompression with duraplasty. Notably, there was no difference in long-term clinical outcome between the two groups, nor was there a difference in syrinx resolution or improvement. Like previous studies [8, 16], we found the PFDO group had shorter hospital stay lengths and shorter operative times than the PFDD group. Based on an average daily charge of $1,812 at SLCH, charges for PFDO patients were on average almost $2,200 less than for patients undergoing PFDD, though this difference may be larger if operative time and reoperation for complications are considered. In addition, overall CSF-related complications were significantly more common in the PFDD group and there was a trend toward improved CCOS complication score in the PFDO group.

In recent years, investigators have published several series examining the outcome from extradural decompression for CM1 compared to the more-invasive decompression with duraplasty [8, 13, 16, 17, 22–24]. Such studies have focused primarily on operative morbidity, postoperative complications rates, and repeat surgery for persistent disease. Frequently, these investigations have found PFDO is associated with decreased operative morbidity and length of hospital stay, as well as lower rates of CSF-related complications [6, 8, 13, 15, 17, 25]. While some studies, including a meta-analysis by Durham and colleagues [6, 13, 17], have found PFDO associated with higher rates of reoperation for persistent disease, evidence suggesting a difference in symptomatic outcome is mixed [8, 13, 23, 24]. Though not statistically significant in meta-analysis, some findings also show higher rates of syrinx improvement in patients undergoing PFDD [6, 17, 22], leading many physicians to avoid extradural decompression in patients with syringomyelia [20]. However, these previous studies had important limitations: first, these studies lacked treatment groups that were balanced on demographic variables, presenting symptoms, and radiographic findings; second, the methods used to report clinical outcomes were poorly defined and not based on any validated metrics.

The current study addresses these important limitations by providing a balanced treatment comparison that focusses on long-term clinical outcome and radiographic syrinx improvement. Our finding that clinical outcome, judged by the validated CCOS, did not differ between groups is an important contrast to earlier findings suggesting worse clinical outcome in in patients undergoing extradural decompression. Though it has its own limitations, the CCOS provides a more objective comparison of clinical outcome than earlier gestalt methods. Thus, its application here in two groups with similar clinical and radiographic parameters represents an important advance over earlier comparative studies. In addition, the fact all syringes in the PFDO group either improved or resolved suggests that the widespread belief that most CM1 patients with syringes require duraplasty [20] is likely mistaken.

To guide patient management moving forward, we developed a general treatment framework for CM1 patients (Figure 1). We based this framework on several factors: the results presented herein showing long-term comparable outcomes between PFDD and

PFDO; current data and previous studies showing decreased post-operative morbidity and CSF-related complication with extradural decompression [6, 13, 15, 17, 25]; and finally, present and earlier results suggesting cost savings with PFDO [16, 26]. Based on these considerations, we thus recommend that asymptomatic patients without a syrinx receive conservative follow-up as a first step. However, for patients that are either symptomatic or have an asymptomatic syrinx, we recommend extradural decompression as a first step. While the current study suggests that clinical and radiographic outcomes are comparable with both surgeries, we recognize that these data are limited and PFDD could offer some advantage in more severe patients. Thus, for those with rapidly progressive symptoms or severe neurological deficits, we still advise PFDD as the first option. Similarly, if symptoms or syringes fail to improve after PFDO, surgeons should then consider PFDD.

While this study makes important contributions to the CM1 treatment literature, it has several limitations. First, like earlier investigations, the retrospective nature of this study increases the potential for error and bias in data collection. Second, the limited number of treating surgeons is both a limitation and a benefit. Specifically, having only two treating physicians increases the potential for referral bias and potentially decreases the generalizability of these results. However, having only two surgeons perform all operations increases the consistency between the PFDD and PFDO groups and helps minimize bias in treatment decisions for postoperative complications. Finally, while the sample size in our study is comparable to or larger than most earlier series, the restricted number of patients in each treatment group is also a limitation.

CONCLUSION

Extradural decompression for CM1 leads to comparable clinical and radiographic improvement compared to traditional decompression with duraplasty, but offers decreased postoperative morbidity and lower costs. Based on these considerations, PFDO represents an attractive first-line treatment option for most CM1 patients, as outlined in our treatment algorithm. A prospective, randomized trial is needed to corroborate these findings.

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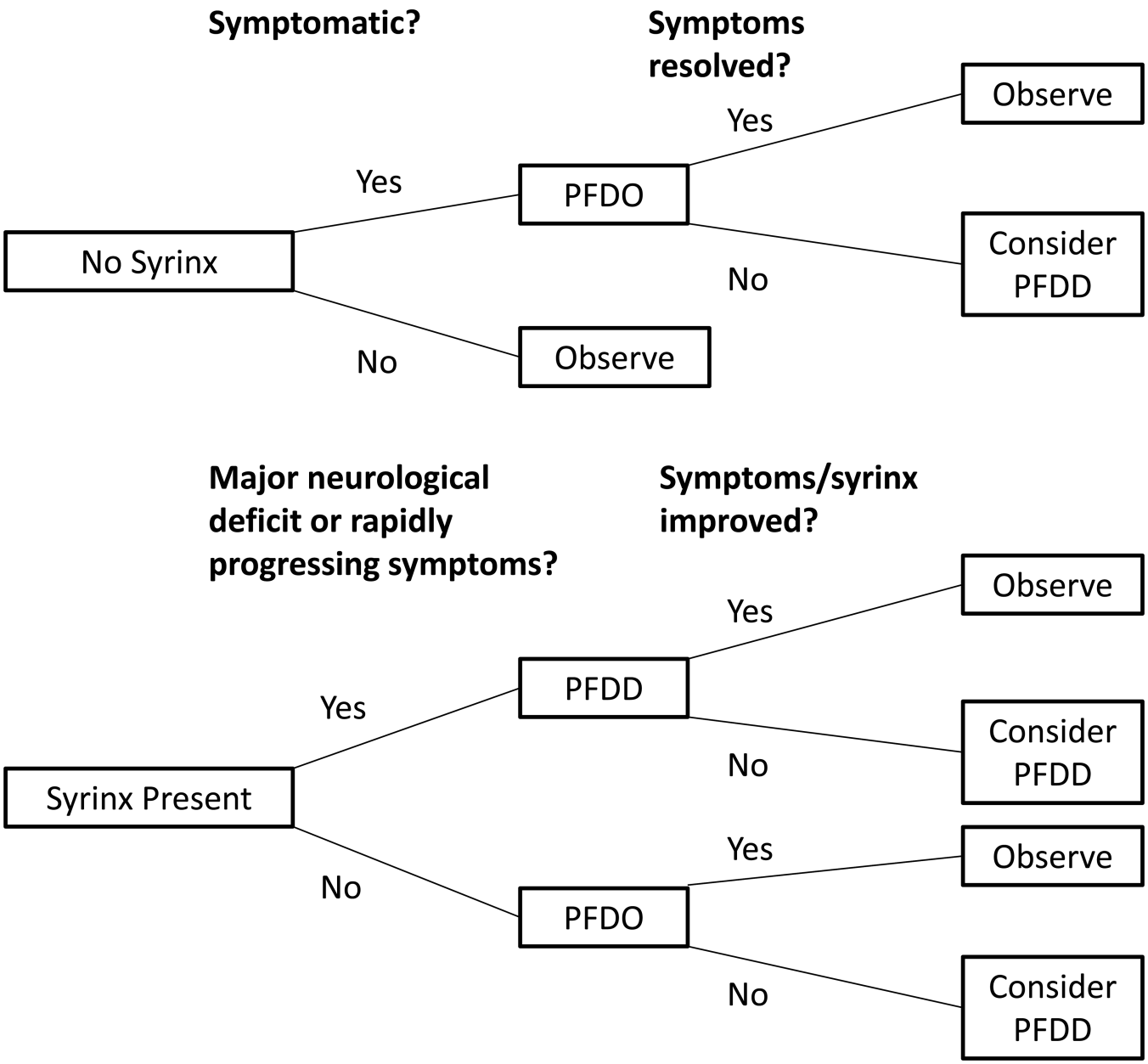

Figure 1.

A framework describing the management of patients with CM1. PFDD = posterior fossa decompression with duraplasty. PFDO = extradural, osseous-only, decompression

Table 1

Patient demographics, frequent presenting symptoms, and preoperative radiographic characteristics. PFDD = posterior fossa decompression with duraplasty. PFDO = extradural, osseous-only, decompression.

	All Subjects (n=65)	PFDD (n=36)	PFDO (n=29)	P-value*
Demographics				
Mean age in years (SD)	9.5 (5.2)	9.9 (5.3)	8.9 (5.2)	0.45
Number (%) male	31 (48%)	16 (44%)	15 (52%)	.62
Presenting Symptoms (%)				
Headache	53 (82%)	28 (78%)	25 (86%)	.52
Post-tussive headache	12 (18%)	6 (17%)	6 (21%)	.75
Neck pain	16 (25%)	7 (19%)	9 (31%)	.39
Coordination/ataxia	14 (22%)	10 (28%)	4 (14%)	.23
Sensory changes	17 (26%)	14 (39%)	3 (10%)	.01
Bulbar dysfunction	11 (17%)	6 (17%)	5 (17%)	1.00
Weakness/atrophy	10 (15%)	7 (19%)	3 (10%)	.49
Visual complaints	9 (14%)	5 (14%)	4 (14%)	1.00
Preoperative MRI				
Tonsillar descent in mm (SD)	11.8 (5.0)	11.5 (5.6)	12.3 (4.3)	.51
Presence of a syrinx (%)	23 (36%)	15 (43%)	8 (28%)	.30
Maximum syrinx diameter in mm (SD) [a]	5.5 (2.5)	5.9 (2.5)	4.9 (2.4)	.37
Mean number of syrinx levels (SD) [a]	10.5 (6.7)	12.0 (7.0)	7.6 (5.3)	.14

*P-value comparing PFDD and PFDO. Significance testing done using Student's T-Test, Mann-Whitney U Test, or Fisher's Exact test, as appropriate.

[a]Calculations for syrinx diameter and number of levels only include those patients with a syrinx present preoperatively.

Table 2

Characteristics of initial hospital admission and postoperative complications. PFDD = posterior fossa decompression with duraplasty. PFDO = extradural, osseous-only, decompression. CSF = cerebrospinal fluid.

	All Subjects (n=65)	PFDD (n=36)	PFDO (n=29)	P-value*
Initial surgery and hospital stay				
Estimated blood loss in ml (SD) [a]	64 (80)a	78 (103)	47 (37)	.39
Length of surgery in hours (SD)	2.3 (0.9)	2.8 (0.7)	1.5 (0.4)	< 0.001
Length of hospital stay in days (SD) [b]	2.7 (0.7)	3.3 (1.1)	2.1 (0.5)	< 0.001
Complications (%)				
CSF-related	7 (11)	7 (19.4)	0 (0)	0.014
Aseptic meningitis	3 (5)	3 (8)	0 (0)	0.25
Reoperation for CSF Leak/Pseudomeningocele	4 (6)	4 (11)	0 (0)	0.12
Revision duraplasty	3 (5)	1 (3)	2 (7)	0.58
Wound infection	1 (2)	0 (0)	1 (3)	0.45

*P-value comparing PFDD and PFDO. Significance testing done using Student's T-Test, Mann-Whitney U Test, or Fisher's Exact test, as appropriate.

[a] 2 subjects had missing data regarding estimated blood loss.

[b] 15% trimmed mean (SD) are reported for length of hospital stay due to the presence of two extreme outliers.

Table 3

Clinical and radiographic outcome measures. PFDD = posterior fossa decompression with duraplasty. PFDO = extradural, osseous-only, decompression. CCOS = Chicago Chiari Outcome Scale.

	All Subjects (n=44)	PFDD (n=25)	PFDO (n=19)	P-value*
Mean number of months of clinical follow-up (SD)	23.6 (5.7)	23.5 (5.8)	23.7 (5.6)	.90
Composite CCOS Score (SD)	14.7 (1.1)	14.6 (1.2)	14.7 (1.1)	.70
Pain subscore (SD)	3.52 (0.51)	3.52 (0.51)	3.53 (0.51)	1.00
Non-pain subscore (SD)	3.66 (0.53)	3.72 (0.46)	3.58 (0.61)	.47
Functionality (SD)	3.75 (0.53)	3.76 (0.52)	3.74 (0.56)	.92
Complications (SD)	3.73 (0.50)	3.60 (0.58)	3.89 (0.32)	.053
Patients with follow-up syrinx imaging	21/23 (91%)	13/15 (87%)	8/8 (100%)	.53
Number (%) syringes qualitatively improved or resolved	18 (86%)	10 (77%)	8 (100%)	.26
Mean reduction (SD) in syrinx diameter (mm)	2.7 (1.8)	2.9 (1.7)	2.3 (2.0)	.42
Mean reduction in number of syrinx levels (SD)	6.4 (6.3)	7.8 (7.1)	4.1 (4.2)	.20