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ORIGINAL ARTICLE

Evaluation of knee proprioception and effects of proprioception exercise in patients with benign joint hypermobility syndrome

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Abstract The first aim is to show if there is a disorder in proprioception in cases with benign joint hypermobility syndrome (BJHS) when compared to healthy subjects. The second aim is to evaluate the effect of proprioception exercise in BJHS cases. To evaluate the proprioceptive sensibility of the knee joint with 40 BJHS and 30 healthy subjects enrolled in the study. Then, cases with BJHS were randomized into two groups; proprioceptive exercises were applied to 15 patients for 8 weeks in clinic and 25 patients were taken as controls. Outcome measures included proprioceptive sensation, AIMS2 and VAS. Proprioception is significantly impaired in cases with BJHS. In BJHS group, significant decreases in VAS levels were detected in cases who did exercise compared with cases who did not, and statistically significant improvements were detected in occupational activity. For this reason proprioception exercises cause decrease in pain and improvement of functional status in BJHS group.

Keywords Proprioception · Hypermobility syndrome · Exercise · Knee · Isokinetic

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Introduction

Hypermobility syndrome (HMS) is an inherited disease due to variations or mutations of the genes coding collagen, elastin, fibrillin and tenascin; it affects multiple organs and causes musculoskeletal system complaints. It is seen in 0.6–31% of adults and is found to be five times more frequent in women than that in men [1–4]. As it does not have life-threatening complications, it is defined as "benign joint hypermobility syndrome" (BJHS). There is no laboratory or radiologic diagnostic criteria, and present diagnosis is based on generalized laxity and specific joint involvement. Carter–Wilkinson, Beighton, Bulbena and Rotes have worked to identify diagnostic criteria, and Brighton's criteria have been accepted presently [1, 5].

Proprioceptive system plays a critical role in maintenance of joint stability, including sensation of both position and movements of joint, under dynamic conditions. Muscle and joint receptors are main sources for proprioception [6]. Impairment in proprioception was defined in BJHS cases, and this is first shown in knee and proximal interphalangeal joints [7, 8]. Inactive status, joint instability, debility in

muscle strength, decreased endurance can be attributed to weakness of proprioception and frequent injuries. In these patients proprioception deficit that affects functional parameters make them more vulnerable to minor traumas compared to normal people [1, 3, 9].

It was shown that exercises which stabilize the joint decrease the hypermobility and relieve the pain in short time [3, 10, 11].

In this study the aim was first to show the proprioception impairment in cases with BJHS and secondly to investigate the effect of proprioception exercises done towards the impairment.

Materials and methods

Subjects

For the first step of the study, 40 cases diagnosed with BJHS, admitted to outpatient clinic of Physical Medicine and Rehabilitation Department and 30 healthy individuals were enrolled. For the second step of the study, after selecting randomly among cases with BJHS, 15 cases were taken to exercise group and 25 cases were taken to control group. Patient group was composed of cases who the diagnostic criteria according to the Brighton 1998 diagnostic criteria were Beighton scores of 4/9 or above plus one major or two minor symptoms, aged between 20 and 45, having knee pain, having no contraindication to make exercise and not having osteoarthritis or ligament damage at knee joint. Control group was selected among age and gender matched hospital staff. Cases with severe knee trauma, having osteoarthritis or ligament damage and additional disease were not included in the study. After describing the planned procedures to all cases, informed consent and acceptance of ethical committee were obtained. Patients were evaluated in the beginning and in the eighth week of the treatment with visual analog scale (VAS, movement, resting) for pain severity, with Arthritis Impact Measurement Scales-2 (AIMS-2) for function and with Biodex System 3Pro Multijoint System Isokinetic Dynamometer (Biodex Medical Inc, Shirley, NY, USA) for measurement of proprioception sensibility.

Evaluation criteria and protocol

Study design

In this study proprioception measurements of cases with BJHS and control cases were done in the first step. In the second step, after dividing BJHS cases into two groups, one group received proprioception exercise for 8 weeks and the other group did not receive exercise as in the control group. Both groups were evaluated with VAS, AIMS-2 and proprioception measurement in the first and eighth weeks. Exercises were done under control of a doctor in clinic for 8 weeks and for 3 days in a week.

1. Brighton criteria. Major criteria: Beighton score should be 4/9 or above, arthralgia in four or more joints for longer than 3 months. Minor criteria: Beighton score 1, 2 or 3/9, arthralgia in one to three joints; back pain for longer than 3 or more months, spondylosis, spondylolisthesis/spondylolysis; dislocation or subluxation in more than one joint; three or more soft tissue lesions (epicondylitis, tenosynovitis, bursitis); Marfanoid habitus (tall, slim, ratio of upper and lower segment <0.89, arachnodactyly); abnormal skin: striae, thin skin, ocular signs; dropping eye lids, myopia, antimongoloid slant; varicose veins, hernia, or uterine or rectal prolapse, mitral valve prolapse [5].

2. Proprioception testing. Active–active method in sitting position, which is one of the reproduction tests evaluating joint position sense, was used. Reproduction tests evaluate the ability of patients to reproduce the knee angles actively or passively which were previously determined actively or passively. In this test, first an angle is produced in the joint of the patient actively or passively, after that neutral position is provided. In the last step patient is asked to repeat the first angle position actively or passively [6]. In active–active method, patient is asked to actively bend his/her extremity to a predetermined angle position and then to form this angle actively again [6, 12]. In the previously used methods every researcher applied different angle degrees and numbers [6, 12–14]. Biodex System 3Pro Multijoint System Isokinetic Dynamometer was used for the calculation of knee proprioception sense. Patients and control group cases were asked to reproduce the fixed knee angle (5°, 15°, 30°, 45°, 60°, 75°) that was actively positioned for 10 s between 5° and 75° angles. After application of this procedure to BJHS group (both exercise and control groups) before and after the exercise, and to the healthy control group only once, the mean of every angle position was measured. The testing was performed in a silent room with air conditioner and constant temperature while the cases were blindfolded. "Absolute angular error" values were obtained for every angle, later the mean of absolute angle error of six angles was calculated as "average absolute angle error". The average absolute angle error value is conversely correlated to proprioceptive sensibility [15–17].

3. Evaluation of pain. Patients were evaluated according to the knee pain that developed during movement and resting position with VAS of 0–10 mm (0 point: no pain, 10 point: severe pain).

4. Evaluation of functional status. AIMS-2 is a questionnaire evaluating health condition and quality of life. It includes 15 scale and 77 questions measuring the mobility, walking and bending, hand and finger functions, arm

functions, self-care, house activities, social activities, family friend support, arthralgia, occupational activity, emotional status, level of tension, ability, time period the patients want to get better and effect of arthritis. Every scale has 4 to 12 questions and physical, mental, social, emotional and working health status are measured with the answers. The score of every subscale is between 0 (best score) and 10 (worst score) [18, 19].

Proprioception exercises

Kinesthesia and exercises of balance comprised this program. In the first week: walking backward (30 s), heel walking (30 s), walking on fingertips (30 s), walking with eyes closed (30 s), standing on one extremity (30 s), bending to forward–backward-sides on one extremity (eyes open), bending forward–backward-sides on one extremity (eyes closed) (30 s), slowly sitting on a high chair and standing up (20 times); exercises added in the second week: exercise with rocker bottom wood (2 to 3 min), slowly sitting on a low chair and standing up (10 times), plyometric exercises (passing through a 15 cm height by jumping) (10 times), 8 exercises (slow walk–broad circle, fast walk–broad circle, slow walk–narrow circle, fast walk–narrow circle) (5 times); exercises added in the third week: exercise with Biomechanical Ankle Platform System (BAPS) board balance wood (2–3 min), minitrompolin exercise (jumping) (30 times) [20, 21].

Statistical analysis

SPSS program 12.0 version was used for statistical analysis. Mann–Whitney *U* test and Wilcoxon analysis were used to compare both groups. Significance was accepted at $P < 0.05$.

Results

The BJHS cases whose mean age was 26.90 ± 7.15 (6 men and 34 women) included in the study. The mean age of the control group was 26.37 ± 6.13 (4 men and 26 women). Among BJHS cases, the mean age of exercise group was 25.60 ± 5.98, (3 men and 12 women) and the mean age of control cases was 27.68 ± 7.77 (3 men and 17 women). Of the BJHS group, 80% of exercise group cases and 72% of control cases had knee hyperextension. None of the healthy controls had knee hyperextension. With respect to age, height and weight there was no statistically significant difference among all the groups ($P > 0.05$) (Table 1). Before exercise, no significant difference was present in none of AIMS-2 subscales, resting VAS (VASr) and movement VAS (VASm) between exercise group and control groups in cases with BJHS ($P > 0.05$).

Table 1 Characteristic properties of exercise and control group of patients with BJHS

Group	Exercise ($n = 15$)	Control ($n = 25$)	P
Age, years	25.60 ± 5.98	27.68 ± 7.77	0.380
Height, cm	167.07 ± 8.10	165.36 ± 8.25	0.528
Weight, kg	64.60 ± 10.11	61.44 ± 8.15	0.285

Values are the mean ± SD

First step: proprioception measurement in BJHS

When the proprioception measurements of healthy subjects ($n = 30$) and cases with BJHS ($n = 40$) were evaluated, the increase in average absolute angle error values in both knees was statistically significantly higher in BJHS cases than that in healthy subjects ($P < 0.001$). There was highly positive significant correlation between right knee and left knee with respect to average absolute angle error value in BJHS group and healthy controls ($P = 0.007$, $r = 0.423$ for BJHS; $P = 0.000$, $r = 0.620$ for healthy subjects) (Table 2).

Second step: effect of proprioception exercises in BJHS

When the exercise and control groups of BJHS group were compared, no significant difference was detected between right and left extremity of two groups before the exercise ($P > 0.05$). During postexercise, when the proprioception measurements were evaluated, the increase in average absolute angle error values was detected as statistically highly significant in both knees in exercise group ($P < 0.001$). However, in BJHS control group no statistically significant difference was found between two knees with respect to average absolute angle error value measurements done in the first and eighth weeks ($P > 0.05$) (Table 3).

Alterations in VAS

After exercise, there was a statistically significant drop in VASr and VASm values of exercise group ($P < 0.05$).

Table 2 Average absolute angle error value (AAAEV) in right and left extremity of BJHS cases and healthy individuals

AAAEV	BJHS ($n = 40$)	Control ($n = 30$)	$P^{\ddagger}$
Right extremity	2.40 ± 0.80	1.25 ± 0.41	0.000*
Left extremity	2.36 ± 0.76	1.23 ± 0.38	0.000*
$P^{\ddagger}$	0.391	0.715	

Values are the mean of six different fixed knee flexion angles

‡ By Mann–Whitney *U* test

* Statistically significant

Table 3 Average absolute angle error values (AAAEV) in right and left extremity of exercise and control group among cases with BJHS

BJHS	1-week AAAEV	8-week AAAEV	$P^{\ddagger}$
Exercise group (n = 15)			
Right extremity	2.73 ± 0.80	1.83 ± 0.72	0.000*
Left extremity	2.42 ± 0.68	1.63 ± 0.63	0.001*
Control group (n = 25)			
Right extremity	2.38 ± 0.59	2.41 ± 0.59	0.586
Left extremity	2.40 ± 0.74	2.41 ± 0.69	0.743

Values are the mean of six different fixed knee flexion angles

‡ By Wilcoxon signed rank test

* Statistically significant

However, in control group no statistically significant difference was present in activity and resting VAS values in the end of eighth week ($P > 0.05$) (Table 4).

Alterations on functional status

In exercise group when compared to prior to exercise, statistically significant improvement was detected in AIMS-2 occupational activity of exercise group after the therapy ($P < 0.01$), but there was no significant difference in the other AIMS 2 subscales ($P > 0.05$). In control group no statistically significant difference was present in AIMS-2 in the eighth week ($P > 0.05$) (Table 5).

Discussion

Proprioceptors are the receptors responsible for position and movement sense. Proprioceptive mechanisms effectively assist in coordination of complex movement systems. Proprioception is very important for preventing undesired joints such as hyperextension and hyperflexion, and plays a protective role in injuries. Development of decrease in the muscle tone and tense power of tendons causes insufficiency of proprioception and abnormal motor control. So degeneration of ligaments would contribute to functional impairment by causing proprioceptive feedback loss [6, 12, 22, 23].

Table 4 VAS values in exercise and control group of cases with BJHS

BJHS	1-week VAS	8-week VAS	$P^{\ddagger}$
Exercise group (n = 15)			
VASm	5.87 ± 2.35	1.83 ± 0.72	0.010*
VASr	3.27 ± 2.49	1.40 ± 2.16	0.027*
Control group (n = 25)			
VASm	5.96 ± 1.83	5.88 ± 1.90	0.157
VASr	3.48 ± 2.06	3.44 ± 2.04	0.317

VAS was evaluated between 0 and10 mm

‡ By Wilcoxon signed rank test

* Statistically significant

Table 5 AIMS-2 values in exercise and control group of cases with BJHS

	1 week	8 weeks	$P^{\ddagger}$
BJHS exercise group (15)			
Physical status	1.58 ± 1.31	1.21 ± 1.35	0.358
Emotional status	4.15 ± 2.18	3.85 ± 1.62	0.596
Symptom	3.70 ± 2.69	2.53 ± 2.44	0.206
Social activity status	3.11 ± 1.53	3.05 ± 1.64	0.917
Occupational activity	2.96 ± 2.99	1.12 ± 1.70	0.006*
BJHS control group (15)			
Physical status	1.31 ± 1.14	1.27 ± 1.24	0.891
Emotional status	4.31 ± 1.68	4.64 ± 2.24	0.589
Symptom	4.66 ± 2.91	3.76 ± 2.51	0.185
Social activity status	3.04 ± 1.71	2.96 ± 1.88	0.867
Occupational activity	3.06 ± 1.86	2.14 ± 2.49	0.117

AIMS-2 was health condition and quality of life between 0 and10

‡ By Wilcoxon signed rank test

Cases with BJHS have diffuse musculoskeletal system complaints including many body areas. Pain complaints: acute pain is due to overuse or tissue injury, and chronic diffuse pain has a large spectrum from the pain originating due to disorder in high central pain centers to psychological function pain. Proprioception deficit is one of the important causes of these complaints which is also a factor affecting treatment success in BJHS [1–3, 24, 25]. Furhter, decreased proprioception results in predisposition to development of osteoarthritis due to abnormal position of the joint and contributes to functional insufficiency by affecting functional parameters such as decreased walking speed, shortened step and decreased total walking time [1, 3, 6, 14].

Treatment methods that will be used for hypermobility should be directed to insufficiency and functional limitation. Although exercise does not decrease laxity, as strengthening and proprioception exercises improve muscle structure, they also affect the surrounding joint components [2, 3, 10].

In our study, first it was shown that proprioception was impaired in cases with BJHS compared to healthy individuals. Similar findings were found in previous studies [3, 14, 26]. Hall et al. [14] found in their study, in which they compared age and gender matched 10 female cases with hypermobility syndrome and healthy individuals, that proprioception is worse in cases with hypermobility.

According to these findings, due to insufficiency of proprioception, proprioception exercises should be added to treatment program of cases with BJHS that aim at restoring motor control of lower extremity and assist in maintaining

good balance and enough function [11, 26, 27]. In our study, significant improvement was detected in proprioception by the active–active method, one of the reproduction tests that measures it. Also with this exercise program significant decrease was detected in VAS values. It is controversial whether decrease in pain is only due to improvement in proprioception or not. But, decreases in traumas that develop because of proprioception deficit, improvement in kinesthesia and joint position have positive effects on pain [1]. Also exercise affects decondition and immobilization positively which is one of the reasons for the pain in hypermobility [1, 10, 28]. Barton and Bird [11], Ferrell et al. [24] followed the results with some parameters after giving proprioception and stabilization exercise particularly to every joint for 6–8 weeks in cases with BJHS. After the treatment, their detection of significant decrease in VAS which is one of their follow-up parameters is similar to the result of our study.

In both groups, significant improvements were not observed in subscales present in AIMS-2, except in occupational activity that showed significant improvement in exercise group. The results of AIMS-2 showed that pain complaint, which is seen prominently in cases with hypermobility, did not affect the physical functions, and they felt minimal trouble due to affected functions. In conclusion, only given proprioception exercises restore the lower extremity motor control. However, all cases enrolled to our study had generalized laxity and studies directed at general exercise programs should be done for total improvement of functional status.

Cases with BJHS are the patients who were examined by many doctors for years, but could not be diagnosed and whose symptoms were thought to be psychological. To give exercise to cases in clinical environment tells them that their disease was understood and by explaining the reason of their complaint, an important psychological support will be given to the patient. As a result of this, we may contribute to decrease in VAS values and improvement of functional status.

The only given proprioception exercise resulted with decrease in VAS values of the cases and although not significant, improvement in functional parameters. General exercise should be given to cases with BJHS and constituting studies directed to this. However, proprioception exercise should be added to every kind of exercise given to patients.

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