



International survey on the management of Chiari 1 malformation and syringomyelia: evolving worldwide opinions

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Abstract

Introduction In 2003, pediatric neurosurgeons were surveyed under the auspices of the education committee of the International Society for Pediatric Neurosurgery (ISPN) to determine prevailing opinions regarding the management of Chiari I malformation (CIM) with and without associated syringomyelia. In the ensuing years, there has been further information from multiple CIM studies, with regards to indications, success rates of different surgical interventions, and complications. The purpose of this study was to re-evaluate current opinions and practices in pediatric CIM.

Materials and methods Pediatric neurosurgeons worldwide were surveyed, using an e-mail list provided by the ISPN communication committee chairperson. Respondents were given scenarios similar to the 2003 CIM survey in order to determine opinions regarding whether to surgically intervene, and if so, with which operations.

Results Of 300 surveys electronically distributed, 122 responses were received (40.6% response rate)—an improvement over the 30.8% response rate in 2003. Pediatric neurosurgeons from 34 different countries responded. There was broad consensus that non-operative management is appropriate in asymptomatic CIM (> 90%) as well as asymptomatic CIM with a small syrinx (> 65%). With a large syrinx, a majority (almost 80%) recommended surgical intervention. Scoliotic patients with CIM were generally offered surgery only when there was a large syrinx. There has been a shift in the surgical management over the past decade, with a bone-only decompression now being offered more commonly. There remains, however, great variability in the operation offered.

Conclusion This survey, with a relatively strong response rate, and with broad geographic representation, summarizes current worldwide expert opinion regarding management of pediatric CIM. Asymptomatic CIM and CIM with a small syrinx are generally managed non-operatively. When an operation is indicated, there has been a shift towards less invasive surgical approaches.

Keywords Chiari 1 malformation · Syringomyelia · International survey · Treatment · Patterns of practice

Introduction

A survey of pediatric neurosurgeons' opinions regarding Chiari 1 malformation (CIM) management was conducted

in 2003 [1]. At that time, a series of clinical vignettes were presented, and opinions were sought as to indications for surgical intervention, and which interventions would be performed in operated cases.

Since the publication of that study, there has been a substantial volume of published literature, including natural history studies of Chiari malformation/syrinx, and surgical series [2–5]. There has been debate as to the best intervention—primarily of bone-only decompression versus dural opening [6–8].

Given the passage of over 10 years, and the continued debate on the indications and technique of surgical intervention for pediatric CIM, the current study sought to reassess expert opinion in this field.

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Material and methods

This study was undertaken under the auspices of the Education and Communications committees of the International Society for Pediatric Neurosurgery (ISPN). A questionnaire was generated—very similar in format and content to the 2003 questionnaire [1]—in order to allow comparison between opinions over time. The questionnaire was administered through the online survey tool, FluidSurvey, hosted by the University of British Columbia, and comprised 21 questions regarding the management of hypothetical cases of pediatric CIM, with and without syringomyelia (Fig. 1). The survey was distributed electronically via the e-mail list provided by ISPN Communications chairperson, and a reminder e-mail was sent 2 weeks later. A total of 300 pediatric neurosurgeons were contacted via e-mail in 2016. Responses were collated and summary statistics generated (Table 1). The variables from the 2003 survey were compared to the 2016 survey using the chi-square and Fisher's exact tests with statistical significance set at $p = 0.05$. Due to multiple comparisons,

the Bonferroni correction was applied to reduce the amount of false positives, resulting in a new statistical significant threshold of $p = 0.0037$.

Results

Of 300 surveys electronically distributed, 122 responses were received (40.6% response rate)—an improvement over the 30.8% response rate in 2003. Pediatric neurosurgeons from 34 different countries responded: USA (32), Brazil (8), Canada (8), Japan (8), UK (8), Argentina (7), Australia (6), Turkey (4), Finland (3), India (3), Italy (3), the Netherlands (3), Denmark (2), Egypt (2), Germany (2), New Zealand (2), Saudi Arabia (2), South Korea (2), Spain (2), Belgium (1), Colombia (1), Costa Rica (1), Croatia (1), France (1), Jordan (1), México (1), Norway (1), Portugal (1), Puerto Rico (1), Singapore (1), South Africa (1), Switzerland (1), Taiwan (1), and United Arab Emirates (1). Among the respondents, 38.7% had greater than 20 years of independent (post-training)

Patient A - T2 Sagittal

A 12 year old male had an MRI completed to investigate learning disabilities. He has a normal neurological examination. The MRI demonstrates the cerebellar tonsils 12mm below the level of the foramen magnum, and no syrinx or signal change in the spinal cord. The remainder of the MRI is normal.

1. Which of the following would you recommend at this time?

- A) Follow-up
 B) Surgery
 Other (please indicate) _____

1b. Which operation would you usually offer in this situation (indicate all that apply)?

For example: Bony decompression + autologous duraplasty would check A + D

- A) Bony decompression of craniocervical junction
 B) Dural expansion/thinning without opening the dura
 C) Opening the dura and leaving it open
 D) Opening the dura + Autologous Duraplasty
 E) Opening the dura + Synthetic Duraplasty
 F) Cerebellar tonsillorhapy (shrinking the tonsils)
 G) Cerebellar tonsil resection
 H) Exploration of the 4th ventricle
 I) Other (please indicate) _____

2. If this same patient had a syrinx from T2-T4 measuring 2mm maximum diameter, which of the following would you recommend at this time?

- A) Follow-up
 B) Surgery
 Other (please indicate) _____

2b. Which operation would you usually offer in this situation (indicate all that apply)?

- A) Bony decompression of craniocervical junction
 B) Dural expansion/thinning without opening the dura
 C) Opening the dura and leaving it open
 D) Opening the dura + Autologous Duraplasty
 E) Opening the dura + Synthetic Duraplasty
 F) Cerebellar tonsillorhapy (shrinking the tonsils)
 G) Cerebellar tonsil resection
 H) Exploration of the 4th ventricle
 I) Other (please indicate) _____

3. If this same patient had a syrinx measuring 8 mm maximum diameter from C4 – T4, which of the following would you recommend at this time?

- A) Follow-up
 B) Surgery
 Other (please indicate) _____

3b. Which operation would you usually offer in this situation (indicate all that apply)?

- A) Bony decompression of craniocervical junction
 B) Dural expansion/thinning without opening the dura
 C) Opening the dura and leaving it open
 D) Opening the dura + Autologous Duraplasty
 E) Opening the dura + Synthetic Duraplasty
 F) Cerebellar tonsillorhapy (shrinking the tonsils)
 G) Cerebellar tonsil resection
 H) Exploration of the 4th ventricle
 I) Other (please indicate) _____

Fig. 1 An example of the set of questions asked for each hypothetical case of CIM. Only respondents who recommend surgery (option B) as a treatment will be presented with a follow-up question asking which operation they would offer in that particular scenario

Table 1 Comparison of responses regarding treatment recommendation between surveys conducted in 2003 and 2016. The data for the 2003 questionnaire is from Schijman et al. [1]

Variable	2003 Questionnaire	2016 Questionnaire	<i>p</i> value*
Response rate (<i>N</i>)	30.8% (76/246)	40.6% (122/300) ^a	0.020
Surgeons recommending surgery			
Patient 1			
No syrinx	8% (6/76)	6.8% (8/118)	0.782
2-mm syrinx from T2–T4	28% (21/76)	31.6% (37/117)	0.631
8-mm syrinx from C4–T4	75% (57/76)	77.6% (90/116)	0.729
Patient 2			
No syrinx	46% (35/76)	63.2% (72/114)	0.025
2-mm syrinx from T2–T4	64% (49/76)	69.9% (79/113)	0.526
8-mm syrinx from C4–T4	90% (68/76)	86.6% (97/112)	0.653
Patient 3			
No syrinx	58% (44/76)	32.1% (36/112)	0.001*
2-mm syrinx from T2–T4	40% (30/76)	43.8% (49/112)	0.652
8-mm syrinx from C4–T4	97% (74/76)	82.7% (91/110)	0.002*
Surgical technique			
Always open dura	76% (58/76)	67% (73/109)	0.0033*
Sometimes open dura	22% (17/76)	16.5% (18/109)	
Never open dura	1% (1/76)	16.5% (18/109)	
Dura management			
Synthetic	27.6% (21/75)	58.2% (53/91) ^b	0.0001*
Autologous	30.3% (23/75)	40.7% (37/91) ^b	0.198
Bovine ^c	6.6% (5/75)		
Cadaveric ^c	15.8% (12/75)		
Left open	14.5% (11/75)	8.8% (8/91) ^b	0.328

* Significant at $p < 0.0037$ ^a Number of surveys submitted^b Some respondents indicated multiple preferences in dura management^c Not offered as a dural substitute in the 2016 survey

practice, 33.9% had 10 to 20 years of practice, 19.4% had 5 to 10 years of practice, and 8.1% had less than 5 years of practice.

The survey's first case presented a 12-year-old male with learning disabilities, normal neurological exam, with an MRI that demonstrated the cerebellar tonsils 12 mm below the level of the foramen magnum (FM), and no syrinx or signal change in the spinal cord (Fig. 2). Surgery was recommended by 6.8% of respondents for the patient's C1M—similar to the 8% who did so in 2003. The following scenario was the same patient as the first, but with a syrinx from T2–T4, with a maximum diameter of 2 mm. Respondents answered similarly to the previous study with 31.6% recommending surgery versus 28% of respondents in 2003. With the presence of an 8-mm syrinx from C4–T4 in the same hypothetical patient, 77.6% recommended surgery; again, similar to the 2003 survey wherein 75% recommended surgery.

The next case was a 9-year-old male with suboccipital headaches for 6 months and a normal neurological exam. The patient had an MRI that showed the descent of flattened tonsils 10 mm into the cervical canal (fig. 3). With no syrinx

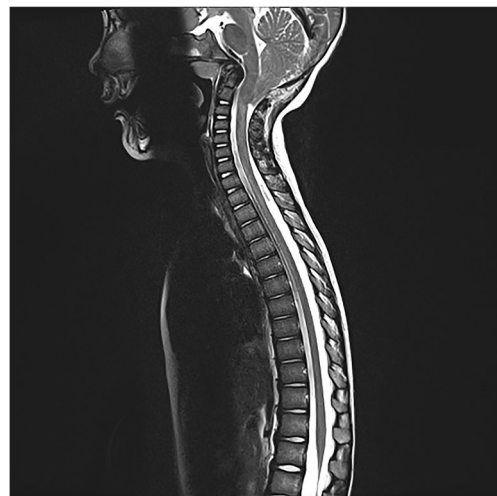


Fig. 2 Patient 1—A 12-year-old male had an MRI investigating learning disabilities. He has a normal neurological examination. The MRI demonstrates the cerebellar tonsils 12 mm below the level of the foramen magnum, and no syrinx or signal change in the spinal cord. Remainder of the MRI is normal

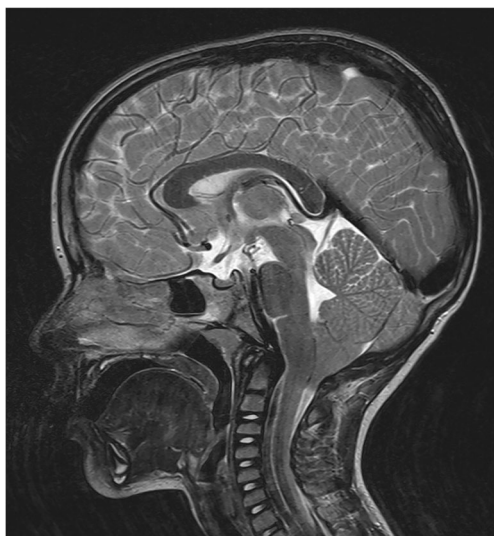


Fig. 3 Patient—A 9-year-old male with suboccipital headaches for 6 months, has an MRI that shows descent of flattened tonsils 10 mm into the cervical canal. His neurological examination is normal

present, 63.2% recommended surgery—an increase from the 46% who did so in 2003. If this patient was found to have a 2-mm syrinx from T2–T4 on MRI, 69.9% of respondents recommended surgery, which was similar to the 64% who indicated so in 2003. Surgery was recommended by 86.6% of respondents if the same patient had an 8-mm syrinx from C4–T4. This result is similar to the previous study wherein 90% recommended surgery.

The last case in the survey was an 11-year-old female with scoliosis with a normal neurological examination, whose MRI showed descent of the cerebellar tonsils 12 mm into the cervical canal with no syringomyelia (Fig. 4). With no syrinx, 32.1% recommended surgery, a decrease from 58% in the prior survey. If a 2-mm syrinx



Fig. 4 Patient 3—An 11-year-old female with scoliosis, has an MRI that shows descent the cerebellar tonsils 12 mm into the cervical canal with no syringomyelia. Her neurological examination is normal

from T2–T4 was found in this patient, 43.8% recommended surgery, which is similar to the previous survey where roughly 40% of respondents would operate on the Chiari malformation. If this patient had an 8-mm syrinx, 82.7% of respondents recommended surgery, compared with 97.0% of respondents indicating so in 2003.

When surgery was offered for CIM, 67% of respondents always opened the dura, 16.5% sometimes opened the dura, and 16.5% never opened the dura—a change from the previous survey, where 76% of respondents always opened the dura, 22% sometimes opened the dura, and 1% never opened the dura. This change in operative recommendation/technique is statistically significant (chi-square statistic 11.43, $p = 0.003$).

For those who did open the dura, 58.2% indicated they used synthetic materials to close, whereas 40.7% used autologous materials. Previously, 27.6% of respondents used synthetic materials and 30.3% used autologous pericranium. In the previous survey, the remainder used either bovine or cadaveric grafts—which were not offered as a dural substitute by any respondent in the current study. Almost 9% of respondents who opened the dura mater left it open at the end of surgery, which was a decrease compared to the previous survey where that number was 15%.

Discussion

Historically, the management of CIM has varied greatly, with some surgeons and centers recommending surgery based primarily upon symptoms or signs, while others recommended surgery based primarily upon radiographic findings. This study confirms a preference for a non-surgical approach when it comes to the management of asymptomatic CIM patients with no syrinx, similar to the results from the 2003 survey done by Schijman et al. [1]. These findings are supported by the literature, which indicates that non-operated asymptomatic patients with CIM rarely deteriorate clinically [2, 3, 5, 9, 10]. For example, Pomeraniec et al. retrospectively reviewed 70 conservatively treated CIM pediatric patients (mean follow-up of 66.3 months); the vast majority (92.9%) did not experience clinical or radiological progression [3]. Novegno et al. had also reported similar findings in their review series with majority (77.3%) of CIM patients showing symptomatic improvements or continued to be asymptomatic [5].

The general consensus on the management of CIM with a small 2-mm syrinx, in an asymptomatic or scoliotic patient, has not changed much since the last survey, with the majority of surgeons (>65%) recommending conservative treatment [1]. Several retrospective natural history reviews have found that non-surgically managed CIM patients (including those with small syrinxes), experienced a benign clinical course and only a small percentage of those required

surgical intervention—largely because of either radiographic progression or symptom severity [3, 5, 10–15]. A majority of surgeons in the current study (>80%) recommended surgical intervention for asymptomatic patients with large syrinxes (8 mm), which is similar to past surveys on CIM management [1, 16]. For symptomatic patients, the majority of surgeons recommended surgical treatment, especially in the presence of a syrinx—in keeping with the current literature [2–4, 16, 17].

Opinions regarding which surgical technique to utilize for CIM decompression appears to have shifted over the past decade with a bone-only decompression now being offered more commonly than in the prior survey. However, dural opening is still more strongly favored [1, 16]. The proportion of surgeons who never open the dura when surgically managing CIM patients significantly increased from 1% in the previous survey to 16.5%. Reasons for this shift towards extradural only operations could be due to a variety of factors. Cerebral spinal fluid (CSF) related post-operative complications are often much higher when opening the dura [6, 8, 18]. A 2015 database study comparing bone-only vs. duraplasty for surgical Chiari malformation suggested duraplasty was associated with higher rates of procedural complication, length of stay, hospital charges, and re-operation rates [18]. However, a majority of surgeons in our survey still prefer to open the dura—perhaps believing that the risks associated with opening the dura can be outweighed by the benefits. As described by Lin et al. in their systematic review, posterior fossa decompression with duraplasty may result in better symptom resolution in patients with syringomyelia and lower recurrence rates [19]. Which surgical procedure is ideal for Chiari malformation remains a matter of some debate. The current study suggests that there has been an opinion trend towards bone-only decompression, but with a majority of surgeons still expressing a preference for duraplasty. The randomized study currently underway by Limbrick et al. might further clarify the role for dural opening in these patients ([ClinicalTrials.gov](https://clinicaltrials.gov/ct2/show/study/NCT02669836) Identifier: NCT02669836).

The management of the dura after decompression is another area where surgeon preferences are evolving. A majority of surgeons surveyed in 2016 (58.2%) indicated they prefer to use synthetic materials when performing a duraplasty—a change from the 2003 survey (27.6%). In addition, more surgeons now prefer to use autologous materials compared to the 2003 survey (40.7 vs. 30.3%) [1]. Although authors in the current literature postulate the ideal duraplasty material should be available in abundance, relatively inexpensive, and non-immunogenic [20–23], there is a lack of consensus on which material is best. Some studies suggest autologous materials are the most ideal choice of duraplasty material as it is non-immunogenic, and inexpensive, whereas other non-autologous materials may cause an immune response resulting in scarring and possibly cause symptoms after decompression and

duraplasty [21–24]. However, a systematic review conducted by Abla et al. in 2010 found no evidence for superiority of autologous or non-autologous materials [22]. Our survey also found fewer surgeons are leaving the dura open at the end of the surgery (8.8%) compared to the 2003 survey (15%). Retrospective reviews have suggested leaving the dura open either helped resolve the patient's symptoms or did not adversely affect outcome [25, 26].

Limitations

There are limitations to performing surveys of expert opinion. With web-based voluntary surveys, response rate and response bias are always a consideration. While it is challenging to mitigate these issues, it is interesting to note that the response rate in 2016 was higher than in 2003 (40.6 vs. 30.8%)—in the context of a larger number of neurosurgeons contacted in 2016. It appears that the ISPN Communications and Education committee platforms are increasingly effective tools to survey pediatric neurosurgeons internationally.

Conclusion

This survey, with a relatively strong response rate, and broad geographic representation, summarizes current worldwide expert opinion regarding management of pediatric CIM. Asymptomatic CIM patients and CIM patients with small syrinxes are generally managed conservatively, with surgery performed on symptomatic patients or patients with a large syrinx. When surgery is indicated, there is a trend over time towards a less invasive approach with more pediatric surgeons offering bone-only decompression now than a decade ago. There continues to be a great deal of variation in the surgical management of pediatric CIM.

Compliance with ethical standards

Ethics approval The study was approved by the University of British Columbia Children's and Women's Research Ethics Board, approval number: H15-03234.

Conflict of interest On behalf of all authors, the corresponding author states that there is no conflict of interest.

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