



# Effects of Benign Joint Hypermobility Syndrome on the Clinical Characteristics of Carpal Tunnel Syndrome in Females

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**Background and Purpose** The study aim was to determine the effects of benign joint hypermobility syndrome (BJHS) on symptom severity and functional capacity in females with carpal tunnel syndrome (CTS) based on the findings of physical examinations.

**Methods** One hundred female patients diagnosed with bilateral CTS in electrophysiological testing were included in this study. The participants were evaluated for BJHS using the Brighton 1998 criteria, and divided into two groups: one consisting of 56 CTS patients with BJHS, and the other comprising 44 CTS patients without BJHS. Tinel's, Phalen's, and reverse Phalen's tests were applied to all patients, and the severity and functional capacity of CTS were evaluated using the Boston Carpal Tunnel Syndrome Questionnaire.

**Results** Symptom severity and functional capacity varied significantly between the two groups in the right hand ( $p=0.037$  and  $p=0.039$ , respectively) and in the left hand ( $p=0.016$  and  $p=0.029$ ). The hypermobile group yielded more positive results on the right side during Tinel's, Phalen's, and reverse Phalen's tests ( $p=0.032$ ,  $p=0.032$ , and  $p=0.018$ , respectively).

**Conclusions** Hypermobility in females exacerbated the symptoms of CTS and led to a further reduction of functional capacity. Therefore, hypermobility should be tested and an intense exercise program should be implemented in BJHS patients, especially in females with CTS.

**Keywords** carpal tunnel syndrome; benign hypermobility syndrome; entrapment neuropathy.

## INTRODUCTION

Carpal tunnel syndrome (CTS) is the most frequent form of entrapment neuropathy, and is caused by entrapment of the median nerve between the carpal bones and the transverse ligament of the wrist. CTS has a prevalence of 10% of global and affects 3% of females and 1% of males.<sup>1</sup>

Benign joint hypermobility syndrome (BJHS) is a disorder without rheumatic features that impacts the locomotor system. It is a connective-tissue disease characterized by joint instability, and leads to numerous symptoms that range from minor skin changes to chronic pain. The reported prevalence of BJHS varies from 4%–13%, and it is more common in females.<sup>2-5</sup>

Only one study reported on in the literature has explored the association between BJHS and CTS.<sup>6</sup> Those authors proposed that BJHS could be a predisposing factor for CTS, and vice versa, but there was a lack of clinical data to support this claim. To the best of our knowledge, the effect of BJHS on the clinical characteristics of CTS has not been assessed previously. Thus, the current study is the first to evaluate the effects of BJHS on symptom sever-

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ity and functional capacity in females with CTS based on the findings of physical examinations.

## METHODS

The methodology used in the current study was consistent with the relevant guidelines and regulations. The Ethics Committee of Harran University approved this study (HRU/19.05.14). Written informed consent was obtained from the study participants.

### Patients

One hundred female patients who had been diagnosed with CTS based on electrophysiological testing were included in the research. Patients with risk factors for CTS, including diabetes mellitus, pregnancy, and rheumatic diseases, were excluded. CTS rarely affects males, and so males were also excluded.

The participants were divided into two groups and evaluated according to the Brighton 1998 criteria for hypermobility:<sup>7</sup> Groups 1 and 2 comprised patients with and without BJHS, respectively. Tinel's, Phalen's, and reverse Phalen's tests were performed. The participants also completed the Boston Carpal Tunnel Syndrome Questionnaire to determine their symptom severity and functional status.<sup>8</sup>

### Instruments to assess benign joint hypermobility syndrome

The Beighton Scoring System criteria were developed to diagnose hypermobility, and they have been widely accepted owing to their ease of use.<sup>9</sup> The Beighton Scoring System measuring instrument consists of five items. The first four items are assessed on the right and left sides. Each hypermobility-related item is scored as either 0 or 1 point, to give total minimum and maximum scores of 0 and 9, respectively. The patient is considered hypermobile if four of the findings are positive.

Since the Beighton Scoring System only documents the degree of hypermobility, Grahame et al.<sup>7</sup> revised the criteria to create a broader evaluation, termed the 1998 Brighton criteria. According to this scale, a diagnosis of hypermobility is made based on the presence of 1) two major criteria, 2) two minor criteria plus one major criterion, 3) four minor criteria, or 4) BJHS in a first-degree relative and two minor positive criteria. The Brighton 1998 criteria were used to diagnose hypermobility in the current study.

### Electrophysiological evaluation

In the current study, a sensorial conduction velocity and distal motor latency of  $\leq 40$  m/s and  $\leq 4$  ms, respectively, signified mild CTS, while values of  $\leq 40$  m/s and  $\geq 4$  ms indicated

moderate CTS.<sup>10</sup> Patients with severe CTS were excluded from the study.

### Provocative tests

#### Tinel's test

To perform Tinel's test, the median nerve of the patient located between the distal part of the transverse carpal ligament and the proximal wrist line was tapped six times. If this induced the experience of tingling, paresthesia, or an electrical shock sensation in the distribution of the median nerve, the test was classified as positive.

#### Phalen's test

To perform Phalen's test, both of the patient's wrists were placed in complete flexion while the dorsal parts made contact for 60 s. If this induced pain or numbness in the distribution of the median nerve, the test was classified as positive.

#### Reverse Phalen's test

To perform the reverse Phalen's test, the wrist was completely extended while the palms of the hands touched for 60 s. If this induced pain or numbness in the distribution of the median nerve, the test was classified as positive.

### Boston Carpal Tunnel Syndrome Questionnaire

The Boston Carpal Tunnel Syndrome Questionnaire consists of a 2-part symptom severity score (11 questions) and a functional status score (8 questions).<sup>8</sup> Each question is scored from 1 to 5, where a score of 1 indicates optimal functional status with mild symptom severity, and a score of 5 indicates poor functional status with severe symptom severity. The total scores for symptom severity score and functional status were determined from the cumulative responses to the corresponding 11 and 8 questions.

### Statistical analysis

SPSS (version 20, IBM Corp., Armonk, NY, USA) was used for the statistical analysis. The distribution of numerical data was evaluated using the Shapiro–Wilk test. The Mann–Whitney U test was used to evaluate numerical data that did not conform to a normal distribution. The numerical data were presented as median (range) values. The chi-square test was utilized to compare categorical data. Results with a probability value of  $p < 0.050$  were considered statistically significant.

## RESULTS

The participants' data are presented in Tables 1 and 2. Group 1 (BJHS) comprised 56 patients aged  $41.96 \pm 10.76$  years (mean  $\pm$

**Table 1.** Comparison of results from electrophysiological evaluations and the Boston Carpal Tunnel Syndrome Questionnaire between group 1 (benign joint hypermobility syndrome [BJHS]) and group 2 (without BJHS)

	Group 1 (n=56)	Group 2 (n=44)	p
Body mass index (kg/m <sup>2</sup> )	30.12 (19.36–41.72)	28.31 (19.36–36.33)	0.033*
Age (yr)	41.96 (24.00–67.00)	46.95 (19.00–67.00)	0.025*
Symptoms duration (mon)	48 (4–336)	48 (1–192)	0.076
Right-hand symptom severity	31.06 (10.00–50.00)	26.79 (12.00–43.00)	0.037*
Left-hand symptom severity	30.67 (12.00–50.00)	26.43 (12.00–43.00)	0.039*
Right-hand functional capacity	24.80 (8.00–39.00)	21.22 (8.00–38.00)	0.016*
Left-hand functional capacity	23.54 (8.00–36.00)	21.00 (8.00–38.00)	0.029*
Right-hand distal motor nerve latency (ms)	4.41 (3.20–10.30)	4.37 (3.17–6.83)	0.336
Left-hand distal motor nerve latency (ms)	4.28 (2.75–9.58)	4.33 (3.25–8.40)	0.945
Right-hand sensory nerve conduction velocity (m/s)	42.90 (10.10–91.40)	40.60 (18.30–65.80)	0.627
Left-hand sensory nerve conduction velocity (m/s)	41.20 (10.10–92.10)	43.86 (17.00–85.30)	0.261

Data are median (range) values. The total scores for symptom severity and functional capacity scores were determined using the Boston Carpal Tunnel Syndrome Questionnaire.<sup>8</sup>

\* $p < 0.050$ .

**Table 2.** Comparison of provocative tests results between groups

	Group 1 (n=56)	Group 2 (n=44)	p
Right-hand Tinel's test			0.032*
Positive	47 (83.9)	29 (65.9)	
Negative	9 (16.1)	15 (34.1)	
Left-hand Tinel's test			0.230
Positive	37 (66.1)	25 (56.8)	
Negative	19 (33.9)	19 (43.2)	
Right-hand Phalen's test			0.032*
Positive	39 (69.6)	18 (40.9)	
Negative	17 (30.4)	26 (59.1)	
Left-hand Phalen's test			0.183
Positive	44 (78.6)	28 (63.6)	
Negative	12 (21.4)	16 (36.4)	
Right-hand reverse Phalen's test			0.018*
Positive	48 (85.7)	29 (65.9)	
Negative	8 (14.3)	15 (34.1)	
Left-hand reverse Phalen's test			0.326
Positive	31 (55.4)	20 (45.5)	
Negative	25 (44.6)	24 (54.5)	

Data are n (%) values.

\* $p < 0.050$ .

SD), and group 2 (without BJHS) consisted of 44 patients aged  $46.95 \pm 11.81$  years ( $p = 0.025$ ). Although body mass index (BMI) was higher than ideal in both groups, it was comparatively significantly higher in the hypermobile group ( $p = 0.033$ ). There was no significant difference in symptom duration between the groups ( $p = 0.076$ ).

There were significant differences between the groups in the increase in symptom severity and the decrease in functional capacity in the right hand ( $p = 0.037$  and  $p = 0.016$ , respectively) and the left hand ( $p = 0.039$  and  $p = 0.029$ ). Symptoms

and functionality were increased and decreased, respectively, in group 1. The findings of Tinel's, Phalen's, and reverse Phalen's tests differed significantly between the groups for the right hand ( $p = 0.032$ ,  $p = 0.032$ , and  $p = 0.018$ , respectively) but not for the left hand. There was no significant intergroup difference in electrophysiological data for the distal motor latency or sensorial conduction of the median nerve.

## DISCUSSION

This study found that BJHS aggravated CTS symptoms and further decreased functional capacity in females.

Various physical examinations are used to identify CTS, the most common of which are Tinel's, Phalen's, and reverse Phalen's tests.<sup>11–13</sup> In the current study, Tinel's test of the right hand produced a positive result in 84% of those in the hypermobile group and 66% of the nonhypermobile group, with this difference being statistically significant. Similarly, Phalen's and reverse Phalen's tests produced significantly more positive findings in the right hand than in the left hand (70% and 86%, respectively, in the hypermobile group, and 41% and 66% in the nonhypermobile group).

The higher rate of positivity identified in the right hands of participants in the BJHS group using Tinel's, Phalen's, and reverse Phalen's tests was attributed to the right hand generally being exposed to more recurrent trauma due the higher prevalence of right-hand dominance and associated greater use of the right hand in daily life.

The exact cause of neurological symptoms in BJHS remains unclear.<sup>14,15</sup> Nerve injury may be induced by recurrent trauma caused by joint laxity. A study of 114 BJHS patients diagnosed 36 patients with CTS neurophysiologically.<sup>16</sup> Another study neurophysiologically diagnosed CTS in only 1 of 15 pa-

tients with hypermobile Ehlers–Danlos Syndrome.<sup>17</sup> A study that evaluated the clinical profiles of BJHS patients identified CTS in only 1 of 84 patients.<sup>18</sup> There is only one report of BJHS possibly being responsible for CTS,<sup>6</sup> indicating that the relationship between CTS and BJHS has not been adequately explored or demonstrated. The present study identified BJHS in 56 (56%) of 100 patients with CTS.

Body weight is a risk factor for the development of both CTS and BJHS.<sup>19–21</sup> BMI in the present study was significantly higher in the hypermobile patients than in the nonhypermobile group, which supports the premise that high BMI is a risk factor for CTS and BJHS.

The study had some limitations. The sample was small, and males were excluded. There is therefore a need for research involving larger samples that include both sexes.

## Conclusion

This study was the first to investigate the effects of BJHS on CTS symptom severity and functional capacity. The findings showed that BJHS increases symptom severity and decreases functional capacity in patients with CTS. Therefore, hypermobility should be tested and an intense exercise program should be implemented, especially in females with CTS. Further studies with large case series are warranted to obtain more information on this subject and confirm the findings of the current study.

## Availability of Data and Material

The datasets generated or analyzed during the study are not publicly available due to privacy and ethical concerns, but are available from the corresponding author on reasonable request.

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## Conflicts of Interest

The authors have no potential conflicts of interest to disclose.

## Funding Statement

None

## REFERENCES

- de Krom MC, Kester AD, Knipschild PG, Spaans F. Risk factors for carpal tunnel syndrome. *Am J Epidemiol* 1990;132:1102–1110.

- Castori M, Morlino S, Celletti C, Ghibellini G, Bruschini M, Grammatico P, et al. Re-writing the natural history of pain and related symptoms in the joint hypermobility syndrome/Ehlers-Danlos syndrome, hypermobility type. *Am J Med Genet A* 2013;161:2989–3004.
- Grahame R, Hakim AJ. Hypermobility. *Curr Opin Rheumatol* 2008;20:106–110.
- Seçkin U, Tur BS, Yılmaz O, Yağcı I, Bodur H, Arasil T. The prevalence of joint hypermobility among high school students. *Rheumatol Int* 2005;25:260–263.
- Reuter PR, Fichthorn KR. Prevalence of generalized joint hypermobility, musculoskeletal injuries, and chronic musculoskeletal pain among American university students. *PeerJ* 2019;7:e7625.
- Aktas I, Ofluoglu D, Albay T. The relationship between benign joint hypermobility syndrome and carpal tunnel syndrome. *Clin Rheumatol* 2008;27:1283–1287.
- Grahame R, Bird HA, Child A. The revised (Brighton 1998) criteria for the diagnosis of benign joint hypermobility syndrome (BJHS). *J Rheumatol* 2000;27:1777–1779.
- Levine DW, Simmons BP, Koris MJ, Daltroy LH, Hohl GG, Fossel AH, et al. A self-administered questionnaire for the assessment of severity of symptoms and functional status in carpal tunnel syndrome. *J Bone Joint Surg Am* 1993;75:1585–1592.
- Beighton P, Solomon L, Soskolne CL. Articular mobility in an African population. *Ann Rheum Dis* 1973;32:413–418.
- Johnson EW. Diagnosis of carpal tunnel syndrome. The gold standard. *Am J Phys Med Rehabil* 1993;72:1.
- Mohamed FI, Hassan AA, Abdel-Magied RA, Wageh RN. Manual therapy intervention in the treatment of patients with carpal tunnel syndrome: median nerve mobilization versus medical treatment. *Egypt Rheumatol Rehabil* 2016;43:27–34.
- Hupaló M, Smigielski J, Fortuniak J, Jaskolski DJ. Value of oxymyography, based on near infrared spectroscopy, in the diagnosis of carpal tunnel syndrome in comparison to provocative clinical diagnostic tests and nerve conduction studies. *Clin Neurophysiol* 2018;129:327–332.
- Lee HJ, Kim IS, Sung JH, Lee SW, Hong JT. Intraoperative dynamic pressure measurements in carpal tunnel syndrome: correlations with clinical signs. *Clin Neurol Neurosurg* 2016;140:33–37.
- March LM, Francis H, Webb J. Benign joint hypermobility with neuropathies: documentation and mechanism of median, sciatic, and common peroneal nerve compression. *Clin Rheumatol* 1988;7:35–40.
- Shapiro SK. A case of Meekrin-Ehlers-Danlos syndrome with neurologic manifestations. *J Nerv Ment Dis* 1952;115:64–71.
- el-Shahaly HA, el-Sherif AK. Is the benign joint hypermobility syndrome benign? *Clin Rheumatol* 1991;10:302–307.
- Granata G, Padua L, Celletti C, Castori M, Saraceni VM, Camerota F. Entrapment neuropathies and polyneuropathies in joint hypermobility syndrome/Ehlers-Danlos syndrome. *Clin Neurophysiol* 2013;124:1689–1694.
- Mullick G, Bhakuni DS, Shanmuganandan K, Garg MK, Vasdev V, Kartik S, et al. Clinical profile of benign joint hypermobility syndrome from a tertiary care military hospital in India. *Int J Rheum Dis* 2013;16:590–594.
- Zyluk A, Dabal L, Szlosser Z. Association of anthropometric factors and predisposition to carpal tunnel syndrome. *Chir Narzadow Ruchu Ortop Pol* 2011;76:193–196.
- Ebrahimzadeh MH, Mashhadinejad H, Moradi A, Kachooei AR. Carpal tunnel release in diabetic and non-diabetic patients. *Arch Bone Jt Surg* 2013;1:23–27.
- Rhee SY, Cho HE, Kim JH, Kim HS. Incidence and reappraisal of known risk factors associated with carpal tunnel syndrome: a nationwide, 11-year, population-based study in South Korea. *J Clin Neurol* 2021;17:524–533.