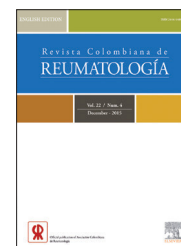




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## Original Investigation

# Relationship Between Generalised Joint Hypermobility and Joint Pain in 4 to 17 Year-old Children in Pasto, Colombia<sup>☆</sup>

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## ABSTRACT

**Introduction:** Generalised joint hypermobility (GJH) and its association with joint pain in children is controversial. So far, there are no publications from Colombia in the literature reviewed.

**Objectives:** The main objective of this study was to identify the association between GJH and joint pain in children aged 4-17 years in Pasto, Colombia.

**Materials and methods:** Descriptive, analytical type, case-control study of children from a school in Pasto, Colombia. GJH was defined using Beighton criteria, considering 5 or more criteria as positive. A questionnaire on joint pain was completed.

**Results:** A total of 576 children, with a mean age of 9.7 years (SD ± 3) were included. GJH prevalence was 52%, with 20% of children referring to joint pain. When GJH was associated with joint pain, the OR was 0.68 with  $p = .07$  (95% CI; 0.44-1.04). The comparison between localised pain in joints and hypermobility in the subjects with and without GJH, the OR was 4.80 with  $p = .0001$  (95% CI; 2.17 to 10.63).

**Conclusions:** The prevalence of GJH was 52%, being higher in girls (66%). The prevalence of pain was 20%, and 17 and 23% of children with and without GJH, respectively, had joint pain.

No association between joint pain and HAG was found in the group evaluated. The result did not change when stratified by age and gender.

It was found that children with GJH were more likely to have pain in joints and an increased range of motion than those who did not have GJH.

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## Relación entre hipermovilidad articular generalizada y dolor articular en niños de 4 a 17 años en Pasto, Colombia

### R E S U M E N

**Palabras clave:**  
Hipermovilidad  
Artralgia  
Estudiantes

**Introducción:** La hipermovilidad articular generalizada (HAG) y su asociación con dolor articular, en pediatría, sigue siendo controvertida. En la literatura revisada no se encontraron publicaciones colombianas al respecto.

**Objetivos:** El objetivo general fue identificar la relación entre HAG y dolor articular, en niños de 4 a 17 años en Pasto, Colombia.

**Materiales y métodos:** Estudio descriptivo, analítico, tipo casos y controles. Se tomó la población de un colegio de Pasto, Colombia, a quienes se les realizó una encuesta sobre dolor articular. Para definir la HAG se aplicaron los criterios de Beighton, considerando 5 o más criterios positivos.

**Resultados:** Se incluyeron 576 estudiantes, edad promedio de 9,7 años ( $\pm 3$  DE). La prevalencia de HAG fue de 52%, el 20% de los estudiantes refirió dolor articular. Al relacionar dolor articular con HAG, se observó: OR = 0,68 con  $p = 0,07$  (IC 95% 0,44-1,04). La comparación entre dolor localizado en articulaciones hipomóviles en estudiantes con HAG y sin HAG mostró OR = 4,80 con  $p = 0,0001$  (IC 95% 2,17-10,63).

**Conclusiones:** La prevalencia de HAG fue de 52%, siendo mayor en mujeres (66%). La prevalencia de dolor fue del 20%. El 17 y 23% de los estudiantes con y sin HAG, respectivamente, presentaron dolor articular.

En el grupo evaluado no se encontró relación entre dolor articular y HAG. El resultado no cambió al estratificar por grupos de edad y sexo.

Se encontró que los estudiantes con HAG tuvieron mayor probabilidad de presentar dolor en articulaciones, con rango de movilidad aumentado, que los que no tuvieron HAG.

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## Introduction

Generalised joint hypermobility (GJH) is a variation of the normal joint mobility, due to genetic alterations in the elastic fibers.<sup>1,2</sup> Its prevalence in children ranges from 4 to 58.7% and may vary depending on the age, the gender, the ethnicity and the classification criteria,<sup>3-5</sup> other studies show a prevalence of up to 58.5%.<sup>6</sup>

GJH is diagnosed using the criteria of Beighton, with a standardized and reproducible protocol, where GJH is present with 5 or more criteria of the 9 evaluated.<sup>1,7</sup> Children with this diagnosis may be asymptomatic,<sup>2</sup> but others may experience moderate or even severe musculoskeletal ailments,<sup>8</sup> and may have joint pain,<sup>9</sup> fatigue,<sup>8</sup> decreased motor activity<sup>9</sup> and muscle weakness.<sup>10</sup>

There are some studies showing a direct association between GJH and joint pain as evidenced by Abujam et al., in Indian children with an odds ratio (OR) of 4.2<sup>6</sup>; however, other studies have not found this association as it was seen in that of Leone et al.,<sup>11</sup> in Italian children. In the literature reviewed there are no studies in Colombia in this regard.

## Materials and Methods

Descriptive, analytical, case-control study. The general objective was to identify the relationship between GJH and joint

pain in children aged between 4 and 17 years, in Pasto, Colombia. The specific objectives included: to identify the prevalence of GJH and joint pain, the frequency of joint pain in children with and without GJH and to identify the relationship between GJH and joint pain, stratifying them by groups of age, gender and location of pain.

All students aged between 4 and 17 years were included; students who stated that they had already been diagnosed with a disease of the musculoskeletal system were excluded.

The sample was collected between September 8 and October 8 of 2014, in a school of basic primary, secondary and middle academic education of the city of Pasto, including students from 4 to 17 years old.

In all students were taken measurements with goniometer of the angles of the joints in extension of: the elbows, the fifth finger of the hands and the knees; bilateral apposition of the thumb to the forearm and the flexion of the trunk maintaining the extension of the knees until placing the palms of the hands on the floor.

The diagnostic criteria for GJH, according to the Beighton criteria, are:

1. Passive approximation of the thumbs over the anterior region of the forearm (right).
2. Passive approximation of the thumbs over the anterior region of the forearm (left).
3. Hyperextension of the fifth finger of the hand until being parallel to the dorsal region of the forearm (right).

4. Hyperextension of the fifth finger of the hand until being parallel to the dorsal region of the forearm (left).
5. Hyperextension of the elbows forming an angle greater than 10° (right).
6. Hyperextension of the elbows forming an angle greater than 10° (left).
7. Hyperextension of the knees forming an angle greater than 10° (right).
8. Hyperextension of the knees forming an angle greater than 10° (left).
9. Flexion of the trunk maintaining the extension of the knees until placing the palms of the hands on the floor.

It was considered positive for GJH, when the student met 5 of more of the 9 criteria, as endorsed by studies such as those of Junge et al.,<sup>1</sup> and Kristensen et al.<sup>9</sup>

It was also conducted a survey interrogating on the history of joint pain, specifically if the student have had pain in the same joint at least on 2 occasions during the last 3 months; history of fractures, sprains, morning stiffness and joint swelling.

The painful joint was specified in the survey and sub-groups were created depending on whether the pain was located in a joint with increased range of motion or in a joint with normal range of motion.

#### Statistical Analysis

All variables were subjected to an exploratory analysis in order to identify their distribution, calculating measures of central tendency and variability for the quantitative variables, and proportions for the qualitative variables.

Age stratification by five-year periods was performed. The categorical variables were compared using the Chi-square test or Fisher's test, according to the case. In the comparisons, significant differences were considered for a value of  $p < 0.05$ .

Subsequently, it was performed a bivariate analysis in order to establish the degree of correlation between the studied variables. Based on the results of the clinical variables, measures of association (OR) were estimated to contrast the hypotheses raised in the study, so that it can be established the degree of relationship between GJH and joint pain, in each group of age, between the Beighton criteria and joint pain;

**Table 1 – Sociodemographic Characteristics**

Characteristic	n (%)
Age in years (range) mean ( $\pm$ SD)	(4-17) 9.7( $\pm$ 3)
Age groups	
4 to 8 years	181 (31)
9 to 13 years	329 (57)
14 to 17 years	66 (12)
Gender	
Female	392 (68)
Male	184 (32)
Total	576 (100)

between the anatomical site of hyperlaxity and the pain in the anatomical site, between joint pain located in the site of hyperlaxity and GJH.

The data were stored in a database in Excel 2007 and later, they were statistically analyzed in the Stata program version 11.0® (College Station, Texas, USA).

## Results

### Sociodemographic information

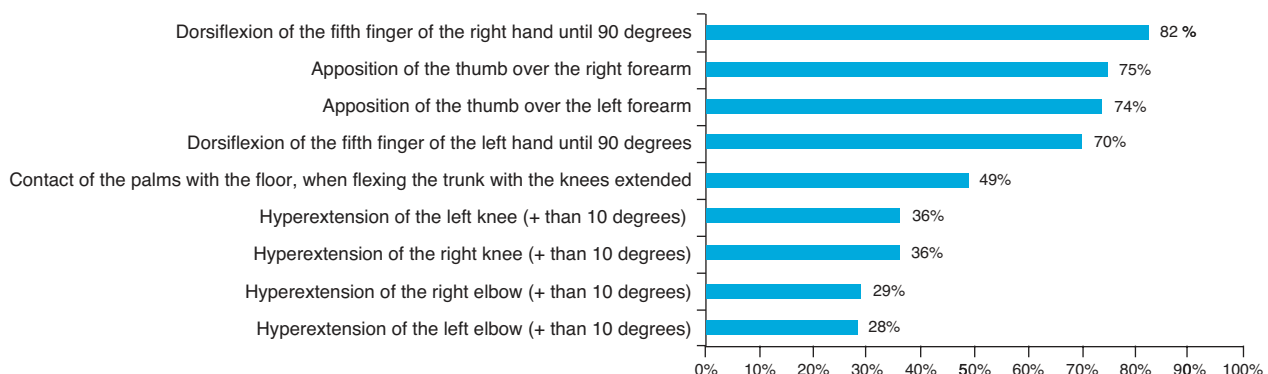
576 children of a school of basic primary, secondary and middle academic education of the city of Pasto, within a range of 4 to 17 years old were included. The sociodemographic characteristics are shown in Table 1.

### Increased Joint Mobility

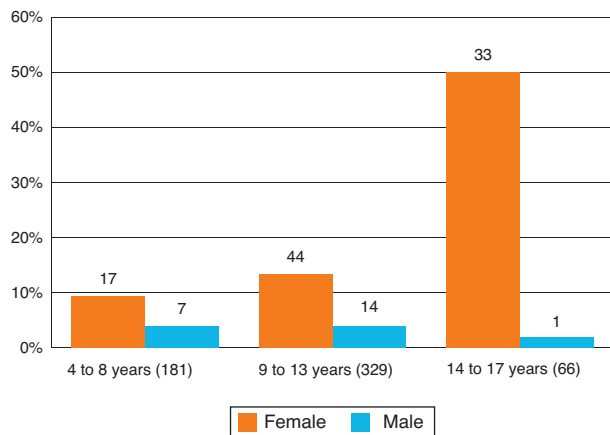
The increased range of motion in each explored joint is described in Figure 1.

### Generalised Joint Hypermobility

The prevalence of GJH in the students was 52% (302/576), being higher in the girls with a 66% (132/201), with no statistically significant differences.



**Figure 1 – Joint with increased range of motion.**



**Figure 2 – Prevalence of joint pain according to age and gender groups.**

33% of the students were diagnosed with 5 criteria, 18% with 6 criteria, 19% with 7 criteria, 12% with 8 criteria, and 18% with 9 criteria.

The prevalence of GJH in the group of 4 to 8 years was 73%, 46% in girls and 27% in boys; in the group of 9 to 13 years it was 43%, 29% in girls and 14% in boys; and in the group of 14 to 17 years it was 42%, 40% in girls and 2% in boys.

#### Joint Pain

Of the total of children interviewed, 20% (116/576) referred that they had joint pain in the last 3 months, on at least 2 occasions in the same joint.

#### Joint Pain According to Age and Gender Groups

The prevalence of joint pain discriminated by age and gender groups is shown in Figure 2.

The prevalence of joint pain increased with age, since in the group of 14-17 years, it was three times higher (52%) compared with the group between 4 and 8 years (13%) ( $p < 0.0001$ ).

Of the students who reported pain, the most painful joints were the knees in 85% (98/116), followed by the wrists 36% (42/116), the elbows 22% (26/116), metacarpophalangeal and interphalangeal joints 10% (16/116) and shoulder and heel, each with 9% (11/116).

Of the 116 children with joint pain, 53% (61/116) said that it occurred at rest and 47% (55/116) after exercise. 3.6% (21/576) of the students reported joint swelling in the last 3 months; most frequently in the knees and heels.

#### Relationship Between Generalised Joint Hypermobility and Joint Pain

52 (17%) out of the 302 students with GJH had joint pain, and 64 (23%) out of the 274 students without GJH had joint pain.

When linking joint pain with GJH, it was observed an OR = 0.68 with a  $p = 0.07$ , (CI 95%; 0.44-1.04).

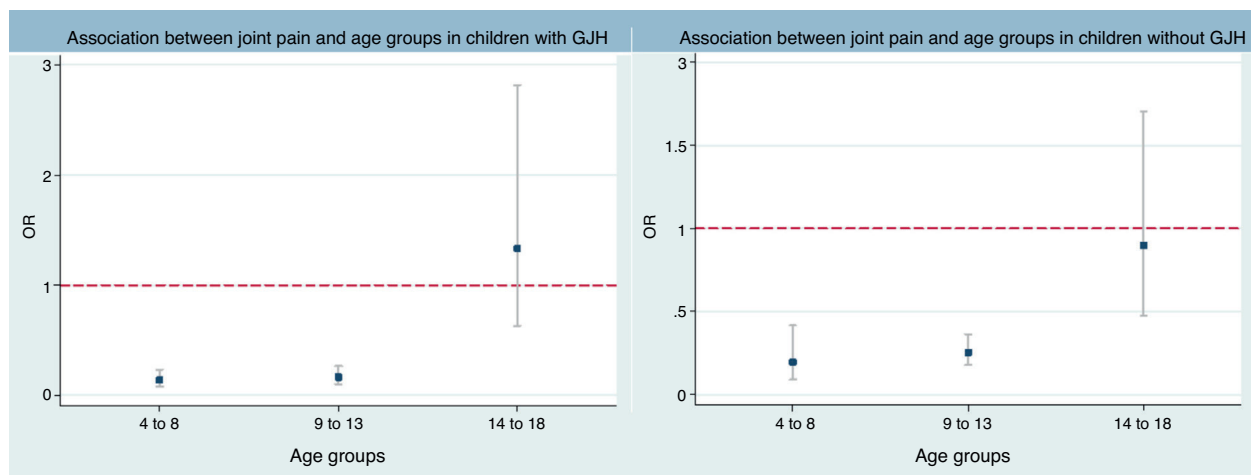
#### Relationship Between Joint Pain, Age Groups and GJH

Figure 3 shows the association between joint pain and age groups (OR- CI 95%) according to GJH.

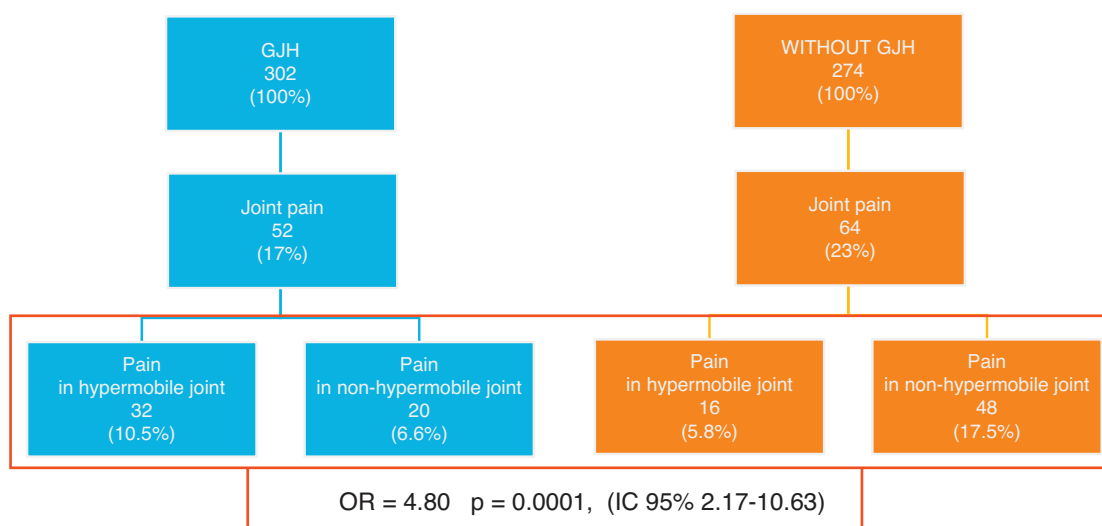
When these groups were stratified by gender it was found: in the group of 4 to 8 years an OR = 1.1 ( $p = 0.76$ ), (CI 95%; 0.53-2.36); in the group of 9 to 13 years an OR = 0.78 ( $p = 0.29$ ) (CI 95%; 0.48-1.3); in the group of 14 to 17 years it was not possible to estimate an association between gender and hyperlaxity, due to the small number of men in this range of age.

#### Relationship Between Pain in a Joint With Increased Range of Motion and GJH

Of the students with pain, 41% (48/116) had pain located in the joints with increased range of motion. Of the students who met criteria for GJH, 10.5% (32/302) had pain in the joints with increased range of motion and 6.6% (20/302) had pain in the joints with normal range of motion. Of the students without GJH, 5.8% (16/274) had pain in the joint with increased range of motion and 17.5% (48/274), had pain in a joint with normal range of motion.



**Figure 3 – Relationship between GJH and joint pain by age groups.**



**Figure 4 – Relationship between pain in joints with increased range of motion and GJH.**

A comparison between the 2 groups was made obtaining an OR = 4.80 with  $p = 0.0001$ , (CI 95%; 2.17-10.63), for pain located in the joints with increased range of motion, in the students with GJH compared with those who had pain in the joints with increased range of motion but without GJH (Fig. 4).

## Discussion

### GJH

GJH represents a variation of the normal joint mobility, with an increase in the range of motion greater than 2 standard deviations.<sup>1,12</sup> It is diagnosed in Pediatrics with the presence of 5 or more positive Beighton criteria.<sup>1,7,12</sup> The scale of Beighton has 5 dichotomous clinical maneuvers (presence or absence, for a range of results from 0 to 9. This scale has been validated and there have been conducted studies that show an adequate interobserver reproducibility.<sup>1,7</sup> It should be noted that some people may have an increased range of motion of one or more joints, but they do not meet the mentioned criteria for GJH. In our study it was taken as a definition of GJH the presence of 5 or more criteria according to the validated studies.

In our case, the prevalence of GJH was 52%, number that resembles studies such as those conducted by de-Inocencio et al.<sup>13</sup> and by Duro et al.,<sup>14</sup> developed in Spain, in which the prevalence of GJH was 55 and 53%, respectively. Indian studies show similar results with prevalence rates of 46<sup>6</sup> and 58.8%.<sup>15</sup>

However, there are reports with a much lower prevalence in countries such as the United States,<sup>16</sup> the United Kingdom<sup>17</sup> and Holland,<sup>5</sup> with prevalence rates of GJH of 12%, 19.2% and 26%, respectively. There are reports in the literature from 7.8% to 58.7%.<sup>5</sup> In all of them the prevalence was higher in women than in men, as shown also in our results where the prevalence of GJH in women was 66%, although this difference was not statistically significant in the general population.

The majority of students with GJH were in the age group of 4-8 years and it decreased in the older age groups, similar to what was found by de Inocencio et al.<sup>13</sup> and by de Abujam et al.<sup>6</sup> Likewise, Leone et al.<sup>11</sup> reported an inverse correlation between the criteria of Beighton and age.

### Joint Pain

Regarding the joint pain we found that its prevalence was 20%, this value fluctuates in the literature depending on the age of the participants in the studies. Studies with similar ages show frequencies of pain from 6.1 to 18%.<sup>6,11,18</sup>

The presence of pain was higher in the group of 14-17 years, similar to what is described in the literature, where the joint pain is more common in adolescents. In our results, the difference in pain between the group of 4-8 years and the group of 14-17 years was statistically significant.

Regarding the most frequent site of pain we found the knee in the first place, in 85% of the students with joint pain, followed by the wrists and the elbows. In the literature the knee is reported as one of the joints in which pain is more frequent.<sup>3,12</sup>

### Relationship Between Joint Pain and GJH

In our study there is no statistically significant relationship between joint pain and GJH, finding an OR of 0.68 but with no statistical significance. When the analysis by age group was performed, it was found an OR between 0 and 1 in the age groups of 4-8 years and of 9-13 years, but this finding was present both in patients with and without hyperlaxity, so there was not a statistically or clinically relevant difference either, similar to what was found in some studies conducted in Spain, the United Kingdom and Holland.<sup>7,12,18</sup> In contrast, there are studies that report a positive association between GJH and joint or musculoskeletal pain in populations similar to the one of this study,<sup>6,19</sup> or specifically in adolescents.<sup>20</sup>



McCluskey et al., in their meta-analysis, did not find a direct association between musculoskeletal pain and GJH; however, it should be clarified that the studies within this meta-analysis were greatly heterogeneous regarding age, ethnicity and criteria used.<sup>21</sup>

When we compared whether the pain was present in a joint with increased range of motion, we found that there was a significant difference, concluding with our data that in the evaluated students, those who have GJH are 4.8 times more likely to have pain in the joints with increased range of motion, compared with the students who do not have GJH. This finding has not been reported in the reviewed literature.

## Conclusions

The prevalence of GJH was 52%, being higher in women, 66%. As for the pain, its prevalence in the general population was 20%. 17% of the children with GJH had pain, while 23% of the students without GJH had pain.

In the evaluated group no relationship between joint pain and GJH was found. When stratification by age and gender groups was performed, no significant association was found either.

This study showed a statistically significant result regarding the presence of pain in the joints with increased range of motion, finding that the students with GJH were more likely to have pain in the joints with increased range of motion than those who do not have GJH.

## Ethical Disclosures

**Protection of people and animals.** The authors declare that no experiments were performed on human beings or animals for this research.

**Data confidentiality.** The authors state that patient data do not appear in this article.

**Right to privacy and informed consent.** The authors state that patient data do not appear in this article.

## Funding

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## Conflict of Interest

The authors declare that they have no conflict of interest.

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