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Treatment of Chiari malformations with craniovertebral junction anomalies: Where do we stand today?

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A R T I C L E I N F O

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A B S T R A C T

Background: Chiari malformation type 1 (CM-1) is characterized by cerebellar tonsil herniation through the foramen magnum and can be associated with additional craniovertebral junction anomalies (CVJA). The pathophysiology and treatment for CM-1 with CVJA (CM-CVJA) is debated.
Objective: To evaluate the trends and outcomes of surgical interventions for patients with CM-CVJA.
Methods: A systematic review of the literature was performed to obtain articles describing surgical interventions for patients with CM-CVJA. Articles included were case series describing surgical approach; reviews were excluded. Variables evaluated included patient characteristics, approach, and postoperative outcomes.
Results: The initial query yielded 403 articles. Twelve articles, published between 1998-2020, met inclusion criteria. From these included articles, 449 patients underwent surgical interventions for CM-CVJA. The most common CVJAs included basilar invagination (BI) (338, 75.3%), atlantoaxial dislocation (68, 15.1%) odontoid process retroflexion (43, 9.6%), and medullary kink (36, 8.0%). Operations described included posterior fossa decompression (PFD), transoral (TO) decompression, and posterior arthrodesis with either occipitocervical fusion (OCF) or atlantoaxial fusion. Early studies described good results using combined ventral and posterior decompression. More recent articles described positive outcomes with PFD or posterior arthrodesis in combination or alone. Treatment failure was described in patients with PFD alone that later required posterior arthrodesis. Additionally, reports of treatment success with posterior arthrodesis without PFD was seen.
Conclusion: Patients with CM-CVJA appear to benefit from posterior arthrodesis with or without decompressive procedures. Further definition of the pathophysiology of craniocervical anomalies is warranted to identify patient selection criteria and ideal level of fixation.

1. Introduction

Chiari malformation type I (CM-1) was first described in 1891 by Hans Chiari after observing elongation of the cerebellum in postmortem examination of patients with hydrocephalus.[1] Today, it is characterized by tonsillar herniation through the foramen magnum, although the definition and diagnostic criteria remain debated.[1–3] There are many theories to explain the pathophysiology of CM-1, including reduced volume of the posterior fossa, atlantoaxial instability or dislocation, or intraspinal hypotension.[3–6]

It is common for CM-1 to be associated with a concomitant CVJA–studies have cited incidences as high as 74%.[7] Abnormalities of the CVJ include basilar invagination, platybasia, or atlantoaxial dislocation. In addition to neurological dysfunction secondary to brainstem compression, patients with concomitant CVJ deformity are likely to have cervical and occipital pain, instability, and refractory symptoms; timely and appropriate treatment is warranted in this population.[6,8,9]

With evolving definitions, multitude of presentations, and a lack of

Abbreviations: CM-1, Chiari Malformation Type I; CVJ, craniovertebral junction; CVJA, craniovertebral junction anomaly; BI, basilar invagination; PFD, posterior fossa decompression; OCF, occipitocervical fusion; TO, transoral.

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guidelines, it follows that treatment for CM-CVJA is debated.[10,11] Studies have reported success with anterior decompression and transoral approaches, posterior decompression with or without instrumentation, craniocervical fusion, and C1/C2 joint distraction and fixation.[12–16] The literature on the treatment for CM-CVJA is heterogeneous; varying populations, interventions, and measured outcomes render it difficult to claim an ideal method. We therefore sought to systematically review the literature to evaluate the trends and outcomes of surgical interventions for patients with CM-CVJA abnormalities.

2. Methods

A query of the Pubmed, Embase, and Web of Science databases was performed on 9/30/2022 in accordance with the Preferred Reporting Items for Systematic Reviews and Meta-Analysis (PRISMA)[17] guidelines. Search terms (Chiari AND adult AND ("cervical kyphosis" OR "platybasia" OR "basilar invagination" OR "basilar impression" OR "cervical instability")) were used. There was no limitation for time of publication. Duplicate articles were removed and inclusion and exclusion criteria were applied for selection.

2.1. Eligibility and selection process

The titles and abstracts of all articles were screened for adequacy by two authors (S.V and J.F.D.). The following criteria was used:

Inclusion

(1) Patients had both CM-1 and CVJA
(2) Surgical intervention described

Exclusion

(1) Non-English
(2) Letter, comment, abstract, or otherwise not full publication
(3) General review
(4) $N \leq 3$

2.2. Data collection and variables

Data was extracted from articles by two authors (S.V. and A.D.) and confirmed by a third (S.S.). Variables of interest were entered into a Microsoft Excel (*Microsoft Excel* [Computer Software]. Version 16.66.1, Redmond, VA: Microsoft; 2022) data sheet. Article information of year of publication, author, title, and study design were collected. Other variables included age of patient, symptomatology, imaging characteristics, interventions, and post-operative courses.

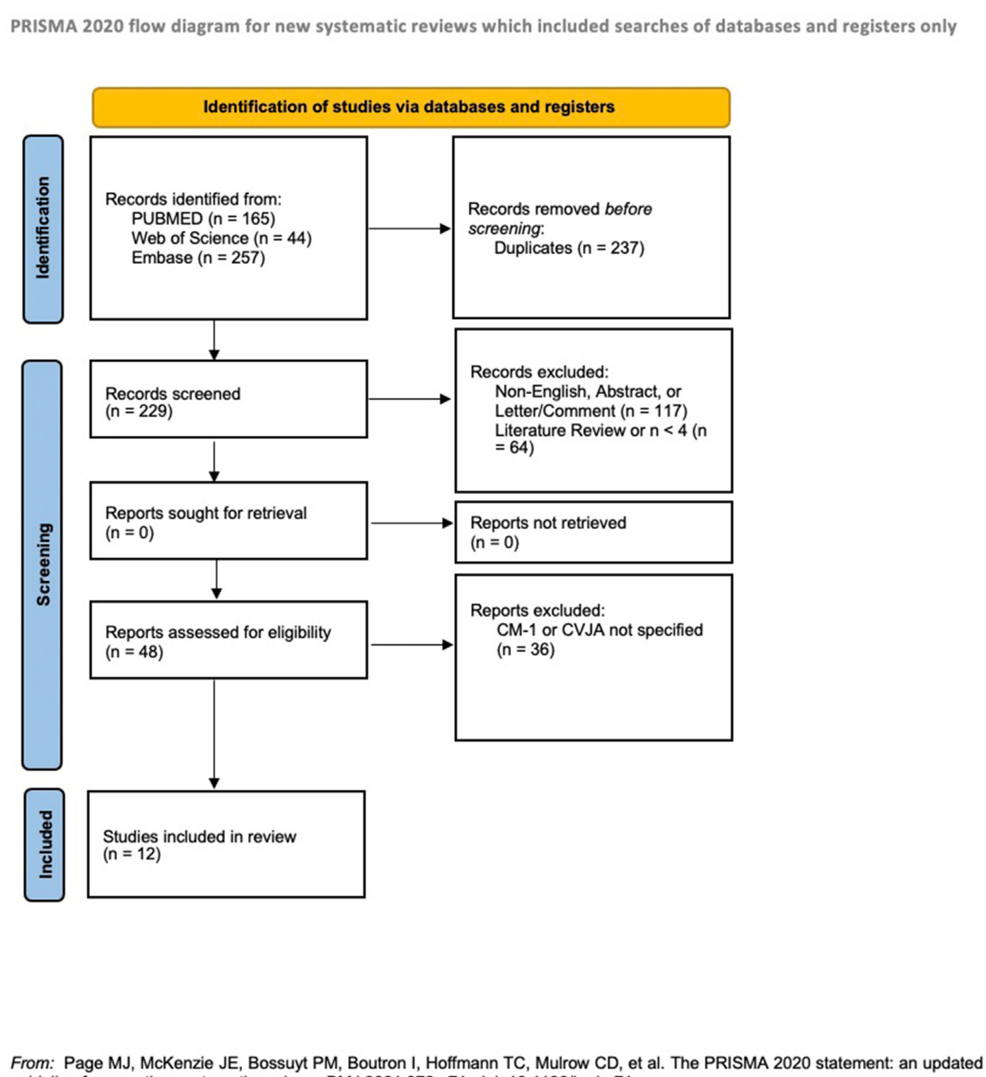

Fig. 1. PRISMA diagram of search query.

2.3. Study risk of bias assessment

Two reviewers (S.V.) and (J.F.D.) independently assessed the risk of bias using the Joanna Briggs Institute (JBI) Critical Appraisal Checklist for Case Series.[18] High risk was defined as answering yes for less than 50% of the questions included. Moderate risk was an answer of yes for 50–69%, and low risk was 70% or greater (Supplementary Table 2).

3. Results

3.1. Characteristics of included articles

Twelve articles published between 1998 and 2020 met inclusion/exclusion criteria (Fig. 1). Eleven (91.7%) were retrospective chart reviews and one (8.3%) was a combined retrospective and prospective study. Patients who underwent surgical intervention from the years 1950–2020 were included in the articles. Detailed characteristics of included articles can be found in Supplementary Table 1.

A total of 449 patients underwent surgical interventions for CM-CVJA amongst the 12 articles. The number of patients in each article ranged from 9 to 298, with an average of 66. CVJAs included basilar invagination (BI) (338, 75.3%), atlantoaxial dislocation (68, 15.1%) odontoid process retroflexion (43, 9.6%),and medullary kink (36, 8.0%). CM-1 with platybasia, assimilated C1 arch, or BI was seen in 11 (2.4%) and CM-1 with atlas assimilation was present in eight (1.8) patients (Table 1).

3.2. Symptoms

Occipital/neck pain was reported in seven articles: 192 of 328 patients (58.5%) had symptoms of occipital/neck pain.[4,5,7,12,15,19,20] In the six articles that reported paresthesias, 221 of 307 patients (72.0%) were affected.[4,5,7,12,15,19] Five articles reported weakness. In these articles, 252 of 298 (84.6%) patients had weakness.[4,5,12,15,19] Four articles reported rates of ataxia: 113 of 200 patients (56.5%) had symptoms of ataxia.[7,12,15,19] (Table 2).

3.3. Outcomes studied, length of follow-up

Clinical improvement was assessed in six (50.0%) of the articles, and Japanese Orthopedic Association (JOA) scores were outcomes in three (25.0%) studies.[4,5,14,15,19–22] Syrinx regression or collapse, delayed fusion, Nurick grades, and Visual Analogue Scale (VAS) neck pain scores were outcomes measured in one (8.3%) study each.[4,12,16,23] (Table 3). Average follow-up ranged from 19 months to 3.7 years (Supplementary Table 1).

3.4. Interventions (Table 4, Fig. 2)

3.4.1. Posterior decompression, 1950–2013

Four (33.3%) of the included articles discuss PFD as an operative strategy for patients with CM-1 and BI.[15,16,19] One study of 190 patients found that patients with CM-1 and BI who underwent PFD had improved clinical outcomes compared to those who underwent TO.[19] A retrospective review including 46 patients with CM-1 and BI found that posterior decompression without stabilization was adequate to relieve occipital pain and improve gait ataxia in patients without anterior compression.[15] PFD was also reported as appropriate in patients with CM-1 and associated BI, atlas assimilation, and atlantoaxial instability without significant brainstem compression.[16]

Table 1
Distribution of CVJAs in the patients included in the articles.

CVJA	No of patients (%) (n = 449)
Basilar invagination	338 (75.3%)
Atlantoaxial dislocation	68 (15.1%)
Odontoid process retroflexion	43 (9.6%)
Medullary kink	36 (8.0%)
Platybasia, assimilated C1 arch, and basilar invagination	11 (2.4%)
Atlas assimilation	8 (1.8%)

Table 1: Distribution of CVJAs in the patients included in the articles.

Table 2
Symptoms described in CM-CVJA patients.

Symptoms	No of patients (%)
Weakness	252/298 (84.6%)
Paresthesia	221/307 (72.0%)
Occipital/Neck Pain	192/328 (58.5%)
Ataxia	113/200 (56.5%)

Table 2: Symptoms described in the included articles.

Table 3
Outcomes reported in the included articles.

Outcomes	No of studies (%) (n = 12)
Clinical improvement	6 (50.0%)
JOA scores	3 (25.0%)
Syringomyelia regression	1 (8.3%)
Delayed fusion	1 (8.3%)
VAS neck pain scores	1 (8.3%)
Nurick grades	1 (8.3%)

Table 3: Outcomes reported in the included articles.
JOA = Japanese Orthopedic Association.

Table 4
Interventions described, number of articles focusing on the intervention, years the intervention was undertaken, and population in which the intervention was deemed successful.

Intervention	No. of Articles (%) (n = 12)	Years Studied	Populations Studied
PFD	4 (33.3%)	1950–2013	CM + BI, without significant compression CM + BI + atlas assimilation + atlantoaxial instability
TO + OCF ± PFD	4 (33.3%)	1984–2016	CM + BI with ventral compression CM + BI with ventral and dorsal compression
Atlantoaxial stabilization	4 (33.3%)	2010–2018	CM + BI CM + BI + atlantoaxial dislocation, who failed suboccipital decompression
PFD + stabilization	3 (25.0%)	1985–2013	CM + BI with ventral compression CM + BI CM + BI + atlantoaxial dislocation + syringomyelia

Table 4: Interventions described, number of articles focusing on the intervention, years the intervention was undertaken, and population in which the intervention was deemed successful.

3.4.2. Combined anterior and posterior approaches, 1984–2016

TO decompression with OCF was discussed in three (25.0%) of the included articles.[7,16,21] All three studies found this intervention to be suitable for patients with CM-1, BI, and ventral compression.[7,16,21] One study of 298 patients with CM-CVJA assessed patient outcomes after undergoing traction and transpalatopharyngeal cervicomedullary decompression with PFD and OCF.[21] In a retrospective review, eight of nine patients with CM-1 and BI who underwent TO dens resection with PFD and OCF experienced neurologic improvement.[7] Another cohort of patients with CM-1 and BI, atlas assimilation, atlantoaxial instability, and brainstem compression with CSF obstruction experienced effective relief

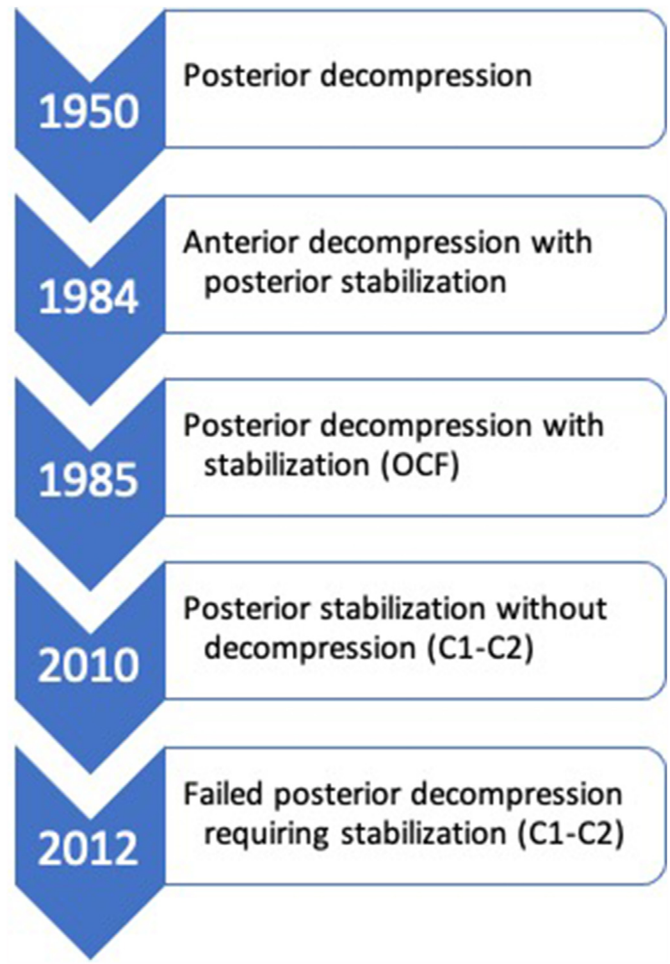

Fig. 2. Visual timeline of the reported interventions for patients with CM-CVJA.

of obstruction with anterior decompression and posterior fixation.[16]

One study assessed the feasibility and safety of endoscopic endonasal odontoid resection with PFD and fusion in patients between 2013-2016.[22] This study found this technique to be safe and feasible in patients with CM-1 and BI, and concludes similar outcomes than patients undergoing open TO decompression in the literature.[22]

3.4.3. Posterior decompression with stabilization, 1985–2013

Posterior decompression with stabilization was discussed in three (25.0%) articles.[12,15,22] In a cohort of 46 patients with CM-1 and BI, posterior fusion was necessary in patients with craniospinal instability or ventral compression due to upper cervical spine misalignment.[15] Another study found that patients with CM-1, BI, or clivoaxial angle <125° may need OCF as an adjunct to PFD.[23] In this study, all patients with CM-1 and BI failed PFD alone and required OCF.[23] In 43 patients with CM-1, reducible atlantoaxial dislocation (AAD) seen on pre-operative flexion-extension cervical x-rays, and BI, there was a decrease in Nurick grades and diminished syringomyelia in patients who underwent PFD and stabilization with occipital cortical screws and C2/C3 lateral mass cortical screw fixation devices.[12]

3.4.4. Posterior stabilization without decompression, 2010–2018

Three studies assessed the effectiveness of stabilization without PFD.[4,5,14] Two studies by the same first authors found improved JOA and VAS neck pain scores in patients with CM-1 and BI who underwent atlantoaxial fixation.[4,5] The authors conclude that CM is related to atlantoaxial instability, and stabilization is an effective treatment for patients with BI with or without CM[5]. In a series of patients with CM-1 and platybasia, assimilated C1 arch, or BI, eight out of 11 patients who underwent C1–C2 distraction and fusion without PFD had neurological improvement.[14] Three of the 11 patients had syrinx progression, increased herniation, or symptom progression following posterior fusion without PFD.[14] The authors note that outcomes after posterior fusion are similar to those cited in the literature after PFD in patients with CM with or without apparent instability. Patients with AAD were excluded from the study.[14]

3.4.5. Failed posterior decompression requiring fixation, 2012–2017

Posterior atlantoaxial facet joint reduction, fixation, and fusion was successful in improving JOA scores in a cohort of patients with CM-1, BI, and AAD who had previously undergone simple PFD.[20] These patients presented for revision surgery with occiput/neck pain, extremities weakness, paresthesia, and ataxia.[20]

4. Discussion

Published results of treatment of CM-CVJA describe a variety of combinations of surgical approaches and results. Posterior decompression appears to be the most accepted treatment, likely because of the initial thought paradigm that defined CM-1 as a product of reduced posterior fossa volume.[8,24] Ventral decompression was reported in earlier populations but seems to have lost favor, as the most recent study of open ventral decompression included patients up to 2007.[7] The use of primary or salvage posterior arthrodesis was used as far back as 1950 and has become more widely accepted as studies have shown success with or without concomitant PFD.[4,5,20]

Decompression of the posterior fossa - through ventral or posterior approach - was not universally performed on patients identified by this systematic review. Ventral decompression is a seldom-used surgical approach, as it is a challenging technique with high risks of complications.[22,25] The literature included in this review do not show convincing results that it is necessary in the setting of CM-CVJA, as every article that approached ventrally also performed PFD.[7,16,21,22] Additionally, the open TO approach was not described in more recent literature.

Interestingly, the use of PFD alone is challenged as a necessary technique by some authors.[12,15,23] Studies consistently found that patients with associated CVJA required stabilization as an adjunct to PFD.[12,15,23] This may be explained by the theory that instability, rather than reduced posterior fossa volume, is the pathophysiology of tonsillar herniation in patients with CM-CVJA.[4] Series reporting improved symptoms, JOA, and VAS neck pain scores with the use of posterior fusion without decompression support these explanations.[5,20] Furthermore, repeat surgery after PFD to incorporate posterior arthrodesis due to increased neck pain or instability has been reported in the literature.[20,26,27] In certain CM-CVJA patients, correction of malalignment of the CVJ may serve to indirectly decompress the neural elements, therefore obviating the need for PFD.

These descriptions of surgical treatment of CM-CVJA assist in the elucidation of the anatomical pathophysiology involved. Selection of surgical techniques suggest that CVJA confers a degree of instability that is benefited by posterior arthrodesis.[4,20,28] The resolution of neurological symptoms in these patients suggests resolution of compression or trauma to the brainstem or cervicomedullary junction that is caused by the instability. The level contributing most to instability is not completely clear, as some series show good results with OCF while others show the same with only atlantoaxial fusion.[4,12,23] However, it is important to note that the majority of the CM-CVJA population is pediatric. Fusion at the time of decompression is likely not possible given the requirement for growth. More studies exploring long-term outcomes and delayed stabilization through posterior arthrodesis in pediatric patients undergoing PFD for CM-CVJA are warranted.

This systematic review is not without limitations. First, the findings represent retrospective descriptions of cases in a specific population with CM-CVJA. Our findings are therefore only applicable to this patient population; patients with isolated tonsillar herniation or CVJA may benefit from different treatment paradigms that address the specific etiology of their symptoms. Furthermore, there are no direct comparisons of fusion with or without PFD among all different types of CVJA. It is important for physicians to assess the cause of neurological dysfunction to best address the patient's condition, rather than assign treatment based on the eponymous term of "Chiari malformation".[1,10,29] Different variables and outcomes are described in the articles and this makes it difficult to perform statistical analysis to develop associations. There was also a large range in follow-up timelines, with the longest follow-up described as 3.7 years. Given the effects of degenerative disease and instability that may be exacerbated over time, this follow-up may be inadequate to assess long-term outcomes in this patient population. The heterogeneity of the outcomes defined and different surgical approaches are likely the result of varying definitions, recognition, and understanding of both CM-1 and CVJA. Additionally, neither indications for delayed posterior arthrodesis nor the term 'instability' or severity of 'instability' are clearly defined in the articles. Finally, although all articles were deemed low risk for bias, selection and publication bias cannot be ruled out in the reported series of the articles.

5. Conclusion

Instability can be present in patients with CM-CVJA and may contribute to morbidity. Decompression of the posterior fossa in CM-CVJA patients does not address instability and may exacerbate it. Definitive treatment for durable results is provided by posterior arthrodesis. The need for incorporation of the upper portion of the fixation to the occiput versus the atlantoaxial spine is unclear. Further definition of the pathophysiology of craniocervical anomalies is warranted to identify patient selection criteria.

CRediT authorship contribution statement

Sima Vazquez: Conceptualization, Data curation, Formal analysis, Methodology, Writing – original draft, Writing – review & editing. **Jose F. Dominguez:** Conceptualization, Methodology, Writing – original draft, Writing – review & editing. **Ankita Das:** Data curation, Writing – original draft. **Sauson Soldozy:** Conceptualization, Methodology. **Merritt D. Kinon:** Supervision. **John Ragheb:** Supervision. **Simon J. Hanft:** Supervision. **Ricardo J. Komotar:** Supervision. **Jacques J. Morcos:** Supervision.

Declaration of competing interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.wnsx.2023.100221.

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