Original Article

The Response of Rehabilitation Management in Patients Presenting with Locomotor Symptoms of Benign Joint Hypermobility Syndrome

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Abstract

The benign joint hypermobility syndrome (BJHS) was first addressed by Kirk as a distinct pathology in 1967, as the presence of rheumatic symptoms with generalised joint laxity in the absence of any demonstrable systemic rheumatic disease.

In this prospective, longitudinal, analytical study, we tried to find out the response of rehabilitation therapy in patients presenting with locomotor symptoms of BJHS and selected 61 patients randomly. The rehabilitation protocol followed: Explanation and reassurance, teaching of joint protection techniques and work modification, isometric muscle strengthening exercise (both extensor and flexor muscles), endurance exercise. Clinically most of the patients showed significant overall response quantitatively, in all the parameters.

It can be concluded that the rehabilitation protocol prescribed here is very much suitable both quantitatively and qualitatively for the patients of BJHS.

Key words: **Benign joint hypermobility syndrome, locomotor symptoms rehabilitation theropy.**

Introduction:

Hypermobility of the joint as health entity has been recognised in the ancient human society since before the Christ era, even at the time of Hippocrates[1,2]. He commented on the disadvantage of this looseness of joint of Scythians in drawing the bowstring or hurl a javelin. Kirk *et al*[3,4] addressed it as a distinct pathology, in 1967, as the presence of rheumatic symptoms with generalised joint laxity in the absence of any demonstrable systemic rheumatic disease. Beighton[5] identified this, for the first time, as a syndrome, in the International Nosology of Heritable Disorder of Connective Tissue and declared 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. Now-a-days the term benign joint hypermobility syndrome (BJHS)is gaining popularity and is used worldwide.

The BJHS has been described[6] as an autosomal dominant trait of distinct familial disorder but there are examples of autosomal recessive cases[7] as well as sex linked hereditary cases[8]. About 4% - 7% in general population are having lax joints[9].

Diagnosis is made by the Beighton's scale[8,10,11] because this scale is easy to perform in clinic and cover small, big and spinal joints and is gaining popularity. It has got total 9 points. A score of 5/9 or more should be the diagnostic criteria for BJHS.

Though it is an age old disease, but till date, it has got no comprehensive full-proof rehabilitation protocol. The rehabilitative management modalities so far have been in practice are targeting mainly on the locomotor

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symptoms[1,8,12] which are mainly the joint pain, muscle cramps, sense of swelling of joint and stiffness.

Review of Literature:

The hypermobility syndrome or (BJHS), has been acornered in a critical position of controversy regarding its etiology, clinical presentation, and management by different researchers with their research works, after being recognised by Kirk *et al* as a syndrome presented by generalised joint laxity with musculoskeletal complaints in an otherwise normal subject.

Graham. *et al*[10,13] from their study suggested that the so called hypermobility syndrome, far from being a benign locomotor disorder of healthy persons, may be a forme fruste of a hereditary disorder of connective tissue. Handler *et al*[14] found an apparent deficiency of collagen type I with over production of collagen Type III from skin biopsy and suggested that this collagen abnormality is also the cause of associated mitral valve prolapse. On the other hand, Jessee *et al*[15] furnished the proof that it is nothing but one extreme of range of normal joint motion and not a systemic connective tissue. This view was supported by the work of Mishra *et al*[16]. Biro *et al*[9] also reserved the same opinion. Charpel . and Marks.[17] from their study found that there is an increased incidence of joint injury and joint degeneration in BJHS. Kirk *et al*[4] from one of their study on Rheumatology OPD patients with hypermobile joints found the occurrence of early onset of degenerative joint disease (even at the age of 30 years) and there was also the evidence of tendonitis, joint effusion, recurrent dislocation, tennis elbow and painful Achilles tendon, though theESR was Below 30 mm in 1st hour. Several researchers[4,12] noted also that high proportion of patients with BJHS had mobile flat feet, hindfoot valgus and hypermobile ankles. Biering-Sorensen[19], who found that men with hypermobile backs are more likely to develop low back pain. It is also found by Pascarelli and Kella[20] with clinical as well as recorded video-tape examination that many cases of cumulative trauma disorders (CTDs) or repetitive stress injury (RS) are related to BJHS. Beighton and Horan[6,7] found that the syndrome is an autosomal dominant familial disorder which is supported by almost all the researchers. They also reported cases with autosomal recessive trait. Frank Biro *et al*[9] observed that about 4 -7% of general population are hypermobile with an male/female ratio of 4:11. Beighton, *etal*[18] found that this laxity diminishes with aging.

The Brighton Score, a modified and revised criterion was introduced by Graham[21,22], which would be suitable for assessing pauciarticular or localised joint hypermobility. It consists of 2 major and 8 minor criteria. The old and time tested Beighton's Scale[11,18] (modification of the scale of Carter and Wilkinson) is popular among the researchers till date which covers the small and big joints as well as the spinal joints. It is easy to perform with in minutes.

Very little work has been done so far, regarding the management of this BJHS, as a result this aspect is less enlightened till date. Sheon *et al*[1] advised a management protocol which includes:

1) Reassurance, 2) joint protection technique education, 3) therapeutic exercises, 4) conditioning activities such as swimming, walking and skating etc, 5) non-steroidal anti-inflammatory drugs.

Aims and Objectives:

To study the response of rehabilitation management in patients presenting with locomotor Symptoms of benign joint hypermobility syndrome.

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, diagnosed as the patients of BJHS with locomotor symptoms.

Sample size: Sixtyone 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).

Study duration : Ten months (Nov 2000 to Aug 2001).

Place of study: Dept. of P.M.R., A.I.I.M.S., New Delhi.

Inclusion criteria :

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

Exclusion criteria:

- Primary inflammatory conditions of joints

- Hereditary disorders of connective tissue
- Significantly raised ESR (>30 mm in 1st hour)
- Presence of rheumatoid factor.

Outline of intervention:

A) Explanation and reassurance-

Explanation of the cause of the disease to the patient

Reassurance regarding the benign nature of the disease

B) Patient education- 1)Modification of ADL 2) joint protection techniques

C) Therapeutic exercise - 1)Muscle strengthening 2) conditioning activities

D) Drug treatment - With NSAID,if necessary for a short period.

Methodology:

All the consecutive patients attending PMR 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.

The age range of the selected patients was kept in between 10 years to 50 years because the children up to the age of 5 years usually show some degree of natural laxity of joints (Biro *et al*),[8] the children in between 5 and10 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 analogue scale because it was easy to perform, even the children or uneducated patients could understand it.

The physical, functional, psychosocial impact : Arthritis impact measurement scale (AIMS) was used for assessment because, though the study was not on arthritis but 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 (grade 0 – III).

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

Intervention protocol followed was the following :

Explanation and Reassurance : The basic cause of the disease was explained to the patients 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. Patient 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 paraspinal muscles and anterior abdominal muscles would be of very much beneficial to protect spine in the same way were also explained to them.

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 non-steroidal 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 post observation, we have applied "paired t test" and "Wilcoxon sign ranktest". For categorical data we have applied x2 test and McNemar-x2 test to see the association between the variables. P=0.05 has been considered as statistical significant level. SPSS – 7.5 has been used for statistical analysis.

Results:

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

Symptomatologically it was found that there was a wide distribution of symptoms among multiple sites (Table-1) with multiple combinations of joint involvement.

Table 1: *Distribution of Sites of Involvement.*

Site	No of patients	Percentage	Single site involvement
Spine	50	81.9 %	Only spine – 4 cases
Small joints	28	45.9 %	Only small joints - 1 case
Big joints	56	91.8 %	Only big joints - 7cases

The pain was found to be present in all the 61 cases (Table 2).

Table 2: *Distritution of Cases according to Visual Analogue Scale (n = 61)*

Visual analogue scale	No.of.Cases (%)
0 - 2	1 (1.6 %)
2 - 4	11 (18 %)
4 - 6	23 (37.7 %)
6 - 8	19 (31.1%)
8 - 10	7 (11.4 %)

Joint swelling was detected in 22 (38.6%) patient out of 57 cases with symptomatic joints. All the swelling were very mild and of grade I only. A majority of the patients (20) were presenting with swelling of big joints (Table-3).

Table 3: *Distribution of Sites of Joint Swelling*

Joints involved	Sites	No of patients	Percentage
Big joints	Knees	16	76.19 %
	Ankles	4	19.00 %
Small joints	P I P Joints	2	09.52 %

One interesting thing was that there was no restriction of range of motion over small or big joint of any of the patients. The spines were definitely showing a very mild degree of restriction of range of motion present only over terminal ranges found to be present in only 16 patients (32.6%) (out of 49 cases with symptomatic spines) and 9 of whom were male and 7 were female. Interestingly only the lumbar spines were involved in all the 16 cases.

The questionnaires of AIMS which were given to the patients for filling up, revealed the facts as depicted in Table - 4.

Table 4: *Distribution of Impact of BJHS on Physical and Psychosocial Activities.*

Parameters	No of Patients Affected	Percentage
Mobility	10	16.3 %
Physical activity	55	90.1 %
Dexterity	9	14.7 %
Household activity	21	34.4 %
Social activity	60	98.3 %
Activities of daily living	2	3.2 %
Pain	61	100 %
Depression	60	98.3 %
Anxiety	61	100 %

Final assessment: The final assessment was done after 6 weeks of the rehabilitation therapy and the result obtained were quite encouraging and as follows:

Overall response to rehabilitation therapy- The overall assessment of pain with the help of visual analogue scale (all of the 61 patients were having joint pain) showed that 49 patient (80.3%) responded with decrease of pain to various degres (Table 5). Only 7 (11.4%) complained of no change in pain status and 5 patients (8.1%) among them said that the pain actually aggravated. Among the responder, 49% of patients reported that quantitatively, there was no pain at all and 68.8% of patients said that more than half of their pain had been disappeared and a decrease of more than 10% of pain was reported by 80.3% of the patients. Quantitatively the response was showing a p-value <0.001.

Regarding joint swelling (out of 57 patients with symptomatic joints, 21 were having joint swelling), 14 patients (66.6 %) showed total disappearance of the swelling and there was no reduction of swelling in 7 patients (33.3 %), even 2 of them actually were showing increasing of swelling from grade I to grade II. The p-value was found to be <0.001.

Stiffness of the spine (out of 50 patients with symptomatic spine, 16 were having stiffness) was relieved in 15 patients (93.7 %) ($p < 0.001$) as shown by the free movement of spine to its extremes of ranges as documented with the help of Schober's test. Only 1 patient (6.2 %) was still having the restriction of movement of spine as before.

Table 5: *Improvement of Joint Pain, Swelling and Stiffness of Spine.*

Parameters	Affected	Improved	Percentage
Pain (V A Scale)	61/61	49	80.3 %
Swelling of joint	21/57	14	66.6 %
Stiffness of spine	16/50	15	93.7 %

The scores obtained from the AIMS scale after the completion of the rehabilitation programe was also favourable and depicted in Tables 6 and 7.

Table 6: *Improvement Asessed with AIMS Scale.*

Parameters	Affected	Improved	Percentage
Mobility	10	7	70 %
Physical activity	55	41	74.5 %
Dexterity	9	8	88.8 %
Household activity	21	15	71.4 %
Social activity	60	33	55 %
Activities of daily living	2	2	100 %
Pain	61	45	73.7 %
Depression	60	41	68.3 %
Anxiety	61	41	67.2 %

Medication: Ibuprofen as non - steroidal anti - inflammatory analgesic was chosen because it was time tested and potent drug. Only 9 patients (male-1, female-8) were instructed to take oral i buprofen-400 mg. thrica daily for 5 days on S O S. basis (in cases with intolerable pain), 6 of them responded well (reported on the first follow up after 3 weeks). The remaining 3 patients were advised to continue the same medication for another 5 days with no improvement in the pain status even after 10 days of medication (reported on an early S O S visit).

Quantification of the impact of BJHS on the patients with the different scoring systems applied before and after the therapy also showed favourable outcome. The number of patients responding, 10 %, 50 % or 100 % quantitatively to the rehabilitation therapy in different activity components of AIMS scale and also to pain, joint swelling and range of motion were analysed and found to be significantly indicative of the success of the therapy. The Tables 6 to 8 are showing the detailed accounts of the responses in different sectors after therapy.

Table 7: *Quantitative Response with Pre and Post Therapy Differences.*

Parameters	No of cases	Mean + SD	p values
Mobility	61	Pre 4.26 ± 0.68 Post 4.16 ± 0.60	0.090
Physical activity	61	Pre 7.40 ± 1.18 Post 6.09 ± 1.35	0.001
Dexterity	61	Pre 5.22 ± 0.64 Post 5.14 ± 0.81	0.552
Household activity	61	Pre 7.98 ± 1.63 Post 7.45 ± 1.28	0.007
Social activity	61	Pre 11.73±4.34 Post 10.27±3.76	0.001
Activities of daily living	61	Pre 4.04 ± 0.28 Post 4.00 ± 0.00	0.182
Pain	61	Pre 15.49±3.58 Post 8.34 ±5.87	0.001
Depression	61	Pre 17.55± 6.13 Post 11.62±7.25	0.001
Anxiety	61	Pre 19.09 ± 5.23 Post12.34±6.46	0.001
Pain (V A Scale)	61	Pre 6.00 + 1.91 Post2.36 + 3.15	0.001*
Swelling	57	Pre 0.36 + 0.48 Post0.14 + 0.39	0.001*
Spine range of motion	50	Schober's test	0.001#

(* Paired t-test / # McNemar & x2 test)

Table 8: *Amount of Improvement Assessed with the Help of AIMS Scale.*

Parameters	No of Patients improved	10% or more improved	50% or more improved	100% improved
Mobility	7/10	7 (100%)	7 (100%)	7 (100%)
Physical activity	41/55	41 (100%)	39 (95.1%)	24 (58.5%)
Dexterity	8/9	8 (100%)	8 (100%)	8 (100%)
Household activity	15/21	15 (100%)	14 (93.3%)	10 (66.6%)
Social activity	33/60	32 (96.9%)	8 (24.2%)	0 (00%)
Activities of daily living	2/2	2 (100%)	2 (100%)	2 (100%)
Pain	45/61	44 (97.7%)	41 (91.1%)	27 (60%)
Depression	41/60	41(100%)	36 (87.8%)	2 (4.8%)
Anxiety	41/61	40 (97.5%)	39 (95.1%)	8 (19.5%)
Pain (Visual analogue scale)	49/61	49(100%)	42 (68.8%)	30 (49%)
Joint swelling	14/21	14 (100%)	14 (100%)	14 (100%)
Stiffness of spine	15/16	15 (100%)	15 (100%)	15 (100%)

Discussion:

The genuineness of any rehabilitation protocol always demands a positive outcome, whatever the quantity of response may be. As a result, the quantification of the impact of the disease is very important along with the quantification of the response of the rehabilitation management, which helps to assess the success of the therapy. Our effort in this study was to highlight this aspect of BJHS, which is still lying untouched by researchers. Unfortunately not a single study has been available so far with quantitative assessment of the locomotor symptoms of BJHS patients with its management protocol along with the quantification of the response of that protocol. Only the epidemiological data which, fortunately were collected by previous researchers, were available to compare our data.

In this study the findings of female predominance (60.6%) was consistent with the findings of previous workers[1,9,23]. We found a lower Beighton's score in older patients and higher in youngers and this is also consistent with the previous findings[18,24,25] indicating loss of joint hypermobility with increasing age. Beighton *et al* [5,6,18] found from their previous survey that a lower Beighton score is more predominant among the hypermobile subjects and the number of subject is inversely proportional to the Beighton's score which we found to be true in our subject group also. We found that the BJHS is predominant among the members of economically well to do families with sedentary work habits and this fact is supporting the views of previous workers[1].

In our study, joints were found involved in combination and a single type of joint (small joint or big joint or spine) involvement were found comparatively less in number and the similar observation was also found by Larsson *et al*[25]. We found a good number of subjects affected in the age group of 21- 30 years, this is probably the most appropriate age group in our country when a person becomes engaged in work for the first time (in a new job or in female as house-maker after marriage) and it supports the views of Sheon *etal*[1] who observed that the symptoms usually start on initiation of work by a person previously engaged in sedentary work only.

All of our patients were complaining of pain of varying degree around the joints as was observed by almost all the previous researchers. Typical morning stiffness that was described by previous observers[1] was a very common complaint reported by our patients also, which usually was lasting for ½ hour to 1 hour on an average as described by them. The pain also was found typically after sudden over-use in otherwise sedentary type of individual or in the patients who had to maintain a particular posture for an abnormally prolonged period and was typically found decreasing on rest. The pattern of joint involvement was variable regarding the number and site of joint and the intensity or amount of pain. The degree (amount, measured both by visual analogue scale and by AIMS scale) of pain was also variable with no joint specificity, no relation with Beighton score and no relation with age or sex as well.

Previous studies[1,8] showed that the incidence of synovial swelling or effusion was very rare and swelling of joint was present in very few cases (and that also with milder degree). This was supported by our findings of about less than 1/3rd cases (36.8%) of joint swelling and all of them were within degree I only. Mainly the big joints were involved (knee-16, ankle-4 and small joint-2 only). The relatively lower involvement of small joint swelling is probably due to the frequent movement of those joints in daily activities causing failure of oedema fluid to accumulate.

The restriction of range of motion was not demonstrated by any of our patients but in very few cases there was a terminal voluntary resistance due to anticipation of pain, which overcome later on with reassurance. Theoretically this was expected, as the ailment itself was due to laxity of joint and until or unless the joint tissue injury was quite severe one should not expect a clear and demonstrable restriction. In one of his case reports Russek [26]also explained the similar finding in the same way.

Though 49 cases were having pain over spine, only 16 patients were showing stiffness or restriction of spinal mobility and all of them were over lumbar spine. We assume, unlike other weight bearing big joints, the spine; especially the lumber spine is very much susceptible and sensitive to injury because it is the main pillar of weight bearing with a multi-axial and complex kinesiology, making it vulnerable to easy ligamentous injury, which was reflected in the spinal stiffness of the above patients.

The areas of impact of BJHS, measured by AIMS scale showed variable amount of involvement. Only 2 patients indicated a very mild degree of difficulty in activities of daily living. There was negligible degree of involvement in the sector of mobility and dexterity but the number of patients suffering were a little more, 10 and 9 respectively. Previous studies[1,8] also did not mention such involvement. A little more number (n=21) of patients was seen affected in the sector of household activities but the amount of involvement was negligible and was seen mild difficulty in mopping, washing, etc. A large number of cases (n=55) were seen complaining of variable degrees of difficulties in physical activities and most of them were showing quite a higher score. Difficulties in stair climbing, bending or stooping of body, lifting heavy objects, running or other such vigorous activities were seen contributing for the higher score. Social activities were affected in almost all of the patients (n=60) with variable degree of severity and we assume from thorough examination and detailed counselling that it was indirectly contributed due to the presence of higher degree of depression and anxiety associated with BJHS. We found all of the 61 cases were having anxiety and 60 were suffering from depression. This finding was consistent with the reports published by Bulbena etal[21]and Russek[8]. It is very much rational that any patient suffering from a disease which is, though milder in form but having a chronic course and undiagnosed by majority of the attending physician will suffer from anxiety along with a good degree of reactive depression and BJHS fulfills all of the above factors.

After 6 weeks of rehabilitation therapy as advised was completed by the patients, we found an encouraging outcome in overall response. Though the mobility, dexterity, household activity and activities of daily living were found statistically not significant due to less number of patients affected, the response of therapy was remarkable both in number of responders as well as in amount of response. We expect from this trend that if the sample size would be a little more, all the above results would be significant statistically. Rest of all other parameters were also found responded very well in both degree of response and number of responders and were statistically very much significant.

The reason of getting a statistically significant result in case of pain when measured in AIMS scale but not in visual analogue 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 benign joint hypermobility syndrome.

Conclusions:

The inferences made from the study are as follows :

- Benign joint hypermobility syndrome(BJHS)is more predominant in female.
- Hypermobility declines with age.
- A higher Beighton's score is relatively less among subjects of BJHS.
- Younger patients usually have higher Beighton's score.
- Affects the joints mostly in combination of various joints.
- Predominantly affected age range is 21years to 30years.
- All the patients were having pain, which may be of varying degrees.
- Few patients were having swelling of joints of grade I only and majority had big joints involved, mainly the knees, few had ankles and rarely proximal interphalangeal joints.
- No incidence of restriction of range of motion except few cases of lumbar spine mobility restriction was documented.
- Clinically most of the patients showed remarkable overall response quantitatively to rehabilitation therapy in all the parameters.

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