Efficacy and Safety of Duroplasty in Chiari Malformation Type 1 (CM-1) Patients Associated with Syringomyelia

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Abstract

Background Data:

Chiari malformation type-1 (CM-1) is a challenging subject to wrap our hands around table. Chiari symptoms often range from, unexplained, and/or occipital Valsalva type headache, chronic fatigue syndrome, lower cranial nerve abnormalities, brain stem compression, till severe neurological insult which augmented by syringomyelia, or srynigobulbia. Exact diagnostic and prognostic tools carry a great controversy which ranged from simple MRI study to MR imaging–based CSF velocity measurements, morphological, dynamic craniocervical junction assessments, subarachnoid pressure recordings, and compliance calculations were compared before and after surgical treatment.

Purpose:

This study aimed to estimate the efficacy and safely of duroplasty in CM-1 patients associated with syringomyelia in Sohag University Hospital

Study Design:

A retrospective descriptive study.

Patients and Methods:

The study was conducted on (23) consecutive adults with CM-1 associated with syringomyelia. They underwent surgical treatments at Sohag University Hospital from February 2011 to May 2015.

Results:

The current study was applied to (23) patients; (10) males (43.5%) and (13) females (56.5%), aged 18-64 with a mean age of (41) years. The duration of symptoms before presentation varied from 1 month up to 20 years. Clinical outcome was classified according to Glasgow outcome scale, 21 patients (91.3%) were graded V, but 2 patients only (8.7%) were grade IV, and no patient graded I. Radiologically MRI craniocervical junction suggested that decompression of the posterior fossa was achieved in all patients. Post operatively illustrated that no deterioration occurred in any of them. Improvement occurred and increased gradually on post-operative period.

Conclusion:

Craniocervical decompression with duroplasty is effective in treating patients with CM-1 with syringomyelia without the need for putting a shunt tube in the syrinx, and both clinical and radiologic improvement was documented on the follow up period

Keywords:

Chiari malformation, duroplasty, Cranio-cervical, Syringomyelia, syringobulbia.

Introduction

Chiari malformation type-1 (CM-1) is a challenging subject to wrap our hands around table

Firstly, the true incidence among population, vary from 0.35 to 1.85% with a mean 0.75% of the population, and this anomaly is associated with syringomyelia revealed wide range of patients 45%–70%[19].

Secondly, lack of agreement as regards the constitutional criteria in (CM-1),with many authors accepting 5 mm of tonsillar descent as the minimum criteria, but others suggesting that 0–2 mm, or less [3].

Thirdly, Chiari symptoms often range from, unexplained, and/or occipital Valsalva type headache, chronic fatigue syndrome, lower cranial nerve abnormalities, brain stem compression, till severe neurological insult which augmented by syringomyelia, or syringobulbia[19].

Fourthly, exact diagnostic and prognostic tools carry a great controversy which ranged from simple MRI study to MR imaging–based CSF velocity measurements, morphological, dynamic craniocervical junction assessments, subarachnoid pressure recordings, and compliance calculations were compared before and after surgical treatment[19].

Finally, what is the appropriate treatment for Chiari malformation when surgery is indicated? Single bone-only decompression? Should the dura be opened, or closed, with or without Duroplasty[5,9,11,19]?

Use a combination of different surgical approaches based on presentation such as arachnoid adhesolysis, and 4th ventricular exploration with or without obex plugging or stenting of the ventricle, resection of the tonsils, and various shunting procedures for the syrinx itself, and or cranio-cervical junction fixation[14]?

There is little in the literature to guide ideal algorithm for surgical treatment of Chiari malformation type I (CM-I), there is no consensus among surgeons about which method is preferred [2,5,11].

Aim of the study:

This study aimed to estimate the efficacy and safely of duroplasty in CM-1 patients associated with syringomyelia in Sohag University Hospital

Patients and Methods

- ***Study Design:***

A retrospective descriptive study.

The study was conducted on twenty three consecutive adults with CM-1 associated with syringomyelia. They underwent surgical treatments at Sohag University Hospital from February 2011 to May 2015.

- ***Patient's selection:***

Inclusion criteria were applied to patients with type 1 Chiari malformation with syringomyelia, but patients with type 2 Chiari malformation and those with type 1 without syringomyelia were excluded.

- ***Initial assessment:***

All patients were clinically assessed before treatment by history taking, general and neurological examination. The diagnosis was documented by standard MRI craniocervical junction. In addition, data were collected in standardized patients' sheets.

❖ ***Patients who fulfilled our inclusion criteria were categorized according to:***

* Age group.

* Clinical manifestation.

* The duration of symptoms before presentation.

* Presence of syringobulbia.

* The level of tonsillar descent.

* Syrinx size preoperative measured at the widest point by MRI using computer program measurements.

* Postoperative change of syrinx size was measured by Alkan digital software measurement scales at Sohag University hospital.

* Recovery period.

❖ ***Operative technique:***

The main surgical procedure consisted of a craniocervical decompression by Posterior fossa decompression was performed by a "modest superior extension (approximately 1.5-2.0 cm) and a lateral extension to the lateral most aspect of the foramen magnum and cervical spinal canal, and C1, and C2 laminectomy. Duroplasty was done to all patients using fascia lata graft with water tight closure preceded by adhesolysis of the cerebellar tonsils and plugging of the obex without a need for shunt tube insertion in the syrinx.

❖ ***Post-operative follow-up:***

Patients were on a follow up schedule in the outpatient clinic for one-month intervals after the operation for 6 months. Then it was every year. Follow up MRI craniocervical junction was done for all patients after three months and one year of surgery for at least 2 years up to 6 years.

❖ ***Outcome***

Glasgow outcome scale was used to assess the patients during the follow-up period, as follows:

V: Good recovery, resumption of normal life

IV: Moderate disability, disable but independent

III: Severe disability, dependent for daily support

II: Vegetative state, unresponsive and speechless

I: Death

In addition, syrinx diameter was measured in the follow-up period by MRI by Alkan digital software measurement scales at Sohag University hospital.

Results

The current study was applied to (23) patients; (10) males (43.5%) and (13) females (56.5%), aged 18-64 with a mean age of (41) years. The age group is shown in (table 1).

The duration of symptoms before presentation varied from 1 month up to 20 years (table 2).

They mainly suffered from Neck pain, Sensory disturbance, and intrinsic hand weakness. In addition, there was lower limb heaviness in nine patients (39%), Gait disturbance in twelve patients (52%), Stridor in two patients (8.6%) and bulbar nerves palsy in eight patients (34.7%), and sever combined cranial and spinal manifestations was expected in 21 cases (91.3%) which associated with srynigobulbia (table 3).

Furthermore, eighteen patients (78.3%) were associated with syringomyelia alone, but five (21.7%) revealed additional srynigobulbia. According to MRI findings, the level of tonsillar descent is shown in (table 4).

* From the level of foramen magnum to C1 in (4) patients (17.3%).
* C1-C2 level in (16) cases (69.5%).
* Below C2 level in (3) patients (13.2%).

As regards the duration of recovery of clinical symptoms, thirteen patients (56.5%) showed complete recovery in less than one month, seven patients (30.4%) revealed complete recovery with 1-3 months, and three patients (13.1%) needed 4-12months, with a mean of 6.4 months to show complete recovery.

Via Glasgow outcome scale, Clinical outcome was assessed, 21 patients (91.3%) revealed good recovery, 2 patients only (8.7%) showed moderated disability, and no patients showed severe disability, none of them died (table 5).

Regarding radiological outcome, MRI craniocervical junction suggested that decompression of the posterior fossa was achieved in all patients. The syrinx size preoperatively and postoperatively was assessed. In four patients revealed size less than 5 mm (17.3%) preoperatively, syrinx resolved postoperatively within (1-5 months), with a mean of 2.3 months.

Eleven patients (47.8%) who revealed size 5-10mm preoperatively, the syrinx size measured less than 5 mm postoperatively within (4-9 months), with a mean of 4.2 months.

Eight patients (34.7%) who revealed size more than 10 mm preoperatively, the syrinx size measured (5-10 mm) postoperatively within (6-14 months), with a mean of 6.7 months (table 6 and 7).

Regarding regression of srynigobulbia, all our patients (n=5) revealed nearly complete regression within (3-15 months), with a mean of 7 months.

❖ ***Surgical feasibility & Complications:***

The selected treatment approach was feasible in all cases of our series with no intraoperative complications.

As regards postoperative complications, three patients (13.1%) encountered subcutaneous CSF collection after removing the drain which managed conservatively with diuretics and carbonic anhydrase inhibitors (one of the required to lay prone for 3 days, otherwise no additional interventions). One patient (4.3%) showed wound infection which adequately managed by IV antibiotics, and frequent dressing. Neither additional surgical interventions or lumber drain, nor obvious or serious complication related to our procedure was encountered in our study.

Table 1. Distribution of age groups

Age group	No	%
15-25 years	4	17.4%
25-25 years	12	52.1%
35-45 years	3	13%
45-55 years	2	8.7%
>55 years	2	8.7%
Total	23	100%

Table 2. The duration of symptoms before presentation

Duration	No	%
1 month – 3 months	9	39.1%
3 months – 1 year	6	26%
1 year – 3 years	2	8.7%
3 years – 5 years	4	17.4%
> 5 years	2	8.7%

Table 3. Clinical presentation of 23 of symptomatic adult patients with Chiari malformation-1

Clinical presentation	%
Headache and neck pain	100%
Sensory dysesthesia /numbness	100%
Intrinsic hand weakness	100%
Gait problem	52%
Lower extremity heaviness	39%
Cranial nerve dysfunction	34%
Stridor	8.6%

Table 4. Degree of Tonsillar Herniation by MRI in patients with syringomyelia, and syringobulbia

Tonsillar herniation	Syringomyelia (Type A) (n=23)	Syringobulbia (Type B) (n=5)
Foramen magnum to C1 level	4	1
C1-C2 level	16	3
Below C2 level	3	1

Table 5. Clinical outcome

Duration	Totally improved	Slightly improved	Worse
Syringomyelia (Type A)	22	1	0
Syringobulbia (Type B)	5	0	0

Table 6. Change in syrinx size preoperative and postoperative according to MRI measurements

Syrinx size	Preoperative	Postoperative
Less than 5 mml	4	Normal
5-10 mml	11	Less than 5 mml
More than 10 mml	8	5-10 mml

Table 7. Duration of recovery of clinical symptoms postoperative

Duration	No	%
0 day – 1 month	11	47.8%
1 - 3 months	8	34.7%
3 – 6 months	3	13%
6 – 12 months	1	4.3%
> 12 months	0	0%

Illustrative cases:

Case 1:

Female patient 29 years old present with progressive weakness of the small muscles of the hands with paraesthesia both upper limbs and spastic gait with frequent falls, on examination hand grip power was G2, with wasting of the hand muscles, dissociated sensory loss with hypertonia and hyper reflexia both lower limbs.

Surgery was done clinical improvement occurs in the upper and lower limbs, power become G4a and improved gait and sensation

Case 2

Male patient 24 years old presented with stridor, difficulty in swallowing spastic gait, on examination there were lost gag and palatal reflexes, with hypertonia and hyperreflexia both lower limbs and right hemihyposthesia.

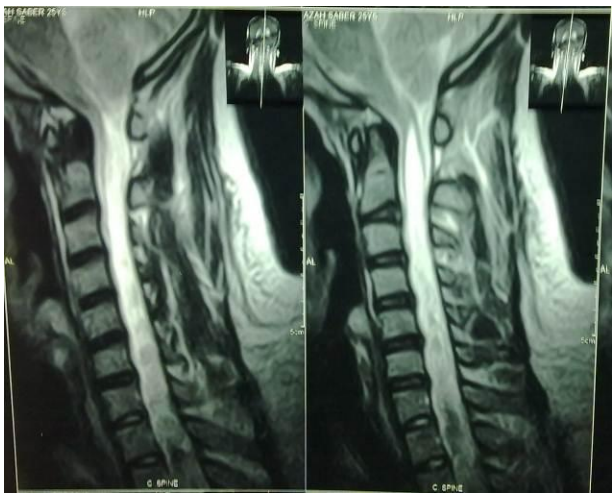
Surgery was done to him and on follow up improved bulbar symptoms and gait.

Case 3

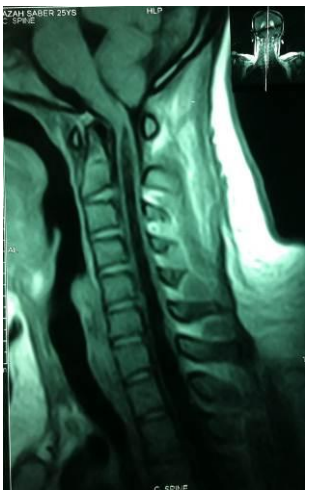
Male patient 18 years old presented with difficulty in swallowing, nasal tone of voice, and regurgitation of fluids, on examination there were lower cranial nerve palsy.

Surgery was done to him and on follow up improved bulbar symptoms completely.

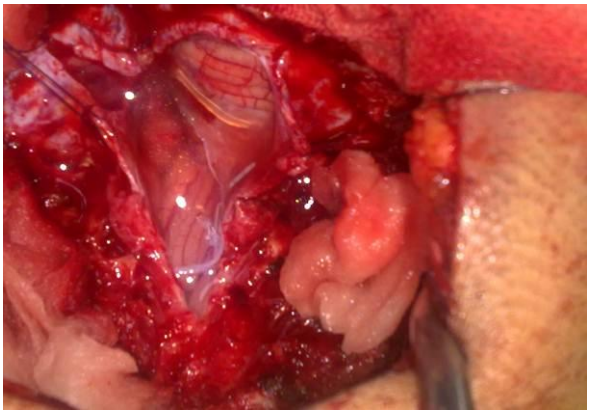
Case 1:



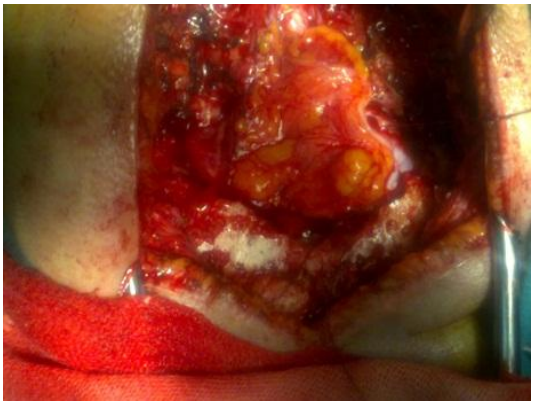
Preoperative MRI-T2



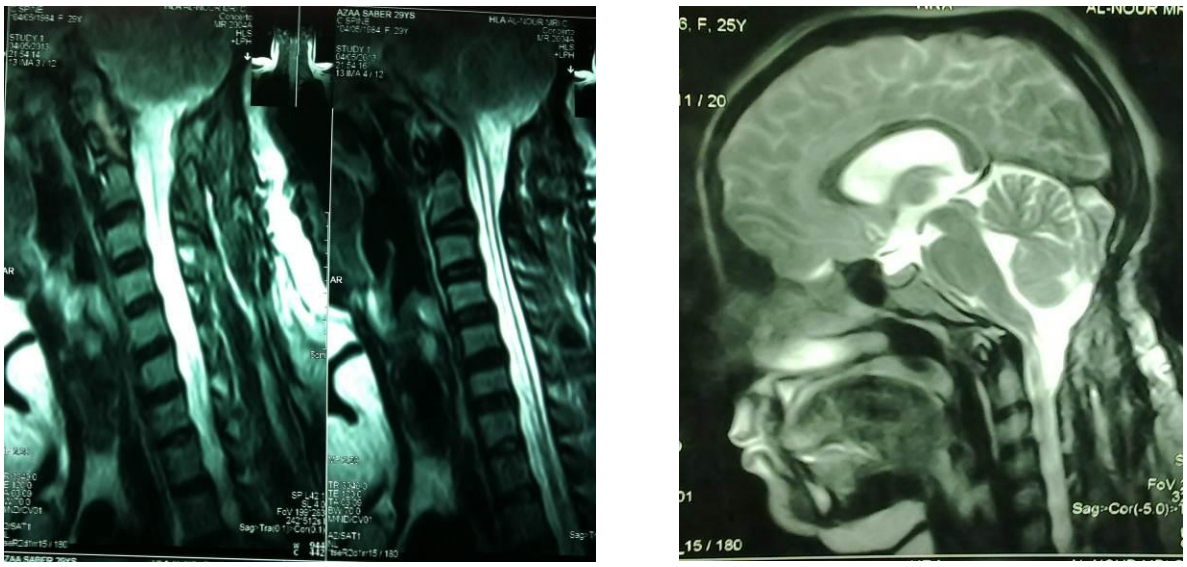
Preoperative MRI-T1



Intraoperative view before duroplasty

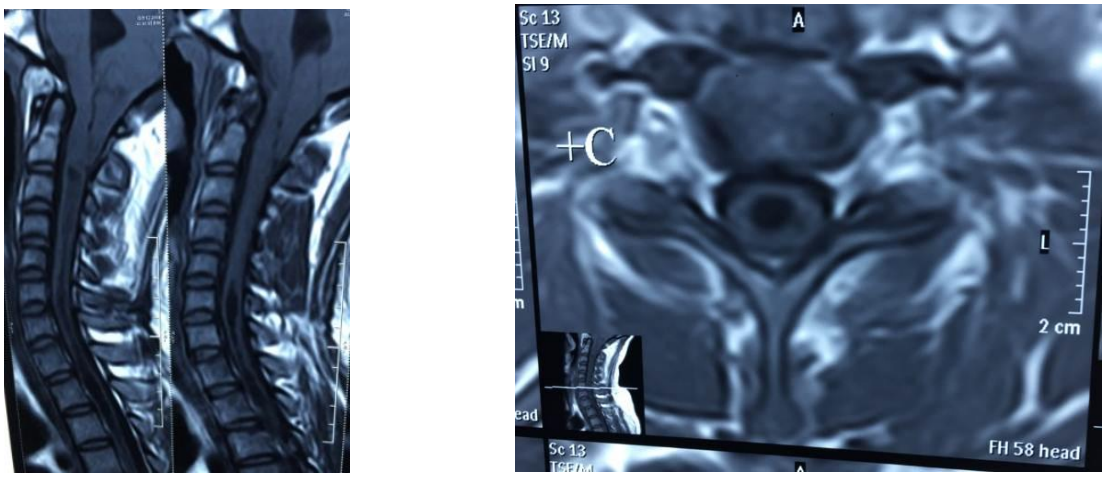


Intraoperative view after duroplasty

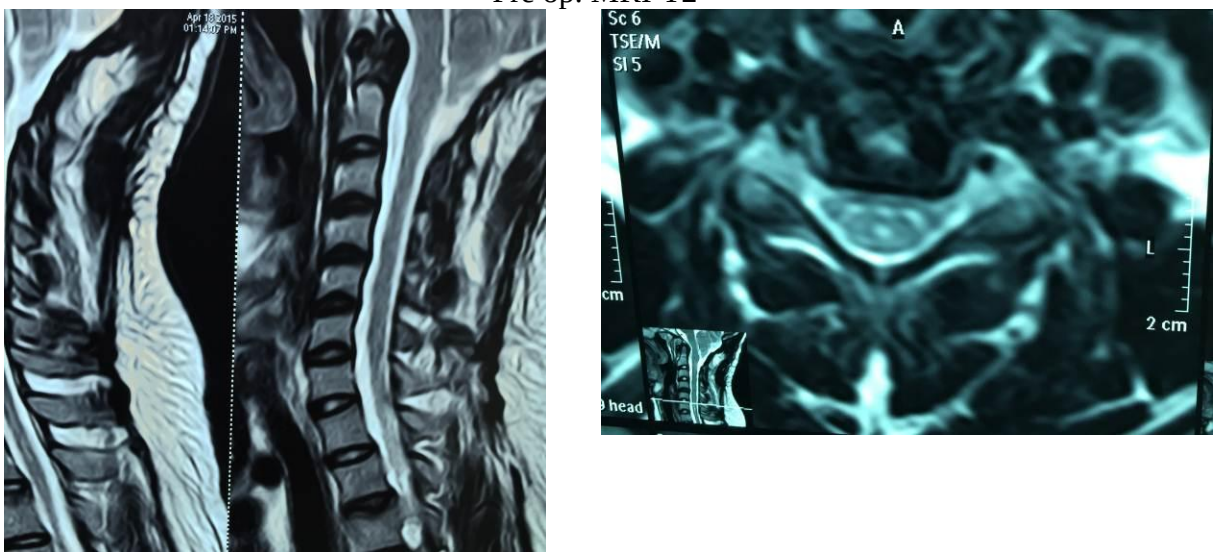


Post operative MRI T2 after 1 year

Case 2

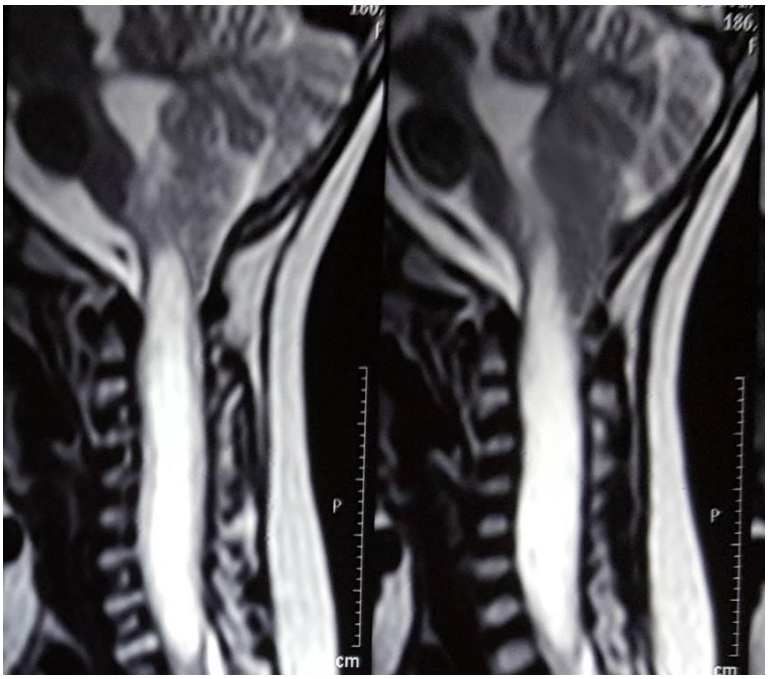


Pre op. MRI-T2

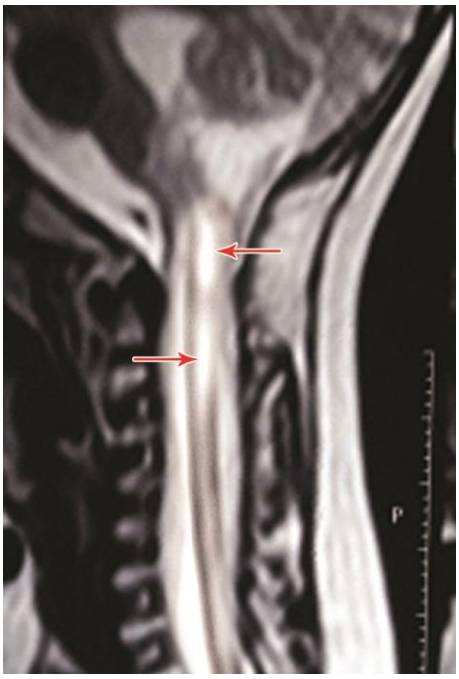


Post op. MRI –T1 after 6 months

Case 3



Preoperative MRI showing the extent of the syringobulbia



Postoperative MRI showing regression of the syringobulbia (red arrows) after one year

Discussion

This first sentence in the study by Yilmaz et al. on the surgical treatment of CM-1, encapsulates the treatment dilemma that surgeons face with this disorder[20].

Overall, results confirm that posterior fossa decompression gives high rates of clinical and radiological improvement and a low complication rate[7,11,16].

This study checked the efficacy of duroplasty in treatment the patients of CM-1 associated with syringomyelia, and surgical outcome.

All patients clinically improved but with varying degrees during the follow up period. Symptoms duration and age were correlated with the postoperative clinical and radiological improvement[2]. This was approved in our series. About 69.5% of the patients aged below 35 years and those who reveled short term duration not more than 1 year (56.2%) revealed postoperative clinical and radiological improvement in (82.6%), and (93.5%), respectively within 3 months postoperative.

Milhorat, et al, 1999 reported that the mean age at onset of syringomyelia in CM-I was 25 years, with women representing (75%) of the patients[16].

Alfeiri et al., 2012 found that the population's mean age was 45.9±13.7 years (18–77 years), with females representing (58.8%) of the patients[2].

In a study covering (177) patients, Batzdorf, et al., 2013 reported that male patients were (45) (25.4%) and female ones were (132) (74.5%). They aged 5-78, with a mean of 37.92 years [4].

This study covered 10 were male patients and 13 female ones, aged 18-69 and a mean age of (43.5 years), denoting that syringomyelia with CM-I is more common in females and young than males and middle-aged adults.

Alfeiri et al., 2012 reported that because clinical signs and symptoms are complex and vary from one patient to another; they can be simplified into cranial (due to brainstem compression or hydrocephalus) and spinal (caused by the syrinx). In addition, that their patients mainly had signs or symptoms related to the syrinx, but only 22.9% of them experienced cranial symptoms. A combination of spinal and cranial symptoms was assessed in 16 individuals. Furthermore, clinical spinal disturbances included: sensory loss, dysesthetic pain, segmental dissociated anesthesia, limb weakness, spasticity, amyotrophy, and sphincter deficiency. Cranial symptoms of brainstem compression illustrated cranial nerve neuropathies, uncontrollable hypertension, and tinnitus[2].

Baisden 2012 reported that CMI patients were often young adults with a multiplicity of vague complaints, such as: headache, neck pain, generalized discomfort with nausea, vomiting, dizziness, fluctuating hearing loss, visual disturbances, paresthesias, weakness, fatigue, and gait difficulties. Physical examination might vary from one office visit to another. Although it sometimes reveals no focal abnormalities, it may demonstrate nystagmus, cerebellar signs, or frank myelopathy[3].

Deng et al., 2014 reported that they believe that the dominant side of tonsillar herniation determined syrinx deviation, which in turn determined clinical presentations[5].

In our series, it is found that neck pain, intrinsic hand weakness, and sensory disturbance appeared in all patients and that 5 patients suffered from lower limb heaviness. In addition, gait disturbance was manifested in 8 patients and bulbar nerves palsy in 4 patients of them stridor was found in 3 patients. The physical signs were a motor weakness in patients, sensory affection in the form of dissociated sensory loss in patients, hemihyposthesia in patients, bulbar palsy with the loss of gag palatal reflexes in the patients.

Regarding different modalities of management, Aghakhani et al., 2009 believed that many authors performed different procedures (Posterior fossa decompression and shunting), and that their results could not be easily distinguished. Most importantly, there were no clear or reproducible clinical scores to analyze patients' outcome[1].

Ellenbogen, et al., 2000 claimed that successful treatment required reestablishing CSF pathways by a surgical procedure appropriate for the presumed origin and nature of the obstruction[8].

Baisden., 2012 reported that the bony extent of the cranial decompression, the need for duroplasty, the choice for the Dural substitute material and/or Dural sealant used, and the need for an expansible cranioplasty remained controversial. However, shunting of the obex and primary shunting for syringomyelia associated with CMI became disapproved. He concluded that the primary pathology in CM-1 was attributed to CSF flow obstruction, rather than the absolute location of tonsillar descent below the foramen magnum. Therefore, this surgery basically aimed at restoring normal CSF flow dynamics[3].

Heiss et al., 2010 concluded that persistent blockage of the CSF pathways at the foramen magnum caused increased pulsation of the cerebellar tonsils, acting on a partially enclosed cervical subarachnoid space to create elevated cervical CSF pressure waves. It, in turn, affected the external surface of the spinal cord to force CSF into the spinal cord through the Virchow-Robin spaces and to propel the syrinx fluid caudally, leading to syrinx progression. A surgical procedure of reestablishing the CSF pathways at the foramen magnum reversed this pathophysiological mechanism and resolved syringomyelia[12].

Heiss et al., (2012) also found that a spinal subarachnoid block increased spinal subarachnoid pulse pressure above the block. This produced a differential pressure across the obstructed segment of the SAS, which resulted in syrinx formation and progression[12].

Alfieri et al., (2012) reported that surgical decompression with durotomy, arachnoid opening, tonsillar shrinkage, and re-creation of the cisterna magna was a safe and effective procedure. Prognosis

was excellent, with global clinical and radiological improvement in more than 90% and 80% of patients, respectively. Definite clinical predictors of poor clinical and radiological prognosis are age at the time of surgery and symptoms duration[2].

Sindou, (2009) reported that patients undergoing foramen magnum decompression with incision in the outer layer of dura or complete dural opening followed by duroplasty were significantly better (P <0.05) than those patients having foramen magnum decompression with dural and arachnoid opening but without duroplasty[17].

From a completely different point of view, Goel, (2015) claimed that Given our experience, we conclude that CM, with or without basilar invagination, was associated with instability at the atlantoaxial joint, even if such instability was not clinically manifested or demonstrated on radiological imaging. He adopted stabilization of the atlantoaxial joint as the treatment. Foramen magnum bone or dural decompression was unnecessary. In addition, both syringomyelia and CM were secondary natural events related to long-standing atlantoaxial dislocation and there was no need for direct surgical manipulation[16].

Kennedy et al., (2015) reported, in their series of outcomes after posterior fossa decompression (PFD) without dural opening, that (40) out of (57) patients (70%) demonstrated radiographic improvement, (13) patients (23%) remained stable, and (4) patients (7%) developed increased syrinx size or a new one. Furthermore, (8) patients (20%) with a syrinx underwent reoperation for persistent or recurrent symptoms or progression of associated scoliosis. They recommend applying PFD without opening the dura for most symptomatic patients with CM-I. However, it had rapidly progressive neurological deficits, rapidly progressive scoliosis with the syrinx, and craniovertebral instability requiring fusion. Based on these results, dural opening surgery if preoperative MRI suggests that partial C-2 laminectomy was required to achieve adequate decompression of the tonsils[13].

Regarding duroplasty, Yilmaz et al, (2011) In a 82 consecutive patients study of CM-I, Post-operative outcome was assessed by the Japanese Orthopedics Association scores and recovery rate. Average follow-up was 9 months. Outcome was analyzed by group (duraplasty or non-duraplasty), revealed that duraplasty seem to be the essential surgical treatment of this condition[20]

Mutchnick IS et al, (2010) in a recent comparative study included 121 patient of CM-1, 56 of the patients underwent surgery without duroplasty while 64 underwent surgery with duraplasty. The primary criteria for deciding which procedure the patients had been the presence of a syrinx. Patients with a syrinx were given a duraplasty, whereas for those without a syrinx, the dura was generally not opened, as expected, the duraplasty group had a lower rate of surgical recurrence (meaning a patient required additional surgery), at 3.1% versus 12.5% for the no duraplasty group. Also as expected, the no duraplasty group had a lower complication rate. In fact there were no complications among the no duraplasty group versus 2 pseudomeningoceles and one superficial wound issue in the duraplasty group[17].

Lal Rehman et al, (2015) reported a case series of 21 patients to evaluate the symptomatic outcome after Posterior Fossa Decompression with duraplasty in Chiari-1 malformations, revealed that Posterior fossa decompression with duraplasty is the best treatment option for Chiari-1 malformations because of symptomatic improvement and less chances of complications[14].

In a large case series study, conducted by Dickerman RD et al, (2008), to evaluate if Duraplasty is required for Chiari decompression or not? They concluded that the literature does not support bony decompression alone for any Chiari patient, and it is up to the neurosurgeon to decide between duraplasty alone or duraplasty with cerebellar tonsillar shrinkage. In addition, they recommend preoperative cine MRI on all Chiari patients[6].

A precious study by Flávio R et al, (2010) discussed What is the best choice in patients with Chiari type 1 malformation associated with syrnigomyelia?, and try to delineate the surgical outcomes in Suboccipital craniotomy with and without duraplasty, and raised a lot of challenge because It is difficult to draw conclusions from a series of limited size; however, there is a suggestion that patients with syringomyelia may have a higher likelihood of improvement after undergoing duraplasty, and Further studies are needed in order to better characterize these patients and to determine which patients with Chiari I malformation are better served with bone decompression alone, and which patients will require duraplasty to resolve their syringomyelia[9].

In our experience, duraplasty in symptomatic adult patients with CM-I is successful in all cases with minimal complications. We believe that the risk of CSF leak has to be accepted as a downside of the essential procedure, which is opening the dura and widening the cisternal space.

Magnetic resonance imaging has revolutionized early detection, and provided a greater understanding of the pathology, genesis, and manifestations of CM-I, and has also transformed outcome studies[3].

Limitations

The current study is not a comparative study. In addition, a small number of patients and a longer period of follow up is needed to detect a late possibility of syringomyelia recurrence, with no dynamic plane for atlanto-axial segment.

Conclusion

Craniocervical decompression with duroplasty is an effective treatment modality in CM-1 with syrnigomyelia without shunting of the syrinx cavity. Both clinical and radiologic improvement has been addressed on the follow up period.

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الملخص العربى

فعالية وسلامة ترقيع الأم الجافية في علاج تشوه شياري النوع الأول المرتبط بتكهف النخاع

تمهيد:

يعد التشوه الخلقي شياري النوع الأول تحديا كبيرا لجراح المخ والأعصاب. وغالبا ما تتراوح أعراض هذا المرض من الصداع الشديد غير المبرر، ومتلازمة التعب المزمن، وتشوهات الأعصاب الدماغية السفلى، وضغط جذع الدماغ، حتى الإصابة العصبية الحادة والتي تضاف إليها تكهف النخاع، أو تكهف مقلة العين. ولا تزال أدوات التشخيص والمتابعة تثير قدراً من الجدل، وهي تتراوح من دراسة التصوير بالرنين المغناطيسي البسيط إلى قياسات سرعة السائل النخاعي بواسطة الرنين المغناطيسي، إلى وتقييم المورفولوجية، وديناميكية اتصال الدماغ بالنخاع الشوكي إلى تسجيلات ضغط ما تحت الأم العنكبوتية إلى حسابات الامتثال قبل وبعد العلاج الجراحي .

الهدف:

تقييم فعالية وسلامة ترقيع الأم الجافية في علاج تشوه شياري النوع الأول المرتبط بتكهف النخاع في جامعه سوهاج

تصميم الدراسه:

دراسة مرجعية

المرضى والطرق:

أجريت دراسة بأثر رجعي على (23) شخصا لديهم تشوه شياري النوع الأول المرتبطة بتكهف النخاع خضعوا لعلاجات جراحية في مستشفى جامعة سوهاج في الفترة من فبراير 2011 إلى مايو 2015.

النتائج:

أجريت الدراسة على عدد (23) من المرضى: (10) ذكور (43.5٪) و (13) إناث (56.5٪)، تتراوح أعمارهم بين 18-64 بمتوسط عمر (41) سنة. مدة الأعراض قبل عرضها تفاوتت من شهر واحد إلى 20 عاما. وقد صنفت النتائج السريرية وفقا لمقياس غلاسكو إلى: 21 مريضا (91.3٪) كانوا في الدرجة الخامسة، ولكن 2 من المرضى فقط (8.7٪) كانوا في الدرجة الرابعة. أفاد التصوير بالرنين المغناطيسي أنه قد تم بنجاح خفض الضغط من الجزء الخلفي من الدماغ في جميع المرضى. لم يحدث تدهور في حالة أي من المرضى بعد الجراحة، وكان التحسن يزداد تدريجيا في فترة ما بعد الجراحة.

الاستنتاج:

عمليه ازاله الضغط عن المنطقه الدماغيه العنقيه مع ترقيع الام الجافيه طريقه مؤثره في علاج مرضى تشوه شيارى النوع الاول المصاحب بتكهف النخاع وذلك دون الحاج ة لوضع انبوبه تحويليه فى التكهف النخاعى. تم توثيق التحسن خلال فتره المتابعه على مستوى الاكلينكي والاشعه.