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DYNAMIC CSF MRI FINDINGS IN THE DIAGNOSIS AND FOLLOW-UP OF ARNOLD CHIARI TYPE 1

ARNOLD CHİARİ TİP 1 TANI VE TAKİBİNDE DİNAMİK BOS MRG BULGULARI

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ABSTRACT

AIM: Chiari malformation is a spectrum of diseases that includes different subtypes, has a wide clinical presentation, and therefore has different follow-up and treatment methods. We aimed to the compare clinical scoring systems and cerebrospinal fluid (CSF) dynamics obtained by magnetic resonance imaging (MRI) in Arnold Chiari Malformation Type-1 (CM-1) patients in the preoperative and postoperative periods.

MATERIAL AND METHOD: Twenty-one patients with CM-1 who underwent CSF MRI between January 2016 and June 2020 were included. In addition, 21 healthy volunteers were evaluated. All the patients had undergone decompression surgery due to CM-1 related symptoms. The relationship between preoperative MRI findings and clinical findings of 21 patients was evaluated. Changes in clinical and imaging findings of 21 patients who underwent surgery were compared.

RESULTS: Measurements were made just below the level of the cerebellar tonsil. CM-1 patients had higher preoperative peak positive velocity anteriorly (PPV: 6.26±2.72 vs 3.89±1.74 cm/s; p=0.001) and peak negative velocity anteriorly (PNV: -7.45±3.36 vs -3.61±2.65 cm/s; p=0.001) than healthy volunteers. Preoperative net flow in the posterior subarachnoid space of CM-1 patients was lower than healthy volunteers (p=0.017). Aliasing of the reverse flow was seen in 13(62%) patients. PPV and PNV were decreased after surgery (PPV: 6.26±2.72 vs 4.73±1.94 cm/s, p=0.017; PNV: -7.45±3.36 vs -4.97±1.97, p=0.005). Net Flow was increased posteriorly (0.32 vs 2.21 ml/min; p=0.053). The postoperative response was inversely proportional to the degree of tonsillar herniation. As the Asgari score of the cases increases, the Peak Positive and Negative Velocity Anterior values also increase with a statistically significant correlation (p=0.003; p=0,032 respectively)

CONCLUSION: Our findings suggest that the determination of flow dynamics and morphology using MR imaging correlates with clinical findings and may be a useful tool in determining the need and timing of surgery.

Keywords: Chiari malformation • cerebrospinal fluid • phase-contrast MRI • craniocervical decompression • Asgari scoring

ÖZET

AMAÇ: Chiari malformasyonu farklı alt tipleri barındıran, geniş bir klinik prezentasyona sahip ve bu nedenle farklı takip ve tedavi yöntemleri olan bir hastalık yelpazesidir. Bu çalışmada Arnold Chiari Malformasyonu Tip-1 (CM-1) tanılı hastalarda preoperatif ve postoperatif dönemde manyetik rezonans görüntüleme (MRG) ile elde edilen beyin omurilik sıvısı (BOS) dinamikleri ve klinik skorlama sistemlerinin karşılaştırılması amaçlanmıştır.

GEREÇ VE YÖNTEM: Ocak 2016 ile Haziran 2020 tarihleri arasında BOS MRG yapılan CM-1 tanılı 21 hasta dâhil edildi. Ayrıca 21 sağlıklı gönüllü değerlendirildi. Tüm hastalara CM-1 ile ilişkili semptomlar nedeniyle dekompresyon cerrahisi uygulanmıştı. 21 hastanın preoperatif MRG bulguları ile klinik bulguları arasındaki ilişki değerlendirildi. Cerrahi uygulanan 21 hastanın klinik ve görüntüleme bulgularındaki değişiklikler karşılaştırıldı.

BULGULAR: Ölçümler serebellar tonsil seviyesinin hemen altından yapıldı. CM-1 hastalarının ameliyat öncesi pik pozitif ve negatif hızları (PPV-PNV) sağlıklı gönüllülerden daha yüksekti (PPV: 6,26±2,72 vs 3,89±1,74 cm/s, p=0.001; PNV: -7,45±3,36 vs -3,61±2,65 cm/s, p=0.001). CM-1 hastalarının posterior subaraknoid alandan yapılan ölçümde net debi değeri sağlıklı gönüllülere göre daha düşüktü (p=0.017). Hastaların 13'ünde (%62) ters akıma bağlı 'aliasing' görüldü. Ameliyat sonrası PPV ve PNV değerleri azaldı (PPV: 6,26±2,72'ye karşı 4,73±1,94 cm/s, p=0.017; PNV: -7,45±3,36'ya karşı -4,97±1,97, p=0.005). Net Akış ise arttı (0.32'ye karşı 2.21 ml/dak; p=0.053). Postoperatif yanıt, tonsiller herniasyon derecesi ile ters orantılı idi. Hastalarda Asgari skoru arttıkça, PPV ve PNV değerleri de artmaktadır (sırasıyla p=0.003 ve p=0,032)

SONUÇ: Bulgularımız, MR görüntüleme kullanılarak elde edilen akış dinamikleri ve morfolojinin klinik bulgularla ilişkili olduğunu, cerrahi ihtiyacı ve zamanlamasını belirlemede yararlı bir araç olabileceğini düşündürmektedir.

Anahtar Kelimeler: Chiari malformasyonu • beyin omurilik sıvısı • faz kontrast MRG • kranioservikal dekompresyon • Asgari skoru

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INTRODUCTION

Chiari malformations are a wide range of diseases that involve an anomaly of hindbrain formation during the development of the central nervous system. Hans Chiari, in his first article on Chiari malformations, described four different types according to the brain structures displaced from the intracranial area. Arnold Chiari malformation type I (CM-1) is defined as a 5 mm or more inferior extension of the cerebellar tonsils from the level of the foramen magnum (FM) (1).

In radiological studies conducted for different reasons, CM is seen at a rate of approximately 1%. Most of them (90%) are asymptomatic and there is not enough information about the long-term natural course (2). The degree of tonsillar herniation shown by imaging alone in Chiari I malformation is not sufficient to explain the severity of clinical symptoms and does not directly correlate with symptoms (3).

In addition, the timing and criteria of surgical intervention in the treatment of Chiari I malformation are controversial. The adequacy of surgical treatment can be followed by the incomplete recovery of symptoms and signs after surgery, or it can be measured by the reflection of clinical relief on imaging (4).

Therefore many studies on CM1 have focused on cerebrospinal fluid (CSF) dynamics (4-6). The CSF volume (mL/min) that moves up or down the foramen magnum is associated with the changing cerebral blood volume during the cardiac cycle. The CSF flow velocity (in cm/s) at the foramen magnum level depends more on the CSF distance and the shape of this area. Any condition that restricts passage within the foramen magnum affects the flow volume and flow velocity (7). Consequently; tonsil herniation, foramen magnum volume, or interventions affecting foramen capacity are expected to affect CSF volume and peak CSF velocity.

In the literature, the altered CSF flow kinetics in Chiari malformation causes signs and symptoms of the disease (8). Phase-contrast Magnetic Resonance (PC MR) studies show the jet flows at the foramen level, heterogeneity in CSF motion, and bi-directional CSF flow. PC MR is also used to demonstrate changes in CSF flow dynamics, CSF flow patterns, and quantitative measurement during each cardiac cycle.

In this study, we evaluated the differences in CSF flow dynamics in the subarachnoid spaces (SAS) in the anterior cervical and posterior cervical, normal control subjects, and patients with CM-1 before and after surgery on axial MR imaging. We aimed to investigate the differences of cerebrospinal fluid (CSF) dynamics obtained by MR imaging in Chiari Malformation Type-1 patients at the preoperative and postoperative period and its relationship with clinical scoring systems.

Abbreviations

CM-1: Chiari malformation type I
CSF: Cerebrospinal fluid
PC MR: Phase contrast Magnetic Resonance
FM: Foramen magnum

MATERIAL AND METHOD

Study Population and Definitions

This retrospective study was carried out in a tertiary care center. Medical records of the hospital for the timeline from January 2016 to December 2020, were reviewed retrospectively.

We noted clinical presentation, neuroimaging findings, surgery, postoperative data, and outcome. Prior to data collection, the Institutional Review Board at our hospital approved the study (Decision number: 2020/0016).

Inclusion criteria were determined as compliance with the following criteria: 1. The MRI, including Phase Contrast sequences, of the patient must be taken at the center of conduct of this research. 2. Sufficient image quality to allow flow dynamics, 3. Tonsillar herniation length 5 mm or more, 4. Patients who have undergone or planned decompression surgery. Criteria for exclusion were set as 1. Patients operated at the research center, but imaging was done in another center, 2. Pediatric population.

The 21 patients diagnosed with Chiari-1 and 21 healthy control groups who underwent CSF MRI were evaluated. **(Figure 1)** All volunteers in the control group declared that they were in good health and had no current or previous spinal or neurological problems. All the patients underwentdecompression surgery due to Chiari malformation.

Clinical information and scores were recorded on a worksheet by the surgical team who were blind to the radiology data. The Asgari score, which measures two different components as cranial nerve involvement and spinal cord dysfunction, was used to evaluate the clinical condition of the patients at the time of first admission (9).

MRI Protocol

MR imaging was performed with two different devices (1.5 Tesla GE Optima MR450w and MR360 (General Electric, Chicago, Illinois, USA)). The imaging protocol included the T1W and T2W scans and cine velocity encoded phase contrast scans, with low-velocity encoding (5-20 cm/s) for imaging the CSF flow at the foramen magnum region. Other typical imaging parameters were matrix 128 × 128, FOV 250 × 250 mm, TR/TE 300/80 ms, and slice thickness of 6 mm.

Phase Contrast Magnetic Resonance and Analysis

The axial section position was chosen as the foramen magnum in normal volunteers. For the patients, the section was placed just below the tonsils where the CSF flow was sufficiently visible. **(Figure 2)** CSF flow data were obtained along with the anterior and posterior subarachnoid space by region of interest (ROI) placement. **(Figure 3)**

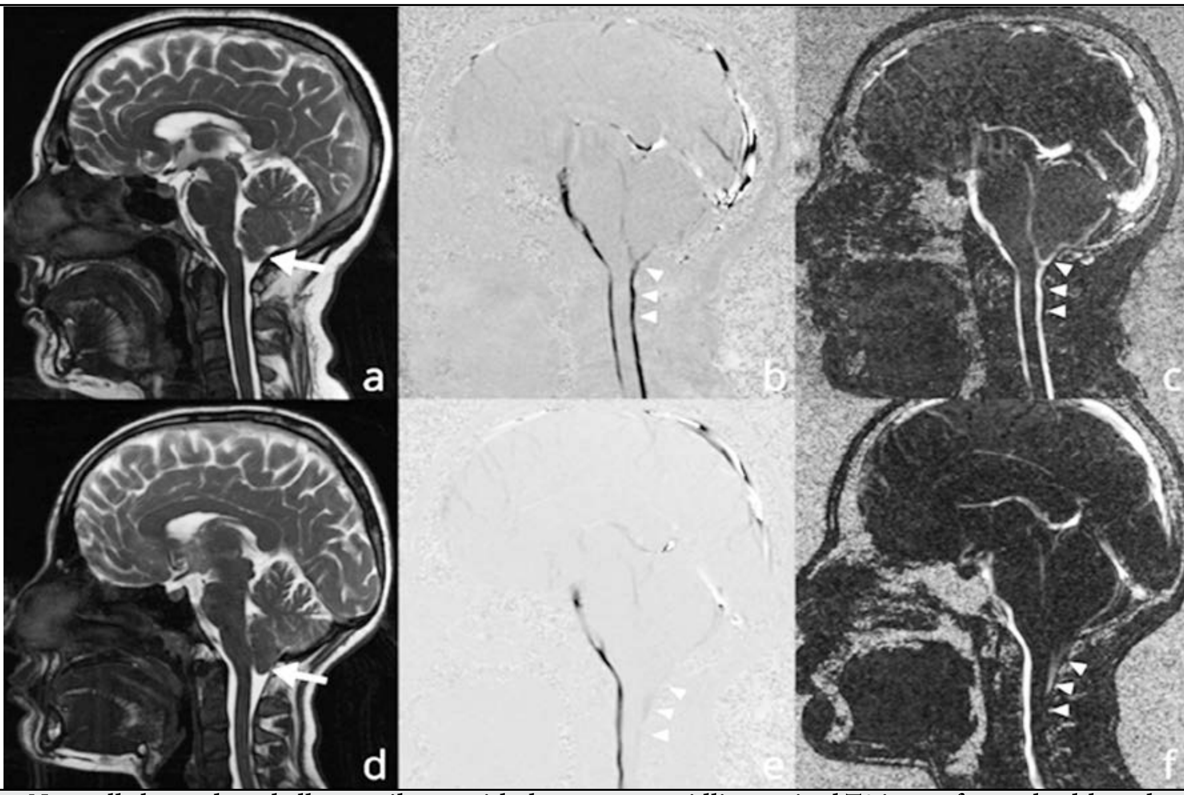

Fig 1. a. Normally located cerebellar tonsil seen with the arrow on midline sagittal T2 image from a healthy volunteer. b, c. Phase contrast and magnitude MRI images showing normal CSF flow marked with arrowheads. d. Caudally located cerebellar tonsil seen with the arrow on midline sagittal T2 image in Chiari 1 patient. e, f. Phase contrast and magnitude MRI images showing reduced CSF flow, marked with arrowheads posteriorly.

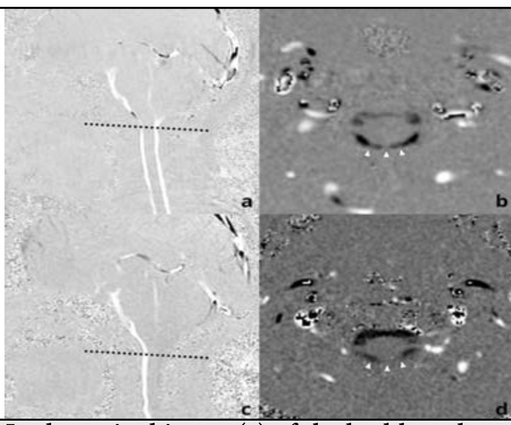

Fig 2. In the sagittal image (a) of the healthy volunteer, the level of the axial phase contrast (b) image taken just below the descending tonsil tip is marked with a dot-by-dot pattern. Normal flow in the posterior subarachnoid space at this level is shown with arrowheads. Similarly, the sagittal phase contrast (c) image and the level of axial phase contrast (d) are shown in Chiari 1 patient. Reduced flow in the posterior subarachnoid space at this level is shown with arrowheads.

Thirty-two image frames were regularly acquired with the wave from a fingertip electrode used for the pulse-gate.

Flow analysis was conducted with commercially available ReportCard software on the GE image processing station (AW VolumeShare 7). The software calculated the flow and velocity in each voxel in the region of interest.

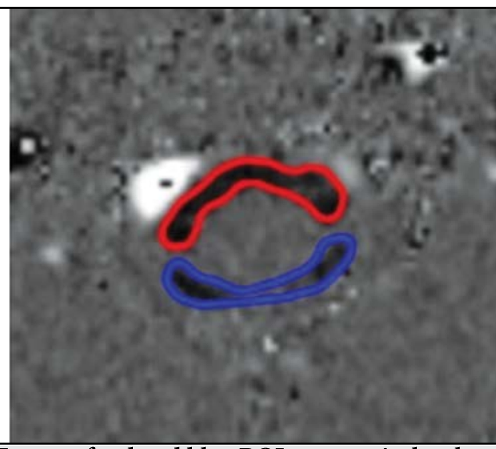

Fig 3. Image of red and blue ROIs, respectively, placed in the anterior and posterior subarachnoid spaces.

CSF dynamics were measured preoperatively and after 1 month to 1 year postoperatively to see the changes in CSF flow. The relation between preoperative MR findings and the clinic of all 21 patients included in the study was evaluated. The changes in clinical and imaging findings of 21 patients who have been operated on so far were compared.

Surgical Technique

Surgical Indications and Technique: After explaining the risks and benefits of the surgery, informed consent was obtained from the patients. Then we performed craniocervical decompression in symptomatic patients. The indications for decompressive CM surgery were symptoms like headache, myelopathic symptoms, paresthesias, syringomyelia, and scoliosis, which were consistent with published reports in similar populations (10,11). Standard decompression surgery was performed on CM-1 patients.

Follow-up and Outcomes

Neurological examination and radiological imaging were performed on the first postoperative day. The patients were followed up every three months for the first year, and then at annual intervals, adding neurophysiological tests if necessary. The outcome of surgery was evaluated by the improvement in the neurological findings of the patients and their lifestyles (12,13).

Statistical Analysis

Mean, standard deviation (SD), and the correlation coefficient were calculated for each group of patients and healthy subjects using SPSS software version 23 (IBM SPSS Inc., Chicago, Illinois, United States). Postoperative changes in CSF dynamic values of the patients were compared using the Wilcoxon Signed-Rank test. The relationship of dynamic CSF values with clinical scores was examined with Spearman's correlation analysis. Pearson chi-square test and Fisher's exact test were used to comparing qualitative data. The p-value of 0.05 was considered statistically significant.

RESULTS

Patient Characteristics and Clinical Presentation

Twenty-seven female and fifteen male participants were included in this study. The mean age at diagnosis was 37.8 years (range 18-65 years). The mean symptom duration of the patients included in the study ranged from 1 month to 9 years (mean: 24 months). The follow-up period was between 1 and 5 years. Suboccipital headache (95.2%), numbness in upper extremities (61.9%), disability in upper extremities (14.3%), disability in lower extremities (33.3%), loss of sensation (33.3%), difficulty swallowing (19.1%) were the most relevant clinical features. Other findings including syringomyelia and basilar invagination are summarized in **Table 1.**

MRI Findings

In the patients, it was found that the cerebellar tonsil herniated from the foramen magnum in the mean of 12.5 mm in midline sagittal MRI. There is a statistically significant positive correlation between the Asgari scale and tonsillar herniation (the Asgari scale increases as the herniation increases) ($p < 0.01$). As the Asgari score of the cases increases, the Peak Positive and Negative Velocity Anterior values also increase with a statistically significant correlation ($p=0.003$; $p=0,032$ respectively). There is a negative relationship between the Asgari scale and net flow, but it is not statistically significant. There was no correlation between other dynamic values of CSF and clinical score.

At the end of cerebellar tonsil level peak positive and negative velocity (PPV-PNV) of CM-1 patients were higher than healthy volunteers pre-surgery. Preoperative net flow in the posterior subarachnoid space of CM-1 patients was lower than in healthy volunteers ($p=0.017$). **(Table 2)** Preop 'Positive Pixel Flow Anterior' value of CM-1 patients were higher than control group also ($p < 0.01$).

PPV and PNV were decreased after surgery ($p=0.017$ and $p=0.005$ respectively). Positive and Negative Pixel Flow was decreased in the anterior segment ($p=0.005$ and $p=0.035$ respectively). **(Figure 4) (Table 3)**

Table 1. General descriptive characteristics and symptoms of the cohort

	Chiari-I N=21	***Control N=21***
Age (Median)	38 (18;56)	35 (18;67)
Gender:		
Female	11 (52.4%)	16 (76.2%)
Male	10 (47.6%)	5 (23.8%)
Symptoms:		
Duration[1]	1 month – 9 years	
Suboccipital headache	20 (95.2%)	
Numbness in upper extremities	13 (61.9%)	
Disability in upper extremities	3 (14.3%)	
Disability in lower extremities	7 (33.3%)	
Loss of sensation[2]	7 (33.3%)	
Difficulty swallowing	4 (19.1%)	
Asgari Score	3.7 (1;8)	
Additional findings:		
Syringomyelia	8 (38.1%)	
Basilar invagination	6 (28.6%)	

[1] Time between the onset of symptoms and admission to the hospital
[2] Cranial nerve involvement

The Net Flow was increased in the posterior segment postoperatively (p=0.053). While CSF flow was observed nonhomogeneous in the patient group, it was homogeneous in the control group.

Aliasing of the reverse flow was seen in 13 (62%) patients.

Table 2. CSF[1] dynamics of samples

Variable	*Normal*	*CM-1[2]*	*p*
Preoperative Tonsiles Herniation (mm)	-	12.61±4.7	
Velocity Anterior			
Peak Positive Velocity Anterior (Mean±SD[3], cm/sec)	3,89±1,74	6,26±2,72	**0.001**
Peak Negative Velocity Anterior (Mean±SD, cm/sec)	3,61±2,65	7,45±3,36	**0.001**
Velocity Posterior			
Peak Positive Velocity Posterior (Mean±SD, cm/sec)	3,31±1,21	4,02±2,22	0.421
Peak Negative Velocity Posterior (Mean±SD, cm/sec)	3,67±2,06	4,16±1,41	0.170
Flow			
Net Flow Anterior (Mean±SD, ml/min)	2,60±5,24	3,84±9,96	0.618
Net Flow Posterior (Mean±SD, ml/min)	3,67±4,71	0,32±3,97	**0.017**
Pixel Flow Anterior			
Positive Pixel Flow Anterior (Mean±SD, ml/min)	17,66±6,15	23,03±9,43	**0.001**
Negative Pixel Flow Anterior (Mean±SD, ml/min)	-15,05±5,60	-19,18±9,06	0.084
Pixel Flow Posterior			
Positive Pixel Flow Posterior (Mean±SD, ml/min)	12,42±5,89	9,59±5,18	0.106
Negative Pixel Flow Posterior (Mean±SD, ml/min)	-8,74±4,39	-9,26±6,68	0.767

[1] CSF: Cerebrospinal fluid
[2] CM-1: Chiari malformation type 1
[3] SD: Standart deviation

Table 3. Post-operative changes of CSF[1] dynamics

Variable	*CM-1 Pre[2]*	*CM-1 Post[3]*	*p*
Velocity Anterior			
Peak Positive Velocity Anterior (Mean±SD[4], cm/sec)	6.24±2.79	4.82±1.94	**0.017**
Peak Negative Velocity Anterior (Mean±SD, cm/sec)	7.53±3.43	5.11±1.92	**0.005**
Velocity Posterior			
Peak Positive Velocity Posterior (Mean±SD, cm/sec)	4.14±2.21	3.74±1.52	0.931
Peak Negative Velocity Posterior (Mean±SD, cm/sec)	4.26±1.37	3.39±1.33	0.131
Flow			
Net Flow Anterior (Mean±SD, ml/min)	3.55±10.13	2.04±6.14	0.533
Net Flow Posterior (Mean±SD, ml/min)	0.26±4.06	2.15±3.42	0.053
Pixel Flow Anterior			
Positive Pixel Flow Anterior (Mean±SD, ml/min)	23,03±9,43	18,24±6,01	**0.035**
Negative Pixel Flow Anterior (Mean±SD, ml/min)	-19,18±9,06	-15.896.91	**0.050**
Pixel Flow Posterior			
Positive Pixel Flow Posterior (Mean±SD, ml/min)	9,59±5,18	9,84±4,82	0.858
Negative Pixel Flow Posterior (Mean±SD, ml/min)	-9,26±6,68	-7,63±3,88	0.291

[1] CSF: Cerebrospinal fluid
[2] CM-1: Chiari malformation type 1, preoperative
[3] CM-1: Chiari malformation type 1, postoperative
[4] SD: Standart deviation

Table 4. CSF[1] dynamics and clinical scores

Variable	*Asgari Score*	
	r	***p***
Preoperative Velocity		
Peak Positive Velocity Anterior (Mean±SD[2], cm/sec)	‡0,452	***0,003***
Peak Negative Velocity Anterior (Mean±SD, cm/sec)	†-0,331	***0,032***
Peak Positive Velocity Posterior (Mean±SD, cm/sec)	‡0,017	*0,916*
Peak Negative Velocity Posterior (Mean±SD, cm/sec)	‡-0,113	*0,474*
Postoperative Velocity		
Peak Positive Velocity Posterior (Mean±SD, cm/sec)	-0,026	*0,912*
Peak Negative Velocity Posterior (Mean±SD, cm/sec)	0,232	*0,312*
Peak Positive Velocity Posterior (Mean±SD, cm/sec)	0,119	*0,608*
Peak Negative Velocity Posterior (Mean±SD, cm/sec)	-0,240	*0,294*
Preoperative Flow		
Net Flow Anterior (Mean±SD, ml/min)	†-0,080	*0,616*
Net Flow Posterior (Mean±SD, ml/min)	‡-0,281	*0,071*
Postoperative Flow		
Net Flow Anterior (Mean±SD, ml/min)	0,137	*0,552*
Net Flow Posterior (Mean±SD, ml/min)	-0,179	*0,438*

†r: Pearson Korelasyon Katsayısı ‡r: Spearman's Korelasyon Katsayısı
[1] CSF: Cerebrospinal fluid
[2] SD: Standart deviation

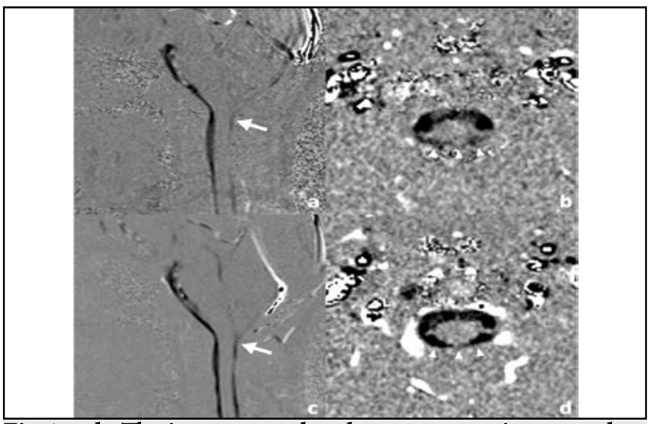

Fig 4. a, b. The images are the phase-contrast images taken from the patient in the sagittal and axial planes in the preoperative period (arrow and arrowheads). c, d. Images from the same patient show the relative change in CSF flow after decompression (arrow and arrowheads).

DISCUSSION

In this study, a correlation was shown between the degree of tonsillar herniation and symptoms, while not showing a significant relationship between the flow rate of CSF and the degree of symptoms. We found that the maximum CSF velocities were higher in CM-1 patients below the cerebellar tonsillar level compared to the control group (14). The positive and negative peak velocities decreased and net flow increased after craniocervical decompression surgery in CM1 patients. These results support the abnormal CSF flow pattern in CM1 patients (15-17).

In CM-1 patients, the maximum systolic and diastolic velocities were 6.26 ± 2.72 and 7.45 ± 3.36 (cm / s), respectively. These values are similar to those of Gholampour et al (5). However, many ranges below and above these values have been reported in the literature (6,7,18). The reason for this wide range can be thought to be that the level of measurement, the ROI area, and the devices are different. The net flow rate was found to be 0.32 ml/min in the patient group. Contrary to the variability of the CSF velocity, it is found that the velocity values after decompression decrease in almost all studies in the literature (15,19).

There is a statistically significant positive correlation between the Asgari scale and tonsillar herniation (the Asgari scale score increases as the herniation increases). As the Asgari score of the cases increases, the Peak Positive and Negative Velocity Anterior values also increase The reason for this is the difficulty of CSF flow in the posterior subarachnoid space in patients with increased tonsillar herniation. While this increases the symptoms, it also shifts the CSF flow anteriorly. With the widening of the posterior subarachnoid space after surgery, the net flow increased and approached normal. As a result of the posterior shift of the flow, the peak velocity values decreased and approached normal in the anterior.

Statistically significant changes and a correlation were found in clinical scores and CSF MRI flow values before and after surgery. This is the first study to be conducted in this context, aiming to reconcile the quantitative MR findings with the quantitative values obtained by examination.

Our most important limitation was that the measurement location and technique of CSF dynamics differed between studies. The retrospective clinical scoring and the limited number of operated patients are other weaknesses.

CONCLUSION

Today, the presence and severity of signs and symptoms are decisive for surgical intervention in Arnold Chiari malformation type I. MRI, on the other hand, plays an increasingly effective role in patient management, as in many other diseases. Our findings suggest that the determination of flow dynamics and morphology using MRI findings correlates with clinical findings and may be a useful tool in determining the need and timing of surgery.

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