

Study of the Comparative Efficacy of Strengthening Exercise between Extensor Group of Muscles and Both Flexor and Extensor Groups of Muscles in Patient presenting with Symptoms in Affected Joints of Benign Joint Hypermobility Syndrome

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ABSTRACT

In the management of benign joint hypermobility syndrome (BJHS), there is no clear-cut idea regarding the types of exercises which will be appropriate to incorporate in the management protocol of this unique condition. According to some the extensor muscles group should be strengthened and some other opined to strengthen both the extensor and flexor groups of muscles. We presume that strengthening of both the extensor and flexor will be more beneficial because it will provide balance between two opposing groups of muscle and hence, give more stability to the joint and the isometric strengthening exercise would be the exercise of choice. In our study for therapeutic exercise, the 61 patients were grouped into two randomly. Group I (30 patients) were instructed to perform only isometric strengthening exercise for only extensor group of muscles. Group II (31 patients) were advised to do isometric strengthening exercise for both the extensors and flexors group of muscles. After 6 weeks of therapy we found group II is showing a superior result.

Keywords: Arthritis Impact Measurement Scale, Beighton scale, Joint laxity, Visual analog scale.

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INTRODUCTION

In 1967, Kirk et al^{1,2} found this ailment as a distinct pathology, with the presence of rheumatic symptoms with

generalized joint laxity in the absence of any demonstrable systemic rheumatic disease.

Previously it was not recognized as a syndrome, but refer to the manifestation of joint hypermobility or articular hypermobility. Beighton^{1,3} declared this, for the first time, as a syndrome, in the International Nosology of Heritable Disorder of Connective Tissue and addressed it as a familial articular hypermobility syndrome and excluded genetic diseases that include joint hypermobility as an associated finding, such as Ehlers–Danlos syndrome, osteogenesis imperfecta, and Marfan syndrome.

This syndrome has been identified by a variety of names: “Hypermobility syndrome,” “Hypermobility joint syndrome,” “BJHS,” etc.

Typically the patient presents the history⁴ of joint pain aggravated by use and often associated with a sensation of joint swelling that lasts for hours and a sensation of stiffness. Symptoms frequently involve the knees, finger, and elbows. The diagnosis is very often made by exclusion of other diseases, probably due to the lack of laboratory or radiological findings.

Hip and shoulder are also involved and acute intermittent low back pain without structural abnormality may also be frequently present. Morning foot pain is also a common complaint.

Diagnosis is made by the Beighton’s scale^{1,5,6} because this scale is easy to perform in clinic and cover small, big spinal joints and is gaining popularity. It has got total 9 points. A score of 4 or 5/9 should be the diagnostic criteria for BJHS. The Brighton’s score is nowadays gaining popularity as well.

Very often there is a positive family history and there is a female^{7,8} preponderance. The BJHS has been described⁹ as an autosomal dominant trait of distinct familial disorder, but there are examples of autosomal recessive cases¹⁰ as well as sex-linked hereditary cases.¹ About 4 to 7% in general population are having lax joints.⁷

The rehabilitative management modalities so far have been in practice are targeting mainly on the locomotor symptoms,^{1,4,11} which are mainly the joint pain, muscle

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cramps or fibromyalgia like symptoms, sense of swelling of joint and stiffness.

Management Approaches

- Believe the patient's complaints with empathy (the absence of physical findings may lead to an erroneous diagnosis of neurosis)
- Reassurance of the patient and proper explanation of the disease to the patient
- Exploration of the aggravating and relieving factors and modification of activity of daily living accordingly
- Education of the patient regarding joint protection techniques
- Therapeutic exercise: (a) Aerobic exercises (for conditioning of body, to reduce the body weight). (b) Comprehensive muscle strengthening exercises accordingly to compensate the laxity of ligaments of joints in protecting the joints
- Nonsteroidal anti-inflammatory drugs and/or anti-depressants or anxiolytics if required
- Use of protective and/or assistive devices
- Vocational modification if required
- Workplace rearrangement accordingly if required

But there is a dilemma in the therapeutic exercise part of the management as most of the literatures have not given much idea of the type of the exercise and the groups of muscles to be strengthened and on the contrary some authors⁴ prefer the strengthening of extensor groups of muscles only, we presume that strengthening of both the extensor and flexor will be more beneficial because it will provide balance between two opposing groups of muscle and hence, give more stability to the joint. And last but not the least, the exercises, which may lead, directly or indirectly, to stretching of tendon or joint ligament should be avoided and in this context, the isometric strengthening exercise would be the exercise of choice.

REVIEW OF LITERATURE

The hypermobility syndrome or BJHS has been cornered in a critical position of controversy regarding its etiology, clinical presentation, and management by different researchers with their research works, after being recognized by Kirk et al² as a syndrome presented by generalized joint laxity with musculoskeletal complaints in an otherwise normal subject, and this is nothing but an indication of the demand of more and more works on this topic.

Jessee et al⁸ furnished the proof that it is nothing but one extreme range of normal joint motion and not a systemic connective tissue disorder.

Kirk et al² from one of their study on rheumatology outpatient department (OPD) patients with hypermobile joints found the occurrence of early onset of degenerative joint disease (even at the age of 30 years).

About 80% of all humans will at some point of time in life be affected by low back pain; Biering-Sørensen,¹² found that men with hypermobile backs are more likely to develop low back pain.

In the modern era, few new diseases are emerging as a curse of modernization and one of these is again related to this BJHS and is seen among computer users, named as cumulative trauma disorders or repetitive stress injury.

Beighton et al¹³ study found that females are more hypermobile than males of the same age and this laxity diminishes with aging, falling rapidly as childhood progressed and more slowly during adult life and was described by Lewkonia¹⁴ that it sometimes became problematic to diagnose the cause of joint pain in the apparent absence of frank joint laxity in a case, with a past history of joint hypermobility.

Assessment Scales

There is no single universally accepted diagnostic criteria or scale for BJHS. Bulbena et al¹⁵ studied the existing scales made by Carter and Wilkinson,¹⁶ Beighton et al,¹³ and a basic set of criteria to define hypermobility was proposed, which could show better internal reliability and homogeneity and be more suitable for screening studies. Bird¹⁷ in his report said about the formation of hypermobile patient support group. In that meeting another new criteria was discussed by Grahame and Bird.¹⁸ The Brighton Score, a modified and revised criterion was introduced by Graham,^{19,20} which would be suitable for assessing pauciarticular or localized joint hypermobility. It consists of two major and eight minor criteria. Though in its preliminary studies it has been showing validity and reliability but till now it is not widely used and the old and time tested Beighton's scale^{6,13} (modification of the scale of Carter and Wilkinson) is still popular among the researchers. Baum and Larsson²⁰ are supporting the use of Brighton scale for the assessment of BJHS. Bird and Barton²¹ also found it as easily applicable, quicker to perform, and suitable for generalized joint laxity cases with expending less time (approximately only 1 minute per subject), can be applied for large-scale epidemiological study also and it is highly accurate as well. Both system correlated well ($p < 0.001$) with a "global index" of joint laxity derived by adding the arcs of movement recorded at most joints in the body by the method of the American

Academy of Orthopaedic Surgeons (1965). The Beighton scale of scoring system produced better correlations and is the method of choice for assessing generalized joint laxity.

Assessment of Impact

Arthritis Impact Measurement Scale (AIMS): It was designed and successfully tested by Meenan et al²² with the capability of measuring the health status of any individual with his physical, mental, and social well-being. It is one of the safest yardsticks, used by different workers and useful for evaluating the outcome of arthritis treatment and programs.

Rehabilitation Management

A national survey was conducted by Grahame and Bird²³ among the UK-based consultant rheumatologists regarding their perceptions about BJHS. The result revealed was not encouraging at all. The survey confirmed the previous suspicions that the BJHS is a condition that is underrecognized and underestimated by rheumatologists. There was little sign of awareness of the findings of recent published studies. There was no uniformity and rationality of the treatment as well.

Sheon et al⁴ advised a management protocol which includes

- Reassurance
- Joint protection technique education
- Therapeutic exercises
- Conditioning activities, such as swimming, walking, and skating
- Nonsteroidal anti-inflammatory

Cordery²⁴ described elaborately all the aspect of joint protection techniques, range of motion exercise, and muscle strengthening exercise for general endurance and stability of the joints.

Larsson et al²⁵ advocated strengthening exercise for spinal flexor and extensor muscles in case of low back pain, which he found very much helpful.

AIMS AND OBJECTIVES

To study the comparative efficacy of strengthening exercise between extensor group of muscles and both flexor and extensor groups of muscles in patient presenting with symptoms in affected joints of BJHS.

MATERIALS AND METHODS

Type of Study: This was a prospective, longitudinal, analytical study.

Cases: Cases attending directly to the Physical Medicine and Rehabilitation OPD and those referred from other OPDs including Rheumatology Clinic of a tertiary care hospital, diagnosed as the patients of BJHS with locomotor symptoms.

Sample Size: Sixty-one consecutive patients were selected (though in the protocol, we intended to have 40 patients for the study, but due to availability of patients and with an intension to get a statistically significant result, we included few more patients).

Period of Study: Six months

Inclusion Criteria

- Beighton's score 5/9 or above
- Both sexes
- Age range from 10 to 50 years
- Presence of locomotor symptoms
- Informed consent

Exclusion Criteria

- Primary inflammatory conditions of joints
- Hereditary disorders of connective tissue
- Significantly raised erythrocyte sedimentation rate (>30 mm in 1st hour)
- Presence of rheumatoid factor

Outline of Intervention

Explanation and Reassurance

- Explanation of the cause of the disease to the patient
- Reassurance regarding the benign nature of the disease

Patient Education

- Modification of activities of daily living
- Joint protection techniques

Therapeutic Exercise

- Muscle strengthening
- Conditioning activities

Drug Treatment: With nonsteroidal anti-inflammatory drug if necessary for a short period.

MATERIALS AND METHODS

All the consecutive patients attending Physical Medicine and Rehabilitation OPD and those referred from other OPDs including Rheumatology Clinic were screened according to the inclusion criteria and a total of 61 patients were selected after fulfillment of the Beighton's criteria, score of which was considered to be 5/9 or more.

Beighton’s Scale

Item	Score/Point	
a) Passive dorsiflexion of 5th finger (at metacarpophalangeal joint—90° or more)	Left 1	Right 1
b) Passive apposition of thumb to forearm 1	Left 1	Right 1
c) Hyperextension of elbow (10° or more) 1	Left 1	Right 1
d) Hyperextension of knee (10° or more) 1	Left 1	Right 1
e) Touching the palms on floor by Flexing trunk and extending knees	1	
	Total	9

The age range of the selected patients was kept in between 10 and 50 years because the children up to the age of 5 years usually show some degree of natural laxity of joints,⁷ the children in between 5 and 10 years may not be able to understand the different questions of the scoring scales. And the persons around the age of 50 may have some undetected degenerative joint disease.

Assessment of the symptoms and signs was done by the following ways:

Sites: All the affected joints were taken in to the study.

Joint pain and muscle pain: By applying linear analog scale because it was easy to perform, even the children or uneducated patients could understand it.

The physical, functional, psychosocial impact: The AIMS was used for assessment because though the study was not on arthritis the impact of the locomotor symptoms was almost similar to that of arthritis, hence, was considered suitable for this study also.

The swelling of the joint: By grading of swelling of joints clinically.

Degree of Joint Swelling

Grade 0: No swelling

Grade I (Mild): Definite swelling but no blurring of normal skeletal outline

Grade II (Moderate): Definite obscuring of skeletal outline

Grade III (Severe): No discernable skeletal mark

The stiffness of the joint: By assessing the range of motion of affected joints and in case of spine, by modified Schober’s test.

Modified Schober’s Test

Patient is standing or sitting with a maximally possible flexed trunk.

Mark any upper sacral spinous prominence; mark out three 10 cm. Segments up the spine.

Remeasure on erect posture.

- Lower segment should shorten by at least 50%
- Middle segment should shorten by at least 40%
- Upper segment should shorten by at least 30%

Intervention protocol followed was the following:

Explanation and reassurance: The basic cause of the disease was explained to the patient and the patient was reassured about the benign nature of disease.

Patient education: The patients were educated regarding the different techniques of protection of joints, such as avoidance of sitting “Indian Style” with legs crossed to avoid the undue stretching of the collateral ligaments of knee. Patients with genu recurvatum were taught to flex the knees slightly when standing still and to use shoes with arch support where flat foot with pain was detected. The patients, with the jobs requiring repetitive finger motion, were advised to interrupt the movements frequently to prevent joint injury.

Therapeutic exercises: The patients were trained with demonstrations, the isometric strengthening exercises for the muscles surrounding the affected joints, and were explained to them that this would in turn compensate the deficiency of the function of the ligaments. Isometric strengthening exercises for para spinal muscles and anterior abdominal muscles would be of very much beneficial to protect spine in the same way were also explained to them.

For therapeutic exercise, the 61 patients were grouped into two groups randomly. Group I (30 patients)—the patients of this group were instructed to perform isometric strengthening exercise for extensor group of muscles. Group II (31 patients)—these patients were advised to do isometric strengthening exercise for both the extensors as well as flexors.

The exercises instructed were isometric contraction for 5 seconds followed by 5 seconds of relaxation, each cycle would be repeated for 15 times per sitting, thrice a day and at least 5 days a week for a period of 6 weeks.

Conditioning exercise for increasing endurance by daily walking for about 30 minutes was also advised.

Medication: Treatment with nonsteroidal anti-inflammatory analgesics was used initially for those selected patients who complained the pain as unbearable. Ibuprofen was actually prescribed for a very short period as per need.

Assessment: It was a 3-point study. First follow-up was done after 3 weeks and final assessment was performed after 6 weeks. Since the response was not very appreciatively different from the initial assessment, the results were depicted only after 6 weeks of intervention.

Statistical Analysis

Descriptive statistics has been calculated. The standard deviation and frequency distribution have been found out. To see the significant differences within the variables for pre- and postobservation, we have applied Paired t-test and Wilcoxon sign rank test. And to see the difference between the responses to the strengthening exercises of extensor groups of muscles vs that of combined extensor and flexor groups of muscles we have applied Student's t test (unpaired). For categorical data we have applied χ^2 test and McNemar- χ^2 test to see the association between the variables. $p = 0.05$ has been considered as statistical significant level. Statistical Package for the Social Sciences version 7.5 has been used for statistical analysis.

OBSERVATIONS AND RESULTS

A total of 61 patients were selected for this study of whom 24 were male (39.34%) and 37 were female (60.65%). The age distribution was as follows: 8 (13.1%) patients were found between the age of 10 and 12 years, 31 (50.8%) were between the age of 21 and 30 years, 12 (19.6%) were between 31 and 40 years, and 10 (16.3%) were found to be between 41 and 50 years of age (Table 1).

Comparison of Responses between Groups I and II

On comparison between the results of the two groups, i.e., patients, who have done the strengthening exercise of extensor group of muscles (group I) vs those with the strengthening exercise of combination of both extensor and flexor groups of muscles (group II), it has been found that there was a definite difference between the two groups, but except for the pain in AIMS scale, all other items were statistically not significant (p-value obtained by χ^2 test with Yate's correction factors) (Tables 2 and 3).

So far the number of responders was concerned, it has been well documented from the above tables that 28 patients out of 31 from group II with pain (measured in visual analog scale), responded to therapy (90.3%). The affected patients of group I were 30 in number and responders were 21 (70%). The difference between the groups were found not significant ($p = 0.09$)

Reduction of joint swelling was documented by 8 patients out of 13 (61.5%) in group I and 6 patients out

Table 1: Age distribution among the cases

Age group (years)	No. of patients	Percentage
10–20	8	13.1
21–30	31	50.8
31–40	12	19.6
41–50	10	16.3

Table 2: Differences of results between the two groups in pain, swelling, and stiffness

Ailment	Extensors	Extensors + flexors	p-value
Pain relieved (visual analog scale)	21 (in 30), i.e., 70%	28 (in 31), i.e., 90.3%	0.09
No change of pain	5 (16%)	2 (6.45%)	
Increase of pain	4 (13.3%)	2 (3.2%)	
Reduction of swelling	8 (in 13), i.e., 61.5%	6 (in 8), i.e., 75%	0.65
No change in swelling	3 (23%)	2 (25%)	
Increase of swelling	2 (15.3%)	0 (00%)	
Spine stiffness relieved	7 (in 8), i.e., 87.5%	8 (in 8), i.e., 100%	1.00
No relieve of stiffness	1 (12.5%)	0 (00%)	
Increase of stiffness	0 (00%)	0 (00%)	

Table 3: Difference between the results of two groups assessed by AIMS scale

Ailments	Extensors	Extensors + flexors	p-value
Mobility	3 in 6 (50%)	4 in 4 (100%)	0.20
Physical activity	16 in 25 (64%)	25 in 30 (83.3%)	0.18
Dexterity	1 in 2 (50%)	7 in 7 (100%)	0.22
Household activity	4 in 7 (57.1%)	11 in 14 (78.5%)	0.61
Social activity	14 in 29 (48.2%)	19 in 31 (61.2%)	0.45
Activities of daily living	1 in 1 (100%)	1 in 1 (100%)	1.00
Pain	18 in 30 (58%)	27 in 31 (90%)	0.03
Depression	18 in 30 (60%)	23 in 30 (76.6%)	0.27
Anxiety	18 in 30 (60%)	23 in 31 (74.1%)	0.36

of 8 (75%) in group II and the difference showed a non-significant "p-value" of 0.65.

In group I, the stiffness of spine was shown relieved in 7 cases out of 8 (87.5%) while all of the 8 patients of group II responded (100%) but a "p-value" of 1 was obtained.

The differences assessed by AIMS scale between the two groups were found as follows:

Mobility—In group I, 3 out of 6 (50%) were responded and in group II, all of the affected 4 patients showed response (100%). The "p-value" was 0.20.

Physical activity—In group I, 16 out of 25 (64%) and in group II, 25 out of 30 (83.3%) responded with $p = 0.18$.

Dexterity—Responder was 1 out of involved 2 patient (50%) in group I and all of the involved 7 of group II responded to therapy (100%) with $p = 0.22$.

Household activity—In group I, 4 out of 7 (57.1%) and in group II, 11 out of 14 patients (78.5%) showed response with $p = 0.61$.

Social activity—In group I, 14 out of 29 (48.2%) responded while 19 out of 31 (61.2%) of group showed response with a "p-value" of 0.45.

Activities of daily living—Only one patient in each group was affected and both of them responded to therapy with $p = 1.00$.

Pain—Only in this parameter, assessed by AIMS scale, showed a significant difference in response between the two groups with a “p-value” of 0.03. The responders were found to be 18 out of 30 in group I (58%) and 27 out of 31 (90%) in group II.

Depression—Out of affected 30 patients, 18 patients showed improvement (60%) in group I, and in group II, 23 patients out of 30 (76.6%) responded to therapy with $p = 0.27$.

Anxiety—In group I, 18 patients showed response out of involved 30 (60%) while 23 were found responded out of 31 (74.1%) in group II and the difference showed a “p-value” of 0.36.

DISCUSSION

So far the comparison between the two groups (patients performed strengthening exercise of extensor muscles only and those exercised for both extensor and flexor muscles) was concerned, the group that strengthened both the extensor and flexor muscles demonstrated relatively better result. Though quantitatively the response was not so remarkable, the number of responders was definitely more than that of the other group.

In this comparative study, the difference between the two groups was found not significant statistically (except pain measured by AIMS scale) but the trend, definitely is suggestive of superiority of exercise of extensor and flexor muscles in combination. A larger sample size probably would be required to have a statistically significant result.

The reason of getting a statistically significant result in case of pain when measured in AIMS scale but not in visual analog scale might be due to the difference in sensitivity between the scales because both the scales were based on subjective perceptions of the individual patient.

So we can say that the outcome of this study is clearly indicating that the rehabilitation protocol prescribed here is very much suitable both quantitatively and qualitatively for the patients of BJHS and isometric strengthening exercise for both extensor and flexor muscles is superior than that of strengthening exercise of extensor muscles only.

CONCLUSION

The response to therapy showed by the group, that performed strengthening exercise of both the extensor and flexor group of muscles were found better than the group that exercised their extensor group of muscles only in the rehabilitation of the patients presenting with locomotor symptoms of BJHS.

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