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ORIGINAL PAPER

CSF complications following intradural spinal surgeries in children

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Abstract
Background Cerebrospinal fluid (CSF) leakage is a complication of intradural spinal surgery and is associated with poor wound healing and infection. The incidence of CSF leak is reported at ~16 % in adults, but little information is available in children.
Purpose The aim of this study is to determine the CSF leak rate and predisposing factors after intradural pediatric spinal surgeries.
Methods This study was a retrospective chart review of 638 intradural spinal operations at BC Children's Hospital. CSF leak was defined as pseudomeningocele or CSF leak through incision. Primary operations to untether lipomyelomeningoceles, myelomeningocele/meningocele closure, and Chiari decompressions were excluded.
Results CSF leaks occurred in 7.1 %, with 3 % having overt CSF leaks through skin (OCSF leak). CSF leaks, specifically OCSF leaks, were associated with postoperative wound infection ($P=0.0016$). Sixteen of 45 cases of CSF leak required reoperation. The type of dural suture used, site of operation, or use of fibrin glue did not affect CSF leak rates. Previous spinal surgery ($P<0.0001$), use of dural graft ($P=0.0043$), method of dural suturing ($P=0.0023$), and procedure performed ($P<0.001$) were associated with postoperative CSF leakage. Patients with CSF leak were older than those without leak (98 vs. 72 months, $P=0.002$).
Conclusions Our results provide evidence on intraoperative factors that may predispose to CSF leaks after spinal intradural surgery and may help guide surgical practice. This study confirms that the pediatric population shares many of the same risk factors for CSF leak as in adult populations. Further research is needed to explain how specific factors are associated with CSF leaks.

Keywords Pseudomeningocele · CSF leak · Spinal surgery · Children · Technique · Outcome · Complication

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Introduction

Cerebrospinal fluid (CSF) leakage is a well-recognized complication after intradural spinal surgery and may manifest as a pseudomeningocele or as an overt CSF leak (OCSF leak) through the skin incision. In spinal procedures, CSF leak is associated with significant postoperative complications including poor wound healing and/or wound dehiscence, wound infection, postural headaches, chronic subdural hematomas, radiculopathy, and meningitis [3, 4, 8, 15, 17]. Revision surgeries may be required to repair the CSF leak in cases where conservative management fails, posing an additional surgical risk to patients. The particularly high risk of CSF leak in intradural spinal procedures is thought to be related to high intradural hydrostatic pressures at the operative site [17].

The rate of CSF leak through the skin incision after intradural spinal procedures is reported in the adult literature to be about 12–16 % [8, 16, 17]. A number of factors have been associated with the rate of CSF leak [5, 10, 16, 17, 19], including the site of operation in the spine—cervical, thoracic, or lumbar, the diagnosis or pathology being treated, the method of dural closure including the type of suturing (continuous non-locked, continuous locked, or interrupted), use of fibrin glue, use of artificial dura or duraplasty, method of wound closure, and the age of the patient.

The purpose of this study was to determine the rate of CSF leak after intradural spinal procedures in a pediatric neurosurgical population to identify associated risk factors, and then determine how these factors may differ from those of adult patients. This study attempts to establish a baseline leak rate for monitoring complications in the future. Identifying the factors that predispose to CSF leaks will help modify surgical practice and postoperative management to reduce the incidence of CSF leak and, thus, its associated complications. Identification of patients who are at higher than normal risk of CSF leak preoperatively also allows for appropriate operative risk counseling for patients and their families.

Methods

This study was a single-center retrospective chart review of 638 operations in 589 patients who underwent intradural spinal surgery at BC Children's Hospital, a tertiary care institution, from August 26, 1982 to January 6, 2012. Patients were identified from a prospectively maintained database in the Division of Pediatric Neurosurgery. Patients' charts (including operative reports, admission notes, discharge summaries, and follow-up clinic visits) were reviewed to identify patient age at the time of surgery, initial diagnosis, history of past spinal surgery, site of operation (cervical, thoracic, or lumbar spine), type of procedure performed, method and type of dural suturing, the use of fibrin glue, the use of a dural graft, operating neurosurgeon, and any postoperative complications that occurred (e.g., CSF leak, pseudomeningocele, wound infection, meningitis, or neurological worsening). Any available postoperative imaging was also reviewed.

The principal outcome of interest, CSF leak, was defined as the presence of a clinically apparent pseudomeningocele according to the evaluating physician or CSF leakage through the surgical wound (OCSF leak). The rationale for this definition was that both a pseudomeningocele and a CSF leak through incision would be indicators of significant CSF leak though the dural closure. If a CSF leak occurred, the onset of the leak after closure was documented and any interventions (conservative management or operative management) were noted.

Operations for the following conditions were excluded: primary closures of myelomeningoceles, meningoceles, lipomyelomeningoceles, conus lipomas with a dural defect, and Chiari decompressions. These were excluded on the basis of preexisting defect in the dural layer by nature of the diagnosis. Thus, these dural closures were not based on opening and subsequent repair of the iatrogenic dural defect.

Categorical variables were summarized as frequencies and evaluated using either Fisher's exact test or a chi-square test. Continuous variables (age at time of surgery) were analyzed using an unpaired two-sample *t* test. To account for multiple comparisons, a Bonferroni correction ($n=10$) was applied, and P values less than 0.005 were considered statistically significant.

Results

Six hundred thirty-eight intradural spinal operations in 589 patients were reviewed. A CSF leak developed in 45 operations with an incidence of 7.1 %. Of the CSF leaks, 27 were pseudomeningoceles, 13 were OCSF leak, and 5 cases had both. Reoperation was required in 8 of 18 cases of OCSF leak compared to 8 of 27 cases of pseudomeningocele only. Nonoperative management was successful in the remainder of the patients. Conservative management of a CSF leak consisted of observation and bed rest with the head of bed down in most cases. None of the patients were managed with a lumbar drain. However, six patients with OCSF leak had additional sutures placed on the ward, and this was reported as conservative management. When operations were performed, either for OCSF leak or for pseudomeningocele only, the dural defect was repaired by resuturing with or without a graft. Fibrin glue was used by surgeon preference for dural repair. The indications for fibrin glue were unable to be analyzed retrospectively.

The various factors that were analyzed are presented in Table 1. The average age of patients in the study was 73.5 months (range from 1 day to 19 years, SD 56.2 months). The lumbar spine was the most common site of operation with selective dorsal rhizotomies and cord untetherings being the most commonly performed procedures.

Rates of wound infection but not postoperative neurological worsening were significantly higher in patients who developed a CSF leak (Table 2). All six cases of wound infection occurred in patients with OCSF leak. No wound infection occurred in patients with a pseudomeningocele in the absence of OCSF leak. None of the patients in our study developed bacterial meningitis.

The operative factors are summarized in Table 3. History of previous spinal surgery, method of dural suturing, and use of a dural graft were associated with CSF leakage. Patients who had previously undergone spinal surgery had a significantly higher rate of CSF leak compared to patients with no previous spinal surgery (16.0 vs. 4.3 %, $P<0.0001$). Dural closure with continuous suturing had a significantly higher leak rate than dural closures with a continuous locked method (7.4 vs. 0.0125 %; $P=0.0023$). Patients who had dural closure using a graft had a significantly higher leak rate compared to not using a dural graft (18.8 vs. 6.2 %; $P=0.0043$). The use of

Table 1 Summary of patient data

Variable		No. of operations (%)
Site of operation	Cervical	28 (4.4)
	Thoracic	48 (7.5)
	Lumbosacral	562 (88.1)
Procedure performed	Excision of spinal tumor	85 (13.3)
	Cord untethering—sectioning of the filum only	113 (17.7)
	Cord untethering—other than filum sectioning	153 (24.0)
	Dorsal rhizotomy	230 (36.1)
	Excision of dermal sinus tract	32 (5.0)
	Excision of arachnoid cyst	16 (2.5)
	Other	9 (1.4)
Previous spinal surgery	No	488 (76.5)
	Yes	150 (23.5)
Use of fibrin glue	No	514 (80.5)
	Yes	119 (18.7)
	Unknown	5 (0.8)
Use of dural graft	No	584 (91.5)
	Yes	48 (7.5)
	Unknown	6 (1)
Type of suture material used for dural closure	Vicryl	571 (89.5)
	Other (nylon, silk, polydiaxanone)	45 (7.1)
	Could not be determined	22 (3.4)
Method of dural suturing	Continuous	403 (63.2)
	Interrupted	5 (0.8)
	Continuous locked	160 (25.1)
	Could not be determined	70 (10.9)
Operating Surgeon	1	384 (60.1)
	2	180 (28.2)
	3	8 (1.3
	4	20 (3.1)
	5	11 (1.7)
	6	10 (1.6)
	7	1 (0.2)
	Missing information	24 (3.8)
CSF[a] Leak	No	593 (92.9)
	Yes	45 (7.1)
	Pseudomeningocele only	27 (4.2)
	CSF leak through skin only	13 (2.0)
	Pseudomeningocele and CSF leak through skin	5 (0.8)
Intervention for CSF leak	Non-operative	26
	Operative	16
	Unknown	3
Rate of resolution of leak	Less than or equal to 48 h	9
	Greater than 48 h	20
	Unknown	16

[a] CSF, cerebrospinal fluid

fibrin glue and type of suture material were not associated with the rate of CSF leak. The site of operation on the spine (cervical, thoracic, or lumbosacral) was not associated with different CSF leak rates (chi-square analysis; $P=0.033$). The procedure performed (excision of spinal tumor, cord untethering, cord untethering with sectioning of the filum, rhizotomy, excision of dermal sinus tract, excision of arachnoid cyst, or other) was associated with CSF leakage (chi-square analysis; $P<0.001$) with cord untethering operations having the highest leak rate (Table 3).

Patients who developed a leak were older at the time of surgery than those who did not (98 vs. 72 months, $P=0.002$) (Table 4). A lumbar or wound drain was not used in any of the cases at initial operation.

The operating surgeon was also included as an operative factor (Table 5) to determine if this affected the CSF leak rates associated with the type of dural suturing. Analysis by logistic regression showed no significant difference in CSF leak between surgeons, and that the operating surgeon did not affect the CSF leak rates associated with the type of dural suturing. The majority of the cases were performed by two surgeons.

Discussion

In this study, we defined CSF leak as the formation of a pseudomeningocele or CSF leak through incision. This differs from previous reports, where CSF leak has been defined as leak through the incision and may pose some difficulty when attempting to compare our results to previous studies. However, the rationale for our definition is that in both situations, there is CSF leak outside the dura. We recognize that CSF leak through the incision may have different implications for the patient in terms of management and complications compared to pseudomeningocele alone. However, factors that predispose to CSF leak through the incision should be similar to those predisposing to a pseudomeningocele.

The incidence of CSF leak (both pseudomeningocele and OCSF leak) in our patient population was determined to be 7.1 %. As expected based on previous reports [7, 9, 18], the occurrence of a CSF leak was associated with higher rates of wound infection. In our series, wound infections were associated with CSF leakage only when the leak was through the incision. None of the cases of pseudomeningocele developed a wound infection unless a leak through the incision was also present.

The incidence of postoperative neurological worsening was not significantly greater in patients who developed any type of CSF leak. Most cases of neurological worsening were urinary retention, persistent muscle weakness, or persistent numbness. Only 16 of the 45 cases of CSF leak required reoperation to resolve the complication. This may be explained

Table 2 Complications of cerebrospinal fluid leakage

Rate of complication			
Complication	Patients with CSF[a] leak or pseudomeningocele N=45	Patients with No CSF Leak or pseudomeningocele *N*=593	
Wound infection	6 (13.3 %)	14 (2.4 %)	*P*=0.0016
Meningitis	0 (0 %)	0 (0 %)	
Neurological worsening	7 (15.6 %)	73 (12.3 %)	*P*=0.3469

[a] *CSF* cerebrospinal fluid

by a higher incidence of pseudomeningoceles compared to OCSF leak in our study. An alternate explanation could be that the surgeons at our hospital tended toward conservative treatment and, thus, were less inclined to operative intervention than other institutions. In a previous study by Chern et. al, 11 of 13 (85 %) pediatric patients who developed a CSF leak through incision were reoperated for wound repair [13]. In our study, 56 % of patients who developed a CSF leak through incision resolved with conservative management alone, suggesting that it may be reasonable to initially manage patients in this manner, with surgery being an option in refractory cases. Lumbar drainage is also a reasonable conservative management

Table 3 Comparison of cerebrospinal fluid leak rates based on various operative factors

Operative factors		Rate of complication N	Number of CSF[a] leaks (%)
Use of Fibrin glue for dural closure	Yes	119	6 (5.0 %)
	No	514	39 (7.6 %)
	P value (Fisher's exact test)	0.4294	
Use of dural graft	Yes	48	9 (18.8 %)
	No	584	36 (6.2 %)
	P value (Fisher's exact test)	0.0043	
Type of suture used for dural closure	Vicryl	571	33 (5.8 %)
	Other	45	8 (17.8 %)
	P value (Fisher's exact test)	0.0066	
Method of dural suturing	Continuous non-locked	403	30 (7.4 %)
	Continuous locked	160	2 (0.0125 %)
	P value (Fisher's exact test)	0.0023	
History of past spinal surgery	Yes	150	24 (16.0 %)
	No	488	21 (4.3 %)
	P value (Fisher's exact test)	<0.0001	
Site of operation	Cervical	28	5 (17.9 %)
	Thoracic	48	1 (2.1 %)
	Lumbosacral	562	39 (6.9 %)
	P-value (Chi square analysis)	0.033*	
Procedure	Excision of spinal tumor	85	7 (8.2 %)
	Cord untethering—other than simple filum sectioning	153	20 (13.1 %)
	Cord untethering (with sectioning of the filum only)	113	11 (9.7 %)
	Dorsal rhizotomy	230	2 (0.9 %)
	Excision of dermal sinus tract	32	4 (12.5 %)
	Excision of arachnoid cyst	16	1 (6.3 %)
	Other	9	0 (0 %)
	P value (chi-square analysis)	<0.001*	

[a] *CSF* cerebrospinal fluid

*Contained categories with expected frequencies <5

Table 4 Age at time of surgery in patients who developed a cerebrospinal fluid leak compared to patients who did not develop a cerebrospinal fluid leak

	CSF[a] leak or pseudomeningocele	No CSF leak or pseudomeningocele
Mean age (months)	97.98	71.64
Standard deviation (months)	63.51	55.22
Mean difference (months)	26.3423	
95 % confidence interval (months)	26.34±17.18	
P value	0.002384	

[a] *CSF* cerebrospinal fluid

option prior to operative intervention. For pseudomeningoceles, nonoperative management should be the initial management option.

Our observations regarding fibrin glue were consistent with a previous pediatric study by Chern et al. that had reported no reduction in the rate in CSF leaks in operations for tethered cord release by sectioning of the filum [6]. Our rate of CSF leak through incision (2.8 %) was lower than that of Chern et al. (5.9 %), but our rate of pseudomeningocele was nearly identical despite incorporating a wider variety of spinal procedures in our study [6].

The site of operation on the spine was not a statistically significant factor associated with CSF leak in our study, although this may be due to the decision to use a much lower significance level (Bonferroni correction) than other reports, as cervical operations tended to have a higher leak rate than thoracic or lumbosacral spine operations (17.9 vs. 2.1 vs. 6.9 %, respectively; $P=0.033$). Spinal cord untethering was associated with the highest rate of CSF leak (13.1 % $P<0.001$).

Our results verify that many previously reported factors associated with CSF leak in adult neurosurgery also pertain to the pediatric population. Fibrin glue use in adult spinal surgery is controversial, with one report demonstrating a reduction in CSF leak rates [13], while other more recent studies demonstrated no effect [6, 8]. Because fibrin glue is expensive and has not been shown to reduce CSF leakage in a pediatric population, it cannot be recommended routinely for dural repair. Given that this study is retrospective and fibrin glue was used as per surgeon's choice, it is possible that those patients who received fibrin glue were thought to be at higher than average risk of CSF leak intraoperatively. With that knowledge, it is possible that the use of fibrin glue, while not preventing CSF leak, did decrease the risk of CSF leak to average. This effect, however, was not quantifiable through our retrospective review.

Table 5 Cerebrospinal fluid leak rates by operating surgeon

		N	Number of CSF[a] leaks (%)	*P* value (logistic regression analysis)
Operating surgeon	1	384	15 (3.9 %)	0.564
	2	180	19 (10.6 %)	0.816
	3	8	1 (12.5 %)	0.401
	4	20	2 (10.0 %)	0.967
	5	11	3 (27.3 %)	0.152
	6	10	3 (30.0 %)	0.196
	7	1	0 (0.0 %)	1.000

A reduction in CSF leak rates when using continuous locked method of suturing for dural closure compared to continuous non-locked suturing ($P=0.0023$) was seen. One previous in vitro study demonstrated interrupted suturing to be the most water-tight, but found no difference between continuous non-locked and continuous locked [12]. Type of suture material (e.g., Vicryl or other material) was not associated with CSF leak, but this finding is confounded by the fact that the vast majority of our dural closures (90 %) were done with Vicryl sutures. Due to the overall low rate of CSF leak, our study may not have sufficient power to detect small differences between these groups.

Consistent with multiple prior reports [8, 11], we found that the CSF leak rate was higher with patients who had previous spinal surgery, although our study also included cervical and thoracic procedures while the previous studies mentioned only assessed operations in the lumbar region. In contrast, Sin et al. found no association between history of prior surgery and an increased rate of CSF leak [16]. This difference could be related to their smaller sample size of 77 patients, which may not have been a large enough sample to detect a difference in CSF leak rate in patients with or without previous spinal surgery. It should be noted that their dural repairs were only for incidental durotomies and performed primarily which is different than the intentional dural openings used in our patients. In reviewing the literature, one of the studies with the highest reported rate of CSF leak (17 %) was focused on 22 patients who underwent repeat untethering operations after myelomeningocele or lipomyelomeningocele repair and at least two subsequent untetherings. At least in some situations, repeat spinal surgery appears to be a risk factor for CSF leakage [11].

We reported a leak rate of 18.8 % when using a dural graft, which is higher than previously reported rates in an adult population where a graft was used [1, 14]. CSF leak was significantly higher in operations where a graft was used (18.8 vs. 6.2 %; $P=0.0043$). This is likely because our definition of CSF leak includes pseudomeningoceles, as eight of the nine cases of CSF leak with the use of a dural graft were pseudomeningoceles with only one case being OCSF leak. Some authors believe that synthetic grafts predispose to CSF leaks with one explanation being that grafts are poorly integrated by host tissue [2]. Duraplasty closures were more likely to leak than closures with linear dural sutures alone [12]. To our knowledge, there are no other reports evaluating the incidence of CSF leak when artificial dural grafts have been used in pediatric intradural spinal surgery. This finding, regarding dural grafts, should be interpreted cautiously, given that the indication for using a dural graft (for example, a sizable dural defect that was not amenable to primary repair) might have influenced the leak rate directly.

We found that the average age of patients at the time of surgery was significantly greater in the group who developed a CSF leak compared to the group that did not have a CSF leak (98 vs 72 months, $P=0.002$). This finding is consistent with higher rates of CSF leak in previous studies in adult neurosurgery [16], although we did find this surprising, as we expected younger patients to have a higher CSF leak rate due to their thinner dura. One theoretical explanation is that older patients are typically larger and taller, leading to higher hydrostatic pressures in the spine at the repair site when standing in comparison to younger and shorter patients. This would be consistent with the concept that increased hydrostatic pressure at the incision site may increase the risk of CSF leak in intradural spine surgery.

One major limitation of our study is that certain factors such as the procedure performed do not have an even distribution, such that certain categories have low frequencies (e.g., for site of operation, there were 28 cervical procedures compared to 562 in the lumbosacral region). We were unable to account for interactions between analyzed variables. In addition, expected frequencies in some categories (site of operation and procedure performed) were less than five, which would affect the chi-square analysis. For this reason, we should cautiously interpret the observation that the procedure performed affects leak rate, even though it was statistically significant.

Significant factors associated with CSF leak such as "suturing method" could very well have been confounded by other factors which were not analyzed. There were 45 cases of CSF leak (a relatively low complication rate); this low number of cases makes comparisons among different factors difficult. This factor was further examined by looking at the effect of different surgeons and whether this played a role in the statistical significance. This was examined using logistic regression analysis of the individual surgeons, "suturing method," and the rate of CSF leak. The type of dural suturing remained a significant factor even when accounting for the operating surgeon.

Conclusion

The CSF leak rate after intradural surgery in this pediatric population was 7.1 %. Of the CSF leaks noted, 60 % were pseudomeningoceles, the majority of which resolved without intervention. This suggests that when a pseudomeningocele is noted after such spinal surgery, surgical intervention should be reserved for those in whom the pseudomeningocele does not resolve spontaneously after many weeks; the patient has unacceptable low-pressure symptoms or when the incision line is threatened. The results of our study have identified previous spinal surgery, method of dural suturing, use of dural graft, procedure performed, and age at time of surgery as significant factors associated with postoperative CSF leakage in a pediatric population. Site of operation, use of fibrin glue, and suture material were not associated with CSF leak. Based on these results, we do not recommend the routine use of fibrin glue to prevent CSF leakage for patients undergoing intradural spinal surgery.

Our findings provide useful information to guide surgical practice when performing intradural spinal surgery. This study confirms that the pediatric population shares many of the same important risk factors for CSF leak as in adult populations and advances in either population will improve the management of patients undergoing intradural spinal surgery.

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