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Research Article

A Novel Technique to Assess the Outcome of Adult Chiari Malformation 1 Radiologically Following Stealth Cranioplasty, Posterior Fossa Decompression, and Posterior Fossa Decompression with Duraplasty

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Abstract

Background: This study aimed to assess the radiological outcomes of adult Chiari malformation 1 (CM1) patients with or without syringomyelia (SM) between Stealth cranioplasty (SC), posterior fossa decompression with duraplasty (PFDD) and posterior fossa decompression only (PFD), evaluated by a novel measurement technique that we devised.

Methods: This observational cross-sectional study was conducted from June 2019 to May 2021. Radiological outcomes were measured by changes in the diameter of the foramen magnum (FM) and tonsillar ectopia using our technique, and the changes in the diameter of the SM, and status of Cisterna magna from computed tomography (CT) and/or magnetic resonance imaging (MRI).

Results: 53 symptomatic adult CM1 patients ranging from 18 to 47 years of age were evaluated, 23, 19, and 11 of whom underwent SC, PFDD, and PFD respectively. All the parameters were measured in millimeters. Changes in the diameter of foramen magnum were significantly better in the SC group than in the PFD ($p<0.001$) and PFDD ($p<0.001$) groups. Changes in tonsillar ectopia were also significantly better in the SC group than in the PFD ($p<0.001$) and PFDD ($p<0.001$) groups. A significant reduction in syrinx diameter in the SC group was also seen than in the PFD ($p=0.014$) and PFDD ($p=0.032$) groups. Changes in the status of Cisterna magna were significant as a whole ($p<0.001$) and in the SC group ($p<0.001$) individually.

Conclusion: SC as a technique has several better postoperative radiological outcomes than PFDD or PFD and the new measurement technique is an innovative and effective one.

Keywords: Chiari malformation 1; Cisterna magna; Foramen magnum; McRae's line; Stealth Cranioplasty; Syringomyelia; Tonsillar ectopia

Abbreviations List: AP : Antero-Posterior; Booa: Boogard's Angle; BSMMU: Bangabandhu Sheikh Mujib Medical University; C1: First Cervical Vertebra; C2: Second Cervical Vertebra; CM: Cisterna Magna; CM1: Chiari Malformation 1; CSF: Cerebrospinal Fluid; CT: Computer Tomography ; CVJ: Craniocervical Junction; FM: Foramen Magnum; IRB: Institutional Review Board; Mcrl: Mcrae's Line; MRI: Magnetic Resonance Imaging; NIH : National Institutes Of Health; PFD: Posterior Fossa Decompression; PFDD : Posterior Fossa Decompression With Duraplasty; SC: Stealth Cranioplasty; SM : Syringomyelia; TE: Tonsillar Ectopia; WCCL: Wackenheim's Clivus Canal Line

Introduction

Diversities in presentation, management, and outcome make the Chiari malformation as enigmatic as ever. Chiari Malformation 1 (CM1) is the commonest type of the Chiari spectrum and is often complicated by the presence of Syringomyelia (SM). With time, the pathophysiology is gradually unraveling. However, newer dimensions of the complex nature of CM1 are emerging and an absolute surgical technique for CM1 appears to be a remote possibility. Presently, many surgical procedures are in practice for CM1 with many modifications having their own advantages and disadvantages. Based on magnetic resonance imaging (MRI), CM1 in adults is defined as the descent of the cerebellar tonsils > 5 Millimeters (mm) beyond the foramen magnum [1-4]. The basic technique of surgery for CM1 comprises decompression of the posterior fossa and restoration of regular Cerebrospinal Fluid (CSF) flow and dynamics around the Craniovertebral Junction (CVJ). The 2 most performed surgical techniques are Posterior Fossa Decompression (PFD) and Posterior Fossa Decompression With Duraplasty (PFDD). The posterior arch of the first Cervical Vertebra (C1) is also removed and a variety of modifications in practice have their own advantages and disadvantages. Although the outcomes have improved much lately, they are always not very inspiring [5-8], and a unique and flawless procedure is yet to come. Radiological outcome measurements of CM1 are technically challenging. We developed a novel technique of measurement for evaluation of the postoperative radiological outcomes of CM1 which is easy, simple, uniform, and reliable. In this study, we compared the radiological outcomes between PFD and PFDD with our surgical technique, the "Stealth Cranioplasty" (SC) that we developed few years back, in terms of changes in the diameter of the foramen magnum (FM) and Tonsillar Ectopia (TE) in our novel technique of measurement, and also the changes in the diameter of SM and the status of the Cisterna Magna (CM).

Methodology

Patient Selection

After obtaining approval from the institutional review board (IRB) of Bangabandhu Sheikh Mujib Medical University (BSMMU), this cross-sectional observational study was carried out between June 2019 to May 2021. Radiological follow-ups were done with Computer Tomography (CT) and/or Magnetic Resonance Imaging (MRI) scans. Patients of CM1 with or without SM of more than 18 years of age having symptoms and signs who underwent any of the 3 surgical procedures, PFD, PFDD, or SC and had postoperative CT and/or MRI follow-ups at 3 months were included in the study. CM1 patients having associated anomalies like basilar invagination, atlanto-axial dislocation, or hydrocephalus and those not having a minimum of 3 months of radiological follow-up and refusing to give consent to take part in the study were excluded.

Procedural Details

For surgery, informed written consent was obtained from the patients and/or their legal guardians. Surgical procedures were performed depending on the surgeons' choice. Posterior Fossa Decompression only (PFD) comprised suboccipital craniectomy (3 cm X 3 cm approx.), C1 laminectomy and removal of the dural band only when present and no duraplasty was attempted. In the Posterior Fossa Decompression With Duraplasty (PFDD), the best possible watertight duraplasty with pericranium or fascia lata was accomplished in all the cases following a "Y" shaped dural opening after the suboccipital craniectomy and the C1 laminectomy. During the dural opening, the arachnoid was not opened except for accidental breaches. However, the tonsils were not manipulated. All the cases of the stealth Cranioplasty (SC), which we described in detail a few years back [9-11], comprised 3 cm X 3 cm suboccipital craniectomy with C1 laminectomy (Figure 1A), midline linear arachnoid preserving durotomy (Figure 1B), duraplasty with the superficial layer of the deep cervical fascia (Figure 1C), cranioplasty with pre-shaped 5 cm X 5 cm titanium mesh which is of the shape of the canopy with flat wings of a Stealth bomber (Figure 1D), fixation of the cranioplasty along the margins of the craniectomy with screws to cover the craniectomy gap and tacking of the duraplasty with the titanium mesh to maintain the enlargement of the posterior fossa and the foramen magnum AP diameter (Figure 1E) which can be appreciated well in the postoperative CT scan (Figure 1F & 2).

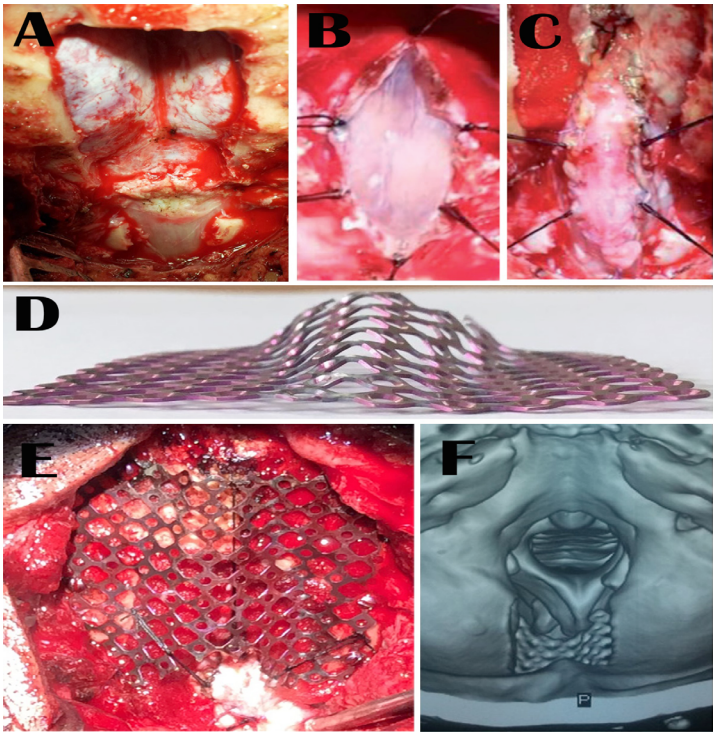

Figure 1: Peroperative picture showing the craniectomy and removal of C1 arch (1A), midline linear arachnoid preserving durotomy with the anchoring of the dural cut margin for tacking (1B), duraplasty with the superficial layer of the deep cervical fascia (1C), the pre-shaped 5 cm X 5 cm titanium mesh shaped like the canopy with flat wings of a Stealth bomber (1D), fixation of the cranioplasty along the margins of the craniectomy with screws and tacking of the duraplasty with the titanium mesh (1E), and postoperative 3D CT scan showing the enlargement of the posterior margin of the foramen magnum (1F).

Radiological evaluations were performed by comparing the preoperative and 3-month postoperative CTs and MRIs. The measurements were done manually with the freely available Java-based public-domain image processing and analysis program developed at the National Institutes of Health (NIH), ImageJ software version 1.53e. Preoperative measurement of the FM diameter or plane can be easily done by drawing the McRae's line (McRL) in both CT and MRI by joining the basion and the opisthion. In SC, the postoperative McRL is drawn on the midsagittal CT bone windows from the basion to the new opisthion formed by the lower margin of the titanium cranioplasty. (Figure 2) which is pretty straightforward as the new opisthion is created by the lower margin of stealth cranioplasty which can be well detected in CT or MRI images. However, in cases of PFD and PFDD, where the posterior bony margin of the foramen magnum is deficient, we adopted a novel technique. A straight line is drawn in the preoperative MRI along the dorsal margin of the clivus down to the basion which corresponds with the Wackenheim's clivus canal line (WCCL). A line from the basion on the WCCL to the opisthion is drawn to mark and measure the McRL which in turn is the antero-posterior (AP) diameter of the foramen magnum. The angle formed between the WCCL and the McRL at the basion is the Boogard's angle (BooA). The BooA is a fixed angle for any individual at any given time as the clivus, the basion, and the opisthion are fixed bony structures. The BooA, measured in the preoperative MRI in every individual CM1 patient is used as the future reference angle to draw and measure the McRL in the postoperative MRI where the opisthion would be missing. As the clivus and basion are the same for any particular individual both preoperatively and postoperatively, the WCCL and basion would be the same as in preoperative MRI, and fixing the BooA on the WCCL at the basion would give the exact trajectory for the McRL. In the postoperative MRI, the BooA is fixed on the WCCL at the basion as measured from the preoperative MRI, and the McRL is drawn by extending the line from the basion up to the posterior-most dural margin marked by the posterior-most margin of the CSF on that line. (Figures 3 & 4).

The descent of the tonsil was measured by drawing a vertical line from the tip of the tonsil on the McRae's line both in preoperative and postoperative MRIs (Figures 3 & 4). Change in syringomyelia diameter was measured by measuring the maximum AP diameter of the SM in the preoperative midsagittal T2WI of MRI and by measuring the AP diameter at the corresponding level in the postoperative T2WI MRI (Figures 3,4&5). The status of cisterna magna was noted in both pre and postoperative T2WI MRIs as well as the in the MR myelograms whenever that was available (Figure 5).

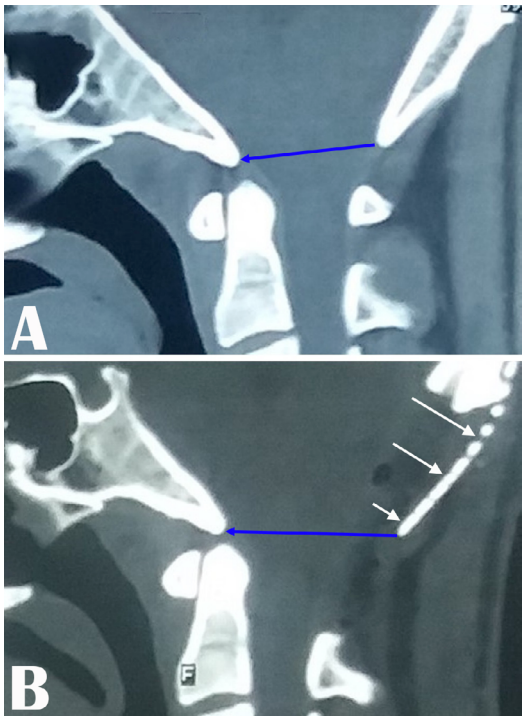

Figure 2: Sagittal CT bone window demonstrating the preoperative (A) and postoperative (B) diameter of the foramen magnum. The enlargement of the newly reconstructed posterior margin of the foramen magnum by titanium mesh in Stealth cranioplasty (B) compared to the preoperative state (A) can be seen well (Blue arrows). The enlarged space of the posterior fossa in the postoperative scan (B) can also be appreciated (White arrows).

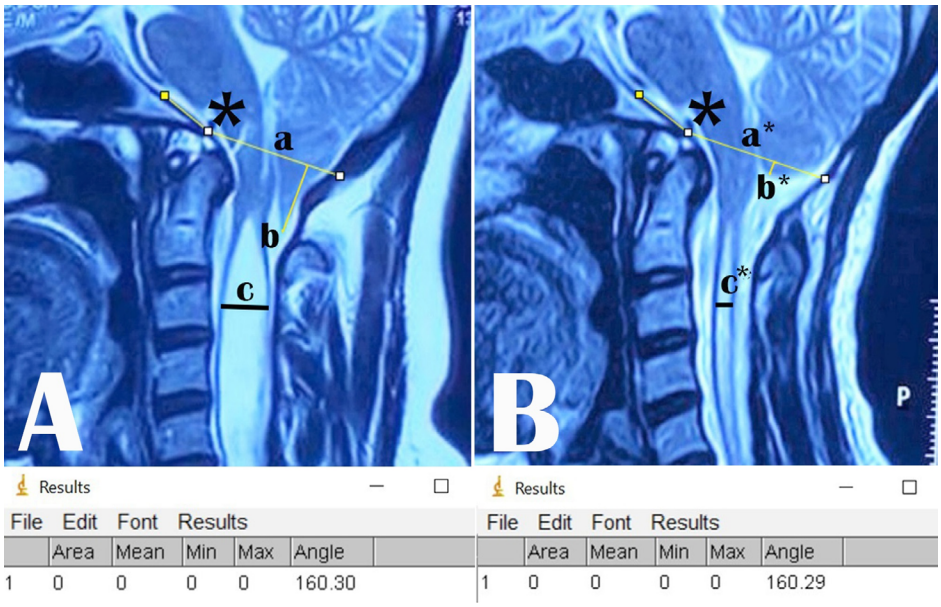

Figure 3: Preoperative (A), and postoperative (B) T2WI MRI following PFDD showing the fixed McRae's line (a & a*), Boogard's angle (big asterisk) with changes in tonsillar ectopia (b & b*), and changes in syrinx diameter (c & c*). The measurements of the Boogard's angle by ImageJ are in the lower panel.

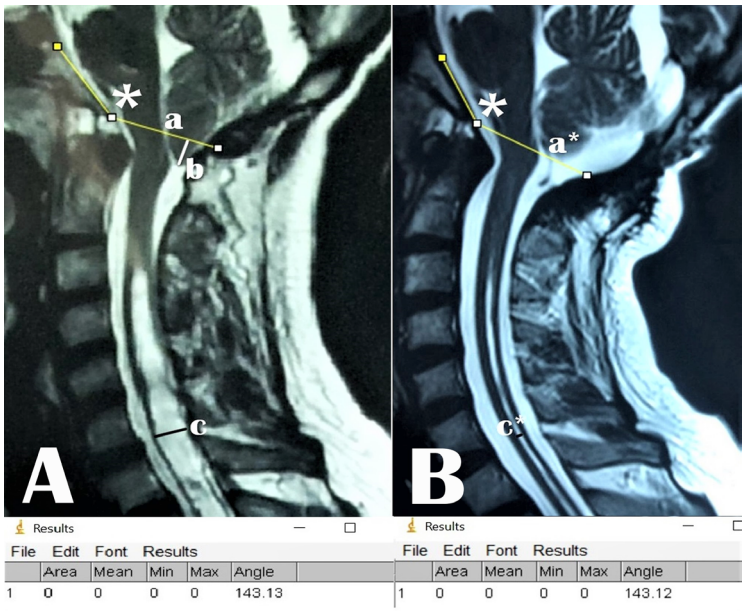

Figure 4: Preoperative (A), and postoperative (B) T2WI MRI following SC showing the fixed McRae's line (a & a*), and Boogard's angle (big asterisk) with changes in tonsillar ectopia (b), and changes in syrinx diameter (c & c*). The measurements of the Boogard's angle by ImageJ are in the lower panel. Note that postoperatively the tonsils ascended beyond the McRae's line.

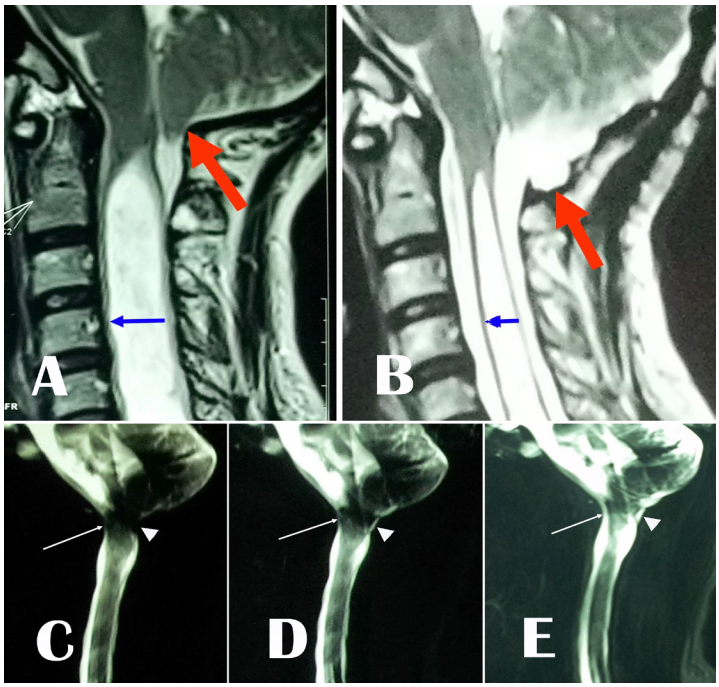

Figure 5: Preoperative (A) and postoperative (B) T2WI MRI shows the appearance of cisterna magna (Red arrows) and reduction of the syrinx (Blue arrows). Preoperative (C), immediate postoperative (D), and late postoperative (E) MR myelograms showing the preoperative absence (C) and gradual postoperative appearance (D & E) of cisterna magna (Arrowheads). The gradual changes in the CSF column anterior to the spinal cord can also be appreciated (White arrows).

Statistical Analysis

Parametric data were expressed as mean ± SD and compared via unpaired t-test. Non-parametric data were expressed as medians and compared via the Chi-square test and McNemar's test. All statistical analysis was performed using SPSS 23 software. A p-value <.05 was considered to be statistically significant.

Results

Overview

This study was conducted on 53 adult symptomatic CM1 patients with or without SM where 11, 19 and 23 patients underwent PFD, PFDD, and SC respectively. The average age of the 37 males and 16 females recruited in the study ranged from 18 to 47 years with a mean age of 30.35±7.49 years. Most of the patients had preoperative and postoperative CT and/or MRI scans to assess radiological outcomes.

Preoperative Data

The preoperative mean AP diameter of the FM (length of McRL) was 30.01±2.70 mm ranging from 22.92 to 35.57 mm. Preoperatively the TE ranged from 5.70 to 14.92 mm with a mean of 9.50±2.91 mm. 51 out of 53 patients had SM (syringomyelia was absent in 2 patients in the PFDD group) which ranged from 2.15 to 13.66 mm in diameter at the widest with a mean diameter of 6.49±2.56 mm. Only 2 patients had the presence of cisterna magna in their preoperative MRIs.

Postoperative Data

The patients were followed up at 3 months postoperatively. Fifty patients in both groups out of total 53 (94.4%) patients had an increase in FM diameter. Measured in millimeters, the changes in FM diameter were significantly better in the SC group than in the changes the PFD (p<0.001), and the PFDD (p<0.001) groups. The mean change in all 3 groups combined was 4.11 ± 2.75 mm while it was 2.46 ± 1.98 mm, 2.89 ± 2.36 mm, and 5.90 ± 2.36 mm in the PFD, PFDD, and SC groups respectively (Table 1). However, a decrease in AP diameter of the FM was observed in 1 patient in the PFD group and 2 patients in the PFDD group. Changes in postoperative TE in millimeters, compared to preoperative ones, the average tonsillar ascent in PFD, PFDD and SC groups were 2.43 ± 0.96, 2.67 ± 1.27 and 5.35 ± 1.91 millimeters respectively and all the patients combined in all 3 groups showed postoperative tonsillar ascent with a mean of 3.78 ± 2.05 mm. The changes were significant when compared between both SC and PFD (p.<0.001), and SC and PFDD (p.<0.001) groups (Table 2). Postoperatively the syrinx diameter changed by 1.90 ± 1.00, 2.05 ± 1.33 and 3.43 ± 2.41 millimeters in the SC, PFD and PFDD groups respectively. Measuring the syrinx diameter pre and postoperatively in millimeters, the changes were observed to be significantly better in the SC group than the PFD (p=0.014) and PFDD (p=0.032) groups. (Table 3) Preoperatively there was no syrinx in 2 patients in the PFDD group. Preoperatively, none of the patients in PFD and PFDD groups had the presence of CM while only 2 patients in the SC group had very small CMs. Postoperatively, the development of CM could be appreciated in only 1 (9.1%) in the PFD group and in 4 (21.1%) in the PFDD group. In the SC group, 14 (66.7%) patients developed CM postoperatively. The appearance of post-operative CM was statistically significant as a whole (p <0.001) and considering the individual groups, it was significant only in the SC group (p <0.001) (Table 4).

Surgery	Foramen Magnum Diameter (in mm)			p-value		
	Preoperative	Postoperative	Change	A vs B	A vs C	B vs C
PFD (A)	29.07 ± 2.41	31.54 ± 2.55	2.46 ± 1.98	0.620	<0.001	<0.001
PFDD (B)	30.59 ± 2.84	33.49 ± 2.72	2.89 ± 2.36			
STEALTH (C)	29.99 ± 2.71	35.90 ± 2.89	5.90 ± 2.36			
Total	30.01 ± 2.70	34.13 ± 3.20	4.11 ± 2.75	-		

Unpaired t test was done

Table 1: Change in Postoperative Diameter of Foramen Magnum in millimeters (mm).

Surgery	Tonsillar Ectopia (in mm)			p-value		
	Preoperative	Postoperative	Change	A vs B	A vs C	B vs C
PFD (A)	8.32 ± 2.58	5.90 ± 3.01	2.43 ± 0.96	0.579	<0.001	<0.001
PFDD (B)	9.26 ± 2.47	6.59 ± 2.27	2.67 ± 1.27			
STEALTH (C)	10.28 ± 3.28	4.93 ± 3.60	5.35 ± 1.91			
Total	9.50 ± 2.91	5.72 ± 3.09	3.78 ± 2.05	-		

Unpaired t test was done

Table 2: Postoperative tonsillar Ectopia in millimeters (mm).

Surgery	Change in Syrinx Diameter (in mm)			p-value		
	Preoperative	Postoperative	Change	A vs B	A vs C	B vs C
PFD (A)	5.90 ± 1.66	4.00 ± 1.22	1.90 ± 1.00	0.755	0.014	0.032
PFDD (B)	5.98 ± 2.46	3.93 ± 1.94	2.05 ± 1.33			
STEALTH (C)	7.20 ± 2.90	3.69 ± 2.20	3.43 ± 2.41			
Total	6.49 ± 2.56	3.83 ± 1.91	2.61 ± 1.94	-		

Unpaired t test was done

Table 3: Postoperative Change in Syrinx Diameter in millimeters (mm).

Surgery	Preoperative	Postoperative	p-value
PFD (A)	0/11 (0.0%)	1/11 (9.1%)	1.000
PFDD (B)	0/19 (0.0%)	4/19 (21.1%)	0.125
STEALTH	2/23 (8.7%)	14/21 (66.7%)	<0.001
Total	2/53 (3.8%)	19/51 (37.3%)	<0.001

McNemar test was done

Table 4: Preoperative and postoperative status of Cisterna magna (CM).

Discussion

The decision of managing CM1 patients surgically depends greatly on presentation as well as meticulously judged clinical and radiological findings. In cases of surgery, PFD or PFDD along with the removal of the C1 posterior arch are the most practiced surgical techniques with the goal to decompress the neural structures and restore normal CSF flow and dynamics around the CVJ. Variations with modifications and combinations of different strategies in addition to PFD and PFDD are also well in practice [2,3,8,12-15]. MRI is the imaging modality of choice for assessing radiological outcomes. Postoperative increase or decrease of foramen magnum diameter, tonsillar ectopia or diameter of the syringomyelia, and appearance of cisterna magna can be well evaluated by MRI [2]. Overall, the rate of postoperative radiological improvement can be as high as 81.1% [13]. Despite different parameters and techniques to measure the radiological outcomes of the surgery for CM1, none has been absolute and researchers have used and modified them according to their convenience. We developed a novel technique to measure some parameters to evaluate the radiological outcomes of surgery of CM1. During the marking and measuring the McRL in our technique an interesting finding was that the Boogards angles were obtuse in most of the cases in all groups which can be a good topic for future research.

The AP diameter as well as the plane of the foramen magnum extends between the basion and the opisthion and can be drawn by joining these on the midsagittal plane of the images which corresponds with McRae's line [16, 17]. Postoperatively, due to the deficient posterior margin of the FM, McRL is generally difficult to draw, while with our technique, the McRL can be easily and reliably drawn and measured. The foramen magnum AP diameter varies between different studies owing to heterogenous study population. Some studies found the AP diameter of the FM larger [18,19] and some found it narrower [20] in CM1 patients than in the normal population. However, no significant difference in AP diameter of the FM in CM1 and the normal population was found in most of the studies [21-24]. We found the AP diameter of the FM slightly smaller than the lower range of most of the studies. This might be due to the ethnic difference of the population in our study. Generally, the people of our geographical area are of shorter stature which might be a reason for the smaller AP diameter of the FM of our study population from an anthropological point of view.

The significant postoperative increase in AP diameter of the FM in SC than PFD and PFDD most likely was due to persistent maintenance of the posterior margin of the newly formed foramen magnum by the Stealth cranioplasty. This support is lacking in cases of PFD and PFDD resulting in filling the decompressed space by muscle bulk and fibrosis causing narrowing of the FM diameter again. Owing to the lack of any effective tool to measure postoperative AP diameter of the FM in absence of the posterior margin the postoperative, changes in foramen magnum diameter and plane have not been evaluated before and literature related to this is scarce. The technique we formulated is an effective technique to evaluate the postoperative FM diameter as well as marking the plane to appraise the status of TE. For the measurement of the preoperative tonsillar ectopia most of the studies used the McRae's line as the reference line [25-27]. However, for the postoperative measurements, various techniques have been applied which do not have much uniformity as different reference points are used pre and postoperatively and measurements of postoperative TE is inconsistent in the literature. For instance, one study described the extent of postoperative change of the tonsils, but the technique of measurement was not clearly stated [28]. Another study measured the changes in tonsillar herniation using several reference lines concurrently like the McRL, a second line drawn through the synchondrosis of the second cervical vertebra (C2), and another line drawn parallel to the lower endplate of the C2. All 3 lines were used for preoperative measurements and only the latter 2 for the postoperative measurements in the MRI [29]. In our technique of measurement, we fixed some constant reference points and lines which give the same scale of measurement both in pre and postoperative MRIs. This makes the measurement of the TE substantially consistent. The TE can be measured easily and reliably by drawing a vertical line on the McRL from the tip of the tonsil on both the images. Whatever the procedure was followed to measure the tonsillar ectopia in the literature [3,18,27,28,30] our findings match with the range of other studies. However, with different techniques of postoperative measurements, changes in tonsillar ectopia varied greatly depending on the measuring techniques and considerable inconsistencies are observed. Interestingly, in one study, the change in tonsillar ectopia was 3.8 mm when measured by the line through the C2 vertebral endplate and 5.4 mm when measured through C2 synchondrosis in the same study population [29]. The differences in measurements in the same set of patients were most likely due to changes in dimensions of the tonsils and the vertebral column in flexion and extension of the neck during scanning as patients cannot be placed in the exact same position for pre and postoperative scans. This is how, our technique proves to be better than the others as each of the WCCL, the basion, and the BooA is constant at any given situation. However, there is still chance of discrepancy in measuring TE owing to possible movements of the tonsils with respiration and cardiac cycle.

The better reversal of tonsillar ectopia in SC than PFD and PFDD can be attributable to comparatively more changes in AP diameter of the FM and better development of the CM with better maintenance of the both. by the cranioplasty. Additionally, the postoperative augmentation in FM diameter and posterior fossa volume helped the tonsils to pull in to the voluminous posterior fossa by the thrust of CSF from below. Furthermore, bigger CM with more CSF keeps the tonsils floating by the buoyancy of CSF. Syringomyelia associated with CM1 is a mysterious issue from its aetiological perspective and many modalities of treatment regimen continue to be practiced with widely varying results. Different studies reported occurrences of syringomyelia in CM1 operative series varying from 20% to 100% in adult preoperative MRIs [3,28-30] The presence of high percentage of syringomyelia in our series could be because of the late presentation of the patients. Most of the patients in this part of the globe typically ignore the early symptoms typically due to lack of awareness, illiteracy or financial constraints and do not consult a physician until the symptoms are debilitating. Another possibility is the ethnic difference of the study population where the smaller FM diameter might play some role in disparity of CSF dynamics across the CVJ to form an early onset SM.

Changes in SM diameter is also highly variable and unfavourable outcome is not infrequent. Various series described postoperative improvement of the SM ranging between 39 and 100% [2,8,13,29,30]. Postoperative worsening of SM ranging between 5.72% and 9.1% have also been described [14,15,29]. No single surgical technique has been found to fare better than the other in SM reduction [14,27]. However, the patients that had

dural opening did better in SM reduction than the patients who did not have a dural opening [13,15]. All the patients having SM in our series had some resolution of the SM. It is worth noting that the follow-up period was only 3 months postoperatively and longer follow-ups might have shown better results. The better SM resolution in the SC group was conceivably due to the better reversal of the CSF dynamics which resulted from creation and maintenance of a larger posterior fossa, wider foramen magnum, and bigger cisterna magna by keeping the soft tissue away with the support of the SC from the craniectomy site. All these together created more chance to have a near normal equilibrium of the CSF circulation. Although we could not do any postoperative CSF flow study, the better resolution of SM and other changes in cases of SC demostrate indirect evidence of better restoration of CSF dynamics and flow around the CVJ.

The absence of cisterna magna is one of the important key features other than the TE and the presence of SM in CM1. The imbalance of CSF dynamics between the cranial and spinal CSF compartments is heralded by the absence of CM in CM1 patients. Hence, reestablishment of CM in the postoperative CM1 patients can be a good sign of therapeutic success. Compression or absence of CM in preoperative MRI have been described in the literature having a range between 55% and 100% [31-33]. One important goal of surgery for CM1 is to reestablish CSF flow around the CVJ by forming or augmenting the CM and several procedures have been described in doing so. Various studies achieved 36.4% to 100% success in creating or augmenting the CM in postoperative MRIs [34-39].The low percentage of development of CM postoperatively in our series, falls in the lower range of the description of the literatures. However, individually, postoperative development of CM was seen significantly higher in number in the SC group. This can be attributed to the fact that in SC, not only the CM is reconstructed and enlarged by duraplasty, but is also maintained using cranioplasty and tenting. Longer follow-ups might have shown better result.

Conclusion

For symptomatic adult Chiari malformation 1, Stealth cranioplasty is a good option relating to some postoperative radiological outcomes. The novel technique that we introduced to evaluate postoperative radiological changes in CM1 patients is a unique, easy and convenient one. We believe, it will be helpful to other researchers to measure the radiological outcomes in a uniform manner. This technique can also pave way for future researchers in discovering newer dimensions of pathophysiology to treat the CMI patients better.

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