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Frequency of Benign Joint Hypermobility Syndrome in Patients Undergoing Inguinal Hernia Repair: A Prospective Study

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The aim of this prospective study is to investigate the frequency and associated clinical findings of benign joint hypermobility syndrome (BJHS) in adult patients who were diagnosed with inguinal hernia. Benign joint hypermobility syndrome (BJHS) is defined as a clinical condition consisting of musculoskeletal symptoms such as arthralgia, pain, recurrent soft tissue disorders, joint dislocation, as well as increased normal range of motion. We hypothesized that the frequency of BHJS may increase in the patients who underwent surgery for groin hernias due to the hypermobility in connective tissue. We evaluated 66 patients in terms of BHJS prospectively in a 6-month period. The patients were divided into 2 groups. The first group consisted of 33 patients who underwent inguinal hernia repair surgery. The second group also included 33 healthy volunteers who did not have inguinal hernia operation. General joint hypermobility (GJH) was assessed using Beighton hypermobility scoring criteria, and BJHS was scored by Brighton scoring criteria. The data were assessed statistically. BJHS was diagnosed in 22 (66%) patients with inguinal hernia and in 4 (12%) healthy volunteers, respectively. Prevalence of BJHS and Beighton scores in patients with inguinal hernia were significantly higher ($P < 0.05$). GJH was found in 20 (60%) of the inguinal hernia patients (Beighton score ≥ 4; $P < 0.05$). BJHS should be considered when evaluating patients with inguinal hernia. We believe that further studies are

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needed in big cohorts to verify the relationship between inguinal hernia in adults and BJHS.

Key words: Hypermobility – Inguinal hernia – Connective tissue – Beighton score – Brighton score

Benign joint hypermobility syndrome (BJHS) is defined as a clinical condition consisting of musculoskeletal symptoms such as arthralgia, pain, recurrent soft tissue disorders, joint dislocation, as well as increased normal range of motion.[1] The term, hypermobility syndrome, was used first by Kirk *et al*.[2] BJHS is characterized by musculoskeletal symptoms in individuals with generalized joint laxity, in the absence of systemic rheumatologic disease.[3–5] General joint hypermobility (GJH) is usually seen in hereditary connective tissue disorders that affect connective tissue matrix proteins, including osteogenesis imperfect (OI), Marfan syndrome (MFS), and Ehlers-Danlos syndrome (EDS). Studies conducted over the last two decades suggest that a mild form of inherited connective tissue disorder may be involved in the etiology of BJHS, a condition showing clinical features similar to those observed with EDS, MFS, and OI.[1,3] There are no laboratory or radiologic diagnostic criteria, or specific joint involvement associated with BJHS.[6,7]

Especially in patients with BJHS, groin hernias are more common in childhood.[8] Inguinal hernia is one of the most common diseases that surgeons encounter in clinics. The treatment may include an elective operation and an emergency surgery due to its complications.[8] Many complications, including hydronephrosis to small bowel obstruction, can occur.[8,9] Based on this we hypothesized that the frequency of BHJS may increase in patients who underwent surgery for groin hernias due to the hypermobility in connective tissue.

The aim of this prospective study is to investigate the frequency and associated clinical findings of BJHS in adult patients who were diagnosed with inguinal hernia. This is the first study evaluating BJHS in adults having inguinal hernia repair.

Materials and Methods

This study was approved by the local ethics committee for clinical investigations. It was performed according to the tenets of the Declaration of Helsinki. The purpose of the study was explained and informed consent was obtained from all subjects.

We evaluated 66 patients in terms of BHJS. The patients were divided into 2 groups. The first group consisted of 33 patients who underwent inguinal hernia repair surgery. The second group included 33 healthy volunteers who did not have an inguinal hernia operation. Physical examinations were performed by 2 physicians on all participants. Afterward, a patient follow-up questionnaire was completed for both groups. They were asked about previous instances of recurrent joint pain, joint swelling, back pain, dislocation, joint sprain, time of gait, and entire medical history. General joint hypermobility (GJH) was assessed using Beighton criteria (Table 1), and BJHS was scored by Brighton criteria (Table 2). In addition, we also evaluated the patients in terms of postoperative complications, such as length of hospital stay, wound dehiscence, and recurrent hernia operation history. Exclusion criteria for analysis were collagen tissue diseases, history of malignancy, neurologic disorders affecting the musculoskeletal system, rheumatologic diseases, or clinical abnormalities. Subjects with systemic diseases such as diabetes mellitus, renal failure, and hepatic failure were excluded.

All data were recorded as the mean ± standard deviation (SD). Before statistical application, the Kolmogrov–Smirnov normal distribution test was applied for the determination of data analysis method. Shapiro–Wilk test was used to check for the normal distribution of values such as age, sex, and Beighton score. Chi-square analysis and the Mann–Whitney U test were used for group comparison. Pearson correlation coefficient was used to evaluate the correlation between Beighton score and age. Statistical analysis of the data was performed using SPSS software for Windows, version 20.0 (SPSS Inc, Chicago, Illinois). $P < 0.05$ was considered statistically significant for all comparisons.

One point may be gained on each side for maneuvers 1–4, so that the total hypermobility score will have a maximum of 9 points if all of them are positive.

Table 1 Beighton hypermobility score[5,7]

Ability	Right	Left
1. Hyperextend the knee to ≥10°.	1	1
2. Hyperextend the elbow to ≥10°.	1	1
3. Oppose the thumb to the volar aspect of the ipsilateral forearm.	1	1
4. Passively dorsiflex the fifth metacarpophalangeal joint to ≥90°.	1	1
5. Place hands flat on the floor without bending the knees.		1

One point may be gained on each side for maneuvers 1–4, so that the total hypermobility score will have a maximum of nine points if all of them are positive.

BJHS is diagnosed in the presence of 2 major criteria or 1 major and 2 minor criteria, or 4 minor criteria. Two minor criteria are sufficient when there is an unequivocally affected first-degree relative.

Results

In this study, the control and patient groups consisted of 2 female and 29 male adults. The mean age and sex of the 2 groups did not determine significantly. Clinical and demographic characteristic of the subjects are summarized in Table 3.

BJHS was diagnosed in 22 (66%) patients with inguinal hernia and in 4 (12%) healthy volunteers, respectively. Prevalence of BJHS and Beighton score in patients with inguinal hernia were significantly higher ($P < 0.05$). GJH was found in 20 (60%) of the inguinal hernia patients (Beighton score $\geq$ 4; $P < 0.05$). A negative relationship between BJHS, Beighton score, and age was found ($P > 0.05$).

Skin hyperextensibility and marfanoid habitus are common in patients having groin hernia surgery. We investigate the surgical procedure and follow-up period in the patients who underwent surgery for inguinal hernia. All patients had undergone Lichtenstein hernia repair procedure. They were followed for 3 months. Mean length of hospital stay was 2.1 days. There were not any recurrences. Three patients had seroma, and 2 of the patients had wound infection. Only 1 patient had wound dehiscence and hematoma.

Discussion

To our knowledge, the frequency of JHS has been reported to be between 10% and 43%.[10–14] This rate varies, depending on conditions such as age, gender, race, and geographic region. Prevalence of JHS is seen more in women than in men.[15,16] In this study we evaluated the patients who underwent groin hernia surgery. Groin hernias are more common in males. Thus, our prevalence of male gender was higher in this study.

In patients with hypermobility syndrome, intrinsic weakness or poor tensile strength in the supporting structures may lead to abdominal and pelvic visceral disorders such as inguinal hernia, urinary incontinence, and pelvic floor prolapsus.[1] Studies in the literature have found a direct relationship between connective tissue disease and inguinal hernias. In a study by Pans *et al*, incidence of inguinal hernia was found to be 25% and 16% in patients with BJHS and in the control group, respectively.[17]

In a study by Nazem *et al*, authors found that the frequency of BJHS was 92% among children with inguinal hernia. In our study, the frequency of BJHS was 66%. We know that incidence of BJHS is decreasing in the elderly population.[15] In children with hypermobility the ratio of type III collagen to type III plus type I collagen is increased. Type III collagen is thin and elastic and is found in relatively greater amounts in extensible connective tissue. This

Table 2 Brighton revised diagnostic criteria for benign joint hypermobility syndrome (BJHS)[5]

Major criteria
1. A Beighton score of 4/9 or greater (either currently or historically)
2. Arthralgia for longer than 3 months in 4 or more joints

Minor criteria
1. A Beighton score of 1, 2, or 3/9 (0, 1, 2, or 3 if aged ≥50)
2. Arthralgia in 1 to three joints or back pain or spondylosis, spondylolysis/spondylolisthesis
3. Skin striae, hyperextensibility, thin skin, or abnormal scarring.
4. Varicose veins or hernia or uterine/rectal prolapse
5. Dislocation in >1 joint, or in 1 joint on more than one occasion
6. Eye signs: drooping eyelids or myopia or antimongoloid slant
7. Three or more soft tissue lesions (e.g., epicondylitis, tenosynovitis, bursitis)
8. Marfanoid habitus (tall, slim, arm span, height; upper segment:lower segment ratio <0.89; arachnodactyly)

Table 3 Demographic and clinical data of the patients and control group

	Hernia group (n = 33)	Control group (n = 33)	*P* value*
Age (year, mean ± SD)	40.3 ± 15.43	40.1 ± 15.08	0.954
Sex (male/female)	31/2	31/2	0.128
Beighton score (mean ± SD)	4.15 ± 3.08	0.87 ± 0.17	0
BJHS positivity	22	4	0
Arthralgia	16	12	0.271
Marfanoid habitus	20	6	0.006
Varicose veins	3	5	0.016
Skin hyperextensibility	20	6	0.003
Recurrent soft tissue lesions	2	1	0.693
Backpain	3	6	0.054
Recurrent dislocation	8	0	0.488
Myopia	8	7	0.500
Drooping eyelids	2	1	0.002

BJHS, benign joint of hypermobility syndrome.

*P value < 0.05 is significant.

is thought to be caused from the depletion of type III collagen in older people, or conversely, the escalated tissue flexibility of childhood.[3,18– 20] In our study we found a higher rate of BJHS in adults. However, very high levels of BJHS were found in a recent study of children.[15] We think that this difference may be due to age. In contrast, another study by Seckin *et al*[18] did not find a significant difference between BJHS and normal students in high school in terms of inguinal hernia. A study reported by Wynne-Davies denoted that abdominal herniation was more common among patients with JHS. In another study by Robinson *et al* prevalence of inguinal hernia was found to be significantly high among patients with Shprintzen-Goldberg syndrome.[21,22] Our results were similar to the literature. To our knowledge, this study is the pioneer study evaluating adult inguinal hernia patients in terms of BJHS.

Our study revealed that BJHS was found more commonly in patients with inguinal hernia compared with the healthy control group similar to the result of the study by Nazem *et al*.[8] However, our study was conducted in adults.

Conclusion

BJHS should be taken into consideration when evaluating patients with inguinal hernia. More studies indicating abnormal collagen metabolism and abdominal hernias should be evaluated in new research. We believe that further studies are needed in big cohorts to verify the relationship between inguinal hernia in adults and BJHS.

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