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ORIGINAL RESEARCH

Clinical Characteristics of 100 Patients With Hypermobility Spectrum Disorders and Shoulder Complaints With or Without Mechanical Symptoms: A Cross-sectional Study

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Abstract

Objective: To describe the clinical characteristics of patients with hypermobility spectrum disorders (HSD) and shoulder complaints with or without mechanical symptoms, and to compare characteristics between these groups.

Design: A cross-sectional study.

Setting: Primary care.

Participants: One-hundred patients with HSD and shoulder complaints for at least 3 months were included from primary care (N=100).

Interventions: Not applicable.

Main Outcome Measures: Medical history, self-reported (shoulder pain and function, discomfort due to other symptoms, fatigue, fear of movement, quality of life) and objective (strength, range of motion, proprioception) characteristics were collected by physiotherapists. Mechanical symptoms (yes/no) were defined as self-reported shoulder instability, subluxation, and/or laxity.

Results: Sixty-seven reported mechanical symptoms. Patients in both groups reported impairments related to shoulder pain, function, fatigue, fear of movement, and quality of life. Patients with mechanical symptoms were younger (mean, 35.1 years [95% CI, 32.3-37.9 years] vs 43.3 years [95% CI, 38.4-48.1 years]), had longer symptom duration (median, 46 months [95% CI, 36-66 months] vs 24 months [95% CI, 9-56 months]), reported a previous shoulder dislocation (25% [95% CI, 16-37] vs 3% [95% CI, 0-16]), experienced that their shoulder was loose (64% [95% CI, 52-76] vs 15% [95% CI, 5-32]), and reported discomfort due to other symptoms (odds ratio, 1.48 [95% CI, 1.17-1.87]). Furthermore, a larger proportion had received supplemental treatment (analgesic medication, steroid injection/surgery).

Conclusions: Both groups with HSD and shoulder complaints presented with substantial shoulder-related impairments. Two-thirds reported mechanical symptoms, were younger, and more severely impaired than those without mechanical symptoms. These findings highlight the importance of managing mechanical shoulder symptoms to fully address the patients' impairments.

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Generalized joint hypermobility (GJH) is a hereditary condition that results in an ability to move the joints beyond their normal range. The prevalence of GJH in the general population reaches 57% depending on race, sex, and diagnostic criteria.[1,2] In 2017, the classification of hypermobility spectrum disorders (HSD) was established to diagnose people as having GJH who experience musculoskeletal complaints, such as persistent joint pain or instability caused by repeated subluxations.[3] Besides the musculoskeletal complaints, patients with HSD further present with chronic pain and fatigue, disability, poor health-related quality of life, and psychological distress.[4,5]

Painful shoulder conditions are experienced by 4 of 5 patients with HSD[1,6-9] and may generally be attributed to pathology in a variety of shoulder structures, for example, the rotator cuff muscles and tendons, the acromial shape, the coracoacromial ligament, and capsular or intra-articular tissues, with no clearly established causality.[10] As an adjunct clinical manifestation to chronic shoulder pain, patients with HSD commonly report having other symptoms such as shoulder instability, subluxations, or laxity, which can be considered as having mechanical shoulder symptoms, although this term has not been used for the shoulder before.[6,11] The term *mechanical shoulder symptoms* is a key feature in shoulder instability—related to a symptomatic extensive translation of the humeral head relative to the glenoid fossa[12]—characterized by pain, discomfort, and loss of function.[13] Shoulder instability represents multiple continuums based on direction (from unidirectional to multidirectional), etiology (traumatic or atraumatic), severity (structural or nonstructural lesions), and frequency (single events to recurrent dislocation).[12,14]

Structural loss (for example, glenoid labrum tear) is considered the main cause for shoulder instability,[15-18] while abnormal muscle activation pattern is also frequently reported in patients with shoulder instability,[19,20] even with the absence of structural loss.[13,20] Furthermore, healthy individuals with shoulder hypermobility can also experience mechanical shoulder symptoms.[21,22]

Mechanical shoulder symptoms may occur involuntarily during arm movements in certain positions, which often results in severe functional impairments, discomfort, and pain, and sometimes these symptoms are even accompanied by a "popping" noise.[13] For some patients with shoulder complaints, nonpositional shoulder instability occurs with the arm in neutral or close to neutral.[13]

Studies on other patient groups (for example, with knee symptoms[23,24]) did not find associations between structural loss and self-reported mechanical symptoms, such as locking, popping, and giving way. However, there is a paucity of knowledge on whether the presence of mechanical shoulder symptoms is a common clinical observation, or it is associated with a higher degree of shoulder dysfunction that needs to be addressed during assessment and management of the painful shoulder in patients with HSD.

Therefore, the aims of this study were (1) to describe the medical history and the self-reported (shoulder pain and function, discomfort due to other symptoms, fatigue, fear of movement, quality of life) and objective (strength, range of motion, proprioception) clinical characteristics in patients with HSD and shoulder complaints; (2) to describe the clinical characteristics between those with or without self-reported mechanical symptoms (shoulder instability, subluxation, and/or laxity); and (3) to investigate whether any of the self-reported and/or objective clinical characteristics differed between the 2 groups.

List of abbreviations:

CIS	**Checklist Individual Strength**
EQ-5D-5L	**European Quality of Life - 5 Dimensions - 5-Level Scale**
5PQ	**5-part questionnaire**
GJH	**generalized joint hypermobility**
HSD	**hypermobility spectrum disorders**
OR	**odds ratio**
RCT	**randomized controlled trial**
TSK-11	**Tampa Scale of Kinesiophobia-11**
WOSI	**Western Ontario Shoulder Instability Index**

Methods

Study design

This cross-sectional study used baseline data from a randomized controlled trial (RCT) on patients with HSD and persistent shoulder complaints (Shoulder-MOBILEX study),[25] approved by the Regional Committees on Health Research Ethics for Southern Denmark (May 31, 2017, S-20170066) and registered with Clinicaltrials.gov (March 8, 2019, NCT03869307). We included a range of potentially relevant clinical characteristics because of the descriptive nature of the study. To reduce bias, these variables were selected and described in our preregistered statistical analysis plan (https://osf.io/pvnku/). The reporting is in accordance with the Strengthening the Reporting of Observational Studies in Epidemiology guidelines for cross-sectional studies.

Setting

The study was conducted in primary care within the Region of Southern Denmark, representing the general patient population in Denmark. The clinical assessments and self-reported measures were completed in person at baseline of the RCT at 2 sites (Esbjerg Municipality Rehabilitation Centre, Esbjerg, Denmark, and University of Southern Denmark, Odense, Denmark), by 1 of 4 blinded physiotherapists.

Participants

Patients were recruited from April 2019 to December 2020 by general practitioners from local medical clinics and physiotherapists from 8 clinics in 3 cities (Odense, Middelfart, and Esbjerg) in Denmark. Patients were asked to answer an online prescreening questionnaire including the 5-part questionnaire (5PQ) for self-reported GJH[26] and questions about what shoulder complaints they had, through Research Electronic Data Capture.[27] The principal investigator (B.L.) contacted patients considered potentially eligible (ie, 5PQ≥2 and having shoulder complaints, and 5PQ=1 was accepted if the patient in addition reported having a hypermobile shoulder) for a physical screening using the Beighton tests to establish whether the patient met the criteria for a clinical diagnosis of HSD (or hypermobile Ehlers-Danlos syndrome).[3,28] All patients had a medical referral to physiotherapy treatment because of their shoulder complaints and provided written informed consent to participate.

Eligibility criteria

Men and women were between 18 and 65 years of age and met the criteria for generalized HSD[28] or historical HSD[26] with concomitant shoulder complaints for at least 3 months[3,28] (complete eligibility criteria in supplementary appendix S1, available online only at http://www.archives-pmr.org/).

Demography and medical history

General demographic information included age, sex, educational level, employment, previous treatment, pain medication, and disease history (eg, previous shoulder dislocation, symptom duration). Anthropometric measurements included weight and height.

Self-reported clinical characteristics

At the physical screening, all patients answered the following question related to the presence of mechanical symptoms at inclusion: "Please indicate which shoulder complaints you have (you can tick off more than 1): (a) Pain, (b) Other symptoms (instability, subluxation, and/or laxity)." The principal investigator (B.L.) was available to offer clarification to the patients during answering. Patients who only selected "Pain" were considered to have no mechanical symptoms. Patients who ticked off "Other symptoms" with or without concurrent pain were considered to have mechanical symptoms.

Shoulder function was measured using the Western Ontario Shoulder Instability Index (WOSI) total score developed for patients with shoulder instability.[29] It has 21 questions, each marked on a scale from 0-100, with 0 being the best score (no limitations related to the shoulder) and 100 representing the worst score,[30] with a total ranging from 0-2100 points. It consists of 4 subscales: physical symptoms (10 questions, 0-1000), sports/recreation/work (4 questions, 0- 400), lifestyle (4 questions, 0- 400), and emotion (3 questions, 0-300).

Shoulder pain intensity was measured using the Numeric Pain Rating Scale, with scores from 0-10 (10=extreme pain).[31] The average, lowest, and highest pain levels was measured for the past week.[31] Similarly, discomfort due to other symptoms (such as shoulder instability, subluxation, and/or laxity) was measured from 0-10 (10=extreme complaints).

Fatigue was assessed with the Checklist Individual Strength (CIS) fatigue subscale, with scores ranging from 8-56 (56=highest level of fatigue).[32] A score of ≥35 is considered as being severely fatigued and associated with more problems and limitations.[33]

To assess generic health status, the Dartmouth Primary Care Cooperative Research Network/World Organization of National Colleges, Academies and Academic Associations of General Practitioners/Family Physicians questionnaire was used. It measures physical fitness, feelings (mental well-being), daily activities, social activities, change in health, and overall health. Categories are rated from 1 (good functional status) to 5 (poorest functional status), with total scores ranging from 6-30.[34,35]

Fear of movement was assessed using the Tampa Scale of Kinesiophobia-11 (TSK-11). The total scores range from 11-44 (44=highest fear of movement).[36] Although no cutoff value for high and low fear of movement exists using the TSK-11 scale, the original TSK-17 cut point[37] may correspond to ≥24.

Quality of life was assessed using the European Quality of Life - 5 Dimensions - 5-Level Scale (EQ-5D-5L).[38] The score ranges from −0.624 to 1 (1=perceived health at the best possible state; <0=worse than death).[38,39] The EQ-5D includes a European Quality Visual Analog Scale where the patients' own health "today" is rated between 0 (worst) and 100 (best). In patients with shoulder instability problems, EQ-5D-5L has shown satisfactory psychometric properties.[39,40]

Objectively measured characteristics

Isometric shoulder torque strength was assessed by measuring maximum isometric voluntary contraction in shoulder scaption, internal rotation, and external rotation using a handheld dynamometer.[41,42,a] Active and passive shoulder range of motion in internal and external rotation with the shoulder at 90 degrees of abduction,[41,43] as well as shoulder proprioception in shoulder flexion angles (low-range and midrange)[44,45] were assessed using a HALO digital goniometer.[b] For each of the objective characteristics, the average of 3 measurements was used in the analyses.

Statistics

The demographic characteristics, medical history, and self-reported clinical and objective characteristics for all patients and by group were presented using descriptive and proportion analyses as mean, median, or proportion with 95% CI (for proportion by use of the cii proportions command). Continuous data were checked for normality using the Shapiro-Wilk test and by visual inspection of histogram and quantile-quantile plot. Group differences in baseline characteristics were presented descriptively using the 95% CIs.

Because the assumptions of the preregistered and intended discriminant analyses were violated, multivariable logistic regression analyses were used to study the association between group (presence of mechanical symptoms or not) as the categorical dependent variable, and the self-reported (model 1) and objectively measured (model 2) characteristics as the independent variables. Backward stepping was conducted to keep variables with P values at a maximum set point of .10 in the multivariable models. We performed an additional analysis (model 3) using variables from the patients' medical history (Beighton score, symptom duration, previous shoulder dislocation, feel shoulder is loose, and have had previous shoulder treatment), and a model (model 4) combining all characteristics with $P<.1$ from the first 3 models. The assumptions for the analyses were met because there was no multicollinearity among the independent variables (inspecting variance inflation factors <10) and no specification error (the logit of the outcome variable was a linear combination of the independent variables).

No power calculation was used for the analyses because the current study population constitutes baseline data from the previously mentioned RCT. The level of significance was defined as $P<.05$. All statistical analyses were performed with Stata Statistical Software: Release 16.[c]

Results

After assessing 279 patients for eligibility, 100 patients (79% female) were included, and 67% reported mechanical shoulder symptoms (61 patients with concurrent pain and 6 patients without pain) (table 1, supplemental fig S2, available online only at http://www.archives-pmr.org/). The mean age for all patients was 37.8 years (95% CI, 35.3-40.3 years), while patients with mechanical symptoms were younger (mean, 35.1 years [95% CI, 32.3-

Table 1 Demographics and self-reported medical history in patients with HSD and persistent shoulder complaints presented for the total group and distributed by presence and absence of mechanical symptoms (shoulder instability, subluxation, and/or laxity)

Variable	All Patients (N=100)	With Mechanical Symptoms (n=67)	Without Mechanical Symptoms (n=33)
Sex (female), %	79	75 (63-84)	88 (72-97)
Age (y)	37.8 (35.3-40.3)	35.1 (32.3-37.9)	43.3 (38.4-48.1)
Weight (kg)	80.3 (76.9-83.7)	81.3 (77.0-85.5)	78.3 (72.3-84.3)
Height (cm)	171.9 (170.1-173.7)	172.4 (170.0-174.8)	170.8 (168.3-173.4)
Hypermobility spectrum disorder			
Beighton score, 0-9*	5.8 (5.5-6.1)	6.0 (5.5-6.4)	5.5 (4.9-6.0)
5PQ, 0-5	3.0 (3.0-3.3)	3.0 (3.0-4.0)	3.1 (2.0-4.0)
Generalized HSD (%)	89	90 (80-96)	88 (72-97)
Historical HSD (%)	11	10 (4-20)	12 (3-28)
Dominant writing hand, right (%)	91	93 (83-98)	88 (72-97)
Symptomatic shoulder, right (%)	57	51 (38-63)	70 (51-84)
Symptom duration (mo)	39* (36-60)	46 (36-66)	24 (9-56)
Previous shoulder dislocation, yes (%)	18	25 (16-37)	3 (0-16)
Feeling shoulder is loose, yes (%)	48	64 (52-76)	15 (5-32)
Education (%)			
Academic education	50	46 (34-59)	58 (39-75)
Skilled	28	28 (18-41)	27 (13-46)
Unskilled	5	7 (2-17)	0 (0-11)†
No education	17	18 (10-29)	15 (5-32)
Employment status (%)			
Full-time	54	52 (40-65)	58 (39-75)
Part-time	10	6 (2-15)	18 (7-35)
Student	17	19.5 (11-31)	12 (3-28)
Unemployed/retired	11	13.5 (6-24)	6 (1-20)
Sick listed	8	9 (3-18)	6 (1-20)
Previous shoulder treatment, yes (%)	68	70 (58-81)	64 (45-80)
Physiotherapy exercise	56	58 (46-70)	52 (34-69)
Physiotherapy passive treatment	44	46 (34-59)	39 (23-58)
Chiropractic	16	18 (10-29)	12 (3-28)
Prescribed analgesic medication	19	22 (13-34)	12 (3-28)
Other treatment (steroid injection, surgery)	21	24 (14-36)	15 (5-32)

NOTE. Continuous data are presented as mean or median with 95% CI, and categorical variables are presented as proportion % (95% CI).
* Median.
† One-sided, 97.5% CI because no patients answered "unskilled."

37.9 years] vs 43.3 years [95% CI, 38.4-48.1]). Patients with mechanical symptoms had longer symptom duration (median, 46 months [95% CI, 36-66 months] vs 24 months [95% CI, 9-56 months]), and a larger proportion reported having had a previous shoulder dislocation (25% [95% CI, 16-37] vs 3% [95% CI, 0-16]) and "feeling shoulder is loose" (64% [95% CI, 52-76] vs 15% [95% CI, 5-32]). Further, a larger proportion of patients with mechanical symptoms had received supplemental prescription of analgesic medication (22% [95% CI, 13-34] vs 12% [95% CI, 3-28]) and other treatment modalities (steroid injection/surgery) (24% [95% CI, 14-36] vs 15% [95% CI, 5-32]) (table 1).

The mean WOSI total score was 1056.8 (95% CI, 984.5-1129.1) for all patients and relatively high for patients with mechanical symptoms with respect to the mean of 1088.2 (95% CI, 993.8-1182.6) compared with a mean of 993.1 (95% CI, 883.2-1102.9) in patients without mechanical symptoms (table 2). Likewise, within the WOSI subdimensions, patients with mechanical symptoms had high scores for physical symptoms and sports/recreation/work. For each item, patients with mechanical symptoms either scored at the same level or somewhat worse (fig 1). However, items number 5 ("How much clicking, cracking, or snapping do you experience in your shoulder?") and 8 ("How much feeling of instability or looseness do you experience in your shoulder?"), which are directly related to experiencing mechanical symptoms showed the largest group differences. Other differences were related to compensating for the shoulder with other muscles (item 9) and participation in sports/recreation/work (items 11-13).

The 2 groups had similar shoulder pain intensities (see table 2), but as expected, patients with mechanical symptoms reported higher levels of discomfort due to other symptoms (mean, 4.0 [95% CI, 3.5-4.5] vs 2.4 (95% CI, 1.7-3.2]). Both groups reported similar fear of movement (TSK-11), fatigue (CIS), generic health status (Dartmouth Primary Care Cooperative Research Network/World Organization of National Colleges, Academies and Academic Associations of General Practitioners/Family Physicians questionnaire), and objective characteristics (eg, strength, range of motion, proprioception). However, patients with mechanical symptoms reported lower quality of life (mean, European Quality Visual Analog Scale: 62.0 [95% CI, 56.8-67.2] vs 70.0 [95% CI, 64.5-75.6]; and EQ-5D-5L index score: 0.68 [95% CI, 0.64-0.71] vs 0.73 [95% CI, 0.69-0.77]). Regarding CIS fatigue, a post hoc analysis showed that 61% in both groups had scores ≥35 points, considered to be severely fatigued (41/67 and 20/33 with and without mechanical symptoms, respectively). Regarding TSK-11, 49%

Table 2 Self-reported and objectively measured characteristics, including clinical tests in patients with generalized hypermobility spectrum disorder and persistent shoulder complaints presented for the total group and distributed by presence and absence of self-reported mechanical symptoms (shoulder instability, subluxation, and/or laxity)

Variable	All Patients, Mean (95% CI) (N=100)	Mechanical Symptoms, Mean (95% CI) (n=67)	No Mechanical Symptoms, Mean (95% CI) (n=33)
Self-reported			
WOSI total score, 0-2100	1056.8 (984.5-1129.1)	1088.2 (993.8-1182.6)	993.1 (883.2-1102.9)
Physical symptoms, 0-1000	471.9 (436.2-507.6)	494.7 (448.5-540.9)	425.6 (372.0-479.3)
Sports/recreation/work, 0-400	201.7 (182.7-220.8)	210.1 (186.6-233.7)	184.6 (151.3-217.9)
Lifestyle, 0-400	180.2 (163.4-197.0)	182.8 (161.2-204.4)	175.0 (147.6-202.4)
Emotions, 0-300	203.0 (191.6-214.3)	200.6 (185.5-215.6)	207.8 (191.0-224.7)
Shoulder pain intensity (NPRS) during the past 7 d			
Lowest, 0-10	2.4 (2.0-2.8)	2.3 (1.8-2.8)	2.5 (1.9-3.2)
Highest, 0-10	6.2 (5.7-6.7)	6.5 (5.8-7.1)	5.7 (5.0-6.4)
Average, 0-10	4.0 (3.6-4.4)	4.1 (3.5-4.7)	3.7 (3.1-4.3)
Discomfort due to other shoulder symptoms (NRS) during the past 7 d			
Lowest, 0-10	2.5 (2.0-2.9)	2.7 (2.1-3.2)	2.0 (1.3-2.7)
Highest, 0-10	4.7 (4.1-5.2)	5.3 (4.7-5.8)	3.4 (2.5-4.3)
Average, 0-10	3.5 (3.0-3.9)	4.0 (3.5-4.5)	2.4 (1.7-3.2)
Tampa Scale of Kinesiophobia, 11-44	22.7 (21.6-23.8)	23.3 (21.9-24.6)	21.7 (19.7-23.7)
CIS, fatigue subscale, 8-56	37.0 (34.9-39.1)	37.4 (34.8-39.9)	36.4 (32.4-40.4)
COOP/WONCA, 6-30	14.4 (13.7-15.2)	14.6 (13.7-15.6)	14.1 (13.1-15.1)
EQ-5D-5L Quality of Life questionnaire			
EQ-VAS, 0-100	64.7 (60.7-68.6)	62.0 (56.8-67.2)	70.0 (64.5;75.6)
Index score, <0-1	0.69 (0.67-0.72)	0.68 (0.64-0.71)	0.73 (0.69-0.77)
Objective			
Isometric shoulder torque strength (N•m/kg)			
Scaption	0.45 (0.41-0.49)	0.46 (0.41-0.52)	0.42 (0.34-0.49)
Internal rotation	0.33 (0.30-0.36)	0.33 (0.30-0.37)	0.32 (0.27-0.38)
External rotation	0.24 (0.22: 0.26)	0.25 (0.23-0.28)	0.23 (0.19-0.27)
Range of motion (˚)			
Internal rotation passive	69 (66-73)	68 (64-73)	72 (65-79)
Internal rotation active	67 (63-70)	65 (61-69)	70 (64-77)
External rotation passive	103 (98-107)	102 (96-108)	104 (96-112)
External rotation active	100 (95-104)	100 (95-105)	99 (91-106)
Proprioception in flexion, error (˚)			
Low range (55˚±10˚)	4.8 (4.2-5.3)	4.7 (4.0-5.4)	4.9 (3.8-6.0)
Midrange (90˚±10˚)	4.0 (3.6-4.5)	4.1 (3.6-4.7)	3.8 (3.0-4.6)

Abbreviations: COOP/WONCA, Dartmouth Primary Care Cooperative Research Network/World Organization of National Colleges, Academies and Academic Associations of General Practitioners/Family Physicians; EQ-VAS, European Quality Visual Analog Scale; NPRS, Numeric Pain Rating Scale; NRS, numeric rating scale; VAS, visual analog scale.

of all patients had scores ≥24 points (37/67 and 12/33 with and without mechanical symptoms, respectively).

The logistic regression analyses for self-reported characteristics (model 1) showed that patients with mechanical symptoms were more likely to have additional discomfort due to other symptoms (odds ratio [OR], 1.48; 95% CI, 1.17-1.87) (table 3). Model 2 including the objective characteristics was nonsignificant, showing no associations with experiencing mechanical symptoms. Model 3 using medical history variables showed that patients with mechanical symptoms were more likely to have a previous shoulder dislocation (OR, 12.88; 95% CI, 1.52-109.03) and to feel shoulder is loose (OR, 10.88; 95% CI, 3.59-33.00). The combined parsimonious model (model 4) was significant, with the same 3 variables remaining significant or marginally significant.

Discussion

All patients with HSD and shoulder complaints presented with severe impairments related to shoulder pain, function, fatigue, fear of movement, and quality of life. Two-thirds of these patients reported mechanical symptoms such as instability, subluxation, and/or laxity, and they were younger than those without mechanical symptoms. Self-reported clinical characteristics were more severe for those with mechanical symptoms, with a higher proportion reporting longer symptom duration, previous shoulder dislocations, feeling the shoulder is loose, an increased odds of reporting additional discomfort, and having had received supplemental treatment.

The high mean WOSI total score of 1056.8 (95% CI, 984.5-1129.1) for both groups is substantially larger than of a healthy

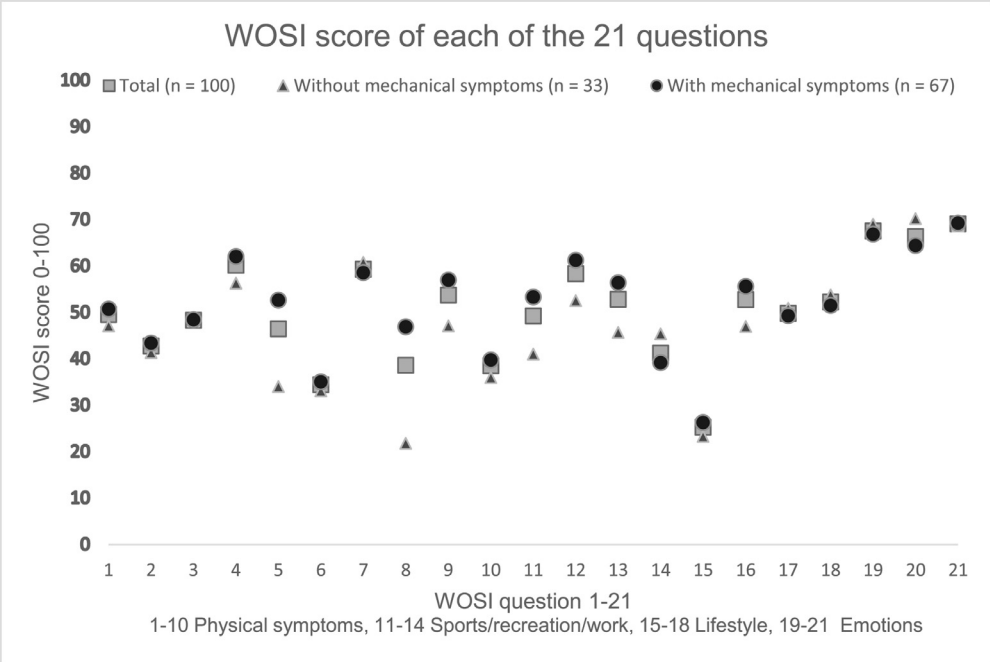

Fig 1 Mean score for each of the 21 WOSI questions (0-100, higher is worse) in patients with hypermobility spectrum disorder and persistent shoulder complaints, presented for the total group and distributed in groups by presence and absence of mechanical symptoms (ie, instability, subluxation, and/or laxity). The actual data are included in supplemental table S3 (available online only at http://www.archives-pmr.org/).

shoulder (84/2100 points[46]) and comparable to previously published data on patients with HSD with shoulder complaints,[47] hypermobile Ehlers-Danlos syndrome,[7] multidirectional instability,[48] and traumatic shoulder dislocation,[49] emphasizing the severity of the symptoms.

The level of shoulder pain (Numeric Pain Rating Scale) (for all patients lowest mean 2.4, highest mean 6.2) was similar in both groups yet comparable with pain levels of the most common shoulder problem—subacromial pain syndrome/rotator cuff –related shoulder pain (ie, mean pain at rest 2.4/2.8 and mean pain during movement 5.7/6.3[50])—representing a patient group responsible for 45%-80% of all shoulder-related contact with the general practitioners.[51,52] Previous studies on hypermobile Ehlers-Danlos syndrome show that moderate to severe pain is known to be an everyday symptom in this patient group and that pain severity is related to hypermobility, dislocations, and previous operations.[8,53] Furthermore, pain severity has been reported to be higher in patients with severe fatigue than in patients with less severe fatigue and to have a larger effect on daily functioning than pain alone.[33] The mean CIS fatigue score (37.0) for both groups

Table 3 Self-reported and objectively measured characteristics, in patients with generalized hypermobility spectrum disorder and persistent shoulder complaints by presence and absence of self-reported mechanical symptoms (shoulder instability, subluxation, and/or laxity)

Variables	OR	95% CI	*P* Value
Model 1: Self-reported characteristics			<.001*,†
Discomfort due to other shoulder symptoms (average past 7 d), 0-10 NRS	1.48	1.17-1.87	.001†
Model 2: Objective characteristics			.121*
Isometric shoulder torque strength (N•m/kg)			
Scaption	8.85	0.77-101.29	.079
Range of motion (˚)			
External rotation passive	0.95	0.91-1.00	.035†
External rotation active	1.04	1.00-1.09	.055
Model 3: Medical history	12.88	1.52-109.03	<.001*,†
Previous shoulder dislocation, yes (%)	10.88	3.59-33.00	.019†
Feeling shoulder is loose, yes (%)			<.001†
Combined model			<.001*,†
Discomfort due to other shoulder symptoms (average past 7 d), 0-10 NRS	1.28	0.97-1.68	.079
Previous shoulder dislocation, yes (%)	10.88	1.25-95.08	.031†
Feeling shoulder is loose, yes (%)	8.45	2.70-26.43	<.001†

NOTE. Results from logistic regression analyses using backward stepping with *P* values used to remove at a set-point of 0.10.
Abbreviation: NRS, numeric rating scale.
* *P* value for model.
† *P*<.05.

was comparable with the mean score of Ehlers-Danlos syndrome (41.7), but the prevalence (61%) of patients above the threshold of being considered severely fatigued (a score ≥35) and having more problems and limitations is lower than for patients with hypermobile Ehlers-Danlos syndrome (84%).[33] Besides looking at the cut-off, assessment of fatigue should also include a clinical interview to determine whether the fatigue is clinically relevant.[32] These findings suggest that pain and fatigue seem to contribute to the burden of disease in this patient group, which is further supported by the current low health status scores (EQ-5D-5L) compared with asymptomatic populations.[54]

Patients from both groups had TSK-11 mean score of 22.7, which is lower than previously reported scores in people with chronic pain.[55] However, half of the patients scored higher than the estimated cut point suggested to indicate high fear of movement, which is more frequent than in a population with recurrent shoulder dislocations.[56] This may indicate that this is a major problem for some of the patients.

Our finding that patients with mechanical symptoms were younger than those without corresponds well with the fact that the prevalence of GJH in both sexes decreases with age.[57,58] Their different medical profiles (eg, regarding previous shoulder dislocations and feeling shoulder is loose) corresponds well with shoulder dislocations being more prevalent in young individuals, which may compromise the glenoid labrum thereby decreasing structural (passive) stability of the joint.[15-18] As a consequence, this may result in recurrent and subsequently chronic instability, repeated episodes of joint sprains, subluxations, and microtrauma, which may explain the longer duration of symptoms and a larger need of supplemental treatment in this subgroup.[59-61] Mechanical symptoms may also be because of poor activity and function of the scapular and rotator cuff muscles potentially affecting the scapulohumeral coordination, in addition to altered movement patterns due to the inherent tissue laxity, a key characteristic of the HSD condition.[62] However, we did not measure muscle activity, recruitment, scapulohumeral coordination, or humeral translation, which may explain why none of the selected objective characteristics could predict the presence of mechanical symptoms.

Patients with shoulder problems are commonly referred to primary and secondary care because of persistent and severe impairments affecting daily function and quality of life. We propose that clinicians examining patients with shoulder complaints rule in or rule out the presence of joint hypermobility (for example, GJH) to capture those patients fulfilling the criteria for HSD. Second, if patients reporting shoulder complaints have HSD, it is suggested that they are interviewed about any mechanical symptoms in their shoulder, in addition to pain, fatigue, and general discomfort. Besides considering age and symptom duration (the current median symptom duration being 39 months), questions related to 1 or more previous episodes of shoulder dislocations and the feeling of the shoulder being loose are suggested to be included in the history taking. When assessing the severity of symptoms, pain assessments should be accompanied by questions specifically focusing on the level of discomfort due to other symptoms because none of the other included self-reported or objective characteristics in this article showed an association with experiencing mechanical symptoms. Future studies should investigate the role of other relevant characteristics (eg, muscle activity, joint translation) in patients with and without self-reported mechanical symptoms, as well as using objective measures of mechanical symptoms to distinguish between these subgroups.

Study limitations

The limitations of this study are attributed to the cross-sectional design making it difficult to draw conclusions about the cause-relationship of the findings. Furthermore, the presence of mechanical symptoms was self-reported without a subsequent objective validation (eg, assessment of positional/nonpositional and/or controllable/noncontrollable instability)[13] or imaging verification of potential structural defects. In addition, the selected variables are only a sample of the many clinical characteristics that potentially can predict the presence of mechanical symptoms.

The strengths are that blinded assessors not involved scientifically in the study collected all data. The main analyses were predefined and published prior to conducting the analyses and supplied with additional analyses when relevant. The reporting follows the Strengthening the Reporting of Observational Studies in Epidemiology guidelines for cross-sectional studies.

Conclusions

All patients with HSD and shoulder complaints presented with substantial impairments related to shoulder pain, function, fatigue, fear of movement, and quality of life. Two-thirds of these patients reported mechanical shoulder symptoms and were younger than those without mechanical symptoms. Self-reported clinical characteristics were more severe for those with mechanical symptoms, with a higher proportion reporting longer symptom duration, previous shoulder dislocations, feeling the shoulder is loose, and an increased odds of reporting additional discomfort and having had received supplemental treatment. These findings highlight the importance of addressing mechanical symptoms in the shoulder during treatment to fully cover and understand the patients' impairments.

Suppliers

a. IsoForce Dynamometer EVO2; Medical Device Solution AG.
b. HALO digital goniometer; Halo Medical Devices.
c. Stata Statistical Software: Release 16; StataCorp.

Keywords

Joint instability; Rehabilitation; Shoulder

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Supplemental appendix S1: Eligibility criteria

Males and females were between 18 and 65 years of age and met the following inclusion criteria:

1. Generalised HSD (G-HSD), defined using a Beighton score cut off ≥ 5/9 for females up to the age of 50 years, and ≥ 4/9 for those > 50 years and all males without age-specific cut-points,[3] or Historical HSD (H-HSD) if the Beighton score was 1 point below the age and sex-specific cut off and the 5PQ was positive (≥ 2/5 positive answers).[2] Although the shoulder is not assessed in the Beighton score, classification of GJH by using the Beighton score builds on the assumption that most joints, including the shoulder, are hypermobile. Therefore, no additional tests for shoulder hypermobility were used as part of the inclusion criteria.
2. One or more of the following self-reported (yes/no) symptomatic musculoskeletal manifestations present[1,3]:
   - Pain in at least one shoulder for at least three months.
   - Recurrent joint dislocations or joint instability without a reported history of trauma, defined as (a) a minimum of three atraumatic dislocations in the same shoulder, (b) a minimum of two atraumatic dislocations in two different joints (a minimum of one in the shoulder) occurring at different times, and/or (c) medical confirmation of joint instability in at least two joints (a minimum of one in the shoulder).

Patients were excluded if they were unable to speak or understand Danish, unable to comply with the study protocol for the RCT, unable to provide informed consent, and if they fulfilled any of the following criteria:

- Clinically suspected referred pain from the cervical spine.
- Diagnosis of systemic inflammatory rheumatic diseases, connective tissue diseases (Marfans, Stickler's or Loeys Dietz syndromes, EDS except hEDS), and/or neurological diseases.
- Pregnancy or childbirth within the past year or planning to get pregnant during the study period, because of increased levels of relaxin.
- Shoulder surgery within the past year.
- Steroid injection in the affected shoulder within the past three months.

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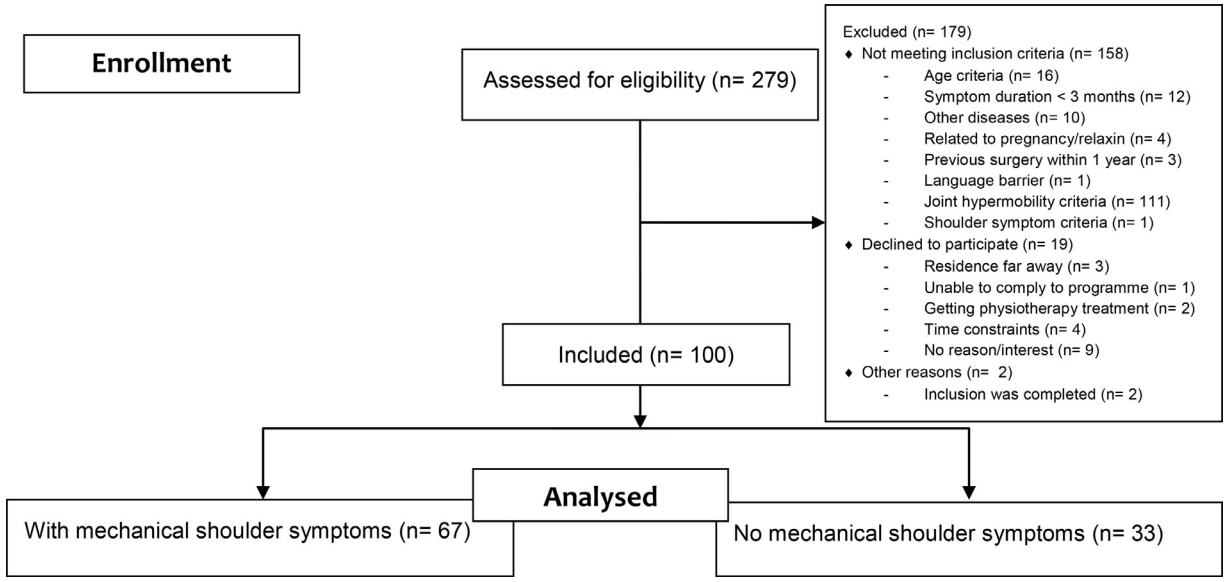
Supplemental Fig S2. Flow diagram



Supplemental Table S3

WOSI question#	WOSI question	Total	95% CI		Without mechanical symptoms	95% CI		With mechanical symptoms	95% CI	
			Lower	Upper		Lower	Upper		Lower	Upper
1	How much pain do you experience in your shoulder with overhead activities?	49.54	44.38	54.70	47.09	39.84	54.35	50.75	43.82	57.67
2	How much aching or throbbing do you experience in your shoulder?	42.76	37.16	48.36	41.36	32.65	50.08	43.4	36.13	50.77
3	How much weakness or lack of strength do you experience in your shoulder?	48.32	43.56	53.08	48.09	39.80	56.38	48.43	42.46	54.40
4	How much fatigue or lack of stamina do you experience in your shoulder?	60.15	55.64	64.66	56.3	48.36	64.25	62.04	56.48	67.61
5	How much clicking, cracking or snapping do you experience in your shoulder?	46.48	40.43	52.53	34.0	23.49	44.45	52.64	45.51	59.77
6	How much stiffness do you experience in your shoulder?	34.4	29.00	39.80	33.12	24.72	41.52	35.03	27.98	42.08
7	How much discomfort do you experience in your neck muscles as a result of your shoulder?	59.3	53.89	64.71	60.79	52.36	69.22	58.57	51.50	65.63
8	How much feeling of instability or looseness do you experience in your shoulder?	38.65	32.87	44.43	21.8	12.89	30.74	46.94	40.25	53.63
9	How much do your compensate for your shoulder with other muscles?	53.76	48.68	58.84	47.12	37.71	56.53	57.0	51.01	63.05
10	How much loss of range of motion do you have in your shoulder?	38.52	33.29	43.75	35.97	26.81	45.13	39.78	33.27	46.28
11	How much has your shoulder limited the amount you can participate in sports or recreational activities?	49.26	42.90	55.62	41.0	31.39	50.61	53.33	45.11	61.54
12	How much has your shoulder affected your ability to	58.37	52.58	64.16	52.55	42.26	62.83	61.24	54.14	68.33

(*continued on next page*)

(*Continued*)

WOSI question#	WOSI question	Total	95% CI		Without mechanical symptoms	95% CI		With mechanical symptoms	95% CI	
			Lower	Upper		Lower	Upper		Lower	Upper
	perform the specific skills required for your sport or work? (If your shoulder affects both sports and work, consider the area that is most affected.)									
13	How much do you feel the need to protect your arm during activities?	52.85	46.96	58.74	45.67	34.67	56.67	56.39	49.41	63.37
14	How much difficulty do you experience lifting heavy objects below shoulder level	41.24	35.72	46.76	45.39	35.73	55.06	39.19	32.35	46.04
15	How much fear do you have of falling on your shoulder?	25.29	19.64	30.94	23.27	14.97	31.58	26.28	18.78	33.79
16	How much difficulty do you experience maintaining your desired level of fitness	52.79	46.72	58.86	47.0	36.78	57.22	55.64	48.02	63.26
17	How much difficulty do you have “roughhousing” or “horsing around” with family or friends	49.85	43.62	56.08	50.91	40.47	61.35	49.33	41.39	57.27
18	How much difficulty do you have sleeping because of your shoulder	52.29	46.27	58.31	53.79	44.25	63.32	51.55	43.73	59.37
19	How conscious are you of your shoulder	67.57	63.09	72.05	69.03	61.96	76.10	66.85	61.03	72.68
20	How concerned are you about your shoulder becoming worse	66.35	61.51	71.19	70.21	63.88	76.55	64.45	57.87	71.03
21	How much frustration do you feel because of your shoulder	69.06	64.09	74.03	68.61	60.42	76.80	69.28	62.92	75.65

Abbreviation: WOSI, The Western Ontario Shoulder Instability Index