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Investigation of Correlations Between Joint Health, Functionality, and MRI Score in Hemophilic Arthropathy of The Elbow Joint: Cross-Sectional Study

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ABSTRACT

Hemophilic arthropathy (HA) of the elbow joint should be examined separately from the knee and ankle because of the anatomical and biomechanical differences of the elbow. The aim is to investigate possible correlations between age, range of motion (ROM), muscle strength, upper extremity functions, joint health and radiological findings.

Twenty-seven joints of 20 patients aged 11-30 years with findings of HA in elbow were evaluated. International Prophylaxis Study Group Magnetic Resonance Imaging (IPSG MRI) score was used in the radiological evaluation. In physical evaluations, ROM and muscle strength were measured by goniometer and digital dynamometer, respectively. Joint health was evaluated with Hemophilia Joint Health Score-Elbow Point (HJHS-EP) and upper extremity functionality with Quick-Disability of Arm Shoulder and Hand (Q-DASH).

The age was strongly correlated with HJHS-EP, loss of extension and pronation ROM and moderately correlated with IPSG MRI score. HJHS-EP showed strong correlation with loss of extension. There were a moderate correlation between Q-DASH and muscle strength of the elbow. The IPSG MRI scores were not significantly correlated with physical examinations.

The elbow joint should be evaluated both radiographically and physically and these assessments cannot be alternatives to each other. Even goniometric assessment of elbow extension alone may provide important information about joint health. Functionality can be improved by increasing muscle strength. Since the deterioration in physical and radiographic examinations with age, it is recommended to conservatively or surgically treat of the elbow joint with physiotherapy at early ages.

Keywords: Hemophilia, elbow joint, joint health, arthropathy, functionality, MRI

Introduction

The elbow joint is prone to recurrent hemarthrosis due to anatomical and biomechanical complexities (1), especially adolescents and young adults (2). Recurrent hemarthrosis restricts the functionality of the upper extremity as it causes pain, muscle weakness, instability, loss of range of motion (ROM) and nerve compression (1). These progressive changes in the musculoskeletal system are seen as the most important cause of morbidity in hemophilic arthropathy (HA) (3). Outcome measures evaluating joint functions and structures in HA are becoming increasingly common in the holistic care of hemophilia (4). A specific detailed evaluation of the joint is required to determine the stage and treatment of HA (3, 5).

Clinical studies are mostly focused on HA of the lower limb joints due to its importance in weight-bearing and mobility (6-10). However, the functionality of the upper extremity and participation in labor has become current issues with increased life expectancy in hemophilia. Heim et al. stated that the hypertrophy of the radial head affects the functionality by causing impingement on the proximal facet and especially

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restriction in supination (1). However, Cuesta-Barriuso et al. reported that early limitations in ROM (flexion-extension) rarely affect functionality because the elbow does not a weight-bearing joint (11). More studies are needed to evaluate the functionality in HA of the elbow joint.

In the guideline for the management of hemophilia, it was emphasized that hemophilia joint health score (HJHS) can be used in physical evaluation and magnetic resonance imaging (MRI) and ultrasound (US) can be used in radiological assessment (12). Poonnose et al. reported that clinical (HJHS) and radiological assessment provide complementary information and should be considered together in the evaluation of joint arthropathy (13). Even if the patient with hemophilia (PwH) does not show any problems on physical evaluation, new signs of HA can be detected by radiological assessment (14). MRI and US are used to detect early soft-tissue abnormalities and osteochondral changes in the joints. Recent studies in the literature have reported that the US is reliable in evaluating the soft tissue abnormalities and osteochondral changes (14, 15). Despite being the trend-topic and advantage of the US, MRI was preferred in our study. Because it is more sensitive to detect soft tissue abnormalities that can be observed at an early age (16) and is useful for analyses of early and moderate stages of HA (17).

In the studies which evaluate HA of the elbow joint, possible relationships between joint status, functionality and MRI score have been relatively under-explored. In addition, it may be important to determine the relationship between parameters where detailed physical examinations cannot be performed adequately. The aim of this study was to investigate the correlation between age, elbow ROM, muscle strength, joint health, functionality and radiological score in adolescents and young adults with HA of the elbow and thus to guide clinicians in physical examination. Our study hypothesized that there is a correlation between joint status, functionality, and MRI scores.

Material and Methods

In this cross-sectional study, 27 elbow joints were evaluated in 20 PwH aged between 11-30 years. This study was performed at Van Yuzuncu Yil University between January 2020 and August 2020. The pain parameter in the study conducted by Cuesta et al. was used as a reference to determine sample size and was found that to be at least 16 PwH (18). Ethics committee approval was obtained. Written and verbal informed consent was received from participants and the parents of PwH age under 18 years. An inclusion criterion was the history of at least one major hemarthrosis in the elbow in the last year. Exclusion criteria were; any frequent bleedings in the shoulder or wrist joints in the same upper extremity; any surgery that may affect upper extremity functionality; and any bleeding in the upper extremity in the last two weeks.

Evaluations: Participants' age, height, weight and hemophilia type were recorded. Physical evaluations were performed by the two non-blind physiotherapists experienced in hemophilia. A universal goniometer was used to measure elbow flexion, extension, supination, and pronation ROM. ROM measurements were made according to anatomical references described American Academy of Orthopaedic Surgeons (19). Biceps and triceps muscle strengths were measured with a digital dynamometer. Muscle strength was evaluated in sitting position, forearm at 90 degrees flexion, and supination. The patient was asked to apply force in the opposite direction of movement (20). Measurement was performed three times at one-minute intervals and the average strength was recorded as pound.

Physical joint assessment is easily accessible and inexpensive, often used to measure structural damage and functionality. HJHS version 2.1 was used to evaluate the joint status (21). Knee, elbow, and ankle joints were evaluated bilaterally. Subheadings of HJHS such as swelling, duration time of swelling, muscle atrophy, crepitation, loss of flexion, loss of extension, joint pain and muscle strength were given point according to the own criteria of HJHS. The maximum score per index joint is 20 points and it indicates the worst joint health. The validity and reliability study of HJHS was conducted in adolescents and young adults (22). We used elbow point of HJHS (HJHS-EP) to evaluate the joint status in HA of the elbow joint.

Quick Disability Arm Shoulder and Hand (Q-DASH) is a specific questionnaire that evaluates the functionality of the upper extremity. The Turkish version of the Q-DASH questionnaire was used to evaluate upper extremity functionality in this study (23). The Q-DASH consisted of 11 questions and a is 5-point Likert scale was used (1=no difficulty, 2=mild difficulty, 3=moderate difficulty, 4=severe difficulty, 5=unable). The total score is calculated according to the formulas given by the questionnaire and was a score

Fig. 1. 2.0 T gradient echo T2- weighted images in the coronal plane of a 20-year-old boy with severe hemophilia. This patient's HJHS was 13, IPSG MRI score was 10 (7+3) and the soft tissue MR score was compatible with HJHS. A: shows surface erosions and subchondral cysts in radial head, B: shows joint effusion (high fluid signal; small arrows) and reveals haemosiderin-laden synovial hypertrophy (large arrows)

between 0-100. A score close to 0 indicates better functionality and symptoms (24).

Twenty-seven elbow joints were imaged in a short protocol based on T2* weighted gradient echo sequences (Fig. 1) in two or three planes (25). MRI scans of PwH were evaluated twice by the same orthopedist, at different times and scored according to IPSG (International Prophylaxis Study Group) MRI scale. This scale is a HA-specific scale that assesses the osteochondral changes and soft tissue abnormalities of the joints (25). When determining the soft tissue score, effusion/hemarthrosis, synovial hypertrophy and hemosiderin are evaluated. The highest score that can be obtained is 9 points and indicates soft tissue abnormalities. Surface erosions, subchondral cysts, and the cartilage degradation are considered to determine the osteochondral score and the highest score is 8 points and indicates poor joint health. The total IPSG MRI score is obtained by adding the soft tissue score and osteochondral score.

Statistics: Median, interquartile ranges and minimum-maximum values were used for descriptive statistical evaluation. Shapiro-Wilk test was used to examine whether the data showed normal distribution. Since the data were not normally distributed, the Spearman correlation test was used to evaluate the relationship between parameters. Statistical analyses were performed using the SPSS 22. The significance level was accepted as $p<0.05$.

Results

In the study, 18 patients had hemophilia A (one had inhibitor) and 2 had hemophilia B of 20 patients. The median, interquartile range, and minimum-maximum values of the variables used in the study were given in Table 1. The r and p values obtained from correlation analysis between age, HJHS-EP, Q-DASH, IPSG MRI scores and other parameters were shown in Table 2.

The results of the statistically significant correlations between the parameters were as follows; age was strongly positive correlated with HJHS-EP and loss of extension and strongly negative correlated with pronation ROM. HJHS-EP showed a strong positive correlation with loss of extension and a moderate negative correlation pronation ROM. There was a moderate negative correlation between Q-DASH and biceps/triceps muscle strengths. Although IPSG MRI total score was strongly positive correlated with the soft tissue score of IPSG MRI, it was moderately positive correlated with the osteochondral score.

In addition, age, ROM, HJHS-EP and IPSG MRI scores of patients whose physical and radiological scores were not compatible were given in Table 3. The IPSG MRI score of patient no:1 was 13 points out of 17 (While 7 points were obtained from soft tissue abnormalities and 6 were obtained from osteochondral changes). The HJHS-EP of this patient was 1 point out of 20. In Figure 1, there are MRI images of a patient whose physical scores are compatible with MRI scores.

Discussion

The elbow joint is the second most common bleeding joint in hemophilia and offers important contributions to upper extremity functionality. Therefore, it is important to evaluate separately from the lower extremity joints. In HA of the elbow joint, it is required to evaluate joint structures and upper extremity functions with both radiographic and detailed physical evaluations. It was found that as the age progressed, loss of elbow extension and HJHS-EP were increased and pronation ROM was decreased, statistically significantly. HJHS-EP was strongly correlated with loss of elbow extension and was moderately correlated with pronation ROM. In addition, a moderate correlation was found between upper extremity functionality and muscle strength of the elbow.

Table 1. Descriptive Statistics of Variables in the Study

Variables	Median (IQR)	Min.	Max.
HJHS-EP (0-20)	5(6)	0	14
Q-DASH (0-100)	20.4 (38)	0	61.3
IPSG MRI Total Score (0-17)	10 (2)	6	14
IPSG Soft Tissue Score (0-9)	6(3)	3	9
IPSG Osteochondral Score (0-8)	4(3)	2	6
Flexion ROM (°)	130 (13)	110	145
Loss of extension ROM(°)	8 (10)	0	40
Supination ROM (°)	90 (5)	60	95
Pronation ROM (°)	80 (15)	40	90
Biceps MS (pound)	21.6 (8.1)	6.9	39.9
Triceps MS (pound)	14.0 (4.1)	6.3	29.2

HJHS-EP: Hemophilia Joint Health Score-Elbow Point, Q-DASH: Quick-Disability of Arm, Shoulder and Hand, ROM: Range Of Motion, MS: Muscle Strength,
IPSG: International Prophylaxis Study Group

Table 2. Correlation Analysis of Data Obtained From Variables

	Age		HJHS-EP		Q-DASH		IPSG MRI STS		IPSG MRI OS		IPSG MRI TS	
	r	P	r	p	r	p	R	P	r	p	r	p
HJHS-EP (0-20)	.60	.00**										
Q-DASH (0-100)	-.21	.28	-.26	.18								
IPSG MRI STS (0-9)	.29	.15	.30	.14	.01	.92						
IPSG MRI OS (0-8)	.26	.19	.14	.49	.12	.54	.24	.23				
IPSG MRI TS (0-17)	.47	.01*	.17	.38	.11	.57	.74	.00**	.40	.04*		
Loss of extension ROM (°)	.66	.00**	.70	.00**	-.19	.32	.31	.12	.05	.79	.30	.13
Supination ROM (°)	.04	.04	-.18	.35	.06	.75	.07	.70	.13	.54	-.08	.67
Pronation ROM (°)	-.65	.00**	-.40	.04*	-.01	.94	.19	.35	.13	.53	-.24	.21
Biceps MS (pound)	.36	.05	.14	.46	-.45	.01*	.10	.60	.25	.21	.14	.46
Triceps MS (pound)	.22	.26	.09	.63	-.48	.01*	.17	.41	.12	.56	.13	.51

Spearman Correlation Analysis, r: Correlation coefficient, * $p<0.05$, ** $p<0.01$.
HJHS-EP: Hemophilia Joint Health Score-Elbow Point, Q-DASH: Quick-Disability of Arm, Shoulder and Hand, MS: Muscle strength, ROM: Range Of Motion, IPSG MRI: International Prophylaxis Study Group Magnetic Resonance Imaging, STS: Soft Tissue Score, OS: Osteocondral Score, TS: Total Score

In the current study, as age increases, especially increased in the loss of elbow extension and restriction in the pronation ROM have a negative effect on joint health. The probable reason for this can be recurrent bleedings over the years causes deterioration of the joint. In the correlation study of Chang et al., they examined PwH between the ages o 4-73 years who had HA in the knee, elbow, and ankle joints and showed that the incidence of HA of the elbow joint increased rapidly from adolescence (26). The results of this study are compatible with our study. These results show that interventions for ROM should be done at an early age for elbow joint.

The use of HJHS, a sensitive measurement tool to detect early changes in HA, is increasing. In our study, the fact that HJHS-EP was strongly positive correlated with loss of extension may indicate that extension of the elbow joint is more affected than rotational movements such as supination and

Table 3. Values of Patient's Physical and Radiological Scores Were Not Compatible

Patient No	Age (Year)	Flexion (°)	Loss of Ext. ROM (°)	Supination ROM(°)	Pronation ROM(°)	HJHS-EP (0-20)	IPSG MRI TS (0-17)
1	16	145	8	90	80	1	13
2	11	130	5	90	85	2	10
3	30	140	0	90	80	2	12
4	13	135	0	90	85	0	12
5	13	135	0	85	85	2	9

HJHS-EP: Hemophilia Joint Health Score-Elbow Point, ROM: Range Of Motion,
IPSG MRI TS: International Prophylaxis Study Group- Magnetic Resonance Imaging Total Score

pronation ROM in early ages in HA of the elbow joint. When we look at the rotational movements of the elbow, the HJHS-EP was moderately correlated with pronation ROM but not with supination ROM. In the current study, pronation ROM was more affected than supination ROM. In the similar studies in HA, there was no study comparing elbow ROM (27, 28). Measurement of extension and pronation ROM in young individuals with elbow HA can provide information about the progression of arthropathy, especially in clinical conditions where HJHS cannot be performed because it takes a lot of time.

In current study, there was a moderate correlation between the Q-DASH and elbow muscle strenght, as expected. But the lack of correlation between elbow ROM and Q-DASH indicates that there is no serious limitation in this age range that may affect functionality. Although there is strong negative correlation between loss of extension and joint health, there is no relationship between loss of extension and upper extremity functionality. If individuals have good health of the elbow joint, a higher upper extremity functionality is expected. However, this study showed unexpected results on it because there may be another factor that may affect this result. Q-DASH scoring system measures functionality of entire upper extremity joints that can compensate for each other. Besides there are some questions in Q-DASH Score such as doing heavy household or having pain involved extremity due to carrying heavy things. These may not assess the precise scoring of upper extremity functions in this study since some participants of this study were children and may not charge with these kinds of activities in daily life. There might be a need for another scoring system to evaluate upper extremity functions for HA patients.

In our study, we found a weak correlation between IPSG MRI soft tissue score and HJHS-EP that was not statistically significant. In addition, as shown in the Table 3, the physical and radiological scores of some patients were not compatible. Poonnoose et al. found that in the HA of the lower limb joints, HJHS was weakly correlated with the soft tissue score of IPSG MRI, and moderate correlated with osteochondral score of IPSG MRI (13). In our study, the correlation of IPSG MRI soft tissue score with HJHS is consistent with this study, but IPSG MRI bone tissue scores are not. This may be because the elbow joint is not a weight-bearing joint, such as lower limb joints, and arthrtitic changes did not affect the physical condition of the joint as much as the lower limb joints. For example, during walking a subchondral cyst in the ankle joint can cause pain that is one of the parameters of HJHS, but a similar osteochondral change can cause less pain in the elbow joint during movement. Nevertheless, it is thought that more studies are needed on this subject.

In our study, there was no correlation between IPSG MRI total score and Q-DASH. No study evaluating the relationship between upper extremity functionality and MRI has been encountered in the literature. Poonnosose et al. reported that radiological scores are useful in evaluating the joint structure and function, but they did not consider the effect of HA on overall musculoskeletal function (29). However, the study performed by Gupta et al. used Functional Independence Score in Hemophilia (FISH) in hemophilic children, a hemophilia-specific functionality scale involving both lower and upper extremities, and found a significant correlation between Petterson (X-ray) score of the elbow joint and FISH score(30). This may be due to the fact that FISH is a general functionality scale and X-ray is used as the imaging method. However, despite the poor radiographic scores in HA, the phenomenon of good function supports this finding (31, 32).

In the current study, IPSG MRI total score is more correlated with IPSG MRI soft tissue score than osteochondral score. This may indicate that

soft tissue changes are more common than bone and cartilage tissue changes in young hemophilic patients. This information may show that conservative treatment options such as physiotherapy and rehabilitation may be effective in the treatment of arthropathy in young patients.

Study Limitations: The number of elbow joints evaluated can be considered as the main limitation of our study. If the number of elbow joints evaluated was larger, the relationship between parameters could be more clearly demonstrated. In addition, the lack of inter-observer reliability among evaluations was another limitation. Therefore, it is thought that better results can be obtained by increasing the number of joints and inter-observer reliability in measurements.

The most remarkable result of our study was the weak or no correlation between MRI scores and joint health/functionality scores in HA of the elbow joint. Therefore, it is necessary to perform a physical evaluation as well as radiological imaging together when assessing the joint health and functionality. Even if there are no MRI findings, physical and functional evaluations should be done together. Loss of elbow extension, which has a strong correlation with HJHS, is thought to provide important information about the joint health when HJHS cannot be performed in the clinic. It is considered that functionality can be improved by increasing muscle strength in HA. Additionally, because joint health worsened with increasing age it was thought that HA of the elbow should be diagnosed at an early age and treatments should not be postponed.

Author Contributions: AMT performed the experimental study, analyzed the data, and wrote the manuscript. SO performed the radiological evaluation. NMT helped with the collection of data and writing the manuscript. AFO and KK organized the clinical study and revised the manuscript.

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Data Sharing Statement: The data that support the findings of this study are available on request from the corresponding author. The data are not publicly available due to privacy or ethical restrictions.

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