# Chiari Malformation Type 1: An Appraisal of Operative Techniques and Outcomes

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**Received:** February 22, 2022, Manuscript No. MRCS-22-51242; **Editor assigned:** February 25, 2022, PreQC No. MRCS-22-51242 (PQ); **Reviewed:** March 14, 2022, QC No. MRCS-22-51242; **Revised:** April 25, 2022, Manuscript No. MRCS-22-51242 (R); **Published:** May 03, 2022, DOI: 10.4172/2572-5130.1000192

## Abstract

**Objective:** Chiari Malformation type 1 is a clinical entity characterized by impaired circulation of the cerebrospinal fluid through the foramen magnum. Traditionally, the syndrome has been characterized by herniation of the cerebellar tonsils into the spinal canal. The treatment is principally surgical. Different operative techniques have been evaluated with decompression of the posterior fossa and foramen magnum.

Materials and methods: This study aims to evaluate surgical treatment options of patients with Chiari malformation type 1using various decompression techniques, and to review the current literature for the reported outcomes. The review was conducted on the basis of database research in PubMed without limitation regarding the year of publication. A search strategy was used including the terms (Chiari I) or (Chiari type 1) and (post-surgical outcome) or (decompression surgery). 90 articles were retrieved of which 50 were directly related to our research objective. By applying exclusion criteria, 20 articles were found to define the benefits of a surgical procedure over conservative treatment in the presence of Chiari type 1 malformation.

**Results:** According to the literature, the main indication for surgical treatment was the presence of clinical findings attributed to Chiari malformation, with respect to the radiological findings. A low middle line sub-occipital craniectomy was always performed, with or without duroplasty. However, according to many authors, the possible complications of the intradural technique must always be taken into consideration before surgical planning.

**Conclusion:** In general, there was a favorable outcome after decompressive surgery associated with syrinx resolution.

**Keywords:** Chiari type 1• Malformation • Decompression surgery Outcomes

## Introduction

Arnold Chiari malformation is a posterior occipital fossa malformation. It was first described in 1891 by Dr. Hans Chiari [1]. This clinical entity is characterized by herniation of t he cerebellar tonsils within the foramen magnum and obstruction of the canal. This results in impaired circulation of Cerebro Spinal Fluid (CSF). The cerebellum controls posture and voluntary movements by influencing the smooth contraction of voluntary muscles. The cortex of the vermis controls the movements of the long axis of the body, while the cerebellar hemispheres contribute to the planning of sequential movement of the body and the conscious assessment of movement errors. Moreover, patients with cerebellar dysfunction may also present with cognitive and language deficits [2]. Six types of Chiari malformation have been described (0, 1, 1.5, 2, 3, and 4) with different radiological and clinical findings. In Chiari type 1, the obstruction of the foramen magnum is due to the extension of the cerebellar tonsils within it (>5 mm). Syringomyelia may coexist in 30-70% of the cases. It is the only type that can be acquired. It is the most common and the least severe of the spectrum, often diagnosed in adulthood. Type 2 (also known as Arnold Chiari malformation) represents a more complex entity, and is less common and more severe, almost invariably associated with myelomeningocele. Hydrocephalus is also very common [3]. Chiari types 3 and 4 are quite rare and almost always incompatible with life, therefore, these malformations are of scant clinical significance. It is not at all clear whether the 4 types of Chiari malformation represent a disease continuum corresponding to a single disorder. The 4 types (particularly types 3 and 4) are increasingly believed to have different pathogenesis and share little in common other than their name. More recently type 0 and type 1.5 have been described in the literature. Chiari 0 is defined by the presence of relative symptoms and a syrinx in the spinal cord, without (or with only marginal) descent of the tonsils. The Chiari 1.5 malformation is defined as a tonsillar herniation within a Chiari I malformation, with additional caudal descent of the brainstem through the foramen magnum (progressed form of $CM^{-1}$). Symptoms vary accordingly, as increased pressure to the brainstem, spinal cord and cerebellum gives rise to different signs. Chiari type 1 may be clinically expressed with various symptoms including: headache and neck pain (worsened by a cough or a valsalva maneuver), myelopathy, cerebellar symptoms, lower brainstem symptoms (e.g., dysarthria, dysphagia, down beat nystagmus), central cord symptoms (e.g., hand weakness, dissociated sensory loss, and cape anesthesia) as a result of the syrinx. Furthermore, symptoms such as drop attacks, dizziness, vertigo, balance issues, blurred vision and sleep apnea may be present [4]. The diagnosis of Chiari type 1 malformation is made by Magnetic Resonance Imaging (MRI), and the treatment is mainly surgical. The operative strategy for Chiari type 1 aims to relieve the symptoms of compression. The surgical technique may vary according to the surgeon's preference, and is personalized to the patients' findings. Decompression of the foramen magnum is always performed and may be accompanied by other surgical options, such as laminectomy, duraplasty, symphysiolysis and coagulation of the cerebellar tonsils [4]. Our study aims to review the literature regarding the different operative techniques and their postoperative outcomes, in patients diagnosed with Chiari type 1 syndrome.

## Surgical technique

Surgery is the main therapeutic approach for symptomatic patients with Chiari type I malformation, with or without syringomyelia. In patients who present with symptoms without hydrocephalus, foramen magnum decompression has been proven successful in relieving the symptoms of compression [5]. Various surgical techniques have been advocated for restoring CSF circulation in and around the foramen magnum. The main surgical techniques include: a simplified craniectomy technique, craniectomy with laminectomy, craniectomy and resection of the fibrous band,

craniectomy with duroplasty; additional methods of symphysiolysis, arachnoid exploration and meninges electrocoagulation, reduction of the tonsils by coagulation, shunting in the case of syrinx performed by a shunt, drainage or puncture with ultrasound guidance.

## Literature Review

This study aims to review the literature for postoperative outcomes in patients who have undergone surgery for Chiari malformation type 1. Our research strategy was based on studies including the following terms: (Chiari I) or (Chiari type 1) and (postsurgical outcome) or (decompression surgery). All the articles retrieved met the inclusion and exclusion criteria (Table 1). In data abstraction of the studies that evaluated, it was mandatory that they included the number of cases involved, the patients' mean age, the presence or absence of syringomyelia, the associated signs and symptoms, and the main postoperative outcomes of each technique used. Case reports were excluded from our study. All these data were collected in relation to the postoperative outcome. This paper aims to evaluate whether there is a consensus about the operative techniques and the postsurgical outcome. (Informed consent has been obtained, that studies have been performed according to the Declaration of Helsinki, and that the procedures have been approved by the local ethics committee)(Table 1).

**Table 1.** Studies and data correlating preoperative symptoms, surgical technique and outcomes in patients with CM[-1].

| Age group | Symptoms | Presence of syrinx | Surgical technique | Outcomes |
|---|---|---|---|---|
| Not defined | Headache | Yes | PFD without duroplasty | Syrinx resolution, No complications |
| Not defined | 78.1% headache | 0.47 | Craniectomy, C1 Laminectomy, durotomy A. dura left open B. closed with graft | Syrinx resolution in 97% with dura left open |
| Not defined | Headache or asymptomatic | N=2 yes | Craniectomy, C1 Laminectomy with ultrasonic bone scalpel Duroplasty with graft. | Excellent clinical outcomes, no neurological complications, CSF leak |
| 17-67 | Headache, Neck pain | No | Craniectomy no dural opening | Improved symptoms In all cases |
| M.A. 7,9 years | N=8 Hydroceph; N=11 without syrinx | N=37; yes | Shunting operation; FMD; Minimal intradural manipulation | Symptoms resolved Syrinx decrease |
| Notdefined | Lordosis level C2-C7 | Yes | PFD | Improved cervical alignment in 85%, and syrinx resolution in 93.3% |
| M.A. 38.7 ± 12.2 y | Physical health problems. | No | Sutureless decompression; Suturable | Both methods effective-significant improvement |
| Neurological deterioration | 0.758 | Yes | PFD w/arachnoid dissection and an alloplastic duroplasty | 82% improved, 2% morbidity, 0.9%mortality |
| 18-68 | Pain, myelopathy, atrophy, sensory loss | Yes | Craniectomy, duroplasty w/dura substitute, no arachnoid manipulation | 80, 5% improved; 11,1% stabile |
| 26 | Headache, Neurological symptomatology | No | Decompression surgery | 75% improved headache |
| Not defined | Cervicalgia, radiculalgia, headache | Yes | PFD, C1 Laminectomy, tonsillar coagulation, duraplasty | Syrinx decreased in 7, resolved in 6. N=11 good outcome, n=2 poor outcome. |
| 11 ± 5 year | Motor symptoms | 80% yes | Decompression surgery | 54% resolution all symptoms improved |
| M.A. 68 m | Head and/or neck pain Upper and lower extremity weakness | 12 yes | FMD by craniectomy and resection of the fibrous band. 21 pt. C1 Laminectomy | Significant improvement of symptoms, Reduced syringomyelia in 50% |
| M.A. 8.2 y | No CSF in cranio cervical junction in 48pt | 24 yes | Limited occipital craniectomy in all, +duroplasty in 21 cases, +tonsillar reduction in 19cases. | Clinical improvement N=12; N=18 improvement. |
| 19360 | Pain, neurological deficits, spinal deformity. | 34 yes | Posterior cranio vertebral decompression and syringosubarachnoid shunt | Excellent outcome in 82%, good outcome in 18%. Normalization of tonsils, |

| | | | | |
|---|---|---|---|---|
| | | | | syrinx resolution except than 2. |
| Not defined | Headache | Yes | PFD and duroplasty a) Neuropatch; b)Fascia lata | N=12 postop fever in A group, N=9 in B, N=1 infected granuloma; N=34 improved |
| 16619 | N=4 Cranial nerve palsy, n=6 nystagmus, N=7 scoliosis, N=1 hydrocephalus, N=5 with cerebellar ataxia and weakness. | 7 Yes | Craniectomy and Dural opening, C1posterior arch decompression,C4,5 laminectomy | 1 mortality |
| | | | syringostomy | Rest improved in their symptoms |
| Adults (N=177) | Symptoms related to syringomyelia | 97 Yes | PFD: Dura and arachnoid opened in 150 pts, Reduction of cerebellar tonsils in 135 pts | Clinical improvement due to enlargement of subarachnoid cisterns |
| Adults (N=121) | Headache, neurologic outcomes | Yes | 270° circumferential decompression | 98% headache improvement |
| | | | | 81% syrinx resolution |
| Pediatric-adult (N=145) | Headache, neurologic outcomes | Yes 65% | 92% PFD with duraplasty, from whom 65% arachnoid was dissected too, 27% Tonsilar resection | 75% improvement of neurological outcomes 37%pediatric complications 11% mortality |
| Pediatric-Adults N=105 | Headache, cranial nerves dysfunction, spinal cord syndrome | Syringomyelia with scoliosis | N=101 PDF with duraplasty | 67.3% improvement 23.9% stabilization of symptoms. |

(M.A. = Mean Age, PFD=posterior fossa decompression, FMD=foramen magnum decompression).

## Results

In the majority of the studies evaluated it was shown that the basic step in each decompression surgery is the removal of the sub occipital bone in order to create more space around the level of the foramen magnum, and restore CSF circulation. Kumar et al, after considering 22 adult patients with the presence of a syrinx, found that a simple craniectomy without duraplasty can achieve good results, while at same time avoiding the complications of intradural techniques [6]. Otherwise, the dura matter can be opened and then closed with an expansion graft, or left intact. In fact, some of the articles indicated simple craniectomy without dural opening as the method of treatment. Koechlin, Tissel and Attenello presented the outcomes of their cases with plain craniectomy [7-9]. They had similar results, reporting that head or neck pain and neurological deterioration were completely resolved after bone-only surgery. However, in these studies, there was no syringomyelia reported as a preoperative finding. On the other hand, reported that syrinx resolution in patients diagnosed with Chiari type 1 is more likely if the dura remains open rather than closed with a graft [10]. Conversely, the studies by Pakzaban, Klekamp and Spena et al. highlighted the importance of duraplasty [11-13]. All these studies included patients with associated syringomyelia. Presenting symptoms were mostly headache and neurological deterioration. In the first study, only two out of six patients had syringomyelia; the second presented seven patients out of 30 with a cyst; in the third study, there was a syrinx present in 75.8% of the group; in the last study by Spena et al, all the patients had syringomyelia. The technique used was the same in all these studies, comprising a sub-occipital craniectomy with duraplasty using a pericranial or other graft. The outcomes were excellent in 80% of their cases, and good for the rest. There were no complications, including no CSF leakage, at follow up. Regarding the closing technique, a study of 34 patients without characteristic symptomatology or syringomyelia by Williams et al. investigated a different surgical method [14]. Half of the patients had sutureless decompression surgery, and the other half had a suturable graft. The authors reported no difference in the effectiveness of the two methods. Another interesting article explored the surgical method of cervical laminectomy (C1 ± C2) for Chiari malformation 1. It may be performed in pateints with extension of the bony structure, but the C1 arch has to be completely removed in order for it to be effective. Massimi et al., presenting 30 cases with head or/and neck pain, upper and lower extremity weakness, ataxia, and/or vertigo, chose to perform foramen magnum decompression by craniectomy and C1 laminectomy in 21 patients out of 30. Syringomyelia was associated in all their cases. Their results refer to significant improvement of symptoms and reduction of syringomyelia in about 50% [15]. Syringomyelia is responsible for many neurological symptoms and may involve the whole cervical spinal cord or even extend into the thoracic cord. The cyst may resolve spontaneously after decompression. Otherwise, the cyst can be drained into the subarachnoid space. In one article by 66 patients, aged 1-53 years and presenting with pain, neurological deficits and spinal deformity were studied. Almost half of them presented with syringomyelia which was treated with a syringosubarachnoid shunt to complement posterior craniovertebral decompression [16]. The outcomes were quite impressive, with 82% excellent and 18% good outcomes. Postoperative MRI imaging showed normalization of tonsillar morphology and syrinx resolution. No complications were reported. El-Chandour et al. comparably performed posterior fossa decompression with a syringosubarachnoid shunt in 32 out of 46 patients. All the patients improved clinically, while there was syrinx resolution in 21 cases out of 32 who had received a shunt [17]. According to the different cases presented with syringomyelia, there was syrinx resolution after a simple craniectomy without performing a shunt. According to Prat et al. tonsillar coagulation was performed in 13 patients with Chiari type 1, cervicalgia, radiculalgia and associated syringomyelia. The technique uses tonsillar cauterization with bipolar cautery in order to perform coagulation [18]. The postoperative outcomes were good in 11 patients and poor in the rest of them, and according to the surgeons in this study, tonsillar cauterization is quite effective and safe. Our review indicates that the definitive management for Chiari 1 malformation is surgical in patients with clear symptoms.

A systematic literature review of symptomatic patients who did not undergo surgery found that often headache and nausea still improved, but ataxia and sensory disturbance did

not show spontaneous improvement [19]. On the other hand, in some cases, patients presented with mild symptomatology or even no symptoms at all (incidental finding). In that scenario, conservative treatment with serial follow up is usually adopted. The same review as above showed that approximately 93% of asymptomatic patients with Chiari I remained asymptomatic, even if syringomyelia was present. If the symptoms, or the tonsillar descent, or the size of the cyst have a tendency to progress, then an operation may be indicated. According to Batzdorf et al. in a series of 177 cases diagnosed with CM, symptomatology related to syringomyelia, they aimed to create larger subarachnoid cisterns and reduction of syrinx. They performed PFD with opening of dura and arachnoid membrane in almost all the patients (n=155), and reduction of cerebellar tonsils in more than half of them (n=135). They observed that, according to their research, reduction of cerebellar tonsils with duraplasty is able to achieve the highest goal improving the preoperative symptomatology.

One of the latest reviews published about surgical approach and outcomes is performed by Arnautovic and his colleagues [20]. They studied 121 cases of CM, with preoperative symptomatology related to neurological deterioration. They chose to perform a modified 270 degrees circumferential microsurgical foramen magnum decompression which was resulted to improvement of outcomes and syrinx resolution. In conclusion of this review, they refer an incidental association of BMI with $CM^{-1}$ but further data need to be published about it. Hence, the timing of surgery is very important. There are no strict rules and it usually relies on the surgeon's judgement, but generally depends on the severity and progression of the symptoms, the size of the cyst in the case of syringomyelia, and the presence of other comorbidities.

## Discussion

Chiari type 1 malformation is considered a rare disease; however, the advent of MRI has allowed the identification of many more minimally symptomatic or incidental cases than the representative symptomatic cases. Even though there is a consensus against prophylactic surgery in asymptomatic patients and for surgery in real symptomatic patients, as well as for early surgery when the latter is indicated, there is still considerable controversy in regards to the appropriate surgical strategy [20]. When it has been decided that the patient needs surgery, the surgeon has to choose the best operative technique according to the symptomatology and the imagistic examination of each case. There are a variety of different surgical mechanisms for management of Chiari malformation. The vast majority of the previously published clinical series indicate that sub-occipital craniectomy, removal of the posterior arch of the C1, and augmentative duraplasty represent the baseline surgical approach, which is applicable in most Chiari I patients, and is performed by the majority of the neurosurgeons involved [21].The relation between $CM^{-1}$ and syringomyelia is well known. The presence of syrinx in the cervical or thoracic region is quite frequent. According to the articles included, patients underwent different operative techniques for management of the malformation. It was observed that a simple craniectomy was quite helpful in cases with the formation of a syrinx. In the follow up, there was resolution of the cyst without performing drainage or use of a shunt [22]. The shrinkage or resection of the cerebellar tonsils represents another controversial point. Unfortunately, there are no comparative studies available for evaluating the exact role of this surgical maneuver. In cases of Chiari type 1, where surgery was performed using the method of tonsillar reduction by electro coagulation, there was an impressive number of patients with complete resolution of symptoms. Still, according to the literature, this method is not performed very often due to damage to nervous tissue which may be linked to other complications, such as nausea, vomiting and adhesion development. Surgical management is always associated with increased risk of complications. Complications are an important factor when the surgeon is choosing the right operative procedure in each case. As in all surgeries, infection is one of the most frequent complications. Meningitis can be extremely severe if it remains untreated. Patients become feverish with cervical pain and stiffness. One rare and severe complication which can be fatal is aseptic meningitis. This refers to infection without a bacterial agent, where there is leakage of the cerebrospinal fluid which comes in contact with inflammatory proteins and develops pyocytes. The culture is negative but the clinical image of the patient is similar to bacterial meningitis. Other complications include vascular lesions of the arteries and nerve injuries. Pseudomeningocele is also one of the complications presented, that is, a cyst with liquid surrounded by soft tissues and not meningeal membranes. This is the difference from meningocele, where in this case the cyst formation is surrounded by meninges. Treatment of the cyst is conservative, but in some cases neurological invasion is necessary [23]. Moreover, there is significant variety in the surgical methods used, in each patient's symptomatology, and even in the anatomy presented. The goal in each case remains the same, and decompression surgery needs to be performed [24-32]. There is no consensus regarding the best strategy, but there is a general approach which refers to early surgical intervention. The idea is to act in good time and repair the malformation before severe symptomatology occurs. This review presents the functional outcomes, in different age groups, with various symptoms, and the use of different operative techniques. Still, there are many issues which need to be well-defined. Many studies are underway for addressing all these controversial issues.

## Conclusion

In Chiari 1 malformation, surgical treatment is only advocated in symptomatic patients, since the natural history of the disease seems to be quite benign in asymptomatic cases. There is consensus that in cases where an indication for surgery exists, early surgery provides better functional outcomes, with neurological improvement and syrinx resolution. There are several different surgical approaches for these patients, indicating precisely that none of them can be universally employed, and surgical treatment should be individualized. Sub-occipital craniectomy along with C1 laminectomy and expansible duraplasty seems to be the most frequently performed procedure. The exact clinical importance of arachnoid dissection and/or coagulation/shrinkage of the cerebellar tonsils remain incompletely elucidated. Similarly, the most suitable surgical approach for patients with Chiari 1 and syringomyelia remains to be defined, albeit the majority of neurosurgeons seem to favor foramen magnum decompression alone (without directly addressing the syrinx) as the first-line treatment. Large-scale, multi-center, prospective studies are required to shed more light on these controversial issues. Already Known on this Topic: Various surgical approaches, such as craniectomy with duraplasty, laminectomy, tonsillectomy and many more, are in use for the treatment of Chiari malformation type 1. Universal consensus on which surgical approach should be practiced has not yet been reached, nor which is the safest and least invasive, while still providing sufficient space and allowing adequate CSF circulation. Study adds this review presents preoperative symptoms and postoperative outcomes of all the techniques used and reaches the conclusion that sub-occipital craniectomy along with C1 laminectomy and expansible duraplasty seems to be the most frequently performed procedure.

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**Cite this article:** Troupis T, et al. "Chiari Malformation Type 1: An Appraisal of Operative Techniques and Outcomes". Med Case Stud. 2022, 7(5), 001-005.