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Exploring predictors of surgery and comparing operative treatment approaches for pediatric intracranial arachnoid cysts: a case series of 83 patients

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OBJECT Although intracranial arachnoid cysts are a common incidental finding on pediatric brain imaging, only a subset of patients require surgery for them. For the minority who undergo surgery, the comparative effectiveness of various surgical approaches is debated. The authors explored predictors of surgery and compared operative techniques for pediatric patients with an intracranial arachnoid cyst seen at a tertiary care center.

METHODS The authors reviewed records of pediatric patients with an intracranial arachnoid cyst. For each patient, data on baseline characteristics, the method of intervention, and surgical outcomes for the initial surgery were extracted, and cyst size at diagnosis was calculated (anteroposterior × craniocaudal × mediolateral). Baseline variables were analyzed as predictors of surgery by using logistic regression modeling, excluding patients whose surgery was not related to cyst size (i.e., those with obstructive hydrocephalus secondary to the cyst compressing a narrow CSF flow pathway or cyst rupture/hemorrhage). Data collected regarding surgical outcomes were analyzed descriptively.

RESULTS Among 83 pediatric patients with an intracranial arachnoid cyst seen over a 25-year period (1989–2013), 27 (33%) underwent surgery; all had at least 1 cyst-attributed symptom/finding. In the multivariate model, age at presentation and cyst size at diagnosis were independent predictors of surgery. Cyst size had greater predictive value; specifically, the area under the curve for the receiver-operating-characteristic curve was 0.89 (95% CI 0.82–0.97), with an ideal cutoff point of ≥ 68 cm^3. This cutoff point had 100% sensitivity (95% CI 79%–100%), 75% specificity (95% CI 61%–85%), a 53% positive predictive value (95% CI 36%–70%), and a 100% negative predictive value (95% CI 91%–100%); the positive likelihood ratio was 4.0 (95% CI 2.5–6.3), and the negative likelihood ratio was 0 (95% CI 0–0.3). Although the multivariate model excluded 7 patients who underwent surgery (based on prespecified criteria), excluding these 7 cases did not change the overall findings, as shown in a sensitivity analysis that included all the cases. Descriptive results regarding surgical outcomes did not indicate any salient differences among the surgical techniques (endoscopic fenestration, cystoperitoneal shunting, or craniotomy-based procedures) in terms of symptom resolution within 6 months, need for reoperation to date, cyst-size change from before the operation, morbidity, or mortality.

CONCLUSIONS The results of these exploratory analyses suggest that pediatric patients with an intracranial arachnoid cyst are more likely to undergo surgery if the cyst is large, compresses a narrow CSF flow pathway to cause hydrocephalus, or has ruptured/hemorrhaged. There were no salient differences among the 3 surgical techniques for several clinically important outcomes. A prospective multicenter study is required to enable more robust analyses, which could ultimately provide a decision-making framework for surgical indications and clarify any differences in the comparative effectiveness of surgical approaches to treating pediatric intracranial arachnoid cysts.

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KEY WORDS arachnoid cysts; craniotomy; cystoperitoneal shunt; endoscopic fenestration; predictors; surgery; congenital

ABBREVIATIONS AUC = area under the curve; CP = cystoperitoneal; IQR = interquartile range; ROC = receiver operating characteristic

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Intracranial arachnoid cysts are collections of CSF within the subarachnoid space of the meninges, which may become contained when the arachnoid membrane forms septations.[15] These lesions are common among the pediatric population and are most often congenital, developing during the cortical migration phase of fetal development, although acquired cysts have been described.[8,13]

The reported prevalence of pediatric intracranial arachnoid cysts has increased with the growing use of intracranial imaging, particularly CT and MRI. A recent estimate for prevalence among 11,738 patients aged ≤ 18 years who underwent MRI at a single institution over an 11-year period was 2.6% for intracranial arachnoid cysts.[1] The majority of these lesions were incidental and asymptomatic, observations that are consistent with those in previous research.[11,12,18]

When children and adolescents with an intracranial arachnoid cyst become symptomatic, it is often due to cyst growth, the underlying mechanisms of which are unknown. Several mechanisms have been proposed, including active fluid secretion by cells in the cyst wall, expansion after formation of an osmotic gradient, and unidirectional valve mechanisms.[4,7,16,19] Cyst growth can lead to clinically important symptoms through increased intracranial pressure and/or compression of surrounding brain structures, including a CSF flow pathway. Symptoms can also arise suddenly after cyst rupture or hemorrhage into a cyst, and for both cases (cyst growth and cyst rupture/hemorrhage), symptom type and severity tend to depend on cyst location.[19]

Most children and adolescents with an intracranial arachnoid cyst will remain asymptomatic and will not require surgery.[1] Identifying predictors for surgery, therefore, can inform decision-making about clinical management. There is a paucity of literature addressing the predictors of surgery among these patients, with only 1 recent study suggesting that larger cyst size at diagnosis and a younger age at presentation are statistically significant predictors.[1] In addition, for the minority of children and adolescents with an intracranial arachnoid cyst who undergo surgery, the comparative effectiveness of various surgical approaches for treating pediatric intracranial arachnoid cysts (e.g., endoscopic fenestration, cystoperitoneal [CP] shunting, and craniotomy-based procedures) is debated.[3,10,14,17,20] Given these areas of uncertainty, we undertook a case-series study among patients referred to our pediatric neurosurgery practice, which is based at a tertiary care center. The primary objectives were to explore predictors of surgery and compare outcomes among the surgical techniques for pediatric intracranial arachnoid cysts.

Methods

Data Collection

We retrospectively reviewed all records (electronic and paper) of patients aged ≤ 18 years with a radiologically confirmed intracranial arachnoid cyst, as identified in August 2011 using the electronic medical record system at McMaster Children's Hospital in Hamilton, Ontario, Canada. To achieve high search sensitivity, we broadly queried the system with the terms "brain" and "cyst" and subsequently reviewed all records found to identify patients with an intracranial arachnoid cyst. In addition, from August 2011 through September 2013, eligible patients were prospectively identified, because ours is the only pediatric neurosurgical practice in the region.

We reviewed all neurosurgical records for all the identified patients. Demographic data, including age at presentation and sex, were extracted for each patient. The following clinical and radiological data were also extracted: cyst-attributed symptoms and findings (including hydrocephalus and cyst growth), cyst location, and, when available, cyst size at diagnosis and at the most recent follow-up visit (based on CT or MRI). Cyst growth was defined, as reported by the radiologist, as an increase in cyst size between the initial CT/MR images and subsequent CT/MR images (acquired by using the same imaging modality per patient). Cyst size was recorded as the reported maximal anteroposterior, craniocaudal, and mediolateral dimensions (in centimeters) and was calculated in terms of volume (in cubic centimeters) by multiplying the reported dimensions (i.e., anteroposterior × craniocaudal × mediolateral). For patients with a reoperation, cyst size at follow-up corresponded to the most recent size noted before the second surgery.

Throughout the study period, the decision to operate and the specific surgical technique used were based on surgeon preference. For patients who underwent surgery, data were extracted for only the first arachnoid cyst–related surgery. These data included the surgical technique used and the following surgical outcomes: symptom resolution within 6 months, need for reoperation to date, length of stay in hospital, cyst-size change from before the operation, morbidity (CSF leak, infection, and/or hemorrhage), and mortality.

The study protocol was approved by the Hamilton Health Sciences/Faculty of Health Sciences research ethics board at McMaster University in Hamilton, Ontario, Canada.

Statistical Analysis

Baseline characteristics (i.e., sex, age at presentation, cyst size at diagnosis, and cyst location) and number of cyst-attributed symptoms/findings were summarized for patients managed conservatively and for those managed surgically. Categorical variables were reported as frequencies or relative frequencies and compared by using the Pearson chi-square test or Fisher's exact test, as applicable; the Monte Carlo test was used when the expected counts were < 5. Continuous variables were reported as means with the SD or, if not normally distributed, as medians with the interquartile range (IQR) and compared by using the Student t-test or Mann-Whitney U-test, respectively.

To explore predictors of surgery, baseline variables with a p value of < 0.1 in univariate analyses (i.e., age at presentation and cyst size at diagnosis) were entered into a multivariate stepwise logistic regression model. We used 10 patients (i.e., those who underwent surgery) and 10 controls (i.e., those who did not undergo surgery) per parameter, as a rule of thumb regarding statistical power and fit for multivariate logistic regression modeling.[9] The

specific or overall numbers of cyst-attributed symptoms/findings were not considered baseline variables, because neither all symptoms nor findings (e.g., cyst growth) had developed at diagnosis. The multivariate analysis excluded the small minority of patients whose surgery was not related to cyst size, namely, those who experienced sudden symptoms as a result of cyst rupture/hemorrhage or had obstructive hydrocephalus secondary to the cyst growing to compress a narrow CSF flow pathway (e.g., cerebral aqueduct, foramina of Monro). In such cases, larger cyst sizes (in absolute terms) are not related to the need for surgery and thus would have diminished the predictive value. However, to investigate whether these exclusions changed the overall findings, we conducted a sensitivity analysis that included all cases in the multivariate model. For all regression analyses, odds ratios with 95% confidence intervals are reported; model fit was assessed by using the Hosmer-Lemeshow goodness-of-fit test.

We plotted a receiver-operating-characteristic (ROC) curve based on the optimal predictor, evaluated by the area-under-the-curve (AUC) statistic. The optimal cutoff (in terms of sensitivity and specificity) was calculated from the ROC curve.

Data collected for 2 other areas of importance regarding the clinical and surgical management of pediatric intracranial arachnoid cysts were analyzed descriptively because of the small sample size. These areas were 1) reasons why patients who were initially followed conservatively eventually underwent surgery and 2) surgical outcomes for all patients and per surgical technique (endoscopic fenestration, CP shunting, and craniotomy-based procedures).

All statistical tests were 2-sided, and a p value of < 0.05 was considered statistically significant. Stata 13.1 (StataCorp.) was used for data analysis.

Results

Among 337 records retrospectively reviewed (earliest from February 1989) and eligible cases included prospectively through September 2013, 83 pediatric patients with an intracranial arachnoid cyst were identified. MRI was available at our institution throughout this 25-year period. Twenty-seven (33%) patients underwent surgery, all of whom had at least 1 cyst-attributed symptom or finding (Fig. 1); the most common cyst-attributed symptoms/findings among these patients were headache/irritability (44%), macrocephaly (37%), and cyst growth (37%). The median age at presentation for patients with cyst growth was < 1 month (IQR 20 months); the oldest patient was 46 months old. All but 1 patient managed conservatively had no cyst-attributed symptom/finding; the exceptional patient had macrocephaly.

Sixty-two (75%) patients were male. In terms of cyst location, 46 (55%) patients had a cyst in the middle fossa, and 18 (22%) had a cyst in the posterior fossa. In univariate analyses among the children managed conservatively versus those managed surgically, the age at presentation (median ± IQR 87 ± 101 vs 18 ± 67 months, respectively; $p = 0.002$), cyst size at diagnosis (median ± IQR 26 ± 63 vs 92 ± 439 cm^3, respectively; $p < 0.001$), and proportion of patients with ≥ 1 cyst-attributed symptom/finding (2% vs 100%, respectively; $p < 0.001$) were statistically significantly different; sex (77% vs 70% male, respectively; $p = 0.59$) and cyst location were not (Table 1).

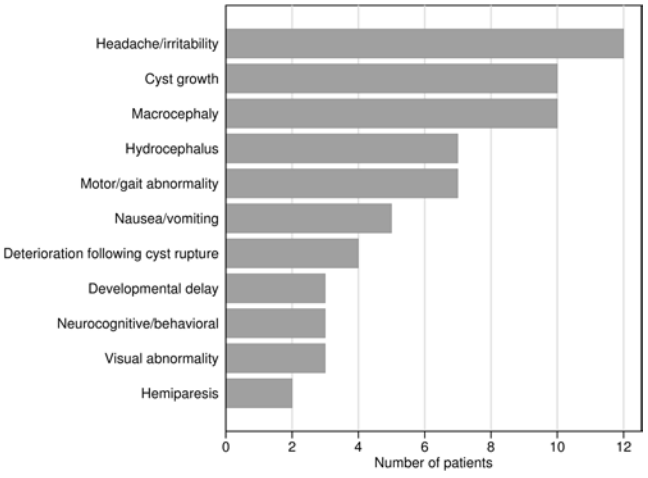

FIG. 1. Individual breakdown of cyst-attributed symptoms and findings for 27 patients who underwent surgery (among 83 pediatric patients with an intracranial arachnoid cyst). None of the patients had seizures or endocrinopathy.

In a multivariate analysis, age at presentation and cyst size at diagnosis were independent and statistically significant predictors of surgery without interaction ($p = 0.71$). Specifically, the OR was 0.984 per month (95% CI 0.971–0.998 per month; $p = 0.02$) for age at presentation and 1.008 per cm^3 (95% CI 1.003–1.012 per cm^3; $p = 0.001$) for cyst size at diagnosis. Results of the Hosmer-Lemeshow goodness-of-fit test indicated adequate model fit ($p = 0.58$). Seven patients who underwent surgery were excluded from the multivariate model based on prespecified criteria: 5 patients had obstructive hydrocephalus related to an intracranial arachnoid cyst compressing a narrow CSF flow pathway (cerebral aqueduct [3 patients], foramina of Monro [2 patients]), and 2 patients experienced sudden clinical deterioration after cyst rupture (1 patient had a recent history of head trauma). Excluding these 7 cases did not change our overall findings; in a sensitivity analysis that included all cases, age at presentation and cyst size at diagnosis remained independent and statistically significant predictors of surgery without interaction (Table 2). In addition, surgery for 3 other patients (2 with hydrocephalus and 1 with sudden deterioration after cyst rupture) was indicated by large cyst size; thus, these cases were included in the multivariate analysis per prespecified criteria.

Cyst size at diagnosis was the most optimal predictor of surgery (AUC 0.89; 95% CI 0.82–0.97) (Fig. 2). The ideal cutoff point was ≥ 68 cm^3, which had 100% sensitivity (95% CI 79%–100%), 75% specificity (95% CI 61%–85%), a 53% positive predictive value (95% CI 36%–70%), and a 100% negative predictive value (95% CI 91%–100%), corresponding to a positive likelihood ratio of 4.0 (95% CI 2.5–6.3) and a negative likelihood ratio of 0 (95% CI 0–0.3). Recent MR images of 4 patients are shown in Fig. 3 to provide a visual depiction of large (Fig. 3A and B) and small (Fig. 3C and D) cysts relative to this 68-cm^3 cutoff point.

Five patients who were initially managed conserva-

TABLE 1. Univariate analyses between patients managed conservatively and those managed surgically

Patient or Cyst Characteristic	Conservative Treatment (n = 56)	Surgery (n = 27)	p Value*
Male (no. [%])	43 (77)	19 (70)	0.59
Age at presentation, mos (median [IQR])	87 (101)	18 (67)	0.002
Cyst size at diagnosis, cm^3 (median [IQR])†	26 (63)	92 (439)	<0.0001
No. (%) w/ ≥1 cyst-attributed symptom	1 (2)	27 (100)	<0.0001
Cyst location (no. [%])‡			
Middle fossa	34 (74)	12 (26)	0.24
Posterior fossa	13 (72)	5 (28)	0.78
Intraventricular	3 (60)	2 (40)	0.66
Interhemispheric	3 (60)	2 (40)	0.66
Quadrigeminal plate	3 (60)	2 (40)	0.66
Sellar/suprasellar	0 (0)	3 (100)	0.06
Cerebral convexity	0 (0)	1 (100)	0.33

* Based on Pearson's chi-square statistic, Mann-Whitney U-test, and Fisher's exact test, as applicable. The Monte Carlo test was used when expected counts were < 5.

† Cyst size was calculated by multiplying reported dimensions (anteroposterior × craniocaudal × mediolateral). For 4 cases in the surgical group, however, neither these data nor corresponding MR/CT images were accessible.

‡ The denominator for each of these proportions is all patients (conservatively or surgically managed) with a cyst in that location.

tively and eventually underwent surgery are characterized descriptively in Table 3. For 3 cases, the primary indication for surgery was cyst growth, as noted in serial MRI.

Surgical follow-up and outcomes of all 27 patients and according to 3 common surgical techniques for intracranial arachnoid cysts are summarized descriptively in Table 4. The median follow-up time among the surgical techniques was > 1 year. All but 1 patient were followed up for > 6 months; the exceptional case was followed up for only 1 month (for this study) because his surgery occurred 1 month before the end of the study period. Eighteen (67%) patients experienced symptom resolution within 6 months; most of them were children who had a CP shunt (8 of 10 [80%] patients). However, 11 (41%) patients required at least 1 reoperation of their initial surgery, of whom 6 (22%) have required > 1 reoperation to date. Reoperations were also most common in children with CP shunts (5 of 10 [50%] patients), 3 (60%) of which were revised as a result of shunt malfunctioning.

The most common surgical morbidity was CSF leak, which occurred in 5 (19%) patients, 3 of whom had a craniotomy-based procedure. Three (11%) patients had a postoperative infection; 1 patient had a CP shunt, another had an endoscopic fenestration that was accompanied by a CSF leak, and 1 underwent bur hole drainage. Finally, 1 patient experienced a mild asymptomatic hemorrhage (chronic subdural hematoma) after endoscopic fenestration, ostensibly because a decrease in intracranial pressure after a rapid cyst-size decrease (from 192 cm^3 to 57 cm^3 within 1 month) caused bridging subdural veins to rupture. No deaths occurred.

In terms of cyst-size change from before the operation, 18 (67%) patients' cysts decreased in size (by more than 10 cm^3) and 6 (22%) patients' cysts increased in size (by more than 10 cm^3), whereas 3 (11%) patients' cysts were unchanged (within 10 cm^3). Four of the 6 patients whose cyst had grown postoperatively were aged ≤ 2 years at presentation; 2 of 6 (both of whom were aged ≤ 2 years at presentation) underwent reoperation during the study period, 1 related to CP shunt malfunction and the other related to closure of fenestration. Cyst-size change was not associated with postoperative symptom resolution within 6 months (p = 0.67; data not shown). The median length of hospital stay was 5 days (IQR 3 days); the patients who underwent endoscopic fenestration stayed for the shortest duration on average (median ± IQR 3 ± 1 days).

TABLE 2. Sensitivity analysis of multivariate logistic regression models for predictors of surgery*

Model & Predictors	OR (95% CI)	p Value	AUC (95% CI)†
All cases (n = 79)			
Age at presentation (mos)	0.982 (0.971–0.994)	0.003	0.73 (0.61–0.86)
Cyst size at diagnosis (cm^3)	1.006 (1.002–1.010)	0.004	0.80 (0.69–0.91)
All ops related to cyst size (n = 72)			
Age at presentation (mos)	0.984 (0.971–0.998)	0.02	0.72 (0.58–0.86)
Cyst size at diagnosis (cm^3)	1.008 (1.003–1.012)	0.001	0.89 (0.82–0.97)

* Model fit, as assessed by using the Hosmer-Lemeshow goodness-of-fit test, was adequate for both models—for all cases (p = 0.38) and for surgeries related to cyst size (p = 0.58). Interactions between the predictors were nonsignificant (p = 0.82 for all cases, p = 0.71 for surgeries related to cyst size) and therefore were not included in either model.

† The AUC reported pertains to only the predictor of interest rather than to a model with multiple predictors.

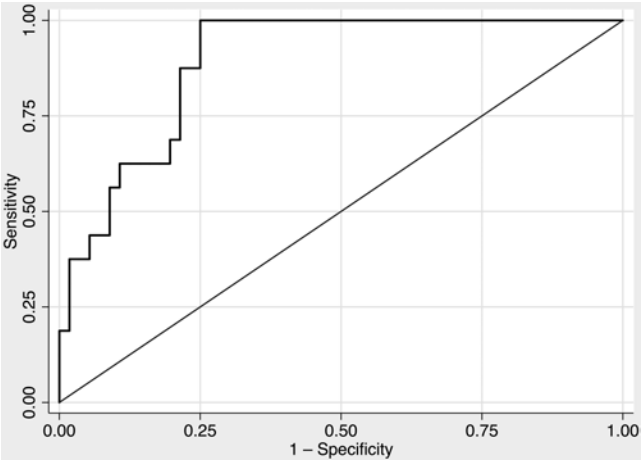

FIG. 2. ROC curve of cyst size at diagnosis as a predictor of surgery (n = 72; AUC 0.89; 95% CI 0.82–0.97).

Discussion

In this study, conducted at the sole regional pediatric neurosurgery center at a tertiary children's hospital, we identified 83 patients with intracranial arachnoid cysts seen over a 25-year period (February 1989 to September 2013), 27 (33%) of whom underwent surgery. Each patient who underwent surgery had at least 1 cyst-attributed symptom/finding, whereas none of the patients managed conservatively (except for 1 patient with macrocephaly) had cyst-attributed symptoms/findings. The number of cyst-attributed symptoms/findings, age at presentation, and cyst size at diagnosis were statistically significant in univariate analyses among patients managed conservatively versus those who were managed surgically, findings which are concordant with those of a previous study.[1] However, in contrast to previous research that found positive correlations of anterior fossa and quadrigeminal plate cysts and an inverse correlation of posterior fossa cysts to be predictors of surgery,[1] cyst location was not statistically significant in our analyses, which may be a result of our relatively smaller sample size.

In a multivariate exploratory analysis, we also found that both age at presentation and cyst size at diagnosis were independent predictors of surgery. Of these predictors, cyst size at diagnosis had greater predictive value in terms of the AUC.

Seven patients who underwent surgery were excluded from the multivariate model based on prespecified criteria: 5 patients had obstructive hydrocephalus related to an intracranial arachnoid cyst compressing a narrow CSF flow pathway (cerebral aqueduct [3 patients], foramina of Monro [2 patients]), and 2 patients experienced sudden clinical deterioration after cyst rupture (1 patient had a recent history of head trauma). In addition, surgery for 3 other patients (2 with hydrocephalus and 1 with sudden deterioration after cyst rupture) was indicated by large cyst size, and thus their cases were included in the multivariate analysis per the prespecified criteria. We believe that excluding the patients whose surgery was not related to cyst size is a clinically appropriate mode of analysis, because although surgery for patients with an intracranial arachnoid cyst is often prompted by clinical symptoms (presumably because of a relatively large cyst that causes mass effect or raises intracranial pressure), in the minority of cases sudden symptoms can arise with cyst rupture/hemorrhage or obstructive hydrocephalus caused by a cyst growing to compress a narrow CSF flow pathway (e.g., cerebral aqueduct, foramina of Monro). In such cases, a larger cyst size (in absolute terms) is not related to the need for surgery and thus would have diminished the predictive value. Nevertheless, we conducted a sensitivity analysis that included all cases, which showed that excluding these 7 cases did not change our overall findings: both age at presentation and cyst size at diagnosis remained independent and statistically significant predictors of surgery, and cyst size at diagnosis had greater predictive value in terms of the AUC.

Overall, our findings suggest that among children and adolescents with an intracranial arachnoid cyst, 3 groups of patients are more likely to undergo surgery, namely, those who have a cyst that 1) is significantly large (e.g., ≥ 68 cm^3 based on our analyses), 2) compresses a narrow CSF flow pathway (e.g., cerebral aqueduct, foramina of Monro), or 3) has ruptured/hemorrhaged, as noted in a previous study in which approximately 70% of pediatric patients with a ruptured/hemorrhaged arachnoid cyst underwent surgery.[6] Risk factors for pediatric arachnoid cyst rupture/hemorrhage were investigated recently in a matched case-control study of 14 cases, in which larger cyst size (OR 16.5; 95% CI 2.5–∞) and recent history of head trauma including minor falls (OR 25.1; 95% CI 4.0–∞) were identified as risk factors; altitude of residence was not associated with arachnoid cyst rupture or hemorrhage.[6] Altogether, these findings suggest the potential value of using cyst size at diagnosis as a predictor of surgery, which would be convenient for clinical decision-making, because intracranial arachnoid cysts are almost always diagnosed by using CT or MRI.

Furthermore, we identified 5 patients who were initially conservatively managed but eventually underwent surgery. For 3 of these patients, surgery was prompted by significant cyst growth, as noted on serial MRI. It is interesting to note that these 3 patients (all ≤ 2 weeks old) were younger by far than the other 2 patients (28 months and 10 years old), which aligns with the understanding that cyst growth is more likely to occur in younger children, particularly those aged < 4 years.[1] This finding suggests value in the use of serial MRI for young children with an intracranial arachnoid cyst. However, given the limited data, this topic requires additional research, including study of whether there is an age range for which serial MRI is especially valuable for incidentally found pediatric intracranial arachnoid cysts.

Finally, our descriptive results regarding surgical outcomes do not suggest any salient differences among the surgical techniques (endoscopic fenestration, CP shunting, or craniotomy-based procedures). Our rates of surgical revision after endoscopic fenestration (3 of 7 [43%]) and CP shunting (5 of 10 [50%]) are similar to those of other single-center case series of patients with an arachnoid cyst.[2,5] Furthermore, although 6 (22%) patients had cyst growth (> 10 cm^3) compared with before the operation, 4

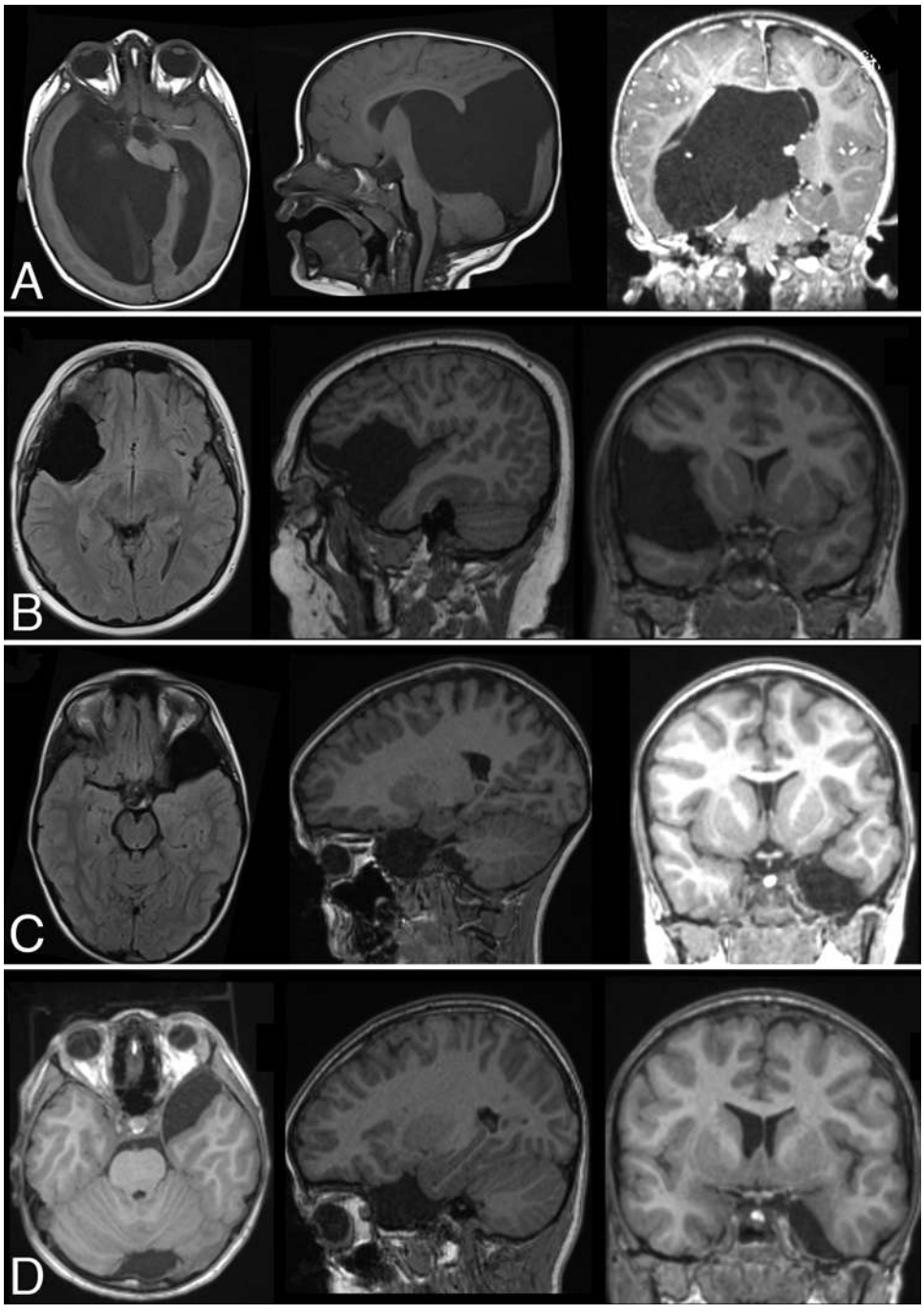

FIG. 3. Axial, sagittal, and coronal T1-weighted MR images (without gadolinium enhancement, except for the coronal image in A) of 4 patients with an intracranial arachnoid cyst. **A and B:** Two patients with a calculated cyst size (anteroposterior × craniocaudal × mediolateral) of > 68 cm^3, a 17-month-old boy (A) (960 cm^3) and a 13-year-old boy (B) (192 cm^3). Both patients underwent surgery, given their cyst-attributed symptoms/findings. **C and D:** Two patients with a calculated cyst size of < 68 cm^3, a 7-year-old girl (C) (64 cm^3) and a 9-year-old boy (D) (46 cm^3). Both patients were asymptomatic and have not undergone surgery to date.

of 6 were aged ≤ 2 years at presentation, an age range in which most head growth occurs (thereby suggesting that the cyst may have enlarged proportionally with the child's growth). In addition, 2 of 6 patients (both of whom were aged ≤ 2 years at presentation) underwent reoperation, 1 related to CP shunt malfunction and the other related to closure of fenestration, which may also partially explain the postoperative cyst enlargement. In terms of surgical techniques, since 1989 (the beginning of our study period), institutions have increasingly moved toward neuroendoscopic approaches for arachnoid cysts. This case series was situated within that period of change, with a solo pediatric neurosurgeon at our institution until 2007 who preferred CP shunting over endoscopic fenestration; hence, a substantial proportion (37% [10 of 27]) of our patients underwent CP shunting. Nevertheless, equipoise

TABLE 3. Reasons why patients who were initially followed conservatively eventually underwent surgery

Patient No.	Sex	Age at Presentation	Cyst Location	Age at Surgery	Primary Indication(s) for Surgery
1	M	6 days	Interhemispheric	8 mos	Cyst growth noted on serial MRI w/ interval macrocephaly (50th–98th percentile)
2	F	2 wks	Lt posterior fossa	8 mos	Cyst growth noted on serial MRI w/ subsequent interference to CSF flow
3	M	2 wks	Suprasellar	16 mos	Cyst growth noted on serial MRI
4	M	28 mos	Lt temporal fossa	7 yrs	Worsening memory & behavioral problems, potentially related to mass effect
5	M	10 yrs	Rt temporal fossa	13 yrs	Worsening headaches & gait abnormalities

continues to persist regarding the comparative effectiveness of surgical approaches for treating pediatric intracranial arachnoid cysts, with some studies ascribing benefit to a particular approach (e.g., endoscopic fenestration, CP shunting, craniotomy-based procedures)[10,14,17,20] and other research describing comparable benefits.[3] This area of inquiry also requires additional study.

Our study has several limitations. Because this study was based at a neurosurgical practice, in which the prevalence of surgery-requiring arachnoid cysts is higher than that in the primary care setting, referral bias likely influenced our estimates for predictors of surgery. Thus, we were unable to adjust for factors that primary care practitioners may have accounted for when considering neurosurgical referral for these patients. This may also partially explain the high rate of operation (33% [27 of 83]), because we studied a highly selected group of patients—those referred by primary care practitioners because of a clinical/radiological abnormality. Furthermore, this study was mostly retrospective and thus prone to bias and missing data, the latter of which would have not only lead to underestimation of the total number of cases seen during the study period but also may have contributed to the high rate of operation, given that patients who have had an operation are more likely to be found in a retrospective chart review. This was also a single-center study, thereby resulting in a relatively small sample size and limiting its generalizability. Finally, throughout the study period, the decision to operate and the specific surgical technique used were based on surgeon preference rather than being randomized or performed by using a common protocol. Therefore, our regression analyses of predictors of surgery should be regarded as exploratory and for hypothesis generation. Although it is one of the largest studies of pediatric intracranial arachnoid cysts to date, we also did not have sufficient statistical power to analyze the associations among other variables of interest, such as cyst location and specific symptoms/findings.

Conclusions

Our exploratory analyses suggest that 3 groups of pediatric patients are more likely to undergo surgery for an intracranial arachnoid cyst, namely, patients whose cyst is large, compresses a narrow CSF flow pathway (e.g., cerebral aqueduct, foramina of Monro) to cause hydrocepha-

TABLE 4. Surgical follow-up and outcomes of all patients and per surgical technique

Outcome	All Cases* (n = 27)	Surgical Technique		
		Endoscopic Fenestration (n = 7)	CP Shunt (n = 10)	Craniotomy-Based Procedure (n = 7)
Follow-up time, mos (median [min, max])	61 (1, 207)	21 (1, 107)	75 (23, 207)	21 (7, 126)
Symptom resolution w/in 6 mos (no. [%])	18 (67)	4 (57)	8 (80)	4 (57)
Need for reop to date (no. [%])	11 (41)	3 (43)	5 (50)	2 (29)
>1 reop to date (no. [%])	6 (22)	0 (0)	3 (30)	2 (29)
Complication (no. [%])				
CSF leak	5 (19)	1 (14)	1 (10)	3 (43)
Infection	3 (11)	1 (14)	1 (10)	0 (0)
Hemorrhage	1 (4)	1 (14)	0 (0)	0 (0)
Cyst-size change from before op				
Median size, cm^3 (IQR)	−49 (305)	−51 (115)	−199 (521)	−87 (39)
Decrease in size (>10 cm^3) (no. [%])	18 (67)	5 (71)	7 (70)	6 (86)
Unchanged size (w/in 10 cm^3) (no. [%])	3 (11)	1 (14)	1 (10)	0 (0)
Increase in size (>10 cm^3) (no. [%])	6 (22)	1 (14)	2 (20)	1 (14)
Length of stay, days (median [IQR])	5 (3)	3 (1)	5 (1)	5 (6)

* Includes 2 patients with bur holes and 1 with a ventriculoperitoneal shunt.

lus, or has ruptured/hemorrhaged. Furthermore, among patients who underwent surgery, there were no salient differences among the 3 surgical techniques (endoscopic fenestration, CP shunting, or craniotomy-based procedures) for several clinically important outcomes, including symptom resolution within 6 months, need for reoperation to date, cyst-size change from before the operation, morbidity, and mortality. However, given the limitations in current evidence, a larger prospective multicenter study is required to enable more robust analyses, which could ultimately provide a decision-making framework for surgical indications and clarify any differences in the comparative effectiveness of surgical approaches to treating pediatric intracranial arachnoid cysts.

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Conception and design: Singh, Ali, Farrokhyar. Acquisition of data: Singh, Ali, Bennardo, Zagzoog. Analysis and interpretation of data: Singh, Ali, Almenawer, Smith, Dao, Farrokhyar. Drafting the article: Ali. Critically revising the article: Singh, Ali, Bennardo, Almenawer, Zagzoog, Ajani, Farrokhyar. Reviewed submitted version of manuscript: all authors. Approved the final version of the manuscript on behalf of all authors: Singh. Statistical analysis: Ali, Smith, Dao, Farrokhyar. Administrative/technical/material support: Singh, Bennardo, Almenawer, Zagzoog, Smith, Dao, Ajani, Farrokhyar. Study supervision: Singh, Farrokhyar.

Supplemental Information

Previous Presentation

Portions of this work were presented in poster form at the 82nd Annual Scientific Meeting of the American Association of Neurological Surgeons held in San Francisco, California, on April 5–9, 2014.

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