

Epidemiology of General Joint Hypermobility and Basis for the Proposed Criteria for Benign Joint Hypermobility Syndrome: Review of the Literature

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ABSTRACT. *Objective.* This literature review of generalized joint hypermobility (GJH) syndromes discusses information regarding sex-, age-, and race-related factors from publications that specifically document validated GJH criteria.

Methods. We present an analysis of criterion-referenced connections that identify similarities among major and minor clinical criteria that identify both GJH and benign joint hypermobility syndrome (BJHS). In our search, we found considerable empirical evidence that supports an increased prevalence of hypermobility among children, women, and certain racial groups. Two commonly used clinical assessment tools, the Carter and Wilkinson criteria (≥ 3 positive tests out of 5) and the Beighton method (≥ 4 positive tests out of 9), are the sources of these data. BJHS is diagnosed through a set of major and minor criteria — a combination of symptoms and objective findings — that include arthralgia, back pain, spondylosis, spondylolysis/spondylolisthesis, joint dislocation/subluxation, soft tissue rheumatism, marfanoid habitus, abnormal skin, eye signs, varicose veins or hernia or uterine/rectal prolapse.

Results. Clinically, there is some evidence that arthralgia, the proposed BJHS major criterion, is a major component of alleged hypermobility-related problems. In contrasting, there is no clear evidence that proposed BJHS minor diagnostic criteria are associated with hypermobility-related problems. An empirical correlation between hypermobility and osteoarthritis is possible, but so far unproven. There are no randomized controlled studies regarding effects of existing treatments.

Conclusion. Generalized hypermobility is both sex- and age-related. Racial differences are also identifiable. The existence of BJHS can be accepted using present criteria. (First Release Jan 15 2007; J Rheumatol 2007;34:804–9)

Key Indexing Terms:

JOINT LAXITY HYPERMOBILITY EPIDEMIOLOGY SYNDROME CRITERIA

Musculoskeletal complaints in association with general joint hypermobility (GJH) were, in 1967, labelled as hypermobility syndrome (HS)¹, which now is called benign joint hypermobility syndrome (BJHS)². In a previous article³ focusing on reproducibility and validity of tests and criteria for GJH and BJHS, we concluded that future syndrome-related validity studies will have to be developed on the basis of construct validity using criteria presumed to be part of the syndrome. In this article we focus on 4 items: (1) The epidemiology of GJH,

looking for documentation of age, sex, and racial variations in the prevalence of GJH. (2) Are the major and minor diagnostic criteria for BJHS well enough documented? (3) Is there sufficient evidence to suggest that BJHS leads to an increased prevalence of osteoarthritis (OA)? (4) What scientifically documented prevention and treatment strategies and algorithms are available?

MATERIALS AND METHODS

We searched PubMed, Cochrane Library, and PEDro using the following: joint instability, hypermobility, joint dislocation, back pain, shoulder injuries, sprain, children, age, sports injuries, marfanoid habitus, eye signs, and pregnancy. From the results, we reviewed GJH-related articles that used validated tests and standards synonymous with Carter and Wilkinson's criteria⁴ and Beighton's method⁵. Other publications using limited modifications of the validated tests and criteria are also incorporated.

RESULTS

Epidemiology

Age, sex, and race. Beighton, *et al* demonstrated that the number of positive hypermobility tests was age- and sex-related⁵; the younger the children, the higher the score. Women had higher scores than age-matched men. The findings were confirmed by some authors^{6–10}, but not all^{4,11,12}.

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Table 1. Prevalence of hypermobility among non-Caucasian women and men in various age groups using validated hypermobility tests and criteria.

Study	Race, Age, yrs	Prevalence of Hypermobility	
		Female, % (N)	Male, % (N)
Walker ¹³	Amerindians, 0–19	18 (212)	12 (184)
	Inuit 0–19	32 (165)	29 (133)
Klemp ¹⁴ , New Zealanders	Caucasian > 5	6 (195)	2 (159)
	Maori > 5	9 (256)	2 (182)
El-Garf ²¹ , Egyptians	Arabic 6–15	18 (498)	14 (499)
Pountain ⁶¹ , Oman inhabitants	Mixed Arabic 16–25	29 (178)	9 (131)
Al-Rawi ⁶² , Iraqis	Arabic 20–24	39 (1187)	25 (587)
Al-Rawi ⁵⁰ , Iraqis	Arabic 23–65	18 (76)	—
Birrell ⁶ , Yoruba Africans	Negroid 6–66	57 (116)	35 (88)
Beighton ^{5*} , Tswana Africans	Negoid ≥ 20	20	6

* Beighton did not define any criterion, but mentioned that 80% of females and 94% of males had 0–2 positive tests.

There seems to be an increased prevalence of hypermobility among some racial groups (Table 1). However, only 2 studies were controlled, demonstrating significantly increased prevalence in Igloodik Eskimos compared to Native Americans (Amerindians)¹³, and no difference between Caucasian and Maori groupings¹⁴. The prevalence for Chinese children⁷ and adults (Beighton score $\geq 5/9$)¹⁵ also seems to be increased when compared to Caucasians.

Occupation and sport. A significantly increased prevalence of hypermobility is found among ballet dancers compared to controls^{20,21}. Hypermobility was also present in joints not exposed to stretching exercises, indicating that the hypermobility is hereditary rather than acquired.

In contrast, a palms-to-floor test correlates positively with duration of ballet training²².

American music students and Swedish industrial workers had a relatively high prevalence of hypermobility. No control populations were included in the studies^{23,24}.

Clinical conditions used as hypermobility syndrome criteria
Arthralgia. In 1967, Kirk, *et al*¹ noted that 20 of 24 hypermobile patients with musculoskeletal pain had accompanying joint pain. Later a relatively high correlation was found among Tswana Africans between a hypermobility score and a primitive pain score that included joint pain⁵. In contrast, no correlation was found among Yoruba Africans between joint pain and the hypermobility score⁶. Several later studies pointed towards an increased prevalence of arthralgia in hypermobile populations (Table 2).

Back pain, spondylosis, spondylolysis, and spondylolisthesis. There was no correlation between hypermobility and low

back pain in studies on primary school or high school children^{11,16,26,29}. However, hypermobility in adult populations correlated with back pain in general²⁸, and also with work-related back pain^{23,24}.

Appearance or progression of an idiopathic scoliosis in 10- to 16-year-old girls was not connected to increased joint mobility³⁰, and joint laxity did not relate to the prevalence of spondylolisthesis³¹.

We have not located any studies that discuss the prevalence of facet syndromes/spinal segmental dysfunctions or pelvic dysfunction in a hypermobile population. However, among pregnant South African (“Cape Coloured”) women there was no correlation between peripartum pelvic pain and hypermobility³². Among Caucasian women with pelvic pain the prevalence of hypermobility was 12%³³.

Joint dislocation and subluxation. Using modified Beighton tests, a significantly increased prevalence of ankle sprain was seen among male military recruits²⁷. In contrast, there was no significant increase in the prevalence of joint dislocation among 12³⁴ or 15-year-old hypermobile children²⁶.

Other studies suggest hypermobility connections with congenital hip⁴ and patellar dislocations³⁵. There are conflicting results regarding hypermobility and presence of so-called “pulled elbow” (traction-induced dislocation of the radio-humeral joint)^{36,37} or presence of temporomandibular joint dysfunction^{38,39}.

Soft-tissue rheumatism. A number of studies could not demonstrate a correlation between hypermobility and musculoskeletal pain or myalgia among adolescents^{11,12,26,34}. However, in a 4-year followup study, hypermobility was found to be a strong predictor for pain recurrence in girls⁴⁰. Also, injury

Table 2. Prevalence of arthralgia among hypermobile and non-hypermobile children and adults in various age groups using validated hypermobility tests and criteria.

Study	Patient Population Symptoms Age, yrs	Prevalence of Arthralgia Among:		p
		Hypermobile % (N)	Non-hypermobile % (N)	
Gedalia ²⁵	Students Arthralgia, 1-year prevalence 6–14	40 (53)	17 (52)	< 0.02
El-Garf ¹⁶	Students Arthralgia 6–15	16 (161)	10 (836)	< 0.001
Seçkin ²⁶	High school students Arthralgia Mean 15.4	17 (101)	15 (760)	NS
Diaz ^{27*}	Male military recruits Arthralgia 17	10 (223)	4 (452)	< 0.005
Larsson ²³	Musicians Wrist pain 18–68	5 (198)	18 (462)	< 0.001
Al-Rawi ¹⁸	Students Joint complaints 20–24	13 (528)	12 (1246)	NS
Bravo ²⁸	BJHS + controls Arthralgia Mean 40	57 (230)	27 (64)	< 0.01

* Cut-off level for laxity + hyperlaxity was $\geq 2/5$ Beighton tests. NS: not significant.

totals were significantly increased among hypermobile ballet dancers²² and high school students²⁶.

Muscle strain was not increased in the hypermobile recruits with documented ankle sprain²⁷, and 2 reports on the incidence of tendinitis, bursitis, and fibromyalgia among hypermobile individuals were contradictory^{41,42}. Similarly, hypermobile children had an increased prevalence of fibromyalgia⁴³, in contrast to an adult population (Beighton score $\geq 5/9$)⁴⁴. Finally, looking at patients with fibromyalgia, the prevalence of hypermobility was significantly increased in 3 studies^{43,45,46}, but normal in 2 others^{44,47}.

Abnormal skin. Increased skin extensibility is found among hypermobile children with musculoskeletal pain⁴⁸, using a 10-test scoring system (9 joints + ecchymosis) suggested by Bulbena, *et al*⁴⁹. In contrast, there were no appreciable differences in skin extensibility on the dorsum of the hand among selected, age-matched female patients with or without hypermobility⁵⁰. Other studies demonstrate a significantly increased prevalence of broad scar formation in persons with hyperextensibility in the fifth finger⁵¹ or increased skin extensibility among persons with GJH²¹.

Uterine and rectal prolapse, varicose veins, and hernia. In women > 35 years of age, with pelvic relaxation and urinary incontinence, those with hypermobility (Beighton score $\geq 2/3$) had a significantly increased prevalence of urogenital prolapse, compared to those without hypermobility⁵².

Patients operated on for rectal prolapse had significantly increased mobility in the metacarpophalangeal joint of the fifth finger⁵³, and patients with genital prolapse had a significantly increased hypermobility prevalence compared to an age- and parity-matched gynecologic patient control group¹⁹.

In a cohort of Arabic patients with musculoskeletal pain and general hypermobility, the prevalence of varicose veins was 53% and of hemorrhoids 45%⁵⁴.

Twenty-two percent of patients with endoscopically verified hiatus hernia had a significantly increased prevalence of hypermobility compared with 6% of normal subjects⁵⁵.

Marfanoid habitus, eye signs. Among selected female patients attending a rheumatologic clinic, the patients with hypermobility had an increased incidence of marfanoid habitus⁵⁰. This was also demonstrated recently among consecutively recruited patients with HS²⁸, but could not be demonstrated in a cohort study of high school students²⁶. Similarly, previous information regarding coherence between eyelid laxity and hypermobility⁵⁶ could not be documented²⁶.

Hypermobility and OA

Two early clinical demonstrations of HS, without statistical calculations, suggested an increased prevalence of OA among hypermobile patients^{1,57}. This prevalence was later documented by Bridges, *et al*, who also illustrated a significantly high-

er prevalence of OA among hypermobile individuals (Beighton score $\geq 5/9$)⁴⁴. Also supportive are 2 female cohort studies, both presenting a greater prevalence of hypermobility among those who were generally hypermobile^{58,59}. In contrast, 2 more recent studies concluded that the risk of developing OA is reduced^{60,61}.

Based on the above information, a correlation between hypermobility and OA is possible, but so far unproven.

Treatment studies

In an uncontrolled longitudinal study of 25 selected hypermobile patients, Barton and Bird⁶² demonstrated that a 6-week individualized intervention program positively influenced pain reports and walking distances. However, the patient cohort was not characterized by age and Beighton score, and a cutoff level for GJH was not defined.

Also, in an uncontrolled study, knee joint proprioception training in 20 hypermobile patients brought about a significant improvement of the knee proprioception and balance that also alleviated knee joint symptoms⁶³.

DISCUSSION

Currently available literature documents an increased prevalence of hypermobility among younger females, in spite of some variation among the applied GJH diagnostic criteria (cutoff levels ranging from $\geq 2/3$ to $\geq 7/9$ positive Beighton tests). The literature also supports a decreasing prevalence with aging as well as race-related differences.

The reliability of Beighton's tests and of the cutoff level for GJH was studied among Caucasian adults as discussed previously³. But it is not known from these studies whether the generally used cutoff level (Beighton score $\geq 4/9$) is universally valid. Given the variations discussed above, it would probably be more scientifically accurate to develop and apply differentiated cutoff levels sorted by sex, age, and race. This point of view is supported by Jansson, *et al*¹⁰ and has been applied in at least 2 studies^{11,49}. The following supports this differential concept: Only by increasing the cutoff level for pathological hypermobility to $\geq 7/9$ in adult Arabic populations, could the authors define hypermobile cohorts reporting an increased prevalence of arthralgia^{17,18}.

Vocationally, there is no scientifically solid basis for guiding the hypermobile population toward a choice of career. However, it seems inappropriate to advise against a ballet career^{21,22}.

A variety of BJHS diagnostic criteria have been proposed making use of both subjective reports and clinical signs². There is, however, an unanswered question: Are there good reasons for these choices?

The majority of studies support a connection between hypermobility and the presence of arthralgia. Significantly, the available studies are of populations with different age groups and different races. Also, the studies use different test procedures and cutoff levels (Table 2).

The evidence we were able to locate does not support a general disposition for low back pain in young GJH populations^{11,16,26,29}. In contrast, the prevalence of low back pain was increased in a group of adults with BJHS²⁸ and among hypermobile workers who are seated^{23,24}, indicating that back pain may arise more often in persons with hypermobility. To our knowledge, there are no studies of documented hypermobility in relation to spinal facet syndromes, spinal segmental dysfunctions, or mechanically-based pelvic dysfunction presentations.

An almost equal number of studies argue for and against increased prevalence of joint dislocation and of soft tissue rheumatism among the hypermobile.

Some studies show increased skin extensibility among hypermobile groups. These results can naturally give rise to speculations regarding any differences between BJHS and hypermobility-type Ehlers-Danlos syndrome⁶⁴.

There is one study demonstrating increased prevalence of genitourinary prolapse among hypermobile persons (cutoff level $\geq 2/3$ positive tests). However, among patients with genital prolapse, rectal prolapse, varicose veins, or hemorrhoids there is an apparent increased prevalence of hypermobility that may indicate increased collagen fiber extensibility^{19,53,54}.

Studies of marfanoid habitus and eye signs have given differing information, making the use of these signs as minor criteria for BJHS questionable.

At present, there are no available controlled outcome studies dealing with treatment of this syndrome. Therefore, any recommendations can only be based on theoretical assumptions.

Generalized hypermobility is both sex- and age-related. Racial differences are also identifiable.

Some statistics support the current BJHS major diagnostic criterion of arthralgia, but other data may refute most of the minor criteria. In order to move forward, we recommend that several systematic tasks be considered: Define normal joint range of motion sorted according to age, sex, and race. Develop appropriate hypermobility cutoff levels that accurately portray any group differences. Implement longitudinal and cross-sectional cohort-based diagnostic and treatment studies that identify relevant minor criteria. In the meantime, the existence of the BJHS syndrome can be accepted using present criteria.

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