Translation into Brazilian Portuguese and validation of the five-part questionnaire for identifying hypermobility

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ABSTRACT

Introduction: Joint hypermobility (JH) is an inherited clinical condition with increased joint elasticity in passive movements. In the general population, its frequency, which can be estimated through specific methods, such as the nine-point Beighton hypermobility score (Beighton score) and the self-reported five-part questionnaire for identifying hypermobility (five-part questionnaire), ranges from 10% to 20%. **Objectives:** To validate the Portuguese version of the five-part questionnaire and to determine its sensitivity and specificity when compared with the Beighton score for diagnosing JH. **Methods**: The five-part questionnaire for identifying hypermobility was translated into Portuguese and applied to 2,523 Brazilian university students. Then, a sample with 394 randomly selected students was evaluated by use of the Beighton score, aiming at establishing the JH diagnosis. Finally, the two methods were statistically compared. **Results:** The JH frequency was 37.01% when using the five-part questionnaire, and 34% when using the Beighton score. Considering sex, the JH frequencies according to the five-part questionnaire and Beighton score were 43.54% and 44.26% in females, and 28.44% and 16% in males, respectively. The sensitivity of the self-reported questionnaire was 70.9% and its specificity was 77.4%, with an area under the receiver operating characteristic (ROC) curve of 0.786. **Conclusions:** JH is frequent in Brazilian university students, and more common in women. The self-reported five-part questionnaire for JH identification, translated into Portuguese and validated, was an effective method when compared with the Beighton score for identifying JH.

Keywords: joint instability, validation studies, self-assessment programs, questionnaires.

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INTRODUCTION

Joint hypermobility (JH) is an inherited clinical condition characterized by increased joint elasticity in passive movements and hypermobility in active movements.[1,2] Its prevalence in the general population ranges from 10% to 20%, decreases with age, and is more common in women than men.[3,4,5,6] A wide ethnic variation exists,[3,4,7,8] and JH is more frequent in Asians than in Africans, who are more affected than Caucasians.[3,4,9]

Although most individuals with JH do not have symptoms,[10] over the past years, JH has shown to be one of the major causes of chronic pain.[11] However, many patients with JH and chronic pain remain undiagnosed.[12]

Joint hypermobility can be associated with several symptoms, which are not only related to the musculoskeletal system. Arthralgia is one of the most frequent symptoms, and can affect 31% of the individuals.[13] Patients with JH have a higher frequency of soft tissue rheumatisms (tendinitis, bursitis, fasciitis, and fibromyalgia),[14,15] and chronic fatigue syndrome.[16,17] The associated extra-articular manifestations are as follows: anxiety disorders, such as panic disorder and agoraphobia;[11,18] mitral valve prolapse;[19,20] dysautonomias;

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varices; rectal and uterine prolapses;[15] increased skin elasticity (striae); and increased palpebral elasticity.[13]

Despite its severe impacts on the individuals' quality of life,[21] JH is still controversial for many rheumatologists, and tests for its identification have not yet been included in the routine physical examination. Thus, the JH syndrome is likely to be underdiagnosed and underestimated by most rheumatologists.[22]

The Beighton score is the most used method for diagnosing JH.[1,18,21] Although it is an important score for epidemiological studies, the Beighton score is not an adequate tool for self-assessment. It requires the physical examination of specific joints, does not consider the individual's previous mobility, and its cutoff point for score positivity of four out of nine is arbitrary. In addition, it excludes the assessment of common sites of hypermobility, such as the neck, shoulders, hips, and ankles, generating a significant number of "false negatives".

Aiming at overcoming that difficulty, in 2003, Hakim and Grahame[23] presented the five-part questionnaire for identifying hypermobility. It is a questionnaire with five self-reported questions, in the YES/NO format, in which the presence of at least two affirmative answers identify previous or current JH history. So far, that questionnaire has only been applied to the British population, and its results are in accordance with the Beighton score. The five-part questionnaire for identifying hypermobility correctly identified 84% of the individuals, with sensitivity of 77%-85% and specificity of 89%.

Thus, this study aimed at validating the Portuguese version of the self-reported five-part questionnaire for identifying hypermobility,[23] and at determining its sensitivity and specificity for identifying JH as compared with the Beighton score.

MATERIALS AND METHODS

Validation of the Hakim & Grahame questionnaire in the Portuguese language

Translation of the questionnaire

Initially, the original English version of the five-part questionnaire for identifying hypermobility, shown in Figure 1, was translated into Portuguese by a scientific translator and four Brazilian rheumatologists officially fluent in English. The five versions obtained were compared and discussed by the five translators, and a consensual version was achieved. That consensual version was backtranslated independently by two other scientific translators, one of them native, who did not have access to the original English version. The native translator chose between the two backtranslations the most adequate one. Then the first five translators compared the original English version of the questionnaire with the backtranslated one, and reliability between both versions was observed.

The Portuguese version of the questionnaire was then applied (pilot testing) to a reduced number of individuals (60 students of the medicine, psychology, physical therapy, and occupational therapy courses of the Universidade de São Paulo (USP), Ribeirão Preto campus) to assess the difficulties in understanding that could emerge during the questionnaire application. The suggestions and difficulties in understanding of the participants of the pilot-testing group were considered in the formulation of the final Portuguese version of the questionnaire, and illustrations were added. The adapted version was once again applied to the pilot-testing group, when total understanding of the questions was achieved. Figure 2 shows the Portuguese version of the questionnaire applied in the study with the changes performed after pilot testing.

The final questionnaire containing the changes implemented after pilot testing was sent to the authors of the original questionnaire, Alan Hakim and Rodney Grahame, in England for appreciation. That adapted version was fully approved.

Please mark with a cross the reply you consider correct.

1. Can you now (or could you ever) place your hands flat on the floor without bending your knees?

☐ Yes

☐ No

2. Can you now (or could you ever) bend your thumb to touch your forearm?

☐ Yes

☐ No

3. As a child did you amuse your friends by contorting your body into strange shapes or could you do the splits?

☐ Yes

☐ No

4. As a child or teenager did your shoulder or kneecap dislocate on more than one occasion?

☐ Yes

☐ No

5. Do you consider yourself double-jointed?

☐ Yes

☐ No

Thank you very much for your collaboration. Please feel free to ask if you do not understand some of the questions.

Figure 1
Original self-reported questionnaire for identifying joint hypermobility, in English, created by Alan Hakin and Rodney Grahame.

Perguntas para diagnóstico da Síndrome de Hipermobilidade

Por favor, assinale com um X a resposta que considerar correta.

	1. Você consegue (ou já conseguiu) colocar as palmas das mãos completamente estendidas no chão sem dobrar os joelhos? (Veja figura) ☐ Sim ☐ Não
	2. Você consegue (ou já conseguiu) dobrar para trás o seu polegar até tocar o seu antibraço? (Veja figura) ☐ Sim ☐ Não
	3. Quando criança você divertia seus amigos contorcendo o seu corpo em posições estranhas OU podia abrir completamente as pernas , como bailarina? ☐ Sim ☐ Não
	4. Quando criança ou adolescente você já deslocou ou o ombro ou a patela (a rótula do joelho) em mais de uma ocasião? ☐ Sim ☐ Não
	5. Você se considera uma pessoa mais flexível que o normal? ☐ Sim ☐ Não

Muito obrigado pela sua colaboração. Sinta-se a vontade para perguntar no caso de não compreender alguma das perguntas.

Figure 2
Portuguese version of the self-reported questionnaire for identifying joint hypermobility used in this study with the changes performed after pilot testing.

Application of the instrument (QUESTIONNAIRE)

In the first phase of the study, the final version of the self-reported questionnaire for identifying JH translated into Portuguese was applied to 2,523 individuals, of whom one was an elementary school student and 2,522 attended the following three universities: USP, at the Ribeirão Preto Campus; Universidade de Franca (Unifran); and Centro Universitário Barão de Mauá, in the city of Ribeirão Preto. The university students attended the following courses: medicine (1st to 4th year, 609 students); nursing; psychology; physical therapy; occupational therapy; law; chemistry; medical physics; and speech therapy. The questionnaire was applied collectively in a classroom. The mean time of application was two minutes.

Application of the Beighton score

In the second phase of the study, six months after the application of the self-reported questionnaire for identifying JH, of the 2,523 participants, 394 were randomly selected for the Beighton score application. The sex and age characteristics of such participants matched those of the sample in the first phase.

In this phase, the participants answered once again the self-reported questionnaire for identifying JH, and then the Beighton score was applied at an adequate place. The examiner had no access to the completed questionnaires.

Beighton score

For the Beighton score, patients are attributed a numerical score from 0 to 9, one point being added to the ability to perform each of the following tests. A score of four or more points establishes the diagnosis of JH.[1,24] The tests were as follows:

1. Little finger bending backward past 90°: one point is added for each side (right and left) affected, and a total of two points can be here added.
2. Thumb touching the forearm: one point is added for each side (right and left) affected, and a total of two points can be here added.
3. Elbow bending backward more than 10°: the maneuver should be performed to the right and left sides. If the measure exceeds 10°, one point is added for each side affected, and a total of two points can be here added.
4. Knee bending backward more than 10°: If the measure exceeds 10°, one point is added for each side affected, and a total of two points can be here added.
5. Placing flat hands on the floor with straight legs: when this objective is achieved, one point is added.

Then each participant had his/her achieved Beighton score quantified by the examiner.

The scores of all participants assessed were analyzed and later compared with the data obtained by use of the self-reported questionnaire for identifying JH.

Analysis of the comparison between the self-reported questionnaire for identifying joint hypermobility and the Beighton score

Considering that the Beighton score is an efficient way to diagnose JH, we aimed at establishing the sensitivity and specificity of the self-reported questionnaire for identifying JH for the population studied (test).

Data from the Beighton score and from the self-reported questionnaire for identifying JH were entered in the 9.0.1.1-version of the MedCalc® for Windows (MedCalc Software, Mariakerke, Belgium) program for building a

receiver operating characteristic (ROC) curve, which is a graph to assess the usefulness of a test and to determine its most appropriate cutoff point.[25] Based on that curve, the best cutoff point for the test (self-reported questionnaire for identifying JH) could be established for the population of Brazilian university students.

Regarding the answers obtained in the self-reported questionnaire for identifying JH, the following three statistical methods were used for assessing the agreement of a same participant in both phases of the study: *Kappa* (intragroup agreement); Pearson correlation coefficient; and Spearman rank correlation coefficient.[25]

The project and its written informed consent were submitted to the Committee of Ethics and Research of the HC-FMRP-USP, and both were approved. All participants or their legal representatives provided written informed consent.

RESULTS

First phase

Pilot testing

Sixty individuals participated in the pilot testing of the Portuguese version of the self-reported questionnaire for identifying JH. During questionnaire application, 18 participants had doubts related to performing the maneuver of the second question "Can you now (or could you ever) bend your thumb to touch your forearm?" For the sake of better understanding, the question was then illustrated.

The first question "Can you now (or could you ever) place your hands flat on the floor without bending your knees?" was illustrated, because the pilot-testing participants suggested so to avoid doubts at the occasion of large scale questionnaire application.

The final questionnaire with the changes implemented after pilot testing was sent to the authors of the original questionnaire, Alan Hakim and Rodney Grahame, in England for appreciation. That adapted version was fully approved.

Application of the final questionnaire

The final Portuguese version of the self-reported questionnaire for identifying JH was applied to 2,523 individuals, of whom 2,522 were university students and one was an elementary school student. All participants answered all five questions. Their ages ranged from 17 to 60 years, except for the one elementary school student aged 10 years. The greatest age concentration was between 20 and 24 years (1,476 participants, 58.50%), followed by 17 to 19 years (679 participants, 26.91%). Regarding sex, 1,433 (56.79%) participants were females. (Table 1)

During the questionnaire application, there were doubts relating to the fifth question. In the Portuguese version of that question, the presence of the word "*flexível*" (in English, flexible) led some participants to wonder whether the question referred to "not having radical attitudes", in an allusion to one of the meanings of the word "*flexível*" in Portuguese. The doubt was solved at the occasion of the questionnaire application.

Analysis of the answers to the questionnaire

Of the 2,523 participants, 747 (29.60%) did not answer affirmatively to any question of the self-reported questionnaire for identifying JH (368 females and 379 males), while 1,776 (70.39%) answered affirmatively to at least one question. Of those 1,776 participants, 842 (33.37%) provided affirmative answers to a single question (441 females and 401 males), and 934 (37.0%) answered "Yes" to at least two questions, which, according to the authors of the questionnaire, establishes the diagnosis of JH. The frequency of the disease was 43.54% in the female sex and 28.44% in the male sex.

Most of the 842 (33.37%) affirmative answers to a single question were distributed as follows: question 1 (430, 51%); question 2 (251, 29.8%); question 3 (102, 12%); question 4 (35, 4.1%); and question 5 (24, 2.8%).

Table 1
Comparison of the age groups and sex of participants in the first and second phases of the study in absolute and percentage numbers in relation to the total number of participants in each phase

Age (years)	Number 1st phase (%) / 2nd phase (%)	Female sex 1st phase (%) / 2nd phase (%)	Male sex 1st phase (%) / 2nd phase (%)
10	1 (0.03) / 1 (0.25)	0 (0) / 0 (0)	1 (0.03) / 1 (0.25)
17- 19	679 (26.91) / 80 (20.3)	457 (18.11) / 62 (15.73)	222 (8.79) / 18 (4.56)
20- 24	1.475 (58.46) / 209 (53)	794(31.47) / 132 (33.50)	681 (26.99) / 77 (19.54)
25- 29	248 (9.82) / 55 (13.95)	118 (4.67) / 31 (7.86)	130 (5.15) / 24 (6.09)
30- 34	74 (2.93) / 16 (4)	40 (1.58) / 10 (2.53)	34 (1.34) / 6 (1.52)
35- 39	26 (1) / 13 (3.29)	12 (0.47) / 6 (1.52)	14 (0.55) / 7 (1.77)
40- 44	8 (0.31) / 8 (2)	5 (0.19) / 5 (1.26)	3 (0.11) / 3 (0.76)
> 45	12 (0.47) / 12 (3)	7 (0.27) / 7 (1.77)	5 (0.19) / 5 (1.26)
Total	2523 (100) / 394 (100)	1433(56.79) / 253(64.21)	1090(43.20) / 141(35.78)

Affirmative answers to two questions were provided by 580 participants. The most common combinations were questions 1+2 (33%) and questions 1+3 (31%), and the least common combinations were questions 3+5 (1.3%) and 4+5 (0.5%).

Of the 2,523 participants, 250 (9.9%) provided affirmative answers to three questions. Ten such combinations of answers were provided, but the most common combination was questions 1+2+3 (49.6%), while the least common combinations were questions 1+4+5 (0.8%) and questions 2+4+5 (0.8%).

Of the 2,523 participants, 90 (3.56%) answered affirmatively to four questions, and five combinations of four affirmative answers were observed. The most common combination of four affirmative answers was questions 1+2+3+5 (74.4%). Fourteen (0.55%) participants answered affirmatively to five questions.

For all sums of affirmative answers, most responders were between 20 and 24 years of age, and the female sex predominated.

The participants diagnosed as having JH by use of the Beighton score, and who were asymptomatic, were referred for follow-up at the rheumatology outpatient clinics.

Second phase

Application of the Beighton score

The physical examination for applying the Beighton score was performed in 394 randomly chosen participants, of whom 253 were females and 141 males. The ages of most participants in the second phase ranged from 17 to 24 years. Their comparison with the population of 2,523 responders to the self-reported questionnaire for identifying JH showed that both groups had similar characteristics regarding sex and age (Table 1).

The 394 participants selected for the Beighton score application answered the self-reported questionnaire for identifying JH once again. Data then obtained were compared with those obtained in the first phase, and, as shown in Table 2, similarities in the sum of affirmative answers were observed.

Those analyses lead to the conclusion that the populations of the first and second phases of the study are similar, and that the application of the questionnaire and of the physical examination (Beighton score) for diagnosing JH in one phase can be inferred for the other.

According to the Beighton score, of the 394 participants, 134 (34%) were diagnosed as having JH, and 112 (44.26%) were females and 22 (16%) males.

Self-reported questionnaire for identifying joint hypermobility versus the Beighton score

The Beighton score was performed in 394 participants. Then, the information derived from the previously completed self-reported questionnaire for identifying JH and data from the physical examination were correlated.

When the criterion adopted for diagnosing JH (positive test) was two or more affirmative answers to the self-reported questionnaire for identifying JH, of the 394 participants, a positive test was observed in 156 participants. Of those 156 participants, 93 had a physical examination compatible with the disease, while 63 did not.

Of the 394 participants, a negative test was observed in 238, in whom, the application of the Beighton score showed disease in 41 individuals, and absence of disease in 197 (Table 3).

Based on those data, the sensitivity of the test was 0.69 (93/134), while its specificity was 0.75 (197/260).

Table 2
Comparison of the sum of affirmative questions in each questionnaire according to sex: distribution in absolute and percentage numbers in relation to the total number of participants in each phase

Sum of affirmative questions	Total 1st phase (%) / 2nd phase (%)	Female sex	Male sex
Zero	747 (29.6) / 115 (29.18)	368 / 63	379 / 52
1	842 (33.37) / 23(31.21)	441 / 72	401 / 51
2	580 (22.98) / 90 (22.84)	377 / 67	203 / 23
3	250 (9.9) / 46 (11.67)	180 / 37	70 / 9
4	90 (3.56) / 16 (4.06)	57 / 11	33 / 5
5	14(0.55) / 4 (1.01)	10 / 3	4 / 1
Total	2523 / 394	1433/ 253	1090 / 141

A ROC curve was built with those data with a 0.786-area under the curve (Figure 3). The sensitivity, specificity, positive predictive value (+PV), and negative predictive value (-PV) are shown in Table 4.

It is worth noting that the criterion that provides greater balance between sensitivity and specificity is "more than one affirmative question", as also observed in the original questionnaire by Grahame and Hakim.[23]

Correlation between the questionnaires completed by the same participant in the first and second phases of the study

Of the participants in the second phase of the study, 211 answered once again the self-reported questionnaire for identifying JH. Based on the answers of the same participant on two different occasions, the intragroup agreement (*Kappa*) was calculated for each of the five questions of the questionnaire, in addition to the Pearson correlation coefficient, and the Spearman rank correlation coefficient. Good agreement was observed for questions 1 (*K*: 0.63), 2 (*K*: 0.7), and 3 (*K*: 0.65), while moderate agreement was observed for questions 4 (*K:* 0.57) and 5 (*K*: 0.48) (Table 5).

DISCUSSION

Joint hypermobility is an extremely prevalent hereditary clinical entity, and the Beighton score is the most used method for its identification.

In an attempt to facilitate large scale epidemiological studies on JH, Alan Hakim and Robert Grahame have developed a self-reported questionnaire.[23] The use of a self-reported questionnaire could represent an alternative in situations in which physical examination is not feasible, particularly when the physical examination does not comprise the assessment of all joints and does not take the individual's past flexibility into consideration.

In Brazil, published studies about JH are scarce[26,27] and, in addition to having been performed on children, the number of participants was small. We decided, thus, to validate the self-reported questionnaire for identifying JH.

During pilot testing, on the occasion of the questionnaire application, there were doubts related to the second question. Participants wondered whether the thumb should touch the anterior or posterior part of the forearm. As the translated question into Portuguese used the expression "*dobrar para trás*" (in English, to bend backward), that is a plausible doubt, because, if we take the anatomical position as reference,

Table 3
Application of the test (questionnaire for identifying JH) x diagnosis of the disease (Beighton score)

	Beighton + (%)	Beighton - (%)	Total
Positive questionnaire	93 (59.6)	63 (40.4)	156
Negative questionnaire	41 (17.2)	197 (82.8)	238
Total	134	260	394

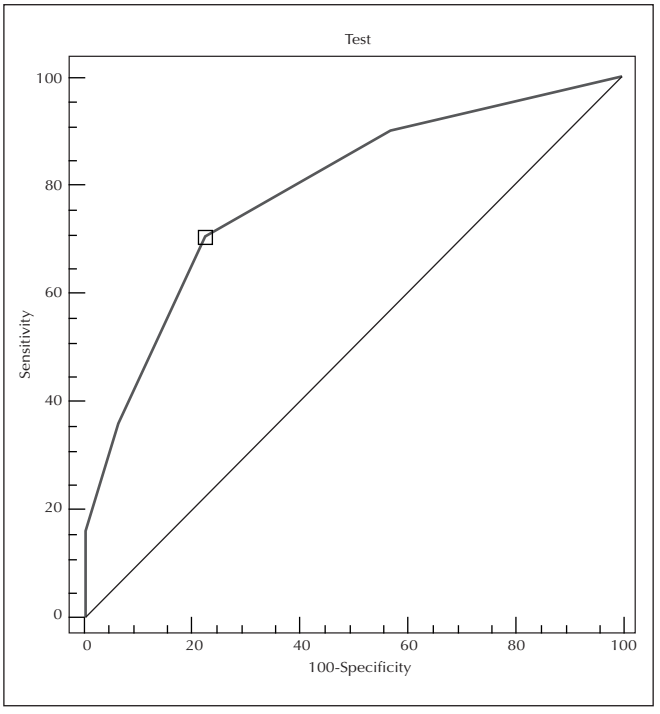

Figure 3
ROC curve obtained after applying the self-reported questionnaire for identifying JH and the Beighton score.

Table 4
Data obtained after building the ROC curve (positive or negative test x diagnosis or non-diagnosis of the disease)

Criterion	Sensitivity (95% CI)	Specificity (95% CI)	+PV	-PV
≥ 0	100 (97.1- 100)	0.0 (0.0- 1.6)	35.6	
> 0	90.6 (84.1- 95.0)	41.7 (35.3- 48.4)	46.2	88.9
> 1	**70.9 (62.1- 78.6)**	**77.4 (71.4- 82.6)**	**63.4**	**82.8**
> 2	36.2 (27.9- 45.2)	93 (88.9- 96)	74.2	72.5
> 3	15.7 (9.9- 23.3)	99.1 (96.9- 99.9)	90.9	68.1
> 4	4.7 (1.8- 10)	100 (98.4- 100)	100	65.5
> 5	0.0 (0.0- 2.9)	100 (98.4- 100)		64.4

Table 5
Statistical analyses of the correlation of the answers obtained for each question of the self-reported questionnaire for identifying JH applied in the first and second phases of the study

Question numbers	Kappa	Pearson correlation coefficient	Spearman rank correlation coefficient
1	0.63	0.6493	0.69
2	0.70	0.6954	0.56
3	0.65	0.6442	0.70
4	0.57	0.4737	0.34
5	0.48	0.50	0.52

"to bend the thumb backwards" could lead the responder to think that the thumb should touch the posterior portion of the forearm, while the question aims at assessing whether the thumb touches the anterior portion of the forearm. For the sake of better understanding, the second question was then illustrated.

The questionnaire translated into Portuguese was illustrated and applied to 2,523 participants, most of whom, university students (2,522), with ages ranging from 17 to 60 years. Considering that age bracket, this is the Brazilian study with the largest population ever assessed and one of the largest in the world. The largest ever carried out and published consisted in the application of the self-reported questionnaire to 2,600 participants in the United Kingdom.[28] In our study, most participants (85.36%) were young (between 17 and 24 years of age), in accordance with most studies performed in different countries. The JH frequency found was 37%.

In a study about the incidence of JH in a population of female twins, obtained by use of a self-reported questionnaire, Hakim *et al.*[23,28] reported that the instrument used was good, because of the following reasons: first, it was understandable, simple, and not ambiguous; second, it was fully filled out by all participants; and third, the sensitivity and specificity of the findings behaved in that population in the same manner that they behaved in the population studied.In our study, however, the final Portuguese version of the questionnaire generated two problems that we had not predicted. The first was identified during the application of the questionnaire and related to the fifth question. During the translation of the fifth question into Portuguese, an equivalent to the original English expression "*double-jointed*" was not found, and the expression was then translated into Portuguese by the word "*flexível*" (in English, flexible), which can have several meanings in Portuguese. According to an authoritative Portuguese dictionary (*Dicionário Houaiss da Língua Portuguesa*), page 1356, the Portuguese word "flexível" can mean "that bends easily; bendable; revealing agility; elastic; elegant; easily managed; tamable; ready to yield to the influence of others; adaptable; tractable, compromising; understandable". The second problem, identified only on the occasion of applying the Beighton score, was related to the second question: "Can you now (or could you ever) bend your thumb to touch your forearm?" Even with the illustration, 95 participants answered negatively to the question, but got one point in the Beighton score regarding that ability (23 with one point, and 72 with two points). It is worth wondering whether the Brazilian population is able to complete that self-reported questionnaire. According to our observations, although the population studied consisted of higher-education students, mistaken interpretations occurred. Two hypotheses were considered to justify those facts. The first is that the questionnaire can be really ambiguous. The second relates to the difficulty of understanding and interpreting of the Brazilian population. This hypothesis is evident in the last question, which, if isolated, could be really ambiguous, but within the context of the other four questions, could have its meaning directed to "bendable, capable of being flexed, elastic".

In the second phase, after applying the Beighton score, the frequency of JH observed was 34%, very close to that obtained by applying the self-reported questionnaire. Data from the self-reported questionnaire and from the Beighton score were applied to a ROC curve, in which, the highest values of sensitivity and specificity were observed simultaneously for the presence of affirmative answers to more than one question. However, in our study, with an area under the curve of 0.786, sensitivity was 70.9% (62.1-78.6), and specificity was 77.4% (71.4-82.6). In the study by Hakim and Grahame, those values were 83% and 89%, respectively.[23]

The frequency of affirmative answers provided by hypermobile individuals in our study and the frequency of affirmative answers provided by the participants in the study by Hakim and Grahame were similar.

Of the participants in the second phase of the study, 211 completed once again the self-reported questionnaire for identifying JH. With the answers provided by the same participant on two different occasions, the intragroup agreement (*Kappa*) was calculated for each of the five questions of the questionnaire. Good agreement was observed for questions 1 (*K*: 0.63), 2 (*K*: 0.7), and 3 (*K*: 0.65), while moderate agreement was observed for questions 4 (*K:* 0.57) and 5 (*K*: 0.48). Pearson correlation coefficient also showed that the data were positively correlated.

Unfortunately, since JH was first described, it has been treated much more as a curiosity than as an entity with clinical significance.[29] However, currently there is plenty of evidence that JH can cause a severe impact on the lives of affected individuals.[21] Despite all musculoskeletal and non-musculoskeletal manifestations that can be associated with JH, such a common entity is still considered controversial by several rheumatologists. This could be confirmed in a survey carried out in England in 1999, in which 92% of the rheumatologists interviewed believed that JH was a clinical entity, but only 39% considered it a distinct pathological entity. Many of them ignored its frequency, diagnostic criteria, and treatment forms. Approximately half of the physicians interviewed believed neither in the significant impact that JH could have on the lives of affected individuals, nor in its association with other rheumatic diseases.[22] These data show the difficulty experienced by physicians in recognizing such entity in their patients. In Brazil, studies about JH are rarely published, and despite its high prevalence, it is totally forgotten in national or regional congresses and meetings, evidencing how JH is neglected.

In conclusion, the self-reported questionnaire for identifying JH translated into Portuguese and validated has proved to be an effective method for diagnosing JH as compared with the traditional Beighton score, providing sensitivity of 70.9% and specificity of 77.4%.

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