Surgical treatment of patients with Chiari I malformation in different age categories

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Abstract

Background: Our intention was to compare surgical treatment of patients with Chiari I malformation in our hospital to treatment in other hospitals in the world and determine the outcome and usability of different surgical techniques.

Methods: We conducted a retrospective analysis of 10 patients who were operated for CM-I at the University Clinical Centre Maribor, Division of Surgery, Department of Neurosurgery, between January 2010 and May 2016. The patients' information was gathered and a descriptive statistic was made. We reviewed English-language reports on the surgical treatment of CM-I in PubMed and Ovid, and compared our results with other series reported in literature.

Results: Between January 2010 and May 2016 we operated on 10 patients, of whom 8 were women and 2 were men, aged between 11 and 57 years. The duration of symptoms before the operation was between 1 month and 29 years. Eight patients experienced headaches, 4 neck pain, 2 vertigo, and one had a feeling of pins and needles in his arms. The duration of the neurological signs before surgery was between 1 and 29 years. Two patients had spastic tetraparesis, two showed sensory deficits and one had tremor of the hands. Five patients had syringomyelia and 2 had scoliosis. All 10 patients underwent surgery. There were no deaths in this group of patients. Two patients had fluid collection in the subcutaneous tissue, one patient also had fever and haematoma, and two patients had discharge from the wound. Patients were followed for 6 months to 6 years and all showed improvement of clinical symptoms.

Conclusion: We have come to the conclusion that our results are comparable with the results of similar studies reported in literature. However, in this respect, it should be pointed out that the number of patients included in our study was relatively small. Obviously, there is no specific entity as to when to decide for a specific type of surgery. Also, outcomes of the same type of surgery vary greatly. Surgical treatment must be individualised for each patient.

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1 Introduction

There are several surgical approaches to treating Chiari I (CM-1) malformation. However, the optimal method remains to be proven. Chiari malformation was first described by Hans Chiari in 1891 as herniation of the cerebellar tonsils below the foramen magnum (1) with or without syringomyelia (2).

The are 5 types of Chiari malformation: 0, I, II, III and IV. The most common one is CM-I, i.e. caudal herniation of the cerebellar tonsils by 5 mm below foramen magnum, which is typically associated with headaches. CM-I is a developmental anomaly of occipital mesoderm somite and is linked to hydromyelia and hydrocephalus (3). There are several explanations for tonsillar herniation and associated hydromyelia: 1. the ratio between the volume of the neural part of posterior cranial fossa and the cranial part is increased; 2. the flow of cerebrospinal fluid (CSF) through the foramen magnum is disturbed during the systole and the diastole of the heart, leading to accelerated tonsillar velocity and pulsation; 3. normal flow of CSF in the spinal canal is altered(2). Failure of rhomboid roof perforation in the fourth ventricle or medial and lateral foramina development results in a non-communicating hydrocephalus. Supratentorially, elevated pressure causes displacement of the tentorium and the development of a shallow posterior fossa. Due to the altered ratio between the volume and the pulsations of CSF, the central spinal canal is forced open, which results in hindbrain herniation, and disturbed CSF outflow during the cardiac cycle. Obstruction at the cervicomedullary junction results in increased CSF pressure during systole and the development of hydromyelia (2).

Symptoms may vary between onsets and remissions. The most common symptom is occipital headache. Characteristically, it is a dull ache that causes discomfort and is precipitated by the Valsalva manoeuvre (coughing, sneezing). While neck pain is also common, the pain is typically non-radicular. It is associated with constant burning deep discomfort in the arms, chest, the back of the head and upper extremities (3).

Signs of damage to the brainstem and the spinal chord include motor and sensory dysfunction, dysarthria, dysphagia and incontinence. CM-I signs evident in a physical examination include lower motor neuron dysfunction in the upper extremities with weakness and hyporeflexia, upper motor neuron dysfunction in the lower extremities with spasticity, hyperreflexion and fasciculations, compression of the rhombencephalon with ataxia, respiratory problems and lower cranial nerve dysfunction with dysarthria, palatal dysfunction, tongue atrophy and the absence of the gag reflex (4).

Diagnosis of CM-I in patients with our without symptoms is made using neuroradiological techniques. The first choice is MRI (3). Radiological criteria for diagnosing CM-I include cerebellar tonsillar herniation by over 5 mm below the foramen magnum, reduced volume of the posterior fossa, decrease in the size of or disappearance of cisterna magna, compression and malformation of the cervical spinal chord and the fourth ventricle a move toward the spinal canal (5). Tonsillar herniation is classified according to the level of descent through the foramen magnum in the MRI: at the first level, tonsils descend between the foramen magnum and C1, at the second level up to C1, at the third level between

C1 and C2 and at the fourth level the tonsils descend to C2 (6). Syringomyelia is diagnosed with a MRI of the entire spine (cervical, thoracic and lumbar). Roentgenograms and computer tomography (CT) is used to determine bone anomalies (3).

One of the options for treating the malformation, which is universally globally accepted, is surgical treatment (7). Only symptomatic patients are treated. According to the Pediatric Section of the American Association of Neurological Surgeons, surgical treatment is not performed on non-symptomatic children due to prophylaxis (8).

The main goals of surgical treatment of CM-I as described by Batzdorf are: resolving craniospinal pressure dissociation, restoring subarachnoid spaces and the cisterna magna in the posterior cranial fossa, eliminating or reducing the syrinx, relieving compression of the brainstem, and relieving or eliminating symptoms and signs of CM-I (9). Two major factors should be assessed in order to predict the outcome of the surgery: improvement of clinical symptoms and signs, and resolving syrignomyelia (7).

In surgical treatment of CM-I, limited suboccipital craniectomy is recommended in order to expand the foramen magnum, and decompress and preserve neural elements in posterior cranial fossa (2). Decompression can be achieved using several surgical techniques (6). Simple suboccipital craniectomy is be performed with C1 (and often also C2) laminectomy. If decompression is insufficient, craniectomy and laminectomy may be accompanied by dural opening. In the latter case, only the superficial layer may be divided or a Y-cut is made through both dural layers (6), with the arachnoid left intact (10). The dural opening is covered by duraplasty (6).

Dural substitutes used in CM-I decompression include autologous pericranium, bovine pericardium, cadaveric dura, synthetic bovine collagen matrix (Duragen), acellular human dermis allograft (Alloderm), autologous fascia lata, expanded polytetrafluoroethylene (e-PTFE) graft, posterior antlantooccipital membrane, splenius capitis muscle flap and porcine small intestinal submucosa (Durasis, Cook Biotech) (11). In case of scarring and adhesion formation around the herniated tonsils (10), dural cut may be accompanied by opening the arachnoid (6). Herniated tonsils may also be reduced by coagulation or partial tonsillectomy (10), and, particularly with hydrocephalus, by opening the Magendi foramen (6). Different types of shunting may be applied to reduce syrongomyelia (3).

In adult patients, the most common techniques include: decompression of the posterior fossa or the foramen magnum, dural opening with duraplasty and arachnoid opening with resection. In paediatric patients, arachnoid opening with tonsillar resection is less common (9).

Following surgical treatment, symptoms improve or the progression of symptoms is stopped in 80–90 % of cases. Most common postoperative complications include bleeding, CSF leak, aseptic or bacterial meningitis and recurrence of the disease (2). Other possible complications include cervical instability and cerebellar subsidence (12).

We will present the surgical treatment of patients with CM-I at the Department of Neurosurgery of the University Clinical Centre Maribor and compare it to surgical treatment elsewhere. We have focused on surgical indications, type of surgery and treatment results.

2 Methods

A retrospective analysis of patients who underwent surgery for CM-I at the Department of Neurosurgery of the University Clinical Centre Maribor between January 2010 and May 2016 was performed. Ten patients were treated during this time. We reviewed the patients' medical documentation and noted demographic data, details on the symptoms and signs before the surgery and information about the surgery, and analysed images taken before and after the surgery, and treatment outcomes.

All surgeries took place in the surgical theatre of the Department of Neurosurgery. The purpose of all surgical treatment was decompression of CM-I. All patients were put in prone position with minimally flexed head placed in the Mayfield head holder. In all cases the cut was made in the midline from the occipital protuberance to the C_3 processus spinosus. This was followed by preparation and then suboccipital craniectomy. The extent of craniectomy was from 1.5 cm under occipital protuberance to foramen magnum. The removed bone fraction measured 3×3 cm. This was followed by C_1 laminectomy in all cases, and laminectomy of C_2 and the upper part of the C_3 lamina with more extensive tonsillar herniations. Thereby, we exposed the dura and separated it into the superficial and inner layer. We could see both tonsils through the transparent inner layer. When sufficient movement of the cerebellar tonsils was noted, the surgery was concluded at this point by decompression. If this was not the case, we proceeded by making a Y-shaped dural incision. We opened the arachnoid, recognised the anatomical structures and performed coagulation and resection of cerebellar tonsils. In case of extensive hydromyelia, we made a fenestration and released hydromyelia. Rinsing was followed by duraplasty. In all cases we used bovine pericardium as dural substitute. We put TachoSil (collagen sponge) or self-adhesive artificial dura over it and always used fibrous adhesive to seal it and prevent CSF leaks. We then closed the operative would layer by layer. We gathered data on patients in tables and performed a descriptive statistics of the sample. We reviewed English-language literature on surgical treatment of CM-I in PubMed and Ovid databases, focusing on indications for surgical therapy and the type of surgery. We compared the results of studies with our findings.

The study was approved by the National Committee of Medical Ethics of the Republic of Slovenia on 28 March 2019 (decision ref. no. UKC-MB-KME-40/19).

3 Results

Between January 2010 and May 2016 10 patients aged between 11 and 57 years received surgical treatment at the Department of Neurosurgery of the University Clinical Centre Maribor. Four patients were under 18 years of age. Medial age was 27.5 years, Eight out of 10 patients were female, and the remainder were male.

Symptoms before surgery had lasted from 1 month to 29 years, and between 1 and 3 years in half of the patients. Eight patients experienced typical occipital headaches, which worsened with straining or coughing. Four patients experienced neck pain, 2 patients had vertigo, while 1 patient reported tingling in the arms.

The signs manifested (Table 1) from 1 year to 29 years before the surgery. Neurological signs did not appear in any patient before 1 year of age. One patient,

with the duration of the disease equal to or longer than 15 years had spastic tetraparesis and tactile, pain and temperature sensory disorder, while another only had spastic tetraparesis. Another patient also experienced sensory issues, while one had hand tremor.

With all patients, CM-I was diagnosed with MRI. Five patients had associated hydromyelia, which was minimal in one case and extensive in another, involving the entire spinal canal. Two patients had scoliosis. No patient had obstructive hydrocephalus.

In all 10 patients, we opted for surgical treatment. The goal of the surgery was to relieve pressure on the brain in the cervicomedullary junction, establish normal CSF flow and reduce hydromyelia. Suboccipital craniectomy and laminectomy were performed on all patients. In 6 patients, laminectomy involved only the lamina of the C1 vertebra, while it included the lamina of C1 and C2 in the remaining 4 patients. In 4 patients, the dura mater was split into the superficial and the deep layer, while in 4 patients, duraplasty with a dural graft was performed. Hydromyelia was fenestrated only in the patient with extensive hydromyelia.

There were no deaths in the group of 10 studied patients. Postoperative complications included subcutaneous collection of the CSF in one patient, elevated body temperature and swelling in the area of the operative wound in one patient, and wound discharge in two patients. Most patients came to a follow-up examination six months after surgery, which included a control MRI of the head and the cervical spine. On average, the patients were followed for three years following surgery. All patients reported improvement of symptoms.

Table 1: Signs of neurological dysfunction and share in % Archive of the Neurosurgery department at the University Medical Centre Maribor.

Signs of neurological dysfunction	Number
No neurological signs	6 (60 %)
Paresis	2 (20 %)
Signs of dysfunction to the cerebellum	1 (10 %)
Lower sensitivity to touch	2 (20 %)

In both affected female patients, scoliosis was corrected so that they did not require orthopaedic surgery. In both patients with spastic tetraparesis improvement included less impaired gait, while the immobile patient was able to stand up with support. Motor deficits improved in both patients, while there was no significant improvement of sensory deficits. Occipital headaches improved in all 8 patients. In one female patient, while headaches were lesser than before surgery, they were still very prominent six months after surgery. In all 4 affected patients, neck pain decreased. Vertigo reported by 2 female patients completely disappeared following surgery. Following surgery, 5 patients had neck pain and decreased mobility.

All patients had an MRI exam a few weeks after surgery, which showed decreased hydromyelia, decreased compression of the brainstem and expanded foramen magnum.

4 Discussion with a review of literature

The main question that we wanted to explore was which type of decompression surgery reported the best results in literature, i.e. the greatest improvement of symptoms and signs and the fewest complications without the need for re-surgery, and which type worked best for our patients.

It should be noted that as our study involved a relatively low number of patients, any conclusions cannot be fully reliable.

4.1 Suboccipital craniectomy with or without duraplasty

There have been many studies comparing two significant types of surgery: decompression of the posterior fossa with and without duraplasty. According to findings, while there is no statistical difference in the surgical outcome and the decrease in hydromyelia between both types of surgery, decompression with duraplasty involves a greater number of complications and decompression without duraplasty leads to a greater number of resurgeries (8,10,13,14,15). Short-term and long-term outcome of the surgeries was the same (13). Taking into account the MRI and tonsillar herniation measurements before the surgery, while there is no statistical difference between decompression with duraplasty or without it at levels 1 and 2, statistically, there is a marked improvement of clinical symptoms and the size of hydromyelia after decompression of the posterior fossa with duraplasty (15). Results were similar in paediatric patients (14,16). Electrophysiological research has also shown that conductivity time did not improve or improved only slightly following decompression of the posterior fossa with duraplasty (14). However, according to the study conducted by McGirt et al., in children with displacement of the tonsils below the inferior border of the arch of the atlas, ultrasonography-indicated osseous decompression alone was associated with a 2-fold risk of symptom recurrence compared to decompression with duraplasty (13). Comparison of our results on decompression with our without duraplasty leads to similar findings. It should be noted, however, that complications following duraplasty could be linked to the additional procedure, i.e. arachnoid opening and syrinx fenestration. In paediatric patients, only decompression without duraplasty was more often performed in the patients without very prominent symptoms or signs or when the symptoms and signs had not been expressed for a very long time. According to literature, first surgical option is usual only decompression in patients with fewer symptoms, while decompression with duraplasty is usually applied in more advanced forms of the disease (16).

4.2 Suboccipital craniectomy with dura-splitting without durotomy

At the craniocervical junction, the dura can be split into two layers: the outer and the inner layer. The outer layer is more fragile, while the inner layer is very plastic and can expand in some stress events (10). We performed dura-splitting without durotomy in four patients operated in our hospital. While their signs improved following surgery, they still remained in a lesser extent. According to literature, this type of surgery should contribute to shorter hospital stays (10), and also complete dural opening should yield better results than just splitting (6). We did not detect any difference in the duration of hospital stays between patients who underwent dural splitting without durotomy and those who also had duraplasty.

4.3 Grafts

Long-term outcomes of decompression with duraplasty also depends on the use of grafts. In their retrospective study, Parker et al. presume that the in-

creased number of complication following decompression of the posterior fossa is due to the use of grafts (8). Compared to allografts, autologous pericranium is linked to lower occurrence of aseptic meningitis, would infection and pseudomeningoceles. Attenello et al. found that e-PTFE is more favourable in comparison with pericranium with regard to improvement of symptoms, reduction of hydromyelia and establishing CSF flow (11). The greatest deficits of non-autologous grafts are: increased possibility of bleeding, transfer or bacteria or viruses, transfer of Creutzfeldt-Jakob disease, eosinophilic aseptic meningitis, extended wound healing, reaction to foreign object and scarring (11). In addition, allografts are linked to greater recurrence of symptoms (11). Pericardium has the advantages of the autologous tissue, as it promotes healing, while the e-PTFE is thought to reduce the adhesions between the graft and nerve tissue and prevent excessive expansion of the tissue (17). Nevertheless, in accordance with literature, there are no greater differences between autologous and non-autologous grafts in duraplasty. However, if available, high-quality pericranium should be preferred (11). We used bovine pericardium as a dural substitute in all patients whenever duraplasty was performed. Therefore, we cannot ascertain any differences in the final outcome of the surgery.

4.4 Extensive or limited suboccipital craniectomy

In suboccipital craniectomy, a decision has to be made on how extensive it should be in order to achieve sufficient decompression of the foramen magnum. If craniectomy is too limited can cause insufficient decompression, while too extensive craniectomy can theoretically cause the cerebellum to descend through (18). According to Klekamp et al less extensive craniectomy yields better results (6). However, suboccipital bone decompression should be large enough to create an artificial cisterna magna when necessary if we wish the herniated tonsils to ascend (18). Extensive suboccipital craniectomy with extreme resection of the posterior rim of foramen magnum promoted by Sindou et al. has not proven to be more effective than standard suboccipital craniectomy (8). In our hospital, we performed standard suboccipital creaniectomy measuring 3×3 cm.

4.5 Arachnoid opening and tonsillar resection

Patients with hydromyelia, previous decompression of the foramen magnum or extensive pathology of the arachnoid envelope sometime need more aggressive decompression (10). Opening the arachnoid exposes the subarachnoid space and increases the risk of adhesive arachnoiditis, aseptic megingitis and CSF leak (18). We opened the arachnoid in 3 patients. We opted for arachnoid opening, as we did not detect sufficient movement of the cerebellar tonsils following durotomy. Two patients experienced post-operative complications, i.e. CSF collection. In literature, some authors advise opening the arachoid only in cases of CM-I with hydromyelia and/or hydrocephalus when simple decompression does not suffice to achieve good CSF flow (6). Others assert that there are no differences in decompression outcomes outside or inside the arachnoid (18). None of the 3 patients with arachnoid incision had hydrocephalus and it was their first decompression surgery. All had a more difficult clinical picture with neurological deficits, having

either extensive herniation and tetraparesis, extensive hydromyelia and sensory deficits or extensive hydromyelia and spastic tetraparesis. While symptoms improved in all three patients, some remain.

If CSF circulation remains insufficient despite arachnoid opening, some authors suggest additional tonsillar resection (6). Tonsillar resection should enable immediate transition from the 4th ventricle and prevent tonsillar repositioning in order to achieve uninterrupted CSF flow from the 4th ventricle, which is one of the goals of decompression (17). In addition, in a bigger number of patients, tonsils are supposed to be firm and gliotic, which would make release only by decompression of the posterior fossa more difficult (17). Tonsillar resection is supposed to reduce the need for resurgery (17). Nevertheless, according to Sindou et al. tonsillar resection does not contribute greatly to improvement (6) but may lead to most complications (7). Compared to duraplasty, its effect on reducing the extent of hydromyelia is much less significant (7). On the contrary, in paediatric population, statistically, the effect of decompression with tonsillar resection on reducing hydromyelia is higher, while the improvement of symptoms and signs is lower (7). None of our 10 patients had tonsillar resection, so we have nothing to add to.

4.6 Obex plugging and shunting

Two other types of surgery that can be performed on CM-I patients are mentioned in literature, i.e. obex plugging and shunting. While our department had no experience in the first type of surgery, we performed shunting together with duraplasty in one patient with prominent symptoms and extensive hydromyelia. Hilda et al. compared the results of suboccipital craniectomy with C1 decompression and syringosubarachnoid (SS) shunting in patients with CM-I and hydromyelia (8). They came to the conclusion that clinical symptoms and radiological exam reports improved faster in the group of patients with SS shunting (8). Aghakhani et al also concluded that ventriculoperitoneal (VP) shunt is the best treatment for hydrocefalomyelia (19). However, Sindou et al assert that shunting is not an appropriate method to treat hydromyelia any more (6). One of the studies showed that shunting with or without decompression yielded the worst results in terms of reduction of clinical symptoms and the size of hydromyelia (7). Following duraplasty and SS shunting, the symptoms and signs improved in our patient and hydromyelia was reduced as desired. We do not attribute the neurological deficits that persisted to a lesser extent to the type of surgery but to the 15 years it had been from the presentation of symptoms to surgery. Comparing this patient with the remaining 4 patients with hydromyelia, where duraplasty was performed in two cases and dura-splitting in two, we can detect no differences, as hydromyelia decreased in all.

4.7 Duration of symptoms

Many authors stress that patients with progressing clinical symptoms and signs should undergo surgery as soon as possible, before the changes to the spine are irreversible (19). In our case, the mean value from the presentation of symptoms to surgery was 18 months, and under one year in half of the cases. There was improvement in all patients, which supports the findings of Aghakani et al.

that late surgery is better than none (19). Excluding the two patients who underwent surgery 15 and 29 years after presentation of symptoms, respectively, we cannot claim the outcome is better if surgery is performed as soon as possible. There was considerable improvement of symptoms and signs following surgery in all cases, even in patients operated on 15 and 29 years after the presentation of symptons, and the remaining patients who underwent surgery in 3 years after first reporting symptoms.

Patients with a long history of CM-I symptoms and signs and hydromyelia have visible atrophy of the upper extremities and increased tone in the lower extremities, symptoms of gait and balance dysfunction and headaches with physical activity more often (17). One of our patients, who had a 15-year history of CM-I and hydromyelia, had notably increase tone in the arms and legs, gait problems and atrophy of the upper extremities.

4.8 Predictive factors

The most significant predictive factor decribed by Nagin is the presence of clinical symptoms and signs, including scoliosis, headache, cervical pain and/or sleep apnea (8). In paediatric population, higher age upon diagnosis should mean more aggravated neurological symptoms (20). Considering the symptoms and the age upon diagnosis of our patients who were younger than 18 years of age when treated in our hospital we have not reached the same conclusion. The oldest paediatric patient, a female who was 15 years old, had only occipital headaches when diagnosed, while a boy younger by two years also had tremor of the hands and neck pain, and a girl younger by 4 years had occipital headache and scoliosis. There was significant improvement in all patients with scoliosis, occipital headache and cervical pain who were operated on in our hospital. Following surgery, scoliosis was absent in both patients and cervical pain dissipated, only one out of 4 affected patients still had occipital headache to a lesser extent.

Valentini et al. concluded that while clinical symptoms are often much worse in children than in adults, the surgical results are much better (8). Our results partly support this conclusion. While surgical results in paediatric population were better than in adults, with significant improvement of symptoms and fewer complications, in our case, the CM-I symptoms in adults were much more prominent.

5 Conclusion

The results of our study are comparable to the results of similar studies reported in literature. In this respect, it should be stressed that the number of patients included is our study is relatively low. No study has clearly defined when a certain type of surgery should be elected, while the reported outcomes vary greatly. No major complications or deaths were recorded in the group of 10 patients treated in our hospital. Only patients who exhibited symptoms underwent surgery. The most common type of surgery in our hospital was suboccipital craniectemy with or without durotomy, while the most commont surgery reported in literature was suboccipital craniectemy with duraplasty. According to experience in Slovenia and elsewhere, there are no major differences in the outcomes between the two types. In accordance with our experience and literature, there are no reliable findings on the clinical use of

more extensive surgery, including arachnoid removal and tonsillar resection, as there have been too few cases. Therefore, surgical treatment of patients should be individualised.

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