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Current management of pediatric chiari type 1 malformations

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ABSTRACT

Objectives: Pediatric Chiari Type 1 Malformations (CM1) are commonly referred for neurosurgical opinion. The ideal management in children regarding surgical and radiographic decision making is not clearly delineated.

Patients and methods: We retrospectively reviewed our cohort of patients age 18 years and younger referred to a single neurosurgeon for CM1. Baseline MRIs of the spine were obtained. Non-operative patients had repeat imaging at 6–12 months. Patients who underwent an operation (decompression with/without duraplasty) had repeat imaging at 6 months.

Results: One hundred and thirty-two patients with mean age of 10 years met inclusion criteria. All patients had post-operative symptomatic improvement. We identified 26 patients with syrinx, 8 with scoliosis, 3 with hydrocephalus, and one had tethered cord. The average tonsillar descent was 8.1 mm in the non-operative group and 11.9 mm in the operative group. Ninety-five patients were managed conservatively (72%). Thirty-seven were offered surgery (28%), and 33 patients underwent intervention; 21 with duraplasty (64%) and 12 without (36%).

Conclusions: Pediatric patients with CM1 require both clinical and radiographic follow-up. Duraplasty may be performed if decompression fails to relieve symptomatology, but is not always needed. CM1 continues to present a challenge in surgical decision making. Adhering to a treatment paradigm may help alleviate difficult decision-making.

1. Introduction

The diagnosis of a Chiari Type 1 Malformation (CM1) remains a controversial topic in the pediatric population [1,2]. CM1 was first described by Dr. Hans Chiari in 1891 as caudal cerebellar herniation through the foramen magnum based upon his work on autopsy specimens, and while unclear, the etiology of this disorder appears to be embryologically tied to aberrancies during development of the craniocervical junction [3–9].

The increasing prevalence of diagnostic imaging has resulted in a growing number of referrals for this condition to pediatric neurosurgeons [1,10,11]. Most patients present with typical findings such as occipital headaches, weakness, and dysphagia, although some are asymptomatic. The diversity in presentation has led to numerous proposed management courses [3,12,13]. In addition, this condition is further complicated by commonly associated findings of syringomyelia and tethered cord, which can drastically affect decision making regardless of presenting symptomology [10,14,15]. Currently, there is no single agreed upon management method [2,3].

Once a child is found to have ≥5 mm herniation of the cerebellar tonsils below the foramen magnum, a radiological diagnosis of a CM1 is made and several important questions arise:

1 Is the patient symptomatic or asymptomatic?
2 Should further imaging be obtained?
3 Should surgery be offered, and if so, what surgery?
4 Finally, should there be radiographic follow-up?

The answers to these questions generally differ among neurosurgeons [13]. There exists a need for a better understanding of which patients are likely to improve with surgical treatment and which are best managed non-operatively, as intervention is not trivial [13,16].

At our center, all pediatric CM1 patients are referred to a single neurosurgeon who utilizes a standardized management paradigm. We performed a retrospective review of this uniformly managed patient population. By reviewing patient outcomes from a single treatment

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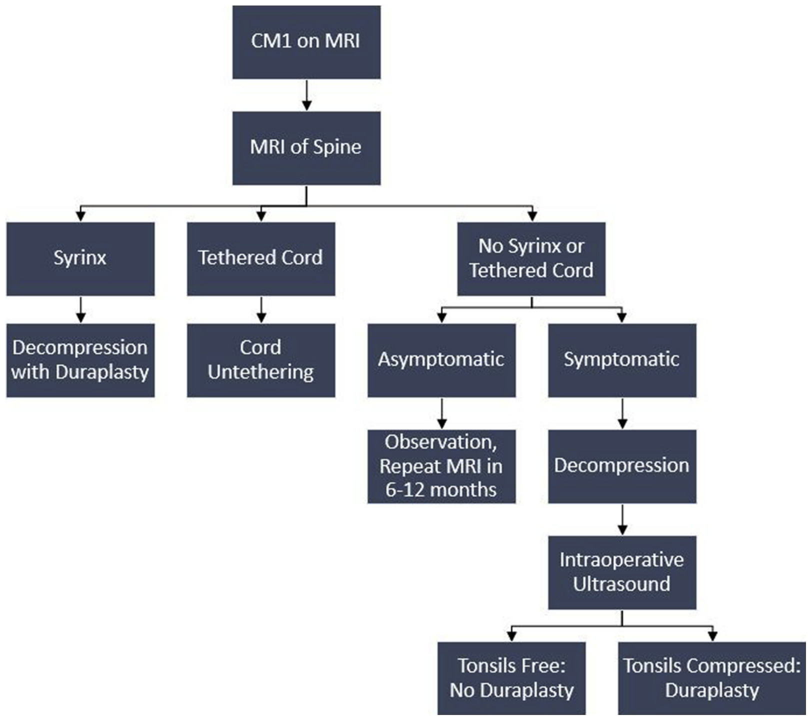

Fig. 1. Treatment paradigm for patients presenting with a Chiari malformation.

model, we hope to better understand which management strategy is best from the standpoint of symptomatic relief and anatomic pathology.

2. Management paradigm

We use a uniform treatment paradigm for all CM1 patients (Fig. 1). All patients found to have ≥5 mm tonsillar herniation below the foramen magnum undergo Magnetic Resonance imaging (MRI) of the entire spine to evaluate for a syrinx and/or tethered cord. We defined tethered cord as presence of conus medullaris terminating caudal to L3, separate from fatty infiltration of the filum. Patients with fat in the filum with the conus at the appropriate level were not considered tethered. Patients with tethered cord are first recommended to undergo untethering regardless of symptoms. Patients with a large and/or multiloculated syrinx are offered suboccipital decompression with duraplasty, also regardless of symptoms. In patients without associated findings, surgical decision-making is more difficult and predicated by the patient's symptomatology. If the clinical picture is felt to be related to tonsillar descent and symptoms are not alleviated by conservative methods, a decompression is offered.

For decompression, suboccipital bone was removed 2–3 cm cranial to and including the foramen magnum using a combination of drill and Kerrison rongeurs, with the foramen widened laterally until the dura was visualized to round out anteriorly. The C1 arch was removed in a similar manner, extending laterally to match the foramen magnum until dura was visualized to be rounding out anteriorly.

After the bone is removed and the ligamentous bands are cut, intraoperative ultrasound is used to evaluate for tonsillar compression. If the tonsils are freely floating with cerebrospinal fluid (CSF) pulsations, decompression is deemed complete. Conversely, if the tonsils appear to remain compressed, conventional duraplasty is performed using a Y-shaped incision and dural substitute [17–20]. Intradural exploration in all patients consisted of mobilization of bilateral cerebellar tonsils, lysis of all adhesions, lateralizing of the tonsils, lysis of all adhesions overlying the fourth ventricle. The fourth ventricle was explored in all patients with special attention to the obex, and lysis of any arachnoid adhesions or veils obstructing the obex. The cerebellar tonsils were then coagulated until they terminated in a more cranial and lateral position, assuring no fourth ventricular outlet obstruction. For asymptomatic patients, a repeat MRI of the cervical spine is recommended at the one-year mark.

3. Patients and methods

Retrospective review of 230 patients referred for CM1 between 2009–2015 was undertaken. All patients who did not meet the radiological diagnosis of CM1 (cerebellar tonsillar descent ≤5 mm, n = 11) were removed. Patients above 18 years of age at initial visit were excluded (n = 49). Those with history of prior decompression (n = 8), who did not show for the initial visit (n = 23), or who did not follow-up (n = 7) were also excluded. This left 132 patients who met inclusion criteria.

Clinical notes were reviewed to record age, reason for initial imaging, symptoms, surgical decision making, surgical procedure (if performed), and clinical course. MRI's were reviewed to obtain length of tonsillar decent (mm), presence of a syrinx or tethered cord (defined by presence of conus on MRI below the L3 level), size and location of syrinx if present, and changes in the syrinx over time.

Several additional measurements were made on initial T1-weighted midsagittal MRI, as described by Tubbs et al. [21]. These included the length of the McRae line, the distance of the obex to a midpoint of the McRae line, tonsillar herniation below the McRae line, odontoid process height, the clival-canal line of Wackenheim and the pB-C2 line (a perpendicular line drawn through the odontoid tip to the ventral dura from a second line drawn from the basion to the inferior-posterior aspect of C2; Fig. 2A–C).

Two odontoid angles were measured; the odontoid retroflexion and the odontoid retroversion angles. The first is defined as the angle between a line, horizontal to the plane of the image, through the midpoint of the dentocentral synchondrosis of C2, with a second through the odontoid process tip. The second is defined as the angle between a horizontal line drawn through the base of C2 and a vertical line drawn through the apex of the odontoid process (Fig. 2D–E). Author JP performed these measurements, subsequently verified by MRG and MAA.

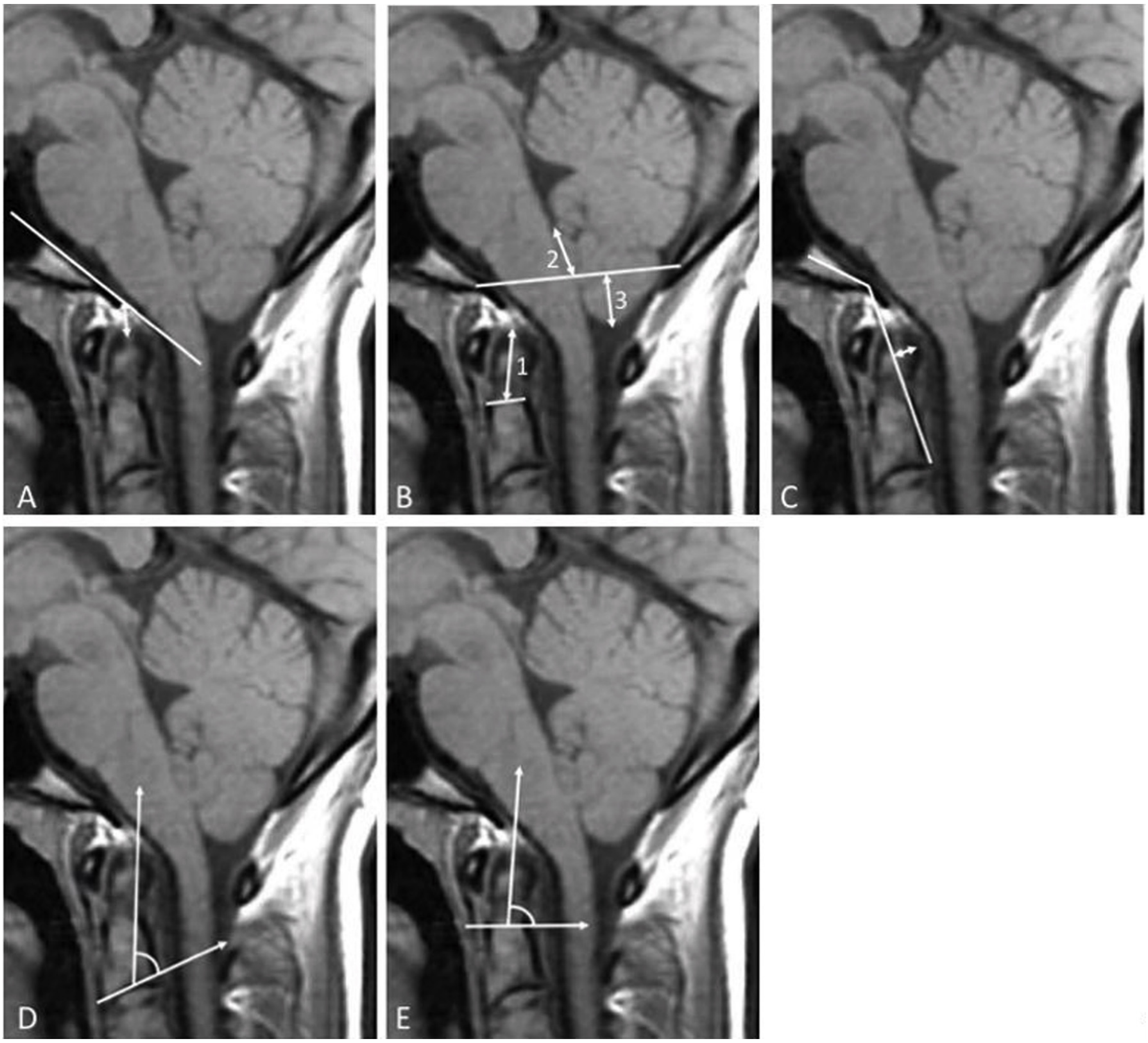

Fig. 2. Craniocervical junction measurements; **A)** Odontoid tip in relation to the clival-canal line of Wackenheim; **B)** 1 – Odontoid process length, 2 – Obex to midpoint of McRae line, 3 – Tonsillar herniation below McRae line; **C)** pB-C2 line; **D)** Retroversion angle; **E)** Retroflexion angle.

Table 1
Presenting symptoms and number of patients where surgery was recommended.

Presenting Symptom	Number of Patients (%)	Surgery recommended	Percentage (%)
Headaches	74 (57%)	21	28%
Scoliosis	8 (6%)	6	75%
Incidental	8 (6%)	0	0%
Pituitary Workup	6 (5%)	1	17%
Eye/vision complaints	6 (5%)	1	17%
Seizure	4 (3%)	0	0%
Dysphagia	3 (2%)	2	67%
Neck pain	3 (2%)	2	67%
Dysarthria	2 (2%)	1	50%
Apnea	2 (2%)	1	50%
Dizziness	2 (2%)	1	50%
Dev. Delay	2 (2%)	1	50%
Dystonia	2 (2%)	1	50%
Weakness	2 (2%)	1	50%
Bell's palsy	2 (2%)	0	0%
Incoordination	1 (1%)	0	0%
Altered mental status	1 (1%)	0	0%
Decreased sensation	1 (1%)	0	0%
Starring spells	1 (1%)	0	0%
Back pain	1 (1%)	0	0%
Toe walking	1 (1%)	0	0%

Patients were classified based upon their initial referral reason (Table 1), namely headaches, neck pain, scoliosis, visual dysfunction, dysphagia, dystonia, apnea, dizziness, and developmental delay. Several were found to have a Chiari malformation incidentally or while undergoing a pituitary work-up.

4. Results

Average initial visit age was 10.5 years (range 6 months to 18 years). On initial imaging, 26 patients had a syrinx (19.7%) and one had tethered cord (0.8%). The average length of follow-up was 1.3 years.

Ninety-five patients were managed conservatively (72%) and 37 were offered operative management (28%). Of those offered surgery, 33 underwent a decompression, 21 with duraplasty (64%) and 12 without (36%). One patient who underwent decompression without duraplasty returned for a re-exploration and subsequent duraplasty 20 months later. The average tonsillar descent was 8.4 mm ($\sigma = 3.3$) in the non-operative group and 12.6 mm ($\sigma = 5.6$) in the operative group ($p = 0.00005$). For those undergoing surgery, average time to surgery was under 6 months. Patients were followed for the presence of symptomology for an average of 18 months post-operatively. Of note, there were no post-operative issues with instability at the craniocervical junction.

Regarding odontoid measurements, the non-surgical group was found to have a significantly greater distance of the obex to the mid-point of the McRae line (6.4 vs 4.1, $p = 0.01$) and pB-C2 line (7.0 vs 6.1, $p = 0.01$). No statistically significant difference between other measurements was found. The surgical group was further subdivided into decompression with duraplasty and those without; no statistically significant difference in outcome was found. Multiple measurements were found to vary significantly between surgical and non-surgical groups, with the odontoid process height and odontoid retroflexion angles approaching statistical significance ($p = 0.09$ and $p = 0.07$, respectively). While these measurements are not validated in the literature, this was an interesting observation and further research could explore this to see if a true correlation can be found.

Twenty-six patients had syringomyelia. Of these, 17 (65%) were offered an operation and 14 underwent surgery (54%). Two patients had an enlarging syrinx after the operation, one requiring placement of a syringo-subarachnoid shunt. The other remained asymptomatic and further imaging revealed stabilization of the syrinx. Three patients did not undergo follow-up imaging to check for syringomyelia but remained asymptomatic. The remaining 9 showed significant reduction or resolution of syrinx in follow-up imaging. Within the syringomyelia subgroup, 12 did not undergo surgery; two showed a spontaneous reduction, two were lost to follow-up, and the remaining 8 reported symptomatic resolution.

5. Discussion

This analysis suggests an important connection between presenting symptoms in patients with CM1 and the likelihood of their progression to surgery. Pediatric patients have a varied symptomatology which make treatment decisions challenging [2]. This population poses an additional challenge as many have nonspecific complaints [22]. Although headaches are the most common symptom (57%), only a comparatively small percentage in our cohort underwent surgery for isolated headaches. Patients who presented with scoliosis, dysphagia, or neck pain were more likely to undergo decompression (Table 1).

It is important to note that not a single patient with incidental diagnosis of CM1 or finding during seizure workup developed symptomology leading to surgical intervention. This suggests that long term follow-up and re-imaging of incidentally found CM1 may not be required, but rather that these patients may return for reevaluation should symptoms arise.

Correlation between extent of cerebellar ectopia and need for surgical intervention is unclear [3]. While our findings suggest that greater ectopia may increase the likelihood of undergoing an operation, both groups exhibited a large spectrum of tonsillar descent, suggesting that the overall clinical picture is of greater importance than the measurement.

This study revealed that most patients who underwent a decompression did so shortly after initial imaging. However, 8% of patients were followed for more than a year before surgical management was offered, illustrating the importance of long term follow-up of pediatric patients. Some patients may have manageable symptoms for years before clinical decline necessitates intervention.

Syringomyelia results from a wide variety of traumatic, inflammatory, or compressive lesions and leads to a slowly progressive deterioration of neurological function [23]. Under [10] normal circumstances, pulsatile flow of blood is propagated by the CSF in the cervical spine and resultant subarachnoid pressure waves dissipate across the spinal canal. The association between CM1 and syringomyelia is well known, and is a result of tonsillar ectopia occluding the cervical subarachnoid space, magnifying these pressures. The "piston-like" movement of the cerebellar tonsils in response to cardiac systole drives the CSF into the Virchow-Robin spaces of the spinal cord, creating a syrinx that may gradually enlarge over time [14].

It is well established that surgical decompression of CM1 usually leads to the resolution of syrinx with the return of normal CSF flow, but it remains unclear whether operative management is always necessary to prevent progression [24,25]. In our cohort, patients with concurrent syringomyelia who did not undergo decompression were those with an incidental diagnosis. These patients exhibited an improvement or resolution of symptoms, or remained asymptomatic at subsequent visits. Thus, in otherwise asymptomatic patients with CM1, the presence of syringomyelia may be monitored for radiographic or symptomatic progression. Our findings are consistent with the literature in that significant regression or resolution of a syrinx is likely following an operation [10,14,25–28]. In our study, two patients (14%) who showed an enlarging syrinx despite surgery but only one required further intervention for clinical decline.

Of particular interest are the results of the craniocervical measurements. The smaller pB-C2 lines found in the surgical patients not only point towards the presence of a relationship between the severity of symptoms and the amount of space at the craniocervical junction, but also provide a quantifiable means of determining likelihood for surgical intervention. Furthermore, those patients requiring a duraplasty were found to have a smaller diameter of the foramen magnum, a value that can be used alongside intraoperative findings when making the decision of performing a duraplasty. While not validated measures and purely observational in this study, further research or trials on focusing on these measurements may help elucidate any validity they have in surgical decision making.

Several questions were raised in the introduction of this manuscript.

1 Is the patient symptomatic or asymptomatic? History taking is of the utmost importance, as not all cases of Chiari I are symptomatic.
2 Should further imaging be obtained? Yes, we recommend imaging of the complete spine to assess for syrinx and tethered cord.
3 Should surgery be offered, and if so, what surgery? If clearly symptomatic then we offer decompression surgery. We perform duraplasty in all cases where there is a syrinx, and in cases where lower brainstem symptoms such as dysphagia and apnea are present, and in all cases where the intraoperative ultrasound demonstrated persistent crowding of the tonsils without elevation of the dura and obvious CSF posterior to the tonsils. In patients with only tussive or exertional headaches we tend to perform the case without duraplasty, unless the previously mentioned ultrasound criteria are present.
4 Should there be radiographic follow-up? We recommend a follow up MRI at 6 months to re-assess the syrinx resolution/decrease in size after decompression. We also repeat imaging of the brain to assess CSF flow if the Chiari symptoms recur or do not improve after decompression.

6. Limitations

There is no clear method for differentiating symptomatic from non-symptomatic pediatric patients. Sometimes symptoms are classic and the decision-making concerning this aspect of the paradigm is clear. However, in many cases the neurosurgeon must interpret this information from the clinical history. In addition, this data is from a single neurosurgeon, and further validation of the management paradigm from additional centers is needed.

7. Conclusion

The choice between conservative and surgical treatment depends upon a set of criteria that varies significantly between surgeons. As a result, it has been difficult to understand the long-term outcomes of these decisions, especially as early operative management limits the potential to understand the natural course of CM1 [2,12,29]. By managing each patient with a single treatment model, we hope to better delineate which patients might benefit from earlier surgical intervention, both from the standpoint of their presenting symptoms and underlying anatomical pathology.

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Disclosure

The authors report no conflict of interest concerning the materials or methods used in this study or the findings specified in this paper.

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