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The effect of generalized joint hypermobility on functional capacity, pulmonary function, respiratory muscle strength, and chest expansion in healthy young adults

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ABSTRACT

Aim: Genetic involvement of connective tissue containing elastin, collagen, and fibrils in joint hypermobility determines the tightness and laxity of the ligaments, thereby increasing the possibility of injuries by affecting the stability of joint capsules and the extensibility of tendons. The aim of this study was to investigate the effects of generalized joint hypermobility (GJH) on respiratory function, respiratory muscle strength, chest expansion, and functional capacity in healthy young adults.

Material and Method: Thirty subjects aged between 18-25 years with a four or higher Beighton Score were included as the GJH group, and 30 healthy age-gender volunteers with three or lower scores were included as the control group. Functional capacity was measured with the 6-Minute Walk Test (6MWT), the quadriceps muscle strength with a digital dynamometer, pulmonary function and respiratory muscle strength with a spirometry, and chest expansion with a tapeline.

Results: There were significant differences in the 6MWT distance ($p=0.017$), FVC ($p=0.001$), FEV_1 ($p=0.001$), and MEP ($p<0.001$) while no significant differences were observed in quadriceps muscle strength, FEV_1/FVC, PEF, and MIP ($p>0.05$). There is a significant difference in the xiphoid ($p<0.001$) and subcostal ($p<0.001$) measurement in the chest expansion value, and no difference in the axillary measurement ($p=0.071$).

Conclusion: The results of this study demonstrated that functional capacity, pulmonary functions, respiratory muscle strength, and chest expansion may be affected in young adults with GJH. In this study, it was found that the values that required a forced expiratory maneuver were affected. This suggests that the abdominal muscles, which play an important role in forced expiration, may also be affected by changes in the muscles due to deterioration in the connective tissue.

Keywords: Generalized joint hypermobility, functional capacity, pulmonary function, respiratory muscle strength, chest expansion

INTRODUCTION

Joint hypermobility is a common condition that increases the hyperelasticity of soft tissues and consequently the range of motion of the joint. When systemic disorders are excluded, it is described as increased mobility of the small and large joints in a particular age, sex, and race (1,2). Generalized joint hypermobility (GJH) is defined as hypermobility affecting multiple joints and classified using the Beighton Score, which is based on nine maneuvers tested for hypermobility, and the persons who get four or higher for a maximum total of nine scores as GJH (2-4). The prevalence of GJH in the general population is 20-26% (5,6), and in the child and adolescent population it varies between 2-55% depending on age, gender, or ethnicity (7,8). It is a global health problem that is seen more in women than men and causes symptoms such as pain, fatigue, discomfort, and joint instability (9). Joint hypermobility and GJH may be inherited as a normal trait with no identifiable genetic variant or also be a part of many different genetic syndromes such as Ehlers-Danlos Syndrome and other heritable disorders of connective tissue (10,11). The hypermobility of the joint changes the body biomechanics and causes compensatory mechanisms such as pain or muscle spasms in other body parts (10). The increased range of motion caused by hypermobility and chronic joint laxity may also cause hyperextension injuries to

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the supporting ligaments and soft tissue. Particularly in people who are already hypermobile, forces from repeated muscle contractions can strain the ligaments of the chest wall, leading to fatigue and pain (12).

Exercise capacity or functional capacity refers to the individual's ability to perform submaximal activities that require the pulmonary, cardiovascular, and skeletal muscle systems to work together and in a healthy manner (13). Decreased muscle strength and exercise capacity was reported in children and adolescent with GJH (14,15). Engelbert et al. (14) reported a reduced exercise capacity in children and adolescents with GJH using a maximal exercise test with an electronically braked cycle ergometer. Another study showed a decreased functional capacity as measured with the 6MWT and a decreased jumping capacity in adolescents with asymptomatic GJH (15). One of the important factors affecting exercise capacity and daily activities is muscle strength. In earlier studies, it was found that children and adolescents with GJH had decreased knee extensor and flexor muscle strength (14,16,17).

The chest wall is an elastic structure and follows the displacement of the lung. Chest wall expansion can be used in clinical practice to evaluate rib cage and wall mobility and can be related to lung volumes in healthy subjects (18). Chest wall expansion is related to respiratory muscle strength in healthy individuals (18,19). A study performed by Reychler et al. (20) revealed inspiratory muscle weakness in Ehlers-Danlos Syndrome, but expiratory muscle strength was not evaluated in the study. The effects of joint hypermobility on fatigue, musculoskeletal pain, headaches, postural dizziness, gastrointestinal system, and pelvic floor insufficiency are frequently mentioned in studies (10,21). The respiratory problems in joint hypermobility were also reported in Ehlers-Danlos Syndrome such as dyspnea, asthma, sleep apnea, pneumothorax, and chest wall abnormalities (pectus excavatum, straight back syndrome) (22). However, the effects of hypermobility on the respiratory system remain in the background and have not been well characterized in GJH. So, this study was planned to investigate the effects of joint hypermobility on respiratory function, respiratory muscle strength, chest expansion, and functional capacity in healthy young adults.

MATERIAL AND METHOD

Study Design and Participants

This study is an observational, analytical type of case-controlled study. The study was carried out with the permission of Nuh Naci Yazgan University Scientific Research and Publication Ethics Committee (Date. 16.09.2022, Decision No: 2022/001-001). All procedures were carried out in accordance with the ethical rules and the principles of the Declaration of Helsinki. Written informed consent was obtained from all subjects before the study. The study was carried out between September 2022 and December 2022 at Nuh Naci Yazgan University. Thirty subjects aged between 18-25 years with a four or higher Beighton Score were included in the GJH group (23), and 30 healthy age-gender volunteers with a score of three or lower were included in the control group. Exclusion criteria were the presence of any neurological and/or orthopedic problems, chronic and/or acute respiratory disease, and medication use that may affect respiratory functions in the last three months for all subjects.

Outcome Measures

The gender, age, height, weight, Beighton Score, smoking, and physical activity status were recorded. Body mass index was calculated using the weight/height2 formula. Functional capacity was measured with the 6-Minute Walk Test (6MWT), the quadriceps muscle strength with a digital dynamometer (Jtech Commander Muscle Tester, USA), pulmonary function and respiratory muscle strength with a spirometry (Cosmed Pony FX Spirometer, Italy), and chest expansion with a tapeline.

Functional capacity was evaluated with the 6MWT according to the criteria of the American Thoracic Society (24). The test is used to assess aerobic capacity and endurance as a sub-maximal exercise test. The distance that the participants walked at their own walking speed for six minutes in a 30-meter-long straight corridor, as fast as possible but without running, was recorded in meters. In addition, SpO2, heart rate, dyspnea, and fatigue levels were evaluated with a pulse oximeter (Beuer pulse oximeter, Beurer GmbH; Germany) before and after the test. The Modified Borg Scale was used to determine dyspnea and fatigue levels (24,25).

The quadriceps muscle strength was evaluated using a digital hand-held dynamometer (Jtech Commander Muscle Tester, USA). The participant sat on the edge of a bed, their feet not touching the floor and their arms crossed in front of the body. They were asked to keep their pelvis on the bed and to extend the knee joint without a swing movement. The transducer was placed on the anterior distal part of the leg. Measurements were performed three times consecutively and the mean values of the right and left side measurements were recorded as force in Newton (N). All muscle strength tests were performed by the same physiotherapist with the same device (26).

The pulmonary function tests of the subjects participating in the study were performed using a spirometer (Cosmed Pony FX Spirometer, Italy) in accordance with the American Thoracic Society/European Respiratory Society criteria (27). The age, gender, and previously measured

height and weight of the participant were recorded while they were resting for 10 minutes before the test. The test began while the participant's feet were in full contact with the ground in a sitting position. Nasal breathing was prevented with a nose clip, and the subjects were asked to inhale as deeply as possible and then quickly perform a deep expiration. It was important to make sure that the spirometer mouthpiece was closed airtight from the lip, the tongue was not inserted into the mouthpiece, the inspiration was completed, the maneuver started quickly and strongly, there was no pause, no other breathing or coughing occurred during expiration, and the expiration continued until a plateau was seen in the volume-time graph (27). Forced vital capacity (FVC), forced expiratory volume in one second (FEV_1), FEV_1/FVC, and peak expiratory flow rate (PEF) were measured.

Respiratory muscle strength was measured (Cosmed Pony FX Spirometer, Italy) in accordance with the American Thoracic Society/European Respiratory Society criteria (28). Before starting the test, the subjects were instructed to close their lips tightly around the mouthpiece to prevent air leaks. The measurement began when the feet were in full contact with the ground in a sitting position. For maximal inspiratory pressure (MIP) measurement, the nasal airway was closed with a latch after the appropriate mouthpiece was fixed. When the subject reached the residual volume, the device's mouthpiece was put in the mouth and maximum inspiration (Müller maneuver) was performed at maximum speed for 1-3 seconds. For maximum expiratory pressure (MEP) measurement, in contrast to the residual volume, it began at total lung capacity and maximal expiration (Valsalva maneuver) was performed at maximum speed for 1-3 seconds. Maximal inspiratory and expiratory performance was put in with one-minute intervals between tests. The measurement was repeated until there was a difference of 10 cmH_2O between the two best measurements and the best result was recorded as cmH_2O (28).

Chest wall expansion was measured using a standard measuring tape at three different levels for chest circumference. With the participant in an upright position, feet shoulder-width apart, arms relaxed at the side of the body, measurements were taken at three sites: Axilla (upper-level), xiphoid (mid-level), and subcostal (lower-level) (29). After placing the tape measure around the chest, the physiotherapist, standing in front of the subject, initially asked them to breathe normally to determine the tidal volume and then to exhale maximally. The difference between the two scales (inhalation-exhalation) was determined as thoracic expansion (chest expansion). Participants were instructed to perform three maneuvers and the average of the obtained values was recorded (18). Measurements were performed by the same physiotherapist to minimize possible errors due to heterogeneity.

Statistical Analysis

Statistical analyses were performed using the SPSS V20.0 statistical program (SPSS, Inc.). The normality of data distribution was determined with the Kolmogorov-Smirnov test. The descriptive data of the demographic variables were expressed as mean-standard deviation, so the data were normally distributed. The differences between the GJH and control groups were tested using an independent sample t-test, and a Chi-square test was used for categorical variables (30).

The G*Power 3.1 (Universitaet Dusseldorf, Germany) software was used for the sample size calculation. In a study performed by Scheper et al. (15), the 6MWT distances of dancers and non-dancers were compared using the Beighton Score ≥ 4 for the classification of GJH. Considering these results, we hypothesized that to obtain a similar difference rate of 90% power and 95% confidence level, a total of 60 participants had to be included in this study.

RESULTS

Thirty young adults with GJH and 30 healthy young adults as the control group were included in the study. The demographic characteristics of the groups are presented in **Table 1**. There were no significant differences between the GJH and the control group in terms of baseline characteristics of the young adults ($p>0.05$), except for the Beighton Score ($p<0.001$).

Table 1. Demographic characteristics of the groups

	GJH group (n=30)	Control group (n=30)	p
Gender (n, %)			0.598
Female	19 (63)	17 (57)	
Male	11 (37)	13 (43)	
Age (years)	22±2.21 [19-25]	22.07±2.32 [18-25]	0.904
Weight (kg)	64.9±13.9 [48-100]	66.8±16.9 [49-120]	0.625
Height (cm)	171.3±8.87 [160-190]	169.5±8.78 [155-186]	0.441
BMI (kg/m^2)	22±3.68 [16.2-32.7]	23±3.8 [18.3-35.1]	0.312
Beighton score	5.57±1.33 [4-8]	0.87±1.2 [0-3]	<0.001
Smokers (n, %)	12 (40)	13 (43)	0.793
Physically active (n, %)	11 (37)	9 (30)	0.584
Infected with COVID-19 (n, %)			0.237
Yes	16 (53)	22 (73)	
No	7 (23)	3 (10)	
Not sure	7 (23)	5 (17)	

Data are presented as mean±SD [min-max], GJH: Generalized joint hypermobility; BMI: Body Mass Index

The mean values of 6MWT distance, quadriceps muscle strength, pulmonary function, respiratory muscle strength, and chest expansion of the two groups, and the comparisons between groups are shown in **Table 2**. There were significant differences in 6MWT distance (p=0.017), FVC (p=0.001), FEV_1 (p=0.001), and MEP (p<0.001) while no significant differences were found in quadriceps muscle strength, FEV_1/FVC, PEF, and MIP (p>0.005). There was a significant difference in the xiphoid (p<0.001) and subcostal (p<0.001) measurement in the chest expansion value, and no difference in the axillary measurement (p=0.071).

Table 2. Comparison of the 6MWT distance, quadriceps muscle strength, pulmonary function, respiratory muscle strength, and chest expansion between groups

	GJH group (n=30)	Control group (n=30)	p
6MWT distance (m)	511.7±69.6	552±56.5	0.017
Quadriceps muscle strength (N)			
Right	298.7±55.6	290.1±72.1	0.638
Left	280.7±57.9	281.4±74.3	0.968
Pulmonary function test			
FVC (L)	3.87±0.6	4.22±0.83	0.064
FVC (% of predicted)	88±6.13	93.4±6.12	0.001
FEV_1 (L)	3.35±0.43	3.87±0.72	0.001
FEV_1 (% of predicted)	89.6±5.07	95.2±7.69	0.001
FEV_1/FVC	87.5±12.4	91.8±4.69	0.076
PEF (L)	6.78±1.94	6.96±2.23	0.736
PEF_%	83.4±18.1	88.2±20.7	0.349
Respiratory muscle strength (cmH_2O)			
MIP	98.88±27.25	105.73±18.99	0.263
MEP	105.87±23.64	135.77±36.23	<0.001
Chest size during inspiration (cm)			
Axillar	68.6±9.78	72.5±12.1	0.175
Xiphoid	77.5±11.8	81.5±12.3	0.211
Subcostal	69.7±11	76.9±12.3	0.021
Chest size during expiration (cm)			
Axillar	80.2±9.03	81.9±9.81	0.471
Xiphoid	71.9±11.2	78±11.5	0.040
Subcostal	68.6±9.78	72.5±12.1	0.175
Chest expansion value (cm)			
Axillar	5.4±3.4	6.7±1.84	0.071
Xiphoid	5.67 ±2.28	3.43±1.81	<0.001
Subcostal	1.17±3.61	4.4±2.11	<0.001

Data are presented as mean±standard deviation, GJH: Generalized joint hypermobility; 6MWT: Six-Minute Walk Test; FVC: forced vital capacity; FEV_1: forced expiratory flow in 1 second; PEF: peak expiratory flow; MIP: maximum inspiratory pressure; MEP: maximum expiratory pressure

DISCUSSION

This research aimed to investigate the effects of joint hypermobility on the functional capacity, pulmonary function, respiratory muscle strength, and chest expansion in healthy young adults. As a result of our study, significant differences in 6MWT distance, FVC, FEV_1, MEP, and chest expansion were observed while no differences were found in MIP and knee extensor muscle strength.

Genetic involvement of connective tissue containing elastin, collagen, and fibrils in joint hypermobility determines the tightness and laxity of the ligaments, thereby increasing the potential for injuries by affecting the stability of joint capsules and the extensibility of tendons (1,31). The hypermobility of the joints change the body biomechanics and causes compensatory mechanisms such as pain or muscle spasm in other body parts. For example, anterior pelvic tilt with lumbal hyperlordosis accompanying genu recurvatum and/or flat feet can be observed (10). Joint hypermobility syndrome can also affect daily life activities, especially prolonged standing activities in patients (4). In addition to the impact on daily activities, there is a decrease in functional capacity and cardiorespiratory fitness in joint hypermobility (14,15). Engelbert et al. (14) reported a reduced exercise capacity in children and adolescents with GJH using a maximal exercise test with an electronically braked cycle ergometer. In a study performed by Scheper et. al (15), decreased functional capacity measured by 6MWT and jumping capacity expressed as reduced walking distance and jumping capacity were reported in adolescents with asymptomatic (pain-free) GJH. In our study, we found reduced functional capacity and walking distance in young adults with GJH compared to the healthy control group. The 6MWT is the most common submaximal test used to evaluate the functional capacity including main cardiovascular, pulmonary, and neuro-musculoskeletal performance in chronic lung diseases such as chronic obstructive pulmonary disease, asthma, or cystic fibrosis. The test evaluates the global and integrated responses of all the systems involved during exercise, including the pulmonary system (24). Moreover, there is a cut-off value for the 6MWT in chronic obstructive pulmonary disease for identifying patients with high mortality risk (32). The reduced walking distance according to the 6MWT in the GJH group compared to the control group in this study suggested that it may be related to decreased lung functions.

Muscle strength is one of the important factors affecting exercise capacity and daily activities. In earlier studies, it was found that children and adolescents with GJH had decreased knee flexor and extensor muscle strength (14,16,17). In a study, lower muscle activity for quadriceps and hamstrings measured by electromyography was observed during stair climbing, an important daily life activity, in women with symptomatic and asymptomatic GJH (33). In contrast, increased knee extensor muscle strength was reported in healthy adolescents with GJH (7). In the present study, there is no significant difference in quadriceps muscle strength between subjects with and without GJH. This decrease in functional capacity may be due to the decreased lung volumes in individuals with GJH. In addition, a reason why there was no difference

between the groups in terms of quadriceps muscle strength in this study may be that the participants were not classified as symptomatic or asymptomatic. Symptoms such as pain prevent individuals from performing physical activity and cause a decrease in muscle strength (15,17). Schubert-Hjalmarsson et al. (34) showed that pain-free hypermobile children appeared to be more physically active in sports. Muscle strength deficit in hypermobile individuals may be related to pain, either directly or indirectly through inactivity due to pain (35). The presence of both symptomatic and asymptomatic participants in this study may have contributed to the lack of difference in quadriceps muscle strength. Therefore, in future studies, it is important to explain the effects of symptoms on muscle strength in individuals with GJH.

One of the determinants of functional capacity is the state of respiratory functions. Respiratory manifestations such as dyspnea, sleep apnea, and decreased respiratory muscle function have frequently been described in classical or hypermobile Ehlers-Danlos Syndrome (22). Respiratory manifestations in hypermobility spectrum disorders have been noted less often in the literature (22). Soyucen and Esen (36) postulated that benign joint hypermobility syndrome may predispose to childhood asthma. This is the first study to investigate the pulmonary function and respiratory muscle strength in young adults with GJH, to the best of our knowledge. We found significantly reduced lung volume (FEV_1 and FVC) and MEP values. Respiratory muscle strength is associated with lung volume and functional capacity (19). Padkao et al. (19) showed that respiratory muscle strength is associated with functional capacity measured by the 6MWT. In this study, decreased respiratory muscle strength in subjects with GJH may be also one of the reasons for the decrease in the 6MWT distance. However, it was observed that subjects with GJH had lower values in maneuvers requiring forced expiration such as FEV_1 and MEP. This suggests that the abdominal muscles, which play an important role in forced expiration, may also be affected due to deterioration in the connective tissue (37,38). Studies in the literature on functional muscle strength in joint hypermobility generally focus on the lower extremity muscles, but more research is required to investigate trunk muscles including abdominals. In addition, the lower lung capacity observed in both groups can be explained by the fact that more than half of the individuals were exposed to COVID-19 infection, as we showed the long-term effects of covid in a previous study (39).

Muscle strength, fatigue, and pain affect functional capacity in individuals with joint hypermobility (15,17). However, chest wall expansion may be also an important contributor to functional status. The chest wall is an elastic structure and follows the displacement of the lung, and the chest wall expansion can be used in clinical practice to evaluate rib cage mobility and wall mobility and it is related to lung volumes in healthy subjects (18). Chest wall expansion is related to respiratory muscle strength and higher chest mobility reflects the greater MIP and MEP value in healthy individuals (18,19). To the best of our knowledge, there is no study investigating chest wall expansion in GJH. In this study, statistical significance was found in chest expansion value in the measurement of xiphoid and subcostal. Subjects with GJH had more chest wall expansion at the xiphoid level and less at the subcostal level compared to the control group. Individuals may have developed different breathing patterns, potentially due to the structural abnormality of connective tissue. Breathing patterns may also be examined in future studies.

The limitations of the present study include the fact that environmental and psychological factors such as social status were not considered in this study. While this information is beyond the scope of the present article, it should be noted that such information may have an impact on the treatment of individuals diagnosed with symptomatic forms of GJH. The possibility of continuing long-term effects of the COVID-19 infection is also a limitation of the study. However, although the majority of the participants had COVID-19 before, the inability to evaluate the effects of the infection on the respiratory functions of the participants is another limitation. Yet another limitation could be that the potential confounders such as pain were not considered in our study. The last limitation, based on the study design, is that these results offer no causative evidence, but future studies designed as longitudinal observational studies may provide evidence.

CONCLUSION

Functional capacity, pulmonary functions, respiratory muscle strength, chest expansion, and quadriceps muscle strength may be affected in young adults with GJH. In this study, it was found that there was an effect on the values that required a forced expiratory maneuver. This suggests that the abdominal muscles, which play an important role in forced expiration, may also be affected due to deterioration in the connective tissue. Considering the changes in chest expansion, patients with GJH may develop different breathing patterns. It is important to know the symptoms and especially the respiratory management strategies in GJH, which have generally been managed using conservative treatment.

ETHICAL DECLARATIONS

Ethics Committee Approval: The study was carried out with the permission of Nuh Naci Yazgan University Scientific Research and Publication Ethics Committee (Date. 16.09.2022, Decision No: 2022/001-001).

Informed Consent: All patients signed the free and informed consent form.

Referee Evaluation Process: Externally peer-reviewed.

Conflict of Interest Statement: The authors have no conflicts of interest to declare.

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Author Contributions: All of the authors declare that they have all participated in the design, execution, and analysis of the paper and that they have approved the final version.

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