

Pain, Balance, Activity, and Participation in Children With Hypermobility Syndrome

Elke Schubert-Hjalmarsson, PT; Anna Öhman, PT, PhD; Märten Kyllerman, PhD; Eva Beckung, PT, PhD

Departments of Physiotherapy (Ms Schubert-Hjalmarsson and Dr Öhman) and Neurology (Dr Kyllerman), Queen Silvia's Children's Hospital, Sahlgrenska University Hospital, and Institute of Neuroscience and Physiology, Occupational Therapy and Physiotherapy (Dr Beckung), Sahlgrenska Academy of Göteborg University, Göteborg, Sweden.

Purpose: To describe hypermobility, balance, pain, activity, and participation in children with hypermobility and compare these characteristics with those of a control group. **Method:** Twenty children aged 8 to 16 years with hypermobility syndrome (HMS) or Ehlers-Danlos syndrome and a control group of 24 children of the same age participated in the study. Hypermobility was assessed according to the Del Mar scale, balance was assessed with the Bruininks-Oseretsky test of motor proficiency, and participation in daily life activities was assessed with the frequency of participation questionnaire. Pain and physical activity were assessed in a diary. **Results:** In comparison with the control group, the children with hypermobility had significantly more hypermobile joints and more pain and scored lower in the balance test, and their activity was affected on a daily basis. **Conclusion:** Pain appears to affect activity and participation in children with HMS. Balance is decreased in children with HMS compared with healthy controls. (*Pediatr Phys Ther* 2012;24:339–344) **Key words:** activities of daily living, adolescent, articular hypermobility syndrome, child, disability evaluation, Ehlers-Danlos syndrome, female, male, motor activity, pain, postural balance, social environment

INTRODUCTION AND PURPOSE

Joints have a varied range of motion (ROM), depending on their function and structures; for example, the shoulder has a large ROM in several directions to allow hand and arm functions, whereas the sacroiliac joint has a

limited ROM because it has a stabilizing function. In general, children have greater ROM in the joints than adults. ROM decreases with increasing age, and girls are found to have a higher degree of ROM than boys.^{1,2} No explanation for the gender difference could be found in the literature.

Hypermobility is defined as follows: “joints which are unduly lax and the range of motion is in excess of the normal in most of the joints examined.”^{3(p419)} Some degree of hypermobility in childhood seems to be natural and does not necessarily reduce the motor competence and physical activity in children.⁴ The reported prevalence for children with hypermobility is 5.3% to 64.6%.^{5,6} The rather high variation between the reported incidences may be due to differences in performing the assessment and the age of the participants. Hypermobility as a symptom exists in several disorders, which can affect children, for example, Marfan syndrome and osteogenesis imperfect.⁷

Hypermobility syndrome (HMS) is a hereditary disorder of the connective tissues characterized by hypermobility, which affects multiple joints and is combined with pain from muscles and joints. There is no systemic inflammatory joint disease such as rheumatoid arthritis involved.⁸ In HMS, symptoms often persist and become generalized, which can cause functional problems.⁷ The prevalence is reported to be 5%.⁹

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Correspondence: Elke Schubert-Hjalmarsson, PT, Department of Physiotherapy, Queen Silvia's Children's Hospital, 41685, Göteborg, Sweden (elke.schubert@vgregion.se).

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Ehlers-Danlos syndrome (EDS) is a heterogeneous group of hereditary disorders of the connective tissues. There are different types of EDS; the most common is type III, the hypermobility type, with a prevalence of 1:10 000, characterized by hypermobility (one of the major symptoms), increased skin elasticity, and tissue fragility caused by a genetic defect in the collagen structure. It is common to be associated with chronic progressive pain, hypermobile joints, hyperelastic and/or smooth, velvety skin, easy bruising, and molluscoid pseudotumors.¹⁰ Muscle diseases may be suspected on the basis of the symptoms of muscle hypotonia. Ehlers-Danlos syndrome can also easily be mistaken for rheumatic pain. The symptoms can vary between different individuals and also within individuals in the same family.¹⁰

It is believed that HMS and EDS, hypermobility type, are the same condition.¹¹ For children and youth, functional problems can occur when they have HMS, including limited physical capacity, joint problems (often feet and knees), and delayed motor development.¹⁰ There is also an increased risk of dislocation of the joints, especially the shoulder and the patella.⁷ Adults with EDS have been found to have deficits in balance,¹² and that reduced physical activity may have negative effect on balance.¹³ Children with HMS can be affected by problems in everyday living, for example, in physical education at school and other sport activities such as running.⁷ Our clinical experience is that children with HMS may experience very high fatigue after daily activities.

A study has shown that long-term pain is more common in adults with HMS (100%) than in a control group (37%).¹⁴ Only a few studies examined how HMS might affect activity and participation in activities of daily living.¹³ Some studies and case reports have indicated that physiotherapy has a positive effect on children with HMS.¹⁵ Increased muscle strength can result in better stability of the joints, and this can decrease the pain.¹⁵ More knowledge about HMS is needed to optimize physiotherapy treatment.

The aim of this study was to describe hypermobility, balance, pain, activity, and participation in children with HMS and compare these characteristics with those of a control group.

METHODS

Design

A cross-sectional study comparing the 2 groups (HMS and control) was performed.

Subjects

The caregivers of children with HMS aged 8 to 16 years who were being or who had been treated in the local health care system were asked to allow their children to participate in this study. Inclusion criteria were children aged 8 to 16 years, diagnosed with HMS or EDS, hypermobility type. Both diagnoses form the HMS group. Exclusion criteria were all other known disorders. The

children were selected by approaching caregivers or parents through local schools and hospital staff. Ninety-seven children, born between 1994 and 2001, were identified with a diagnosis of HMS or EDS. Twenty-two were excluded because of other diagnoses and 14 families had moved from the area. Sixty-one families were invited to participate. Twenty children aged 8 to 15 years (mean = 11.2 years, SD = 1.9), 8 girls and 12 boys, chose to participate. The control group consisted of 24 children aged 8 to 15 years (mean = 11.4 years, SD = 2), 11 girls and 13 boys.

The assessments were done between February and April 2009. The local ethical committee approved the study and the parents gave their informed consent.

Measurement Instruments

Hypermobility was assessed according to the Hospital Del Mar criteria for hypermobility (Del Mar scale).¹⁶ Passive ROM of spinal flexion, lateral rotation of the shoulder, elbow extension, thumb to the forearm, fifth finger extension, hip abduction, knee flexion and extension, patella movement, dorsal flexion of the ankle, and extension of the first toe was examined. The Del Mar scale has good reliability in adults.¹⁶ The scale consists of 11 items of which 5 were taken from the Beighton scale. The Beighton scale has been shown to have satisfactory validity and reliability for children.⁵

Balance was assessed with the Bruininks-Oseretsky test of motor proficiency (BOT-2). The BOT-2 assesses children's motor performance. The balance subtest is composed of 8 items: standing with feet apart on a line; walking forward on a line; standing on one leg on a line; standing with feet apart on a line with eyes closed; walking forward heel-to-toe on a line; standing on one leg on a line with eyes closed; standing on one leg on a balance beam; standing heel-to-toe on a balance beam; and standing on one leg on a balance beam with eyes closed. The stationary items are assessed in seconds (maximum, 10 seconds). The other items are measured by number of steps (maximum, 6 steps). The test has good validity and reliability.¹⁷

Over a 2-week period, pain was reported by the children or their families in a diary on 4 separate occasions each day. The intensity of the pain was documented on a scale with 5 levels (0 = no pain; 1 = pain, I am only aware of it if I pay attention to it; 2 = pain, but I can ignore it at times; 3 = pain, it is difficult for me to concentrate. I can only do easy activities; 4 = pain, such that I can't do anything). The pain diary has been proven to have good validity for children aged 9 to 17 years, with headaches.¹⁸ The pain diary is used at our clinic to measure joint and muscle pain; however, it is not validated for hypermobility.

The children also reported to what extent the pain limited their activity during the day. Pain was also assessed by pain drawings, where the patient made marks on the picture of a body to show the place and the type of pain, for example, burning or oppressive.¹⁹ No information about

validity and reliability of pain drawings has been found in the literature.

Participation in daily life activities at school and during leisure time was assessed with the Frequency of Participation Questionnaire (FPQ). The FPQ (see the Appendix, Supplemental Digital Content 1, available at <http://links.lww.com/PPT/A34>) is composed of 14 questions about frequencies of participation, each with 6 response options for different frequencies.²⁰ The questions are related to the domain “life situations” in the international classification of disabilities and health. The FPQ has been translated to Swedish and is considered to have face validity.²⁰ An additional open question about the child’s need to rest was included. The question was: “How often do you need to rest?”

Physical activity was reported in an activity diary. During a period of 2 weeks, the children reported what activities they performed and when. The families were asked to report activities like riding a bike, playing football, or walking. The activity diary is categorized into 3 effort levels. The activity diary has not been tested for validity and reliability.

Data Collection Procedure

The activity diary, pain diary, and pain figures were sent home to the families after they had agreed to participate in the study. These were completed by the children, with the help of their parents when needed. The families returned them when they came for the assessment. An independent physiotherapist, a certified assessor of the Del Mar scale, assessed all the children. The families came to the department of physiotherapy on one occasion for assessment of hypermobility and balance. The children did not perform stretching or warm-up exercises before the test. At the time of the assessment, the children completed the FPQ, with the help of their parents when needed.

Data Reduction Procedures

The results of the pain diary were analyzed. The overall pain intensity during the 2 weeks, notes on what part of the day pain occurred, and the number of hours with pain were recorded. The overall pain intensity was categorized as follows: 1: no pain; 2: pain on solitary occasions (<60 minutes); 3: a few occasions of low-intensity pain (pain-scale, 1-2); 4: a few occasions of high-intensity pain (pain-scale, 3-4); 5: 75% or more low-intensity pain all the time; and 6: 75% or more high-intensity pain all the time.

The general impression of activity during the 2 weeks was evaluated by 3 persons (2 physiotherapists and 1 occupational therapist). The therapists evaluated each child’s diary individually. The overall activity was categorized as follows: 1: low, minimally physically demanding activities (walking short distance, playing on a sandpit, and playing computer games); 2: moderately physically demanding (walking, swimming, or doing similar activities at least twice a week); and 3: substantially physically demand-

ing (ball sport, jogging, skiing, or doing similar activities at least twice a week). If the evaluators had disagreement about any child’s activity, there were discussions afterward until agreement was reached.

Data Analysis

Descriptive statistics were calculated for gender and age, and for the different categories in the pain diary. Differences between the groups in hypermobility, balance, FPQ, reported pain, and activity were analyzed with the Mann-Whitney *U* test. Multiple regressions with ANCOVA were used to compare the groups with balance, pain, and activity as covariates. SPSS 18.0 was used for statistical analyses. The selected significance level was $\alpha \leq .05$.

RESULTS

A significant difference was found in hypermobility according to the Del Mar scale between the 2 groups ($P > .001$) (Table 1). The mean score for the HMS group was 6.7 (SD = 1.9), and the mean score for the control group was 3.8 (SD = 1.6).

The BOT-2 revealed a significant difference in balance between the groups ($P > .001$). The mean score for the HMS group was 14.7 (SD = 4.0), and the mean score for the control group was 20 (SD = 3.3).

A difference between the groups in the FPQ was found in 4 of the 14 questions. A lower frequency was reported in helping with housework ($P = .004$), riding a bicycle ($P = .001$), and taking part in sport or outdoor games ($P = .05$) in the HMS group. A higher frequency was reported in playing nonsporting games ($P = .03$). The open question showed a significantly higher need to rest in the HMS group ($P = .001$). Ten children in the HMS group had a need to rest daily compared with 2 children in the control group.

Two of the 20 children in the HMS group were excluded from analysis of the pain drawing and activity and

TABLE 1
Hypermobility Evaluated With the Del Mar Scale^a

| Joint | HMS Group (n = 20) | | Control Group (n = 24) | |
|--------------------------------|-----------------------|-----|---------------------------|-----|
| | n | % | n | % |
| Hyperflexion of the thumb | 13 | 65 | 3 | 13 |
| Hyperextension of fifth finger | 9 | 45 | 2 | 8 |
| Hyperextension of the elbow | 14 | 70 | 5 | 21 |
| External rotation of shoulder | 20 | 100 | 18 | 75 |
| Trunk flexion | 0 | 0 | 2 | 8 |
| Hip abduction | 13 | 65 | 11 | 46 |
| Patellar movement | 17 | 85 | 13 | 54 |
| Hyperextension of the knee | 18 | 90 | 8 | 33 |
| Hyperflexion of the knee | 20 | 100 | 24 | 100 |
| Dorsal flexion of the ankle | 1 | 5 | 0 | 0 |
| Hyperextension of the big toe | 8 | 40 | 5 | 21 |

Abbreviation: HMS, hypermobility syndrome.

^aJoints evaluated and the number of children hypermobile in each joint in the 2 groups.

pain diary, because they did not fill in the diary. All children, except 1 in the HMS group, marked different parts of the body on the pain drawing. The most frequently marked body parts were feet and knees, followed by wrists and the shoulder area. In the control group, 11 children marked parts of the body. There was no specific part of the body that was more frequently marked than any other. The control group mostly described muscle aches or pain after minor accidents in various parts of the body.

Pain intensity was significantly different between the groups ($P > .001$). The mean score for the HMS group was 3.6 (SD = 1.6) and that for the control group was 1.9 (SD = 1.0). Distribution of pain during the day is shown in Figure 1.

There were significant differences in pain ($P > .001$), balance ($P > .001$), and activity ($P > .001$) between the groups (Tables 2 and 3).

DISCUSSION

In comparison with the control group, this study showed that the HMS group had significantly more hypermobile joints and more pain, scored lower in the balance test, and their activity on a daily basis was affected.

Balance is needed when performing a range of activities from maintenance of static positions to complex dynamic activities. Children are more active than adults, and dynamic activities are important in the development of motor skills and as part of social life in childhood. The

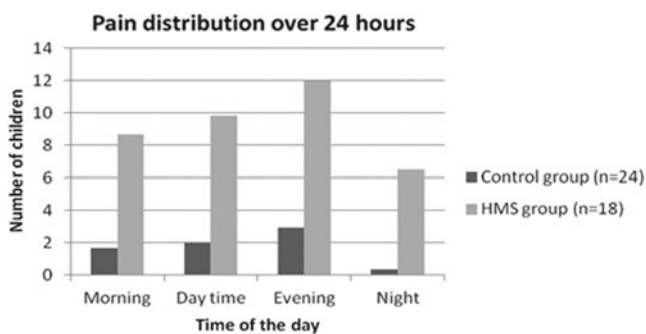


Fig. 1. Pain during different occasions of the day, mean for the 2-week period of pain registration.

HMS group performed significantly poorer in the balance test than the control group. This study supports previous work indicating that balance is affected in individuals with HMS.¹² Rombaut et al¹² found that adults with EDS had deficits in balance, gait impairment, and an increased fall frequency. They discuss that there may be a vicious circle from fear of falling, where self-limited activity results in muscle weakness, postural instability, and walking deficits with an increased risk of falling as a consequence. This could, in turn, lead to an increasing fear of falling. In another study, Rombaut et al¹³ found that reduced physical activity might have a negative effect on motor skills such as balance. Fatoye et al²¹ showed, among others, that children with HMS had an abnormal pattern of knee motion during gait compared with healthy controls. Joint instability in combination with pain may cause reduced balance.

However, Jull-Kristensen et al⁴ found in their study that hypermobility alone does not necessarily cause motor difficulties. Their findings were for children with hypermobility and not with HMS. Those children did not report any pain. Studies have shown that pain is more common for individuals with HMS than in the population without HMS.²²

The pain drawings showed a higher frequency of pain mostly in the feet and knees. Berglund et al²² specifically observed foot pain and disability in adults with EDS, and they showed that daily life activities were strongly restricted by foot pain and related disability in individuals with EDS. Pain reduction is a prerequisite to an increase in activity.

When children do not have the same level of participation in activities, it will affect their social life to a greater or lesser extent. Children in the HMS group also reported a higher frequency of need to rest, which may reduce the possibility for social contacts, for example, after school. Our clinical experience is that children with HMS may experience very high fatigue after daily activities. The result of the open question with the higher frequency of need to rest may indicate that the children in the HMS group have more fatigue. Poor motor function and musculoskeletal joint symptoms may interfere with daily life activities and might result in a less-active lifestyle.

Previous authors have found that it is possible to reduce pain for children with HMS with regular strength

TABLE 2

Mean Values, SD, and Range in the HMS Group and the Control Group Regarding Reported Pain, Pain Intensity, Limitation of Participation in Activity Because of Pain, Hours of Activity, and Level of Activity During a 2-Week Period

| | Mean \pm SD; (Range) | | P |
|------------------------------------------------------------------------------------------------|------------------------|------------------------|-------|
| | HMS Group (n = 18) | Control Group (n = 24) | |
| Number of hours when the child reported pain | 133 \pm 121; (2-336) | 11 \pm 19; (0-74) | >.001 |
| Score on overall pain intensity scale | 3.6 \pm 1.6; (1-6) | 1.9 \pm 1; (1-4) | >.001 |
| Number of days when the child reported limitation of participation in activity because of pain | 7 \pm 6; (0-14) | 1 \pm 1; (0-4) | >.001 |
| Number of hours when the child reported activity | 10 \pm 6; (2-29) | 18 \pm 7; (10-40) | >.001 |
| Score on degree of activity scale | 1.7 \pm 0.7; (1-3) | 2.5 \pm 0.6; (1-3) | >.001 |

Abbreviation: HMS, hypermobility syndrome.

TABLE 3Multiple Regressions, ANCOVA, and the Effects of Balance, Degree of Activity, and Pain^a

| Variable | Standard Error | Beta | P | Confidence Interval |
|--------------------|----------------|--------|-------|---------------------|
| Balance | 0.013 | 0.389 | 0.002 | 0.017, 0.070 |
| Degree of activity | 0.077 | 0.310 | 0.011 | 0.049, 0.359 |
| Pain intensity | 0.038 | -0.305 | 0.011 | -0.176, -0.024 |

^aGroups were used as a dependent variable.

training.¹⁵ Several types of increased physical activity may reduce pain, and there might be a spectrum to choose from for the individual. More studies about physical activities and different types of training to increase stability and strength and reduce pain are needed to find treatment strategies for children with HMS and EDS.

A significant difference was found between the HMS group and the control group in hypermobility. Reference values for the Del Mar scale have been created for adults. In some of the assessed joints with the Del Mar scale (knee flexion and external rotation of the shoulders), only a small difference or no differences was found between the groups. In almost 50% of the control group, hypermobility was found in hip abduction and patellar movement. In contrast, only 2 children, both in the control group, were hypermobile in trunk flexion. This indicates that there might be a need for special reference values for children.

Study Limitations

Because of the low number of participants, the results from this study cannot be generalized. Only a third of the invited children or families chose to participate in the study. It is unknown why some children or families chose not to take part. They may not have been experiencing problems at the time of the study and, because of this, may have been less interested. For others, the reason may have been too many hospital appointments and the child and/or the parents may not have been able to cope with it.

The pain diary was tested for validity, but one problem was that the participants had to remember to fill it in regularly. In future studies, a cell phone call could be used to gather information for the pain diary by sending a short message periodically that would ask the child to respond directly to a question about pain. This may be a better method to evaluate pain in children.²³

Another limitation is the measurement of physical activity using the self-reported activity diary. The activity diary is considered one of the most accurate subjective techniques for adults when investigating activity but has limited use in a pediatric population. Perhaps, in our study, it would have been better to ask the parents to fill in the diary. To limit the evaluator's subjective interpretation of the diary, we decided to let 3 therapists individually evaluate the diaries.

Another limitation was that the definition of physical activity was not clear enough for the children or the

families. Some reported all types of activity, and others reported only sport activities. Important considerations in future studies may be a better definition of activity and/or the use of other measurements to evaluate activity.

CONCLUSION

Pain seems to have a high effect on activity and participation in children with HMS. Balance was significantly decreased for children with HMS compared with the control group. With further research, it is important to find treatments that decrease and limit the pain, and also to investigate the consequence of decreased balance.

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CLINICAL BOTTOM LINE

Commentary on “Pain, Balance, Activity, and Participation in Children With Hypermobility Syndrome”

“How should I apply this information?”

This article provides some evidence that children with hypermobility syndrome (HMS) have problems with hypermobility, pain, activity, participation, and balance. Hence, when examining children with HMS, objective measurement of these areas is warranted. The small number of 20 subjects limits the description of a population. Hence, as the authors say, the results should not be generalized. This means that the differences seen here, between children with the diagnosis and those without it, may not be seen in any individual child with HMS.

This study does not address intervention. Although physical therapists might have something to offer these children, there is no evidence that they will respond in a way that children without this diagnosis might respond. Therefore, relevant objective measures before and after intervention become critical, as does sharing of findings in the form of peer-reviewed publications.

“What should I be mindful about in applying this information?”

The children with HMS were likely aware of their symptoms and those in the control group presumably viewed themselves as able-bodied and therefore the children’s responses may reflect these perceptions, rather than true differences.

The conclusion implies a cause-and-effect relationship between activity level and pain, but this is not supported by the design of the study. What was shown is that children with HMS had more pain and reported less activity than their peers. Unfortunately, the lack of statistical clarity makes the findings of this study difficult to interpret. Covariates were not shown to be significant or insignificant contributors, and neither R nor R^2 values for the regression were reported.

Lastly, the children might not have recorded all applicable activity or might be unreliable in what they did record. For example, they might not have considered walking to and from school an “activity” and hence might not have reported it. Perhaps using a standardized scale like the Activities Scale for Kids¹ would have provided a more valid and reliable measure.

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Lisa Selby-Silverstein, PT, PhD, NCS
Neumann University
Aston, Pennsylvania

Tiffany Atkins, PT, DPT, PCS
Neumann University, Aston, Pennsylvania,
and Kingsway Learning Center, Haddonfield, New Jersey

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